

**Assessing pre-linguistic communication in  
young people with profound intellectual and  
multiple disabilities (PIMD)**

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## **Declaration**

I, Lucy Pepper, 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.

## Abstract

Children with Profound Intellectual and Multiple Disabilities (PIMD) experience profound impairments in communication, remaining pre-linguistic communicators across their lifespan. Whilst the majority will receive speech and language therapy (SLT) input, shortcomings in current approaches to SLT assessment for this group have been identified by existing research and were further explored through a survey study carried out for this thesis. These included a lack of agreement around which prelinguistic skills might be significant and a reliance on indirect methods of information gathering. In response to these issues, this thesis explored the use of structured sampling techniques to measure behaviours relating to joint attention (JA) in children with PIMD and addressed the following questions.

- 1) What patterns of JA behaviour are demonstrated by young people with PIMD?
- 2) Are these patterns of behaviour affected by the ability to fix and shift gaze?
- 3) Are structured probes an effective means of eliciting information about JA behaviours?

A continuum of behaviours underpinning JA was derived from the developmental literature. Structured probes were devised to elicit these behaviours and were administered to seventeen participants with PIMD aged 4-16 on three separate occasions. Since target behaviours involved gaze fixation and gaze shifting between objects and people, a novel measure of these functional vision skills was also administered. Performance on the probes was compared to performance in less structured settings.

Results indicated that young people with PIMD can be differentiated by the profiles of JA behaviours they demonstrate. These profiles were significantly correlated with their functional vision abilities but were not associated with background measures of cognitive and motor skills or with scores on the Communication Matrix (Rowland, 2004). Structured probes were an effective means of assessing JA behaviours, providing a controlled environment and multiple opportunities for participants to demonstrate capacity which was not always revealed by alternative means of information gathering. Findings of this thesis suggest that current approaches to communication assessment for people with PIMD might be enhanced by the increased use of structured sampling and a shared focus on behaviours relating to joint attention.

## Impact statement

People with PIMD represent a small minority of the population but one which is growing, with medical advances leading to improved infant survival and increasing life spans for this group. Despite their increasing numbers, relatively little research is carried out with people who have PIMD, limiting the ability of professionals such as Speech and Language Therapists to engage in evidence-based practice when providing assessment and support. The work reported within this thesis contributes to the knowledge base around communication in PIMD and how this might be assessed, demonstrating how novel measures may be used to identify differential profiles of joint attention (JA) behaviour in this group and providing evidence of the impact which functional vision skills have on JA.

The methods and findings reported in this thesis contribute to academic research in the field by building on the small number of studies which have investigated JA behaviour in people with PIMD to date, consolidating some existing findings and contributing new ones, including evidence of a relationship between functional vision and JA in this population. The novel measures devised for assessing functional vision and eliciting JA behaviours in this study constitute a new methodological approach which may be replicated and used in further research with this population. Preliminary findings from the study have been presented at the early career researcher meeting of the PIMD special interest group of IASSID (the International Association for the Scientific Study of Intellectual Disabilities) (Fribourg, 2017) and through a poster presentation at the Seattle Club conference on intellectual disabilities (Durham University, 2017). It is anticipated that findings will be further disseminated through papers submitted to peer-reviewed journals and presentations at relevant conferences.

This thesis will also have benefits for the clinical practice of speech and language therapy with people who have PIMD. It provides an evidence-based argument for the need to consider JA behaviours when assessing communication in this group, showing how identifying profiles of performance in this area can provide a baseline of ability and highlight suitable approaches to intervention. It also provides evidence supporting the use of structured sampling in assessment, demonstrating how it can provide information about an individual's capacity which is more challenging to obtain through methods such as observation in everyday contexts and caregiver interview.

The evidence presented in this thesis also indicates that structured sampling could improve consistency in assessment, providing a standardised approach which can be replicated by different SLTs over time thus providing a reliable means of measuring progress. The structured probes devised for this study are currently being used in pilot form to assess prelinguistic children with complex needs at a leading children's hospital and further dissemination of the results of the thesis findings is planned through presentation at professional research hubs and conferences for speech and language therapists as well as through publication in *Bulletin*, the professional magazine for Speech and Language Therapists.

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## 1. Introduction

Young people with PIMD may all be pre-linguistic communicators, but their communication profiles are not all the same. Robust assessment of these communication profiles is essential for the designing of appropriate support and intervention as well as for effective monitoring of progress. This thesis explores communication assessment for this population, identifying current issues and carrying out research which contributes to understanding of patterns of communication which might be observed as well as making suggestions for improving clinical practice in assessment.

Speech and Language Therapists play an important role in assessing communication in young people with PIMD. However, research has suggested that there are shortcomings in the approaches they currently use to achieve this (Chadwick, Buell, & Goldbart, 2019). As part of this thesis, these shortcomings are discussed and further explored through a small survey study of SLTs. Combined findings of existing research and this survey study indicate a lack of consistency between practitioners, with current approaches relying heavily on indirect means of information gathering and failing to reflect recent thinking in the field of pre-linguistic development.

Over recent decades evidence and theory in the field of pre-linguistic development has confirmed the significance of a progression from pre-intentional to intentional communication as first described by Elizabeth Bates (e.g. Bates, Camaioni & Volterra, 1975). However, there has also been a growing focus on one aspect of this progression – the development of joint attention (JA). The significance of JA in the typical development of early communication has been established and its definitions discussed at length (e.g. Moore, Dunham, & Bruner, 1995; Seemann, 2011). The fact that JA and the behaviours which underpin it develop during the earliest months of life suggest that it should provide a focus for attention in the assessment of young people with PIMD who might be expected to demonstrate JA behaviours of varying complexity. However, to date, only a limited number of studies have specifically explored the demonstration of JA behaviours in people with PIMD (e.g. Neerinx & Maes, 2016).

The work reported in this thesis aims to extend knowledge in this area, investigating not only the occurrence of the state of JA in a group of young people with PIMD, but also their use of behaviours which might be considered to underpin it. These include attending separately to both people and objects, and, ultimately, integrating this attention by shifting gaze between people and objects whilst using communicative signals. Such behaviours rely heavily on gaze behaviour and vision is known to be subject to impairment in this population (Evenhuis, Theunissen, Denkers, Verschuure, & Kemme, 2001). Therefore, the impact of functional vision impairment on JA behaviours is also investigated.

As mentioned above, SLTs currently rely heavily on indirect forms of information gathering for assessment such as observation in naturalistic settings and interviews with caregivers. Whilst there are benefits to such approaches, they also have their limitations since performance in such settings may not provide opportunities for an individual to demonstrate their full capacity for target behaviours (Cress, Arens, & Zajicek, 2007) and caregiver judgements are subjective (Bradshaw, 2008). Increased use of direct means of information gathering alongside these indirect approaches might be expected to improve the robustness of assessment (Brady & Halle, 1997). Structured sampling is one such means of direct information gathering and, in order to develop knowledge around its use with young people with PIMD, it is investigated as a means of eliciting and measuring JA behaviour for this thesis.

The resulting research was carried out to answer the following research questions.

### **1.1. Principal research questions**

- 1) What patterns of JA behaviour are demonstrated by young people with PIMD?
- 2) Are these patterns of behaviour affected by the functional ability to fix and shift gaze?
- 3) Are structured probes an effective means of eliciting information about JA behaviours?

## 1.2. Structure of the thesis

This thesis consists of eight chapters, beginning with a description of the clinical issues which provided its motivation. A background to the research is then provided. This includes a justification for the need to consider JA in this population and the relevance of functional vision to this area as well as an argument for the use of structured sampling as a means of assessment. The research, in which novel measures were developed to explore JA and functional vision in a sample of young people with PIMD, is then described. Finally, the implications of findings are discussed in relation to their theoretical and clinical implications.

**Chapter Two** defines the population under investigation, outlining terms and the characteristics of people described as having PIMD.

**Chapter Three** states the clinical motivation for this work, including the clinical reflections of the author, existing research into communication assessment with PIMD and a survey study of SLTs which was carried out for this thesis. Findings are then used to evaluate current practice and identify shortcomings which need to be addressed.

**Chapter Four** describes how current literature relating to pre-linguistic communication was appraised to identify significant areas which should be considered in communication assessment. JA is identified as a key factor and is discussed with reference to its definitions, its significance in communication development and factors which might impact upon it including functional vision abilities. Studies investigating JA in atypical populations are reviewed, along with a critical evaluation of existing studies focusing on JA and PIMD. This chapter concludes by describing how the background literature and existing research were used to inform the proposed research to be carried out for this thesis.

**Chapter Five** discusses methodological issues relevant to the proposed research, beginning with a discussion of structured sampling methods, their benefits and relevant areas where they have been used to measure pre-linguistic communication. This is followed by a discussion of general methodological challenges which arise when researching a population with PIMD and the ways in which these were addressed in the design of this research.

**Chapter Six** describes an exploratory phase of work which was carried out prior to the main phase of study, describing how measures of JA and functional vision were developed or selected and tested for feasibility and effectiveness.

**Chapter Seven** describes the phase of study in which participants' functional vision skills, specifically gaze fixation and gaze shifting, were measured. The devising of a measure of functional vision is described, results reported and discussed.

**Chapter Eight** describes the phase of study in which participants' JA behaviours were measured including a description of how the measure of JA was devised, drawing on the exploratory work outlined in Chapter Six. Results are reported and discussed with reference to the research questions.

**Chapter Nine** draws overall conclusions about the findings of this thesis, discussing how findings relate to existing understanding of JA in atypical populations, discussing the benefits of using structured sampling and, finally, considering the potential clinical implications of findings.

## **2. Defining the Population**

People with PIMD represent a unique group within our population. They develop and function in a way which is so different from the typically developing majority that their characteristics and needs are often poorly understood. In this section I outline what is meant by the term PIMD, describing the features, causes and incidence of this condition and briefly present some of the issues people with PIMD face within society.

### **2.1. Terms**

The terms 'Profound Intellectual and Multiple Disabilities' (PIMD) and 'Profound and Multiple Learning Disabilities' (PMLD) are both used to describe this group, with 'PMLD' being used most commonly in the UK (Mansell, 2010). However, the term 'PIMD' is used more widely in an international and academic context and will be the term adopted throughout this thesis.

### **2.2. The characteristics of people with PIMD**

The term 'PIMD' may be viewed as a description rather than a diagnosis (Goldbart, 2016) and, whilst several approaches have been taken to defining its features (see Bellamy, Croot, Bush, Berry, & Smith, 2010 for a review), there is broad agreement that people with PIMD will share the following two characteristics (Nakken & Vlasskamp, 2007):

#### **1. Profound intellectual impairment**

Profound intellectual impairment is a defining feature of PIMD. Attempts to specify the extent of this impairment have included references to standardised intelligence testing, with the International Classification of Diseases-10 (ICD-10) suggesting that people with profound intellectual impairment have an IQ of less than 20 (World Health Organisation, 1992) and the more recent ICD-11 suggesting that they perform at less than the .003 percentile on standardised testing (World Health Organisation, 2019). In practice it is not feasible to measure such impaired profiles of cognitive performance with any accuracy using standardised measures of IQ (Tassé, Luckasson, & Nygren, 2013) and other authors have taken a developmental approach with Hogg (2004) estimating that people with PIMD function at a developmental level

of between 18 and 24 months and Ware (2003) suggesting that the majority function at a developmental level of 12 months or lower.

## 2. Multiple Disabilities

In addition to profound intellectual impairment, people with PIMD will have at least one, and often more than one, additional disability. Severe motor disabilities, often associated with a diagnosis of cerebral palsy, are frequent (Nakken & Vlaskamp, 2007) with the majority of people with PIMD being unable to walk (Mansell, 2010). Visual and hearing impairments are also common with Evenhuis, Theunissen, Denkers, Verschuure, & Kemme, 2001 finding that 50% of the 243 children and adults with PIMD they studied had a visual impairment and that 20% had impairments of both vision and hearing. In terms of vision, impairments may affect either physical aspects of the eye (such as strabismus where the eyes are misaligned) and/or the way that the brain processes visual information, known as cerebral visual impairment or CVI (Salt & Sargent, 2014).

Alongside intellectual impairment and multiple disabilities, people with PIMD very frequently experience health issues and have high support needs. In a study of forty 14-19 year olds with PIMD living in Sheffield, Parrott, Tilley, & Wolstenholme (2008) found that 65.7% had epilepsy, 60% had a chest condition causing respiratory distress, 45% required non-oral approaches to feeding (e.g. via gastrostomy) and none were continent. Perhaps partly as a result of additional health needs, people with PIMD experience atypical patterns of arousal and spend up to 42% of the time in states which are not conducive to engagement, such as drowsiness or agitation (Guess et al., 1990). They may also demonstrate adaptive behaviours which are experienced as challenging by those supporting them (Doukas, Fergusson, Fullerton, & Grace, 2017).



### **2.3. The impact of PIMD on communication**

People with PIMD are non-verbal, pre-linguistic communicators, who use pre-symbolic and informal means of communication such as vocalisations, facial expressions and body movements to express their immediate preferences and emotions. The extent to which such signals are used deliberately as a means of communication varies between individuals, with some showing little evidence of intention (Mansell, 2010). Even where there is intentional communication, signals can be idiosyncratic, meaning that communication needs to be interpreted by those who are familiar with the individual. A critical feature of PIMD is severe impairment in the ability to understand language, although individuals may recognise non-verbal cues in the environment (Bellamy, Croot, Bush, Berry, & Smith, 2010). Since individuals are pre-symbolic, their ability to use alternative means of communication based on systems such as signing, pictures or symbols tends to be limited and is sometimes further affected by physical disabilities which, for example, make it difficult for them to form signs or indicate choices through touching or pointing.

### **2.4. Causes and incidence of PIMD**

Causes of PIMD are varied and can arise from issues arising ante, peri and post-natally as well as from genetic conditions or brain damage. In many cases no cause is identified (Doukas et al., 2017). Advances in medical care resulting in better survival rates for infants with medical conditions causing severe disabilities, as well as in longer life-spans for those known to have PIMD mean that their numbers are growing. Whilst it is difficult to be precise about the incidence of PIMD due to inconsistencies in defining this condition and in collecting data, it has been estimated that there were 14,744 children (aged 0-17) and 16,036 adults with PIMD in the UK in 2008 and that this number was projected to grow by 1.8% each year (Emerson, 2009).

## 2.5. Social issues encountered by people with PIMD

The severity and complexity of impairments experienced by people with PIMD along with their relatively low incidence in the population means that they experience prejudice in many areas of society. The very value of their existence has been questioned, with (McMahan, 2002) comparing them to non-human animals (see Vorhaus, 2016 for a discussion) and legislation such as Valuing People Now (Department of Health, 2009) which aims to improve inclusion and provision of services for people with learning disabilities, failing to have a significant impact for this group (Doukas et al., 2017; Mansell, 2010) who continue to be described as:

“amongst the most isolated and marginalised [groups] in modern society”,

(Doukas et al., 2017, p13)

The marginalisation of people with PIMD has extended to the field of research and they have been described as ‘virtually missing’ from key theoretical and methodological discussions as well as from empirical studies (Mietola, Miettinen, & Vehmas, 2017, p264). It is hoped that the work contained within this thesis will go some way to redressing this balance by enhancing our understanding of the ways in which people with PIMD learn to communicate and improving clinical approaches to supporting this.

### **3. Clinical Assessment of Prelinguistic Communication in People with PIMD: Stating the Problem**

#### **3.1. Clinical reflection**

Shortcomings in communication assessment for people with PIMD provide the motivation for this thesis. These shortcomings initially came to the author's attention in the course of her work as a speech and language therapist (SLT) with clients who had PIMD. Approaches to assessment in this field did not appear to be as well developed as those applied to other client groups. There seemed to be a lack of consistency, with each clinician taking a different approach, and little guidance being provided by published resources or shared assessment frameworks. This situation was particularly problematic given that foundation training for SLTs focused on clients who had at least some degree of speech or language ability, with little attention given to the pre-linguistic communication used by people with PIMD. Since no formal post-graduate training was available in the field, practitioners took their own approaches to acquiring knowledge, learning from their own experience and that of colleagues, a situation which was, doubtless, contributing to the perceived diversity in assessment approaches.

#### **3.2 The preliminary evidence**

Research support for these clinical reflections was initially offered by a conference paper presenting preliminary findings of a study into assessment and intervention practices of SLTs working in the field of PIMD (Goldbart, 2010). Based on a survey study of 55 SLTs working with adults and children with PIMD, results confirmed that diverse and idiosyncratic approaches were, indeed, taken to communication assessment with 80% of participants using personally devised, informal protocols. However, 80% of participants also reported using published assessments in addition to personally devised protocols, the three most commonly cited being the Pre-Verbal Communication Schedule (PVCS) (Kiernan & Reid, 1987) which was used by 46% of participants, the Affective Communication Assessment (ACA) (Coupe, Barton, Collins, Levy, & Murphy, 1985), used by 38% of participants and the Triple-C

(Bloomberg, West, Johnson, & Iacono, 2009), used by 16% of participants. It was notable that the two most frequently used of these assessments had been developed over twenty years previously without being updated and were no longer easily available, with the PVCS being out of print and the complete version of the ACA only being available from a school on request. A poor understanding of the characteristics of PIMD was also evident in a small number of participants who reported using assessments inappropriate for this client group such as the Test for the Reception of Grammar (Bishop, 2003) a standardised assessment designed to test comprehension of grammar at a developmental level of four years and above. It seemed that clinicians took their own approaches to assessment and, where published assessments were used, these were becoming dated and so were unlikely to be based on the most current evidence base relating to prelinguistic communication. Overall these findings added weight to the author's observations that approaches to communication assessment in PIMD were inconsistent and, potentially, lacked a sound basis in up-to-date knowledge relating to prelinguistic communication.

Whilst the preliminary findings reported by Goldbart's conference paper confirmed potential shortcomings in communication assessment approaches for people with PIMD they did not provide detailed information about the personally devised approaches to assessment which SLTs reported taking. The specific areas of communication which SLTs sought to assess and the means they used to achieve this remained unclear. Further investigation was, therefore, undertaken for the current study to explore these issues, and to lay the groundwork for subsequent research reported in this thesis.

### **3.3 An investigation of the key areas targeted by SLTs in communication assessment for people with PIMD and the assessment methods used**

A survey study was used to investigate SLT approaches to communication assessment for people with PIMD by addressing the following research questions:

#### **3.3.1 Research questions for the SLT survey**

- 1) What key areas do SLTs seek to assess in their clients with PIMD?
- 2) What methods do SLTs use to obtain information in relation to these key areas?

#### **3.3.2. Method**

##### **3.3.2.1. Questionnaire Development**

The survey questionnaire was introduced by a definition of PIMD taken from Mansell (2010). This ensured that participants had a clear understanding of the client group under discussion (see Appendix A for a copy of the questionnaire). The term 'Profound and Multiple Learning Disabilities' was used as this is most commonly used within clinical practice in the UK (see section 2.1.).

Participants were then asked to select from options detailing the age of the client group they worked with and the number of years they had been working as an SLT. This provided a simple 'warm-up' phase for the questionnaire (Polgar & Thomas, 2013) as well as gathering demographic data.

Three further questions were then posed to address the aims of the survey study. Given the lack of existing evidence in this area, open-ended questions were used to encourage detailed answers from participants without constraining the range of potential responses (Polgar and Thomas, 2013). Participants were then given the option of making additional comments if they wished (see Table 3.1).

Table 3.1. SLT survey questions

*Table 3.1. SLT survey questions*

|            |  |
|------------|--|
| <b>Q1.</b> | Please detail which key areas of communication and behaviour you consider when assessing clients with Profound and Multiple Learning Disabilities? |
| <b>Q2</b>  | What methods do you use to assess the communication skills of clients who have Profound and Multiple Learning Disabilities?                        |
| <b>Q3</b>  | Are there any other comments you would like to make about assessing clients with Profound and Multiple Learning Disabilities?                      |

The questionnaire was constructed using an online format provided by Survey Monkey ([www.surveymonkey.co.uk](http://www.surveymonkey.co.uk)) so that it could be distributed by email. Use of the design tools provided by this online service also ensured that the questionnaire was easy for participants to complete as well as producing data in an easily accessible format for analysis (Monroe & Adams, 2012).

The questionnaire was piloted with two SLTs working in the author's local area. They reported that it was easy to use and provided suitable responses for analysis. Therefore, no changes were made before circulating it to the full sample of participants.

### **3.3.2.2. Ethical approval**

Ethical approval for the study was granted by the University College London (UCL) Ethics Committee, reference 7565/001.

### **3.3.2.3. Recruitment**

It was not possible to sample a group representative of SLTs working in the field of PIMD since there is no information about their numbers and demographic characteristics (Chadwick et al., 2019). Therefore, a purposive sampling method was used in an attempt to access all the SLTs working with PIMD within nine counties centred around the author's base in London. This allowed her to use her knowledge of the area to help in identifying target contacts.

Targeted counties were as follows:

- Oxfordshire
- Buckinghamshire
- Hampshire
- Wiltshire
- Gloucestershire
- Northamptonshire
- Bedfordshire
- London
- Surrey

Since SLT services may be provided in both health and education settings, a web-based search was used to identify the NHS service providers for each county and any maintained or independent special schools catering for children with severe or profound learning disabilities. The head SLT or lead SLT for learning disability services within each of these settings was then contacted. Repeated and personalised contact is known to improve survey response rates (Dillman, Smyth, Christian, & Dillman, 2009). Consequently, the lead SLTs were approached by telephone, the purpose and nature of the survey study outlined, and an electronic copy of the survey emailed to them if they consented to take part. Where phone contact was not established, an email outlining the study was sent. Taking a snowball sampling approach (Robson & McCartan, 2015), all SLTs who were spoken to on the telephone agreed to be sent a survey and to distribute it to any additional SLTs working with people with PIMD within their service. If responses had not been received within three weeks a further reminder email was sent.

#### 3.3.2.4. Participants

Twenty-five SLTs completed the survey. Of these, 12 had worked as an SLT for over ten years, with only 2 having fewer than two years' experience (see Table 3.2). The majority (17) were working with children, 6 worked with adults and 2 worked with both adults and children (see Table 3.3).

*Table 3.2. Number of years participants had worked as an SLT*

| <b>Number of years working as an SLT</b>     | <b>0-2</b> | <b>2-5</b> | <b>5-10</b> | <b>10+</b> |
|--|------------|------------|-------------|------------|
| <b>Number and Percentage of participants</b> | 3 (12%)    | 5 (20%)    | 5 (20%)     | 12(48%)    |

*Table 3.3. Age of client groups with whom participants were working*

| <b>Age of Clients</b>                        | <b>Children<br/>(0-19 years)</b> | <b>Adults<br/>(19 years +)</b> | <b>Children and<br/>Adults</b> |
|--|----------------------------------|--------------------------------|--------------------------------|
| <b>Number and Percentage of participants</b> | 17 (68%)                         | 6 (24%)                        | 2 (8%)                         |



### **3.3.3. Results**

#### **Key areas of communication and behaviour considered in the assessment of clients with PMLD**

Content analysis was used to identify themes in participant responses to question 1. Using this system, a category or code is applied to describe the meaning of each response. Subsequent responses are either assigned to this category or assigned a new one if their meaning is different. This is undertaken until no new categories are identified (Kumar, 2014). All participants provided responses containing several pieces of information and each of these pieces of information was assigned to a category. As a result, it was possible for each participant's response to be coded with more than one category.

Categories of participant response fell into four main areas; (i) communication related skills and behaviours, (ii) cognitive or developmental skills, (iii) physical and sensory skills and (iv) additional areas, comprising response to previous intervention, establishing preferred stimuli or activities and caregiver perspectives (see Table 3.4).

Table 3.4. Categories of key assessment areas cited by participants

\* Items in italics are examples of responses given in each category.

| Communication Related Skills and Behaviours   |   |
|---|---|
|   | Percentage (and number) of participants citing area |
| <b>Communicative Means</b><br><i>gesture, facial expression, vocalisations, body movements, speech *</i>  | 60%<br>(15)   |
| <b>Comprehension</b><br><i>Response to voice, situational understanding, understanding of objects/photos/symbols</i>  | 44%<br>(11)   |
| <b>Social Behaviours</b><br><i>'Social interaction', engagement with others, reaction to others, turn-taking</i>  | 44%<br>(11)   |
| <b>Communicative functions (expressed intentionally or pre-intentionally)</b><br><i>Requesting/rejecting, making choices, expressing wants, indicating yes/no, showing likes/dislikes</i> | 32%<br>(8)  |
| <b>Degree of intentional communication</b><br><i>Presence of communicative intent, intentional movements, 'level of intentionality' (reflexive, reactive, early intentional etc)</i>      | 28%<br>(7)  |
| <b>Response of others to child</b><br><i>Any communication supports in place, whether others interpret their behaviours consistently</i>  | 12%<br>(3)  |

Table 3.4. Categories of key assessment areas cited by participants (cont.)

| Cognitive or Developmental Skills   |             |
|---|-------------|
| <b>Attention</b><br><i>'Attention and Listening', Orienting, visual and auditory attention, joint referencing, shared attention, how long they can attend for</i> | 40%<br>(10) |
| <b>Cause and effect</b><br><i>Switch operation for cause and effect, anticipation</i>   | 24%<br>(6)  |
| <b>Play</b><br><i>Use of cause and effect toys, sensory play</i>  | 16%<br>(4)  |
| <b>Consistent response to stimuli</b>   | 12%<br>(3)  |
| <b>Object Permanence</b>  | 8%<br>(2)   |
| <b>Cognitive Ability</b>  | 8%<br>(2)   |

Table 3.4. Categories of key assessment areas cited by participants (cont.)

| Physical or Sensory Skills   |             |
|--|-------------|
| <b>Profile of Additional Disabilities</b><br><i>physical skills, sight, hearing, medical diagnoses, epilepsy</i>                                   | 52%<br>(13) |
| <b>Sensory Profile</b><br><i>Sensory processing difficulties, sensory needs</i>  | 16%<br>(4)  |
| <b>Visual behaviours</b><br><i>Transfer of gaze, ability to sustain gaze for eye pointing</i>  | 16%<br>(4)  |
| Additional Areas   |             |
| <b>Response to Potential Intervention Approaches</b><br><i>Historic response to intervention, response to AAC, switches, intensive interaction</i> | 20%<br>(5)  |
| <b>Preferred stimuli/activities</b>  | 20%<br>(5)  |
| <b>Caregiver perspectives</b><br><i>Do parents know what he/she wants, views of family, carers, staff</i>  | 8%<br>(2)   |

There was wide variation in the key assessment areas cited which related directly to communication with no single area being cited by all participants. The most commonly cited area was ‘communicative means’ which was identified as a key area by 60% of

participants. This area involved assessment of the behaviours a client might use to communicate such as vocalisations, body movements and gestures. Assessment of comprehension was cited by 44% of the participants including situational and symbolic understanding as well as the understanding of language. Social aspects of language such as reactions to people were also cited by 44% of SLTs. Thirty two percent of the participants assessed the functions served by client communication, with the specific functions listed being requesting and rejecting, making choices, expressing wants, indicating yes and no and showing likes and dislikes. Smaller numbers of participants also cited assessing the degree of intentional communication used by clients (28%) and the ways in which others responded to the client (12%).

Other assessment areas cited included cognitive and developmental skills which may have an impact on communication, the most frequently cited being 'attention'. Whilst 24% participants referred broadly to assessing 'attention' or 'attention and listening', 16% were more specific and referred to investigating attention span, orienting, shared attention and joint referencing.

Fifty two percent of participants referred to assessing the profile of additional disabilities demonstrated by their clients. These included impairments in physical skills, hearing and sight in addition to medical conditions such as epilepsy. Sixteen percent of participants referred more specifically to assessing visual skills or behaviours, those listed being the ability to transfer gaze and the ability to sustain gaze for eye pointing.

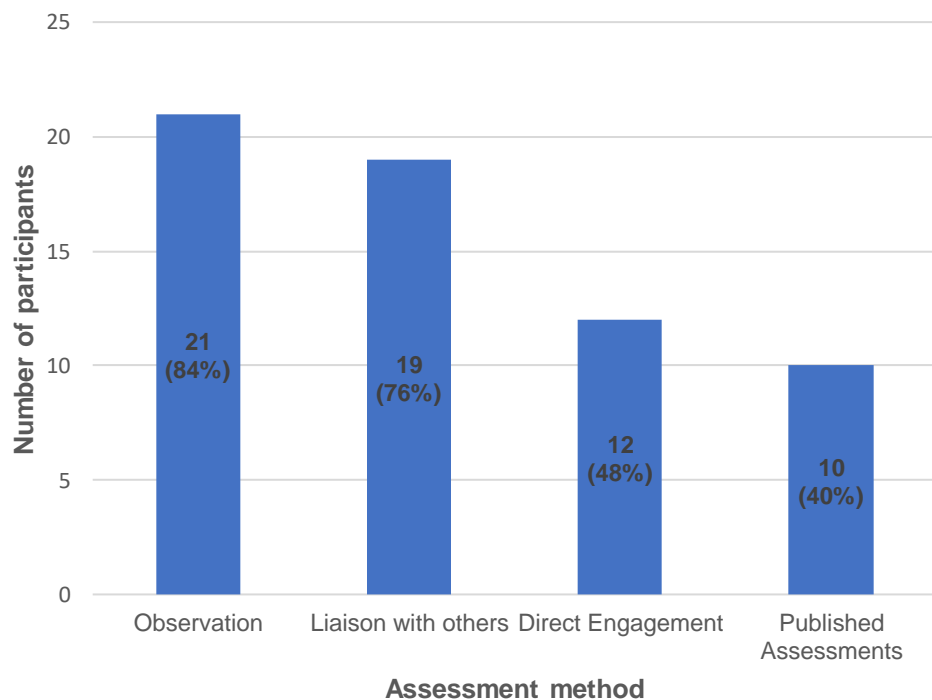
Finally, 20% of participants referred to assessing the client's response to potential intervention approaches (Augmentative and Alternative Communication (AAC), switch use and intensive interaction), 20% assessed the activities which clients preferred and 8% cited assessing the perspectives of caregivers.

### Methods used to assess the communication skills of people with PIMD

The content-analysis approach was applied to Question 2 responses and the following categories of data-gathering methods were identified:

- 1) Observation
- 2) Direct Engagement
- 3) Liaison with others
- 4) Published assessment frameworks

The number and percentage of SLTs who reported using each data gathering method are presented in Figure 3.1. Categories were not mutually exclusive since 92% (23) participants reported using a combination of approaches.



*Figure 3.1. Number and percentage of participants citing each assessment method.*

Observation was the most frequently reported approach used, followed by liaison with others, direct engagement and, finally, use of published assessment frameworks. Further qualitative results relating to each method are reported below.

### **Observation**

Eighty four percent of participants referred to using 'observation' or 'informal observation' and sometimes detailed the context. The following contexts were specified:

- In class
- In everyday situations
- Interactions with others

### **Liaison with Others**

Liaison with others was a method cited by 76% of participants and was generally described as discussion or interview with one or more of the following groups:

- Parents/Family
- Carers
- Teachers
- Specialist teachers
- MDT (multidisciplinary team) professional
- Portage workers

### **Direct Engagement**

Only 48% of participants reported using some form of direct engagement with clients. The type of engagement cited fell mainly into the categories of 'engagement with objects' or 'trailing therapeutic techniques'. Exact descriptions of these approaches can be seen in Table 3.5.

Table 3.5. Types of direct engagement reported by participants

| Type of engagement                      | Percentage and number of participants | Participant description of engagement   |
|---|---------------------------------------|---|
| <b>Engagement with objects</b>          | 20% (5)                               | <p><i>“Sensory items – different items to watch and feel.”</i></p> <p><i>“Real objects for choice making.”</i></p> <p><i>“Responding to multisensory objects and stimuli.”</i></p> <p><i>“Use of people’s own home items, children’s own toys.”</i></p> <p><i>“Enticement with any known motivators or exploring possible motivators.”</i></p> <p><i>“Engaging with the person using sensory/musical toys, may take a big mac switch and a favoured noise maker.”</i></p> |
| <b>Trialling therapeutic techniques</b> | 24% (6)                               | <p><i>“Trial of intensive interaction.”</i></p> <p><i>“Intensive Interaction”</i></p> <p><i>“Intensive interaction as an exploratory activity.”</i></p> <p><i>“Test teach approach with a variety of therapy techniques and strategies.”</i></p> <p><i>“Carrying out therapeutic input/assessment within a functional context to establish the student’s level of ability.”</i></p> <p><i>“Trialling strategies ‘as we go’.”</i></p>                                      |
| <b>Play</b>                             | 4% (1)                                | <i>“Play – sensory, cause and effect.”</i>  |
| <b>Other</b>                            | 4% (1)                                | <i>“Setting tasks in natural settings.”</i>   |



Whilst two participants specified the aim behind their use of objects for assessment – with one investigating choice-making and the other exploring possible motivators – others just reported general engagement with the items. Use of intensive interaction (e.g. Nind & Hewett, 2001) for assessment was reported by three participants with three others referring more broadly to therapeutic strategies or techniques. Finally, one participant referred to the use of play during assessment and one reported 'setting tasks in natural settings' although these were not detailed.

### **Published Assessments**

Ten participants (40%) reported using published frameworks with six of these reporting the use of more than one framework. Each of the frameworks identified was investigated further to ascertain the following information; the type of assessment it comprised, how information is gathered to complete it, evidence for its reliability and validity described in its manual or relevant papers and its cited evidence base (see Table 3.6).

Table 3.6. Published assessment frameworks used by participants

| Assessment   | Number of participants citing assessment | Assessment type                | How information is gathered for assessment  | Evidence for psychometric stability e.g. reliability/validity                                  | Stated evidence base   |
|--|--|--------------------------------|---|--|--|
| <b>Routes for Learning</b><br>(Welsh Assembly Government, 2006)            | 4  | Criterion referenced checklist | Observation<br><br>Liaison with family/teachers/carers.<br><br>Some optional activities for eliciting target behaviours are described | None available   | Communication stages described in the Communication Matrix and (Coupe-O’Kane & Goldbart, 1998)                                   |
| <b>Pre-Verbal Communication Schedule (PVCS)</b><br>(Kiernan and Reid,1987) | 3  | Criterion referenced checklist | Observation<br><br>Liaison with family/teachers/carers<br><br>Specified direct tasks for assessing imitation only                     | Inter-rater reliability and some aspects of construct validity<br><br>(Kiernan and Reid, 1987) | Not specified<br><br>Checklist items are “of significance in communication” (Kiernan and Reid, 1987, p3) but source not detailed |

Table 3.6. Published assessment frameworks used by participants (cont.)

| Assessment  | Number of participants citing assessment | Assessment type                | How information is gathered for assessment             | Evidence for psychometric stability e.g. reliability/validity  | Stated evidence base  |
|---|--|--------------------------------|--|--|---|
| <b>P-Scales</b><br>(Department for Education, 2014) | 2  | Criterion referenced checklist | Observation  | Some evidence for inter-rater reliability<br><br>(Ndaji & Tymms, 2009)   | Not specified   |
| <b>Communication Matrix</b><br>(Rowland, 2004)      | 1  | Criterion referenced checklist | Observation<br><br>Liaison with family/teachers/carers | Evidence for:<br><br>Construct validity<br><br>Inter-rater reliability between parents and professionals<br><br>Test-retest reliability<br><br>(Rowland, 2012) | Light's reasons for communicating (Light, 1988)<br><br>Pragmatic theory (Bates, 1979)<br><br>Theory of symbolic development (Werner & Kaplan, 1963) |

Table 3.6. Published assessment frameworks used by participants (cont.)

| Assessment   | Number of participants citing assessment | Assessment type                | How information is gathered for assessment                    | Evidence for psychometric stability e.g. reliability/validity   | Stated evidence base                                    |
|--|--|--------------------------------|---|---|---|
| <b>Triple C: Checklist of Communicative Competence</b><br>(Bloomberg et al., 2009)             | 1  | Criterion referenced checklist | Observation<br><br>Liaison with family/teachers/carers        | Evidence for:<br><br>Inter-rater reliability<br><br>Internal validity<br><br>(Iacono, West, Bloomberg, & Johnson, 2009) | Not specified.  |
| <b>Affective Communication Assessment (ACA)</b><br>(Coupe, Collins, Levy and Murphy, 1985)     | 1  | Recording sheet                | Systematic presentation of multisensory stimuli to the client | None available  | Bates (1976) theories of pre-intentional communication. |
| <b>Disability Distress Assessment Tool (DISDAT)</b><br>(Northgate and Prudhoe NHS Trust, 2006) | 1  | Criterion referenced checklist | Observation<br><br>Liaison with family/teachers/carers        | Limited evidence for inter-rater reliability<br><br>(Regnard et al., 2007)  | Not specified   |

Table 3.6. Published assessment frameworks used by participants (cont.)

| Assessment   | Number of participants citing assessment  | Assessment type                | How information is gathered for assessment             | Evidence for psychometric stability e.g. reliability/validity | Stated evidence base             |
|--|---|--------------------------------|--|---|----------------------------------|
| <b>Communication Development Profile (CDP)</b><br>(Child, 2006)        | 1   | Criterion referenced checklist | Observation<br><br>Liaison with family/teachers/carers | None available  | Coupe O’Kane and Goldbart (1998) |
| <b>Module 9: Scope AAC Curriculum</b><br>(SCOPE, 2004)                 | 1   | Criterion referenced checklist | Observation<br><br>Liaison with family/teachers/carers | None available  | Coupe O’Kane and Goldbart (1998) |
| <b>Communication Assessment Profile (CASP)</b><br>(Van der Gaag, 2017) | Not described. Designed for verbal clients with mild to severe learning disabilities. |                                |  |   |                                  |

Only three frameworks were cited by more than one participant (the PVCS, Routes for Learning and the P-scales) reflecting the diversity of measures used. The Communication Assessment Profile (Van der Gaag, 2017) was cited by one participant but is not appropriate for the population under discussion since it is described as an assessment for verbal individuals with mild to severe learning disabilities.

All but one of the assessments took the form of criterion-referenced checklists (Crais, 2011) rather than tests, with pre-selected lists or descriptors of skills being given and the assessor being required to judge whether the client demonstrated the skill or not. Similarly, all but one of the assessments required that judgements should be based on observation of the client in everyday settings combined with information about the target skills gathered from parents, teachers or care staff. The exception was the ACA where a set of stimuli (to be selected at the discretion of the assessor) is systematically presented to the client and their responses recorded. The Routes for Learning materials also described some specific tasks which might be optionally used to elicit certain target skills from participants, such as moving an attractive object out of sight whilst the client is looking at it to see if they demonstrate a reaction (Welsh Assembly Government, 2006, p8).

Data regarding psychometric aspects such as inter-rater reliability or construct validity was provided for five of the frameworks with four (the ACA, Routes for Learning, the CDP and the Scope AAC curriculum) having no data to support them in this area.

Finally, five of the frameworks provided information about the theory or evidence upon which they were based with three of these citing Coupe O'Kane' and Goldbart's (1998) book 'Communication Before Speech' as a basis and two referring to Bates's (1976, 1979) theories around pre-verbal communication development.

### **Further Comments Provided by Participants**

Further comments were given by 64% of participants. These focused mainly on the importance of involving a range of people in the assessment process, the need to carry out assessment over time and the challenges involved in assessing this client group.

Four participants emphasized the benefits of liaison with others e.g.:

*“I have found that the most efficient assessment method is to record accurately from information provided by support staff or family members”.*

Three participants referred to the need for assessments to be dynamic and ongoing over time e.g.:

*“Assessment happens most effectively over time, through observation of consistent behaviour and response to intervention.”*

Nine participants referred to challenges they experienced. These included issues in relying on information provided by others e.g.:

*“The quality of information is highly variable and dependent on support workers’ motivation and quality of training as well as skill.”*

*“I [also] think that as carers get to know their students, they ascribe more meaning to their actions which they interpret as progress.”*

Frustrations with current assessment approaches and feelings of being under-skilled were also voiced e.g.:

*“I find the current assessments available (routes for learning, Coupe & Goldbart) very time consuming & an unwieldy way of collecting information.”*

*“I have not found any useful assessments which have allowed me to assess students at this level.”*

*“I am no expert! “*

### **3.3.4. Discussion and conclusions**

Results from this survey study confirmed that SLTs take a diverse approach to assessing clients with PIMD. There was poor agreement around what constitutes key areas for assessment, with the most commonly assessed areas of expressive communication being the means clients used to communicate and the functions these served. Whilst useful for creating a functional description of a client's communication, these areas provide little information in respect of a client's developmental communication level. The means of communication used, in particular, are likely to reflect a client's physical rather than developmental profile in this population, since the ability to use vocalisations, facial expression, gesture and body movements will partly depend on the nature of their physical impairments. Evidence suggests that the extent to which an individual demonstrates intentional communication is a developmental indicator in pre-linguistic communication (e.g. Bates, Camaioni, & Volterra, 1975 and see further discussion in section 4.1 below). However, this was only cited as a key assessment area by 28% of participants.

Informal means of assessment were used by the majority of participants in this survey with only 40% reporting the use of published frameworks. This contrasts with the 80% of Goldbart's (2010) participants who reported using them and may be an indicator that the use of published frameworks is becoming less common in the field since the two studies were carried out several years apart. As in Goldbart's study, a wide range of frameworks was reported with no one framework being cited by a majority of participants. The three most commonly cited frameworks in Goldbart's study, the PVCS, the ACA and the Triple-C, were also cited by participants in this study suggesting that they are still in use despite the PVCS being out of print and the ACA difficult to obtain. Some more recently published assessment frameworks – the Routes for Learning and the Communication Matrix – were cited in the current study suggesting that there have been some developments in the field although further analysis of their evidence base suggests that they may not reflect any changes in theoretical thinking which have evolved over the last twenty years.

Indeed, appraisal of the cited published assessment frameworks confirms that their evidence base may not be up-to-date. There was a high degree of consistency in the underlying evidence base for the assessments with many citing a basis in the socio-linguistic work of Bates (1976, 1979) and Coupe O'Kane and Goldbart's (1998)



'Communication Before Speech', a publication which provides an overview of both socio-linguistic and psycholinguistic theories of pre-linguistic communication, relating them to practice with people who have PIMD. Whilst providing a robust theoretical foundation, these sources are over twenty years old suggesting that these new resources have been slow to incorporate developments in theory.

The preliminary results reported in Goldbart's (2010) conference paper were subsequently published by Chadwick, Buell and Goldbart (2018) in a paper which also provided further details about the rationales SLTs gave for taking their various approaches to assessment. It was found that only one of the fifty-five participants in their study considered the evidence base behind assessments when selecting them, further suggesting a weakness in evidence-based practice in this area. Participants in Chadwick and colleagues' study also gave little consideration to the psychometric robustness of the published assessments they chose, a finding consistent with the current study since four of the nine appropriate frameworks cited had no evidence to support their reliability or validity and one (the DISDAT) had only weak evidence in this area.

A reliance on indirect methods of data gathering was strongly indicated by the results of the current survey. This applied to data gathering for both informal and published assessments. Eighty four percent of participants reported observing clients in everyday contexts and in interaction with others and seventy six percent reported gathering information from parents, teachers or carers. Some participants specifically commented on the need to involve others in assessment. In contrast, only 48% reported using direct engagement as a means of assessment. Where this occurred, it was reported in vague terms and referred mainly to trialling intervention or engaging the client with multisensory objects although the purpose of this was not generally specified. The published frameworks used were almost all checklists which did not require direct engagement with the client, some being specifically designed to be completed in liaison with others familiar with the client or on the basis of observation in everyday contexts (e.g. the Triple-C, the DISDAT, the PVCS, the CDP).

Finally, a number of participants commented on the challenges inherent in this field with some expressing frustration with existing methods and others expressing a lack of expertise in this area.

## **Limitations**

It is unclear how representative the sample of SLTs involved in this survey was since no definitive figures or demographic information about the target population was available. Therefore, the extent to which findings may be generalised is unknown. However, in many respects the findings were consistent with those reported by Chadwick, Buell and Goldbart (2018) which offers some support for their being robust. Using open ended questions allowed participants to generate ideas freely but meant that failing to mention a particular aspect of assessment was not definitive evidence that they never considered it. Additionally, participants provided varying levels of detail in their response so that the underlying meaning of some responses were unclear. For example, one participant identified key areas of assessment as:

“preverbal communication, non-verbal communication occasionally verbal situational (occasionally verbal) comprehension”

Since the terms ‘pre-verbal’ and ‘non-verbal’ are broad and not defined here it is difficult to determine what specific skills this participant is referring to. Further research using more specific questions and a structured interview methodology would provide more accurate results.

## **Summary**

Combined findings from the survey study reported here and those reported by Chadwick, Buell and Goldbart (2018) indicate that the current practice of SLTs assessing communication in people with PIMD is characterised by:

- Idiosyncratic and informal approaches to assessment with poor agreement over what constitutes key assessment areas.
- Published frameworks which are predominantly criterion-referenced checklists. These may not have an up-to-date evidence base and may not be psychometrically robust.
- An evidence base which does not reflect recent developments in the field of prelinguistic communication development.
- Reliance on indirect means of data gathering for assessment such as the use of unstructured observation and liaison with others.

### 3.4. Critical evaluation of current practice

It is important to note that the existing approaches to communication assessment identified above have both positive and negative qualities. For example, taking an informal approach to assessment may be justified by the need to adopt a client-centred approach – a rationale which was voiced by 27% of participants in Chadwick, Buell and Goldbart's (2018) study. Taking an informal approach allows assessment to be tailored to the very specific profiles of each client with PIMD as well as their social and physical environment.

Using multiple approaches, including both formal and informal frameworks and a focus on varying key areas, may be justified by the fact that assessments are carried out for a range of purposes. For example, where the aim of assessment is to decide on suitable school placement, using a formal framework such as the Communication Matrix to focus on developmentally significant skills might be most appropriate, allowing a child to be placed with others of similar ability. Alternatively, where the aim of assessment is to inform intervention for improving peer interaction, informal assessment of functional social skills in the playground may well be more relevant since it provides information about any issues experienced by the child in an everyday context.

Despite having advantages, however, the use of diverse and informal approaches to assessment also leads to inconsistency in practice between individual SLTs. This has significant implications since assessment plays a critical role in establishing baselines and monitoring progress over time (Brinton & Fujiki, 2010). People with learning disabilities, including PIMD have long-term issues with communication and may receive speech and language therapy input across their whole lifespan, a situation which is likely to entail many changes of SLT. If each SLT takes a different approach, focusing on different key areas, using their own informal methods or published frameworks which are difficult to source, there is a risk that the results of repeated assessments will reflect only changes in the assessment approach used rather than real changes in the client's abilities or functioning.

Consistency between practitioners might be improved by facilitating evidence-based practice (EBP) in this field, ensuring that areas of pre-linguistic communication

identified as significant by research, are made known to SLTs and incorporated into shared frameworks for assessment. The move towards better integration of evidence into practice has been strongly promoted in speech and language therapy (Royal College of Speech and Language Therapists website, 2019) as in many fields (Sackett & Sackett, 2000), however, it would seem that EBP has been slow to develop in the area of learning disability, particularly in relation to PIMD. For example, the RCSLT provides a database of evidence relating to different client groups (RCSLT, 2019) but the only studies which focus on PIMD in the learning disabilities database are concerned with eating and drinking difficulties, confirming an ongoing neglect of this client group in terms of collation and dissemination of evidence. The finding that SLTs working in the field rarely referred to the evidence-base when making clinical decisions (Chadwick, Buell and Goldbart, 2018) further suggests an absence of EBP and may, partially, reflect the lack of resources in the field. The development of shared assessment frameworks would contribute to the integration of evidence into practice by providing a research-based structure to direct clinical decision-making. However, the published frameworks listed by SLTs in the survey studies reported above, did not appear to be integrating current evidence into their development. Appraising the current evidence base to identify areas identified as significant in pre-linguistic development would constitute a first step in improving this situation and constituted an aim for this thesis.

In addressing this issue, it is necessary to consider what evidence base might be most relevant to this population. The most prolific sources of evidence around pre-linguistic communication relate to typically developing infants (see (Lock & Zukow-Goldring, 2010 for a review). The trajectory of communication development in people who remain pre-linguistic communicators due to PIMD is the subject of less research and there is a lack of clarity, with some arguing that they, broadly, follow a typical developmental trajectory, albeit at a slower rate (Butterfield, 1991; Iacono, West, Bloomberg, & Johnson, 2009) and others suggesting that they may follow a different developmental path as a result of their complex physical and sensory impairments (e.g. DeVeney, Hoffman, & Cress, 2012; Welsh Assembly Government, 2006). Given the absence of evidence for an alternative pathway for development, it would seem that using knowledge of typical pre-linguistic communication development is the most suitable approach at the present time.

Taking a developmental perspective is also beneficial for effective goal setting (Brinton & Fujiki, 2010). Creating a detailed profile of an individual's current level of functioning enables realistic goals to be identified from those which are at a similar or slightly more advanced level of complexity and may already be within the individual's zone of proximal development (Vygotsky, 1978). Even where limited progress is anticipated, identifying an individual's current level of functioning in developmental terms can be used to ensure that the support they receive is pitched at an appropriate level and that they are given multiple opportunities to use their existing skills within their everyday routines (see Ware, 2003).

In summary, current approaches to communication assessment for people with PIMD are inconsistent. Improving evidence-based practice in this area would help address this consistency and an appraisal of current theory and evidence relating to pre-linguistic communication would constitute an initial step towards achieving this.

A reliance on indirect forms of information-gathering for assessment was clearly apparent in the findings of the survey studies reported above. Again, these are not without their merits. Observing communicative behaviours as they occur in a natural context provides ecologically valid evidence of an individual's everyday functioning (Atkin & Lorch, 2014; Crais, 1995) with some authors emphasizing that assessment should always take place in the context in which intervention is to be carried out (Wetherby, Goldstein, Cleary, Allen, & Kublin, 2003). However, gathering data through observation may offer limited opportunities to see an individual display their full range of communication skills. Individuals with PIMD tend to show a low frequency of communicative behaviours (Cirrin and Rowland, 1985; Ware, 2003) meaning that even a significant period of observation may yield a limited amount of information. In addition, the number of communicative behaviours observed depends, in part, on the opportunities offered by the context, with several studies indicating that prelinguistic communicators with developmental disabilities demonstrate more communicative behaviours in situations which are deliberately structured to offer communicative opportunities than they do in unstructured, natural settings (Cress, Arens, & Zajicek, 2007; Iacono, Waring, & Chan, 1996; Yoder, Warren, & McCathren, 1998). Even where a relatively structured setting is selected for observation, communicative opportunities may be rare. In a study of teaching sessions, Ryan, McGregor and Akermanis (2004) found that situations described by

teachers as being 'structured' did not necessarily offer the expected opportunities for communication or elicit communicative responses.

Therefore, whilst the data gathered through observation may be ecologically valid it is unlikely that, used alone, it will provide sufficient details about an individual's communicative capabilities for a thorough communication profile to be drawn up.

Liaison with others may be viewed as a useful complement to observation. Those familiar with the individual with PIMD are likely to have seen them use their communicative skills on many occasions and in a range of contexts. They are also likely to recognise the informal and idiosyncratic behaviours each individual with PIMD uses to communicate. Within an educational setting the role of teachers is key given the amount of time they spend with students who have PIMD, observing and assessing them during a range of educational, social and care-based contexts. For this reason collaboration between SLTs and teachers is an essential part of effective communication assessment.

However, research has questioned the accuracy and reliability of caregiver judgements about prelinguistic communication skills. For example, parents have been found to over-attribute intention to reflexive responses (Sigafos et al., 2011) while care staff have been found to overestimate the comprehension of clients with learning disabilities and to infer that communicative intent underlies all behaviours (Bradshaw, 2008). In educational settings, judgements about intentional communication in children with severe and multiple disabilities have been found to vary between teachers and SLTs (Carter & Iacono, 2002). Whilst caregivers and teachers have a detailed understanding of the individual with PIMD they are likely to have a more limited knowledge of prelinguistic communicative development resulting in their failing to notice certain behaviours or misinterpreting the relevance of others. In addition, their judgements will be based upon observation of the individual in the natural contexts discussed above, where opportunities to demonstrate a full range of skills may be minimal.

Therefore, whilst seeking information from people who know the individual well is beneficial for gathering information about their unique communicative behaviours and communication in a wider range of settings than the SLT can observe alone, judgements may be unreliable or, at the very least, subjective with no guarantees that

interviewees will have observed the individual's full range of abilities in key areas of prelinguistic communication.

The reliance on indirect methods of information gathering may well reflect conceptual changes in thinking about disability from an 'individual' to a 'social' model (e.g. Oliver, 1983; 2013) which proposes that:

“we are not disabled by our impairments but by the disabling barriers we face in society”  
(Oliver, 2013, p1024)

According to this model, the nature of an individual's disability cannot be determined solely by an assessment of their individual impairments but must include an appraisal of the environment they are in and the responses of those around them. First proposed in the 1970s, (Union of the Physically Impaired Against Segregation, 1975) the social model of disability has been adopted by the World Health Organisation and underpins the International Classification of Functioning, Disability and Health (ICF) (WHO, 2012) as well as being espoused by the RCSLT whose service delivery guidelines state that 'Speech and language therapy service delivery [should] consider communication needs in the context of a social model of disability'. (RCSLT, 2019).

Thus, by focusing on the physical and social environment of the individual with PIMD, SLTs are appropriately reflecting current social and political perspectives but, may be doing so to the detriment of taking individual impairments into account. The ICF includes the qualifiers of 'capacity' and 'performance' where 'the Capacity qualifier ... indicates the highest probable level of functioning of a person in a given domain at a given moment' whereas 'the Performance qualifier describes what an individual does in his or her current environment' (WHO, 2012, p 11).

Determining the gap between capacity and performance, as defined above, can identify the extent and type of environmental manipulations required for an individual to make full use of their capacity (WHO, 2012). Whilst performance currently provides a focus for SLT's approaches to communication assessment for people with PIMD, it would appear that capacity is not being fully investigated.

Assessing capacity involves investigating what an individual can do in a maximally supportive environment (WHO, 2012). In the context of communication assessment this will involve some direct engagement from the SLT to provide suitable activities, creating opportunities for target behaviours and structured degrees of prompting.



Some SLTs did report using direct engagement to assess their clients – often reporting the use of multisensory objects to do so although further investigation would be required to establish the exact nature of such approaches. The fact that they are informal and personally devised by each therapist, however, suggests that they might be difficult for other SLTs to replicate if assessments are repeated over time.

In summary, existing approaches to communication assessment for people with PIMD focus on gathering information through observation and interview with people familiar with the individual. This provides a measure of a client's communication performance but may not reflect their capacity in terms of the highest level of function they can achieve. Therefore, there is a need to develop more direct and replicable methods for assessing capacity.

### 3.5. Addressing the problem

This chapter has presented evidence about SLT's current approaches to communication assessment in PIMD, highlighting shortcomings such as inconsistency, a reliance on indirect methods of information gathering, a tendency to focus on performance rather than capacity and a failure to take recent developments in pre-linguistic theory into account when devising and selecting shared assessment frameworks.

This thesis seeks to address some of these issues through investigating which areas of pre-linguistic communication are identified as significant by current theory and whether capacity for these might be measured by taking a direct and standardised approach to assessment. The following chapter describes the appraisal of current literature around pre-linguistic communication, identifying and discussing the growing focus on joint attention as a key area and identifying the research questions for this study. The subsequent chapter (Chapter 5) will then discuss the use of direct and standardised methods in measuring pre-linguistic communication.

## **4. Using Current Theory and Evidence to Identify Key Areas for Assessment**

This chapter will summarise the evidence base underlying current approaches to assessment in speech and language therapy, evaluating it in respect of more recent changes in perspective, most notably the developing interest in JA. A discussion of theory and evidence around JA will then follow, with particular reference to its definitions, developmental trajectory and variables which might affect its occurrence including the socio-interactive environment and aspects of individual functioning such as visual and motor skills. An overview of current knowledge about the development of JA in atypical populations will be presented, including a detailed evaluation of studies investigating JA in people with PIMD. Finally, an argument for the relevance of JA to communication assessment for people with PIMD will be presented alongside plans for developing knowledge through the work carried out in this thesis.

### **4.1. The underlying evidence base for existing approaches to speech and language therapy assessment and intervention for people with PIMD.**

The foundations for our understanding of prelinguistic communication were laid in the 1970s when the advent of portable video recording enabled detailed observation of infant behaviour (Lock & Zukow-Goldring, 2010). Prior to this time theorists such as Chomsky (1965) had suggested that language acquisition was predominantly innate, paying little attention to the period of development before language emerged. Such ideas were challenged by seminal work carried out by the teams of Elizabeth Bates (e.g. Bates, 1976; Bates, Camaioni, & Volterra, 1975) and Jerome Bruner (e.g. Bruner, 1981; Bruner, 1974). On the basis of detailed, longitudinal studies of infants, these authors identified a clear sequence to pre-linguistic development and developed theories around the social processes which underpinned them. Central to these theories was the concept that pre-linguistic acts were early precursors and pre-requisites for the later social use of language. Later work by authors such as Sugarman (1984) and (Harding & Golinkoff, 1979) provided further support for these ideas.

A key feature of the pre-linguistic developmental trajectory was identified to be a progression from pre-intentional to intentional use of communicative signals (Bates, Camaioni, & Volterra, 1975). Prior to the age of six months, infants produce reflexive reactions to internal states such as crying in response to discomfort (Bates, 1979). Although these do not appear to be produced with deliberate intent to convey a message, caregivers react as if they had been – for example by soothing the crying infant and removing the source of discomfort. This represents the pre-intentional or perlocutionary phase of development. As they develop, scaffolded by the responses of caregivers, infants come to produce increasingly complex signals with the deliberate intention of influencing the behaviour of others, a milestone which is reached around the age of nine months. Bates described the emergence of intentional communication as ‘a moment in the dawn of language’ (Bates, 1979, p33) and identified its defining characteristics to be:

- 1) The alternation of eye contact between the goal [object] and the adult.
- 2) The increasing changing or augmenting of signals depending on the adult’s response.
- 3) The gradual evolution of these signals into short sounds or gestures.

It was further established that initial attempts at intentional communication, known as proto-imperatives, tend to be aimed at obtaining or rejecting items. As intentional communication develops, proto-declaratives emerge as infants begin to communicate about objects or events purely for social purposes, wanting to ‘comment’ or share affect about the item.

This sequence of intentional communication development observed in the typical development of pre-linguistic communication provides a focus for much of the theory and evidence supporting the understanding of communication in PIMD. In their book, ‘Communication Before Speech’ which outlines theories of pre-linguistic communication for practitioners working with PIMD, Coupe O’Kane and Goldbart (1998) describe the following five levels of development:

1. Pre-Intentional: Reflexive Level
2. Pre-intentional: Reactive Level
3. Pre-intentional: Proactive Level
4. Intentional: Primitive Level
5. Intentional: Conventional Level

The framework they describe also integrates understanding around the developing complexity of signals since level 4 includes the use of 'primitive' signals, often motor acts such as 'reaching for a cup while looking at the adult' (Coupe-O'Kane & Goldbart, 1998, p63) and level 5 describes the use of more conventional signals such as "gestures, vocalisations (such as jargon) and verbalisations (for instance protowords)" (Coupe O'Kane & Goldbart, 1998, p65).

Since several of the published assessment frameworks used by SLTs and described in section 3.3.3.2 were based on evidence derived from Bates or Coupe O'Kane and Goldbart, it is unsurprising that they show a similar focus on this sequence of intentional communication development. Both the Triple-C and the Communication Matrix use information about communicative intentionality as well as about the means of communication used, to assign an individual's communication to a particular level with the Triple-C employing the following levels:

1. Unintentional Active
2. Unintentional Passive
3. Intentional Informal
4. Symbolic (basic)
5. Symbolic (established)

Around one quarter of the SLTs taking part in the survey study also referred to assessing intentional communication, suggesting that this area is considered to be of clinical relevance to some practitioners. This is further supported by evidence that intensive interaction and the use of 'micro-switches' are intervention approaches frequently used with this population (Goldbart & Caton, 2010; Goldbart, Chadwick, & Buell, 2014). Both approaches, and particularly the use of microswitches to operate

equipment or 'speak' messages, can be used as a means of developing intentional communication in prelinguistic communicators (Hewett & Nind, 2013; Schweigert, 1989).

It would seem then, that an understanding of the developmental sequence from pre-intentional to intentional communication and the evolution in complexity of communicative signals first identified by Bates and Bruner in the 1970s, continue to underpin SLT approaches to assessment and intervention in relation to pre-linguistic communication in people with PIMD. Taking such developmental approach with this client group is not without issue since the extent to which pre-linguistic communication development follows a typical pathway in people with PIMD is unknown. Their profile of functioning in terms of cognition, motor function, visual, hearing and other sensory processing is understood to differ widely from the typical population and this will inevitably have an impact on their developmental pathway. Research has, for example, highlighted the role played by behaviour states and levels of alertness in pre-linguistic communication for this group (e.g. (Arthur-Kelly, Bochner, Center, & Mok, 2007; Munde, Vlaskamp, Ruijsenaars, & Nakken, 2009). However, as the review of literature above has outlined, the sequence of typical pre-linguistic communication development is increasingly well understood and there is evidence to suggest that at least some individuals with severe and profound disabilities do follow its course in relation to aspects such as intentionality and the development of gestures (see (McLean & Snyder-McLean, 1987). That said, adopting a developmental perspective need not imply that individuals with PIMD are expected to follow a linear path of development through the stages identified from typical development. The Routes for Learning assessment, for example, uses a developmental framework to describe an individual's current level of functioning but acknowledges that learners with PIMD may develop in lateral way within stages or show development in some areas but not others (Welsh Assembly Government, 2006).

Having considered these issues, the work described in this thesis was based on a developmental framework in which evidence and theory derived from research into typical pre-linguistic development are used to explore and describe the skills of children with PIMD. It was felt that taking this approach would provide a strong evidence base for increasing understanding of this client group and describing their existing levels of functioning with reference to an established framework.

Given this approach, the question which needs to be addressed is whether recent understanding in the field of prelinguistic development supports the ongoing relevance of areas identified by Bates and Bruner or whether subsequent research has highlighted the importance of other communicative or cognitive domains which should provide a focus for communication assessment.

A review of research into pre-verbal communication was carried out by Lock and Zukow-Goldring (2010). Their conclusion was that:

“the last decade of work by and large confirms the “developmental milestones” that were previously apparent...What we have got, though is a much finer grain to these observations”

(Lock & Zukow-Goldring, 2010, p397)

Thus, evidence has continued to corroborate the significance of a progression from pre-intentional to intentional communication across the first year of life. However, the research findings summarised by Lock and Zukow-Goldring and a wider appraisal of the literature do suggest a shift in perspective in this area.

As outlined by Bates (1979), above, one defining criterion of intentional communication is that there is combined reference both to another person and to an object or event, frequently manifested through the alternation of eye gaze between the two. This observation has been reiterated by several other authors in an attempt to create operational definitions of intentional communication (e.g. Wetherby & Prizant, 1989). Although always acknowledged as a critical component in the development of intentional communication, such ‘joint referencing’ as it is termed by Coupe O’Kane and Goldbart (1998) has, over recent decades, become a significant focus of interest in its own right, generating theoretical debate and research into the phenomenon conceptualised more broadly, as ‘joint attention’. The prevalence of interest in JA and studies relating to it have prompted Reddy to comment that:

“The empirical picture today is a very different one from that of ten years ago. Reports of infant reactions to others’ gaze and other goal-directed actions drip from every relevant journal and reach into the earliest points in the first year”

(Reddy, 2010, p365)

This growth in understanding around JA, its inextricable links with intentional communication (e.g. Racine, 2011) and its significance during the earliest developmental phases of life provide a persuasive argument for considering its relevance to the development and assessment of prelinguistic communication in people with PIMD. Further discussion around the nature of JA, its definitions, developmental progression and influencing factors will, therefore, now be presented followed by a summary of existing knowledge of JA in atypically developing populations including those with PIMD.



## 4.2. Joint Attention

In broad terms, JA relates to a 'triadic' arrangement where two people coordinate their attention to the same object or event in the environment (Bakeman & Adamson, 1984, Carpenter and Liebal, 2011). In the context of infant development this may arise as a result of the infant following an adult's focus of attention by following their gaze direction or a point. Mundy describes this as 'responding to joint attention' (RJA) (e.g. Mundy & Newell, 2007). Alternatively, joint attention may arise when the infant makes active attempts to direct an adult's attention toward an object or event which is of interest to them, behaviour termed 'initiating joint attention' (IJA) by Mundy.

### 4.2.1. The Role of joint attention in the development of communication and language

There is widespread agreement and a growing body of evidence that JA is essential to the development of communication and language and is a critical factor in the emergence of intentional communication (Adamson and Chance, 1998; Mundy & Newell, 2007; Nunez, 2016; Seemann, 2011; Tomasello & Carpenter, 2007). As outlined above, in order for expressive perlocutionary signals to become intentional forms of communication they must be directed towards a partner through the sharing of attention to the same objects and events (Bates, Camaioni, & Volterra, 1975; Wetherby & Prizant, 1989). Engaging in shared attention to the same objects and events is also critical to the development of the understanding of language as adults accompany shared attention to particular referents with the relevant spoken words. Further support for the significance of JA in language development is provided by evidence that individual differences in the demonstration of behaviours related to JA during the first 18 months of life are associated with a number of later developing social and linguistic factors such as receptive and expressive language (Calandrella & Wilcox, 2000; Morales, Mundy, & Rojas, 1998), the ability to understand and use terms relating to the mental states of others (Brooks & Meltzoff, 2005; 2015), play skills and social relationships (Chiat & Roy, 2008).

Despite agreement around the significance of JA in communication and language development, there has been much theoretical discussion around its exact definition, a situation which has been characterised as the 'rich' versus 'lean' debate (Racine, 2011).

#### **4.2.2. Rich and lean definitions of joint attention**

Under a 'lean' definition, JA is considered to have occurred when two individuals attend to the same object or event, sometimes as a result of an intentional action taken by one them (e.g. Leavens & Racine, 2009). Here, it is possible for attentional focus to be shared without both individuals being aware of this. For example, Bakeman & Adamson (1984) observed infants in play with their mothers engaging in what they termed 'passive joint attention' where a mother might play with a toy whilst looking back and forth between it and the infant. Here, although there is a joint focus of attention, the infant shows no awareness of this.

In contrast, under a 'rich' definition, true JA only occurs when both individuals show awareness of their shared focus of attention by signalling this to each other via means such as a 'sharing look' and/or other communicative signals (Carpenter & Liebal, 2011, Hobson & Hobson, 2007, Tomasello & Carpenter, 2007). Carpenter and Liebal (2011) argue that true JA involves a degree of 'recursive mind reading' in which both partners have a sense of knowing what the other knows so when an infant follows the gaze of an adult it is because he or she understands that the adult is looking at something because they find it interesting or wish to obtain it. The implications of the debate between rich and lean perspectives can be illustrated by considering research into infant gaze following.

Numerous studies have demonstrated that infants will follow the direction of an adult's head turn by looking in the same direction (see Moore, 2008 for a review). An experimental protocol for testing whether infants would follow a head turn was first developed by Scaife and Bruner (1975). In their study infants aged from 2 -12 months were engaged in face-to-face interaction with a researcher. During the course of the interaction, the researcher would turn their head to look at a small light situated to the side and it was observed that the infants would respond by turning their head in the same direction. Although only 30% of 2-4 month old infants demonstrated the response, this increased with age so that, by 11 months all the infants in the study were doing so. Subsequent studies have replicated the finding that infants follow the direction of an adult head turn with increasing consistency over the first year of life (Brooks & Meltzoff, 2014; D'Entremont, Hains, & Muir, 1997; Moore & Corkum, 1998; Senju & Csibra, 2008).

However, ongoing research has suggested that, while the tendency to follow the head turn of an adult may be an early manifestation of a gaze following response they may only be considered to result in JA if a lean rather than rich definition is adopted. Only at 10-11 months of age do infants become more likely to follow the turn of a head with open eyes than one with closed eyes suggesting that the following of head turns in younger infants may occur reflexively and without an understanding that the adult is looking at something (Brooks & Meltzoff, 2005). Thus, whilst the following of gaze in the form of a head turn prior to 10-11 months may result in the infant looking at the same object or event as an adult (JA under a lean definition) this shared focus occurs without the type of mutual understanding (JA under a rich definition) which develops later.

Tomasello suggests that lean joint attention behaviours are the product of basic learning processes i.e. learning that following the gaze of another person may lead to interesting or useful sights, rather than reflecting a more complex desire to share the experiences with another person (e.g. Tomasello, Carpenter, Call, Behne, & Moll, 2005). He also argues that the initiation of JA for proto-imperative (requesting) purposes is another lean form of joint attention since it requires only a mechanistic understanding of other people as providers of objects without requiring an understanding of others' subjective states. Evidence that nonhuman primates demonstrate basic gaze following and proto-imperative JA reinforce this concept that certain types of attention sharing are ontogenetically and phylogenetically less complex than others (Tomasello & Carpenter, 2007, Carpenter and Liebal, 2011).

Perhaps the most useful conclusion which may be drawn from the debate around rich and lean definitions is that JA is not a simple, single entity but a state of shared attention which can reflect varying degrees of socio-cognitive complexity. Indeed Racine concludes that it may be a case of "lean first and rich later" (Racine, 2011, p37) so that there is a developmental sequence in which the behaviours and processes comprising 'lean' JA emerge during early infancy and are precursors to the later-developing and more complex phenomena involved in 'rich' JA. Along the way, Gomez (2007) suggests there may be a 'middle ground' where some intentional reading of behaviour occurs without the attribution of 'unobservable representations' (Gomez, 2007, p793).

Given that the developmental sequence of JA is believed to begin in early infancy and continues to evolve during the first two years of life (Mundy & Gomes, 1998), it is clearly of interest to those focusing on the communication skills of people with PIMD whose developmental abilities are felt to lie within this period. Further discussion of what is known about this developmental trajectory will, therefore, follow. However, before further discussion, it is helpful to draw awareness to the dichotomy between behaviours and states which is implicit in the study of JA in order to avoid what has been termed a 'dual usage' problem.

#### **4.2.3. Behaviours, states and the dual usage problem in the field of joint attention**

A dual usage problem occurs when behaviours of interest are used interchangeably with the function or purpose they serve. It is particularly prevalent in studies in the field of JA where both observable behaviours and postulated mental states are under investigation and have both been labelled as 'joint attention' by different researchers (Tasker & Schmidt, 2008). For example, JA has been described as both 'a complex psychological phenomenon' (Peacocke, 2005, p298) and a 'complex behavioural phenomenon' (Dube, MacDonald, Mansfield, Holcomb, & Ahearn, 2004, p197) and the term 'joint attention' has been used variously as an adjective to describe behaviours and skills or a noun to describe the outcome of such behaviours (Tasker and Schmidt, 2008).

In considering research into JA and its development, therefore, it will be important to differentiate between 'JA behaviours' which are behaviours which may *result* in JA or indicate that JA is occurring (such as alternating eye gaze between people and objects, pointing or following a point) and JA itself which is a state involving two people sharing an attentional focus to the same thing and who may or may not have an awareness of their shared focus. Since the state of JA is internal and can only be inferred through the presence of observable behaviours, both states and behaviours are of interest in any studies of JA.

#### **4.2.4. The developmental trajectory of JA**

The following review of the literature on typical development focuses on the emergence of JA behaviours and outlines how a separate attentional focus on people and then objects is eventually replaced by coordinated attention to both people and objects. This coordinated attention is gradually accompanied by certain additional

signals which are thought to be indicative of 'lean' and then 'rich' JA. The simultaneous development of responding to JA (RJA) in which the infant becomes increasingly more able to follow the attentional focus of an adult is also described.

### **Separate attentional focus on people and objects**

Studies of infant development suggest that, in their early months, infants attend very distinctly to either people or objects. In the earliest months their attention is predominantly directed to people, with studies demonstrating that infants show an over-riding interest in looking at faces from birth (Farroni, Johnson, & Csibra, 2004) and, by three months old, spend half their waking hours in mutual gaze with caregivers (Wolff, 1987). A marked shift then occurs at around the age of five months as time spent in face-to-face interaction decreases and their principal focus of attention becomes objects in the environment (Bates, Camaioni, & Volterra, 1975; Kaye & Fogel, 1980; Lock & Zukow-Goldring, 2010).

Whilst the attentional focus during this period of time is very much separate rather than joint, it is necessary for both attentional interest in both people and objects to be developed before the two can be coordinated. For example, it has been found that children with autism spectrum disorder (ASD) are less likely than typically developing children to develop an attentional focus to faces, a factor which may contribute to their later difficulties developing coordinated attention between people and objects (e.g. Swettenham et al., 1998).

### **Integrated attention to people and objects**

At around the age of 9-10 months infants begin to integrate their attention to people and objects, a transition usually manifested through gaze behaviours (Bates, Camaioni, & Volterra, 1975). For example, whilst playing with an object, a child may look up and make eye contact with their mother before returning gaze to the object (Bruner, 1974). This is termed 'triadic gaze' (e.g. Olswang, Feuerstein, Pinder, & Dowden, 2013). Such looks are significant since they demonstrate that an infant is able and motivated to coordinate their attention between objects and people and suggest a possible interest in the attentional focus of another. However, when used without any additional behaviours the purpose of these looks is unclear. Hobson & Hobson (2007) term these 'checking looks' since they appear to be checking on the focus of another – perhaps to see if they are also looking at the object or to see if a

response will be forthcoming. Whilst such looks may result in an episode of shared attention to the same object or event this could only be considered JA if a lean definition is adopted.

### **Integrated attention to people and objects accompanied by additional behaviours**

As infants continue to develop, they begin to accompany alternating gaze between objects and people with additional signals, most notably, vocalisations (Harding & Golinkoff, 1979). This is where the transition to intentional communication, described by Bates (1979) emerges, as signals are produced with persistence and may be modified until a goal is apparently obtained (Wetherby & Prizant, 1989). As outlined in section 4.1, a distinction between signalling for imperative (requesting) and declarative (commenting) purposes begins to be made at this point, with imperative signalling thought to precede declarative signalling. However, it should be noted that this distinction is often made on the basis of context, since similar observed child behaviours (gaze alternation and signalling) may be interpreted differently in different contexts. For example where an infant looks back and forth between a caregiver and an object which is out of reach whilst persistently vocalising, this may be construed as a request (Bates, Camaioni, & Volterra, 1975) whereas, in the case of an interesting toy which is operating and within reach, gaze alternation between object and adult accompanied by vocalisations is more likely to be construed as a comment since the infant has no need to request in this context (Mundy, Kasari, Sigman, & Ruskin, 1995).

With ongoing development, the behaviours accompanying joint referencing through gaze alternation become more complex from the age of 12 months. One apparently important behaviour which is felt to be an indicator of JA under a rich definition, is the 'sharing look' (Hobson & Hobson, 2007). Defined as:

“looks that human judges can recognize as reflecting intersubjective/personal/affective contact”

(Hobson & Hobson, 2007, p 415)

A sharing look from an infant to an adult is believed, by Hobson and Hobson, to indicate the presence of a rich JA where the adult and infant share affect and understanding about the object or event.

Infants are also observed to use gestures, giving, pointing and, eventually, words to direct the attention of others to their own focus of interest for both proto-imperative or proto-declarative purposes (Lock & Zukow-Goldring, 2010; Tomasello & Farrar, 1986).

### **The development of RJA (Responding to Joint Attention)**

During the period of time in which infants begin to integrate their attention to objects and people as described above, research suggests that they also demonstrate an increasing propensity to follow the focus of another's attention in the form of gaze or point following as outlined in section 4.2.1., above. In summary, studies have suggested that infants begin to follow the gaze direction of an adult somewhere between 2 months (Scaife & Bruner, 1975) and 6 months of age (Morales et al., 1998) with individual differences predicting performance on later measures of language and communication development (Mundy & Newell, 2007). Certain external variables have also been found to influence performance here. For example, 6 months olds were found to be more likely to follow the gaze direction of an adult if the adult preceded their gaze shift by looking directly at the infant and calling their name (Senju & Csibra, 2008). A study by Moore, Angelopoulos, & Bennett (1997) further indicated that younger infants were less likely to follow gaze direction if the active head turn preceding it occurred behind a screen, suggesting that the dynamic aspect of head turning provides a prompt for gaze following in its early stages. Gaze following behaviours continue to develop in complexity into the second year of life so that, by 18 months of age, infants begin to follow the direction of eye gaze alone (Moore et al., 1997) and are also able to follow the point of an adult (Tomasello, Carpenter, & Liszkowski, 2007).

Thus, research into typical infant development indicates a developmental sequence of emerging behaviours which may either facilitate or indicate the presence of JA which is an inextricable component of intentional communication. An overview of this developmental sequence is summarised in Table 4.1.

*Table 4.1. The developmental sequence of behaviours relating to joint attention*

| Age (in months)    | Behaviours relating to JA  |
|--------------------|--|
| <b>0-5 months</b>  | Attentional focus on people<br>Some infants following gaze accompanied by head turn  |
| <b>5-9 months</b>  | Attentional focus objects<br>Increasingly likely to follow gaze accompanied by head turn   |
| <b>9-12 months</b> | Coordinated attention between people and objects (e.g. looking back and forth between them.)<br>Coordination of attention is accompanied by potentially communicative signals such as vocalising (= intentional communication).<br>Coordination of attention is accompanied by sharing looks.<br>Consistent following of gaze accompanied by head turn |
| <b>12 months +</b> | Begins to use pointing, giving and showing to direct adult attention.<br>Follows direction of eye gaze and alone and begins to follow point.   |

#### **4.2.5. The significance of the socio-interactive context**

Engagement in the JA behaviours described above will only lead to the occurrence of a state of JA if this is supported by the socio-interactive context (Moore, 2008; Racine, 2011). Features of the socio-interactive context will not only help to determine whether JA occurs but whether it is imperative or declarative, rich or lean in nature. The role of two factors, the salience of referents and the behaviour of the communication partner are particularly significant and will be considered here.

##### **4.2.5.1. The salience of referents: ‘top-down’ and ‘bottom-up’ joint attention**

Whether rich or lean, JA is a state involving coordinated attention of at least two people to the same referent (an object or event). For this to occur the referent has to be sufficiently interesting, or salient, for at least one of the parties to attend to it in the first place. A significant factor here is whether the referent is initially salient to just one



of the parties or instantly salient to both of them since this leads to qualitatively different episodes of JA, termed ‘top-down’ or ‘bottom-up’ (Carpenter & Liebal, 2011), each of which is presented in the different patterns of behaviour presented in Figure 4.1).

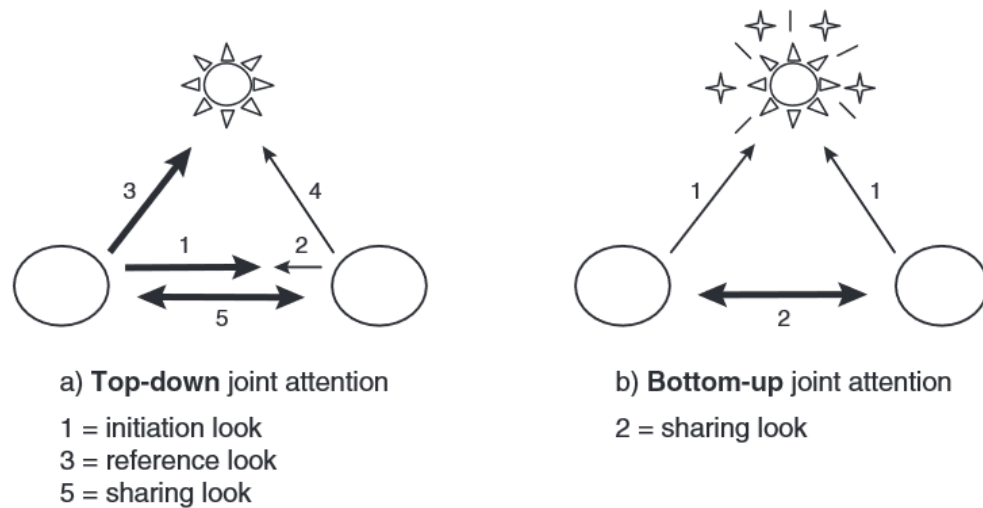


Figure 4.1. The sequence of looks in top-down and bottom-up attention

(Reproduced from Carpenter and Liebal, 2011, p171. Communicative looks are indicated by bold arrows).

Top-down JA occurs when a stimulus is salient to a communicator but is either of no interest to their communication partner or is unseen by them. In order for JA to occur, the communicator must first attract the visual attention of the communication partner with an ‘initiation’ look (possibly achieved by vocalising to get the communication partner’s attention first) then directing their attention to the stimulus using a ‘reference look’. Where a rich episode of JA occurs, the two then look towards each other and engage in a ‘sharing look’, possibly accompanied by other signals such as smiles or speech, through which they communicate their affective reaction to the stimulus. For example, the communicator notices that a saucepan is boiling over, calls the name of their communication partner, engages in mutual eye contact (initiation look) then looks over to the pan (reference look). Both individuals then look back at each other

to share a look of panic (sharing look) and, possibly, additional types of communication.

Bottom-up attention occurs when a referent is simultaneously salient to both communicator and communication partner. Both partners seek to share their reaction with the other so look at each other and engage in a sharing look without either an initiation or reference look being required. For example, the sight and sound of the pan suddenly boiling over engages the attention of both communicator and communication partner who look at it and then simultaneously look at each other to share their panic.

The distinction between 'top-down' and 'bottom-up' episodes of JA is important, partly because it suggests different types of socio-interactive context which might be used to elicit JA but, also, because it results in different patterns of observable behaviour, both of which may denote the presence of JA. Some studies define the occurrence of JA in terms of 'triadic gaze' where gaze must shift from a person, to an object, and back to the person (e.g. Olswang et al., 2013). However, the existence of 'bottom-up' contexts for JA suggests that, while the context is 'triadic' (involving at least two people and a referent) the gaze behaviour need not be and that a single, two-point, gaze shift from an object to a person can be indicative of JA provided affect is then shared in the form of a sharing look or other signals.

#### **4.2.5.2. Communication partner behaviour**

Joint attention, and particularly the type of rich JA involving the sharing of affect, is dependent on the engagement of both partners. For example, when an infant directs a 'checking look' at an adult, this can only develop into a mutual 'sharing look' if the adult is looking at them. In a 'top-down' scenario, an infant may direct an initiating look at the adult. If the adult is not looking back at them, they may either lose interest or use an additional signal to gain the adult's attention. However, if the adult continues to look elsewhere, the potential for JA is lost despite the infant's bid to initiate it. Therefore, when creating or examining contexts where JA might occur, it is important that communication partner behaviour is either monitored or controlled to assess the extent to which it facilitates JA.

In summary, it is important that any examination of JA behaviours or states, including the assessment of JA skills in people with PIMD, not only considers aspects of the individual's behaviour which might lead to JA but also features of the socio-interactive context, specifically the salience of referents and communication partner behaviour.

#### **4.2.6. The relevance of visual and motor skills to JA**

As with many cognitive and communication skills, the development of JA is linked to the maturation of sensory, physical and neural systems (Lock & Zukow-Goldring, 2010; Rowland, 2013). Consideration of the JA behaviours outlined in Table 4.1 demonstrates that visual and motor skills are particularly relevant to this area with the use of alternating eye gaze and gestures such as pointing being universally associated with most accounts of JA development (Reddy, 2010). However, both visual and motor skills undergo a developmental progression of their own which needs to be considered in relation to their impact on the ability to engage in JA behaviours. In establishing how JA might develop in a population with PIMD, consideration of visual and motor development is particularly significant given the prevalence of impairments they experience in these areas.

#### **The typical development of visual skills relating to JA**

In order to demonstrate the JA behaviours outlined in Table 4.1., the infant first needs to be able to fixate their visual attention to single referents in the form of objects or people. To coordinate their attention between an object or person they need to be able to disengage gaze from one stimulus and shift it to another. Such visual skills are not present from birth but develop gradually over the first months of life.

Studies suggest that infants show a strong preference for looking at faces, being able to fixate gaze on them from birth, particularly if the face is directing eye gaze to them (Farroni, Csibra, & Simion, 2002). The ability to fix gaze on objects develops from six weeks and is mainly established by 20 weeks provided objects are no more than 1 metre away (McKenzie & Day, 1972). The distance at which infants can fixate on objects continues to increase with age and is facilitated if additional cues are provided such as the shaking or tapping of objects by a caregiver (Leiba, 2000).

Development of the ability to disengage gaze from a single stimulus and shift it to another has been the subject of much research (see Atkinson & Braddick, 2012 for a discussion). Seminal work in the area was first carried out by Tronick (1972) who

developed a 'fixation shift paradigm' in which a rotating wooden shape was presented centrally, engaging the infant's visual attention. A second stimulus (another wooden shape) then appeared at its side. Although Tronick found that infants aged from 2-3 weeks showed some capacity for shifting their gaze from the first to the second stimulus, this was dependent on the distance between the two stimuli being small and the second stimulus being dynamic (rotating). Subsequent use of this fixation shift paradigm, in which fixation on a first stimulus is followed by the presentation of a second one, has established the existence of a 'competition effect' in infants up to the age of 3-4 months who experience greater difficulty shifting gaze from one stimulus to another if the first stimulus remains in place, rather than being removed, after the second stimulus has appeared (Atkinson and Braddick, 2012). Since typically developing infants have largely mastered the ability to disengage and shift gaze by the age of four months it follows that their functional vision ability is sufficient for them to engage in the type of person-object gaze shifts which underpin JA from this point. However, studies of children with neurodevelopmental disabilities have indicated that their functional vision skills, including the ability to shift gaze, are often subject to permanent impairment.

### **The development of visual skills relating to JA in atypical populations**

Visual behaviours have been extensively studied in children who are developing atypically as a result of brain damage or developmental disabilities including those caused by specific syndromes (Atkinson & Braddick, 2012; Braddick et al., 1992; Cornish, Scerif, & Karmiloff Smith, 2007). Poor performance on measures of visual behaviour, including the fixation shift paradigm, are associated with poorer outcomes in terms of neurological and cognitive development (Mercuri et al., 1999) and persistence of abnormal responses has been found in atypically developing children. For example, the competition effect continued to be observed in the gaze shifting behaviours of children with Williams syndrome up to the age of six years (Atkinson et al., 2003). Patterns of fixation are also affected by atypical social development with children diagnosed with ASD more likely to fixate on non-social referents (objects) than social ones (faces) (Swettenham et al., 1998).

The difficulties with functional vision described above are related to atypical functioning of cortical and subcortical systems within the brain, often described as 'cerebral visual impairment' or CVI (e.g. Sakki, Dale, Sargent, Perez-Roche, &

Bowman, 2018). While these difficulties cannot be attributed to impairments of the eye or optic nerve, these aspects are also frequently impaired in atypical populations (e.g. Evenhuis et al., 2001) leading to issues such as poor visual acuity and strabismus (where the eyes are not aligned). With judgements about higher-order processes, including JA, often being made on the basis of observable visual behaviours (Aslin, 2007), caution needs to be exercised when making such judgements in relation to neurodevelopmentally impaired populations given this prevalence of impairments and atypical patterns in their visual functioning (Venker & Kover, 2015). Clearly, this has implications for the investigations of JA in people with PIMD where care needs to be taken that individuals have the necessary functional visual skills of fixing and shifting gaze if such skills are to be considered indicators of their capacity for JA.

### **The typical development of motor skills relating to JA**

Motor skills begin to be implicated in JA behaviours from the age of around 9 months when gestures may accompany JA as a means of directing adult attention. A developmental trajectory of gestural development has been identified with initial gestures tend to be 'deictic' (e.g. open-hand reaching) and 'referential' gestures (e.g. pointing or putting a cupped hand to mouth to indicate 'drinking'), developing at around 12 months (Crais, Watson, & Baranek, 2009; Tomasello, Carpenter, & Liszkowski, 2007).

### **The development of motor skills relating to JA in atypical populations**

Studies of children and adults with developmental disabilities suggest that many do develop forms of gestural communication and that, for some, the complexity of the gestural forms they use is predictive of their ability to develop language (Brady, Marquis, Fleming, & McLean, 2004). However, for those with profound cognitive impairments, gestural abilities do not seem to be linked to broader cognitive measures (McLean, McLean, Brady, & Etter, 1991) and it is likely that this is due to accompanying motor impairments. As outlined in section 2.2. above, many people with PIMD have severe physical impairments which mean that the production of gestures is more effortful and less accurate than in the typically developing population with some individuals being unable to produce any gestures at all. As a result, the use of visual behaviours to facilitate JA is likely to be even more significant for this group.

In summary, a clear trajectory of development in JA has been established for typically developing infants. However, due to their severe cognitive, sensory and physical impairments it cannot be assumed that JA follows the same trajectory in people with PIMD. Some research has been carried out into the characteristics of JA in atypical populations, including a small number of studies looking specifically at PIMD and their findings will now be considered.

#### **4.2.7. Joint attention in atypical populations**

##### **4.2.7.1. Joint attention in people with Autism Spectrum Disorder (ASD)**

The most developed area of research concerning JA in atypical populations has focused on individuals with ASD. Numerous studies have established that impairments in JA are a defining feature of ASD, to the extent that their presence is a key predictor of ASD in young children (Chiat & Roy, 2008). A specific pattern of JA deficit in ASD has been identified, in which the initiation of JA for declarative (rather than imperative) purposes is disproportionately affected (Summers & Impey, 2011) and, at an individual level, the extent of impairments in JA have also been found to predict subsequent sociolinguistic development (Charman, 2003; Markus, Mundy, Morales, Delgado, & Yale, 2000; McCathren, Yoder, & Warren, 1999; Tomasello & Todd, 1983) all of which evidence underlines the significance of JA impairment in ASD.

Given that such deficits in social reciprocity are a diagnostic criteria for ASD but not for intellectual impairment (American Psychiatric Association, 2013) there is a limited extent to which the findings of this research are relevant to a population with PIMD. However, some studies have included participants with intellectual disabilities (usually moderate-severe) as control groups so that their findings have contributed to knowledge in this field (see below). Additionally, given the known association between ASD and learning disabilities (O'Brien & Pearson, 2004) it is likely that some individuals with a primary diagnosis of PIMD may also show features of social communication disorders with associated impairments in JA.

#### **4.2.7.2. Joint attention in people with intellectual impairment**

Although less prolific than research in the field of ASD, there is small body of evidence relating to JA in populations with specific conditions including intellectual disability such as Down Syndrome (Mundy et al., 1995) or Angelman syndrome (Summers & Impey, 2011) as well as findings contributed by studies in which children described as having 'developmental delay' (DD) are used as control samples for children with ASD (Dawson, Toth, Abbott, & Osterling, 2004; Mundy et al., 1995; Swettenham et al., 1998). Overall results show a somewhat mixed pattern, possibly due to the diversity in measures and developmental ages of the participant groups used in each study. For example, in a study comparing JA in participants with ASD or DD to a group of typically developing (TD) children matched for a mental age of 26 months, Dawson et al. (2004) concluded that children with DD and TD children showed similar JA abilities. In that study, conclusions about JA were drawn from measures of gaze and point following as well as the alternation of gaze between people and objects during structured activities. In a study of free play with objects Swettenham et al. (1998) also found that TD children and those at risk of DD (at 20 months) showed similar patterns of gaze alternation between objects and people.

In contrast, Mundy et al. (1995) discovered deficits in JA behaviours (including alternating eye gaze between people and object, pointing and following a point) in children with Down Syndrome aged from 12-36 where RJA behaviours were particularly affected and deficits appeared to be associated with mental age. Summers & Impey (2011) studied similar JA behaviours to Mundy et al (1995) in four children with Angelman Syndrome chronologically aged between 5 and 10 years old but functioning within a developmental range of 1:3 and 2:3 on the Mullen Scales of Early Learning (Mullen, 1995). They found impairments in behaviours relating to both IJA and RJA in this group with those relating to IJA being more impaired in this group of children although there was great variability between participants. Since Angelman syndrome is known to share characteristics with ASD and three of these participants had received an ASD diagnosis, the identification of disproportionately affected IJA behaviours in this group might be anticipated.

Overall, existing research findings are somewhat inconclusive regarding the impact of intellectual impairment on JA but suggest that patterns of impairment in JA behaviours may be evident in at least some individuals with learning disabilities.

However, the studies discussed involve many participants whose cognitive functioning would be within the upper range of those categorised as PIMD or higher. For example, Dawson et al. report a range of cognitive functioning between 12 and 46 months in their participants as measured by the Mullen Early Language Scales (Mullen, 1995) and participants in Mundy et al.'s (1995) study demonstrated a mental age of between 16-24 months as measured by the Cattell Infant Intelligence scale (Cattell, 1940). In addition, participants in these studies were not reported to have additional sensory or physical impairments and, thus, represent a somewhat different population to individuals with PIMD. If there is, indeed, an inverse relationship between cognitive level and engagement in JA as suggested by Mundy et al. (1995), individuals with PIMD might be expected to show a relatively greater degree of impairment in JA. In addition, it might be expected that impairments in their physical and visual skills, as described above, could limit their capacity for engaging in behaviours necessary for JA.

#### **4.2.7.3. Joint attention in people with PIMD**

A literature search was carried out using the SCOPUS database to identify any studies focusing specifically on JA skills in a PIMD population. Studies were selected if they included both a specified key word relating to JA and a specified key term relating to profound intellectual disability. Since the JA skills under discussion may be described using a range of labels, the specific search terms used in relation to attention were; joint attention, coordinated attention, shared attention and social attention while the search terms used to isolate studies relating to a PIMD population were; profound disability, profound intellectual disability, profound and multiple learning disabilities (PMLD) and profound intellectual and multiple disabilities (PIMD). A small number of studies containing relevant key words were identified and their reference lists examined to identify any further relevant studies. Through the use of this process, seven studies were identified which included JA as a focus and which included participants who meet the criteria for having PIMD (although this term was not always used to describe them). A summary of the studies may be seen in Table 4.2.



Table 4.2. A summary of studies investigating JA in participants with PIMD

| Study                                   | Participants  | Aspect of JA investigated   | Method  | Key findings   |
|---|---|---|---|--|
| <b>Arens, Cress &amp; Marvin (2005)</b> | <p>25 children(C.A: 9-25 months)</p> <p>Developmental disabilities and at risk of being non-speaking due to cerebral palsy or acquired brain injury.</p> <p>Average score of 6 months on Battelle Developmental Inventory (Newborg, Stock, Wnek, Guidibaldi, &amp; Svinicki, 1984)</p> <p>Vision assessed with the Functional Vision Assessment and the Baby Screen Kit (Vision Associates, 1998)</p> <p>All had minimum functional vision but 28% had visual processing difficulties, 8% acuity difficulties and 16% both.</p> | <p><b>Rater judgement of engagement states</b> defined as:</p> <ul style="list-style-type: none"> <li>• Onlooking</li> <li>• Engagement with objects</li> <li>• Engagement with people</li> <li>• Passive JA</li> <li>• Coordinated JA</li> <li>• Combined JA</li> </ul> <p><b>Passive JA</b> = adult and child looking at same object but child not looking at adult</p> <p><b>Two-point gaze shifts</b> = the presence of object-person or preson-object gaze shifts.</p> <p><b>Coordinated JA</b> = the presence of a 3-point (person-obj-person) gaze shift</p> | <p>One video of unstructured play with parent at home. Playing with favoured items.</p> <p>Average length of video 20 minutes</p> | <p>Greatest proportion of time spent in gaze directed to objects (16.9%) or people (14.9%)</p> <p>4.2% of time spent in passive JA</p> <p>Gaze shifts observed in 0.6% of the interaction time coded.</p> <p>2-point gaze shifts used more frequently than 3-point.</p> <p>Gaze shifting not correlated with developmental skills.</p> |

Table 4.2. A summary of studies investigating JA in participants with PIMD (cont.)

| Study   | Participants   | Aspect of JA investigated   | Method   | Key findings   |
|---|--|---|--|--|
| <b>Cress, Arens and Zazicek (2007)</b>                      | As in Arens, Cress & Marvin (2005), above.   | As in Arens, Cress & Marvin (2005), above.  | <p>Participants also videoed in a 'structured' play session with an examiner. Average length 9.8 minutes.</p> <p>Structured play involved use of more objects and strategies to encourage communication e.g pausing during play routines and following child's lead.</p> | Children spent significantly more time engaging in 'coordinated' and 'combined JA' (as evidenced by gaze shifting) in structured than unstructured play session.   |
| <b>Neerinckx, Vos, Van Den Noortgate, &amp; Maes (2014)</b> | <p>28 dyads of adults diagnosed with PIMD and their support workers.</p> <p>71.4% judged to have visual impairment</p> | <p>Rater judgements of:</p> <ul style="list-style-type: none"> <li>• <b>Divergent attention</b> to person or object</li> <li>• <b>Shared attention</b> both partners looking at each other or to object</li> <li>• <b>Joint attention</b> partners have attention to each other and object</li> </ul> | Twenty-minute session of interaction involving client, support worker and objects (only ten minutes coded)   | <p>Shared attention occurred in 51% of coded intervals., JA in 3.15%</p> <p>Shared attention occurred at least once in 27 dyads.</p> <p>Clients with PIMD shown to initiate JA in 8% of intervals coded.</p> |

Table 4.2. A summary of studies investigating JA in participants with PIMD (cont.)

| Study                            | Participants  | Aspect of JA investigated  | Method   | Key findings  |
|----------------------------------|---|--|----------|---|
| Hostyn, Neerinckx, & Maes (2011) | 17 dyads adults with PIMD and support workers<br><br>52.9% judged to have visual impairment | <b>Attention-directing behaviours</b><br>= eliciting the attention of a social partner to direct their attention to an object or event<br><br><b>Rater judgements of:</b> <ul style="list-style-type: none"> <li>• Divergent attention</li> <li>• Shared attention</li> <li>• Joint attention</li> </ul><br>(as described by Neerinckx, Vos, Van Den Noortgate, & Maes, 2014, above) | As above | Shared attention occurred in 37.94% of coded intervals<br><br>Joint attention occurred in 3.14% of coded intervals. Observed in 8/17 dyads.<br><br>Shared attention and joint attention positively correlated<br><br>Only moderate inter-rater agreement ( $k=0.58$ ) for attention directing behaviours<br><br>Clients used few attention-directing behaviours but wide individual variation |

Table 4.2. A summary of studies investigating JA in participants with PIMD (cont.)

| Study                              | Participants   | Aspect of JA investigated   | Method   | Key findings  |
|------------------------------------|--|---|--|---|
| <b>Neerinckx &amp; Maes (2016)</b> | <p>45 adults with PIMD</p> <p>MA less than 24 month on Uzgiris-Hunt Scales (Uzgiris &amp; Hunt, 1975)</p> <p>Severe neuromotor disability.</p> <p>55% judged to have visual impairment</p> <p>Non-symbolic communication</p> | <p><b>RJA Behaviours</b></p> <ul style="list-style-type: none"> <li>following line of regard or pointing.</li> </ul> <p><b>IJA Behaviours</b></p> <ul style="list-style-type: none"> <li>eye contact</li> <li>alternating eye gaze</li> <li>showing an object</li> <li>pointing to an object</li> <li>other person-related behaviours e.g. body movements</li> </ul> <p>(all IJA behaviours had to be intentionally directed to partner and show persistence)</p> | <p>6 x ten minutes sessions of interaction recorded for each client, each varying in the following characteristics:</p> <ul style="list-style-type: none"> <li>Use of preferred/standardised stimuli</li> <li>Interaction with familiar support worker/unfamiliar researcher</li> <li>Structured/standardised interaction with objects</li> </ul> <p>(Standardised interaction = subtests of the Early Social Communication Scales (Mundy et al., 2003))</p> | <p>Between 5% and 20% of interaction time spent engaging in JA behaviours</p> <p>IJA behaviours more common in structured interaction</p> <p>RJA behaviours more common in spontaneous interaction</p> <p>Familiarity of partner did not affect number of JA behaviours</p> <p>Preferred or standardised stimuli did not affect number of JA behaviours</p> |

Table 4.2. A summary of studies investigating JA in participants with PIMD (cont.)

| Study   | Participants   | Aspect of JA investigated  | Method   | Key findings   |
|---|--|--|--|--|
| <b>Olsson (2005)</b>                                    | <p>9 children (CA: 1-6 years) with 'moderate to profound learning disabilities and additional vision or motor disabilities'</p> <p>Cognitive ability rated using parental questionnaire and classification by Piagetian stage.</p> <p>4 judged to have 'severe disabilities in vision'</p> | Rater judgements of behaviours (intentional or pre-intentional) resulting in joint attention i.e. 'judged to have the same function as a comment | Unstructured play with favourite toy with caregiver but setting manipulated so toy was within or outside reach | <p>54% of behaviours observed were judged to serve the function of JA</p> <p>JA more likely to occur when objects were within reach.</p> <p>Incidences of JA positively correlated with measures of cognition and vision but not motor skills (but individual differences)</p> |
| <b>Sandberg, Ehlers, Hagberg, &amp; Gillberg (2000)</b> | <p>8 individuals with Rett Syndrome</p> <p>(CA: 11-36 years)</p> <p>Functioning at 5-13month level on Vineland Scales.</p>   | <p>Rater judgements of:</p> <ul style="list-style-type: none"> <li>• attempts at joint attention</li> <li>• following a point</li> </ul>         | Videoed in a 'structured situation' on one occasion (no further information provided)                          | JA judged to be demonstrated by 3 participants   |

### **Key findings of studies investigating JA in participants with PIMD**

Overall, as might be expected, findings suggested that 'rich' forms of JA (where there is mutual recognition of a shared focus) occur with fairly low frequency in adults and children with PIMD, with studies by Hostyn et al. (2011) and Neerinckx et al., (2014) observing it in around 3% of the time intervals they coded, and Arens, Cress & Marvin (2005) reporting only 0.6% of interaction time being spent in coordinated JA. Some discrepancy arises in the study reported by Olsson (2005) where 54% of behaviours observed were judged to serve the function of JA, however this is likely to be due to differences in the definition of JA used (see further discussion below).

It is notable, however, that JA was not entirely absent from this population. Despite the low incidence, all studies reported some JA or JA-related behaviour with Hostyn et al. (2011) and Neerinckx et al. (2014) observing such behaviour to occur at least once in around half the client/support worker dyads they observed and some authors commented on the individual differences observed in relation to JA (Hostyn, Neericknx & Maes, 2011; Olsson, 2005).

It was also clear that, whilst episodes of rich JA were rare, episodes of lean JA, in the form of shared focus to a single referent, were more common, although findings did vary somewhat here with Neerinckx et al. (2014) observing 51% of coded intervals to be spent in 'shared attention' whilst Arens, Cress and Marvin observed only 4.2% of interaction time to be spent in what they term 'passive JA'. Whilst sharing attentional focus in this way may not be a rich form of JA in itself, it is significant since it could be a first step towards it, a suggestion made by Hostyn, Neerinckx & Maes (2011) who found that shared attention and JA were correlated and that an episode of shared attention preceded many instances of JA.

Arens, Cress and Marvin (2005) analysed the gaze behaviours which they judged indicative of JA and found that 2-point gaze shifts (from an object to a person or from a person to an object), were used more frequently than 3-point gaze shifts (person-object-person). On the basis of this finding they hypothesized that such 2-point gaze shifts may occur more frequently in

people with physical disabilities since they are less complex to perform but may constitute a precursor to the use of 3-point gaze shifts for JA.

Finally, studies suggested a number of factors which may influence the potential for JA to occur. Structured interaction contexts appeared to lead to an increased incidence of JA (Cress, Arens, & Zajicek, 2007; Neerinckx & Maes, 2016) while the use of preferred stimuli and interaction with a familiar communication partner did not have an effect (Neerinckx & Maes, 2016). Having referents within reach also seemed to lead to increased JA (Olsson, 2005). It was unclear whether JA was associated with developmental level with Arens, Cress and Marvin (2005) reporting a correlation between developmental level and the use of gaze shifts but Olsson (2005) finding no link between the cognitive measures developed for her study and the occurrence of JA although she did find that visual and motor skills correlated positively with engagement in JA.

### **Critical Evaluation of Existing Studies of JA in People with PIMD**

It is positive to see the emergence of research acknowledging the importance of JA in communication development for people with PIMD and confirms that it is an area of individual difference. This lends weight to it being a potential means of differentiation between profiles of communication ability in people with PIMD. However, the studies reported had some limitations which mean that it is difficult to compare their findings and to draw firm conclusions from their results.

Firstly, the criteria used to define JA and the aspects of it which were investigated, differed between studies with evidence of some of the 'dual usage' issues discussed in section 4.2.3. above. The majority of studies focused on observable behaviours which may facilitate JA such as gaze shifting or showing of objects. Only two studies additionally reported on the occurrence of the state of JA (Hostyn, Neerinckx & Maes, 2011, Neerinckx & Maes, 2014). Since JA behaviours and the state of JA may both be of interest, a problem mainly arises when the differentiation between the two is not acknowledged. This can lead to conclusions about engagement in the state of JA being made solely on the basis of observed JA behaviours. For example, (Sandberg et al., 2000) concluded that:

“most of the girls [with Rett syndrome] seemed to function at a pre-intentional level with little evidence of joint attention”

(Sandberg et al., 2000, p264)

This is in spite of the fact that the study investigated participant ability to follow a point and ‘attempts at joint attention behaviours’ (Sandberg et al., 2000, p256) and did not explore actual occurrence of JA episodes.

Similarly, Arens, Cress & Marvin (2005) mix judgements about state with judgements about behaviours in their coding scale (see Table 4.3).

*Table 4.3. Coding categories used by Arens, Cress & Marvin (2005)*

| Coding Category |  |
|-----------------|--|
| 1               | Unengaged  |
| 2               | Onlooking  |
| 3               | With Objects   |
| 4               | With persons   |
| 5               | Passive Joint Attention (adult and child look at same object but child does not look at adult) |
| 6               | Coordinated Joint Attention (3-point gaze shifts)  |
| 7               | Combined Joint attention (2-point and 3-point gaze shifts)                                     |

Here, 2-point and 3-point gaze shifts are used synonymously with states described as coordinated or combined JA. Whilst gaze shifts may indicate that a state of JA is occurring, this is not necessarily the case since gaze shifting may occur for non-social reasons. For example, a gaze shift may occur if the child is looking at an adult who then starts to activate a toy. The movement of the toy may cause the child to look at it and, when it stops, to lose interest and shift their gaze back to the adult. Indeed, whilst alternating



eye gaze or gaze shifting were a key focus of analysis in many of the studies, none differentiated between gaze shifts which were socially or non-socially motivated or between the qualitatively different 'checking' and 'sharing' looks described by (Hobson & Hobson, 2007).

In contrast, Olsson (2005) investigates JA defined solely on the basis of judgements by raters. Here, there was no requirement for participants to produce any active or intentional behaviours leading to JA and this may account for the higher occurrence of JA reported in her study.

A second limitation of existing studies is their failure to describe or control for the exact context in which JA states or behaviours occurred. As discussed in section 4.2.5, aspects of the socio-interactive context including the salience of stimuli and the behaviour of the communication partner play a key role in whether and how JA occurs. Although the broad context of interaction is described by most of the studies (generally interaction with another person and objects) it is unclear exactly what events preceded episodes judged to signify JA. This is particularly relevant in relation to the dichotomy between 2-point and 3-point gaze shifts suggested by Arens, Cress & Marvin. Here it is suggested that 2-point shifts may be a precursor to coordinated JA but do not, in themselves, represent it. However, as outlined in section 4.2.5.1., 2-point gaze shifts may be indicative of JA in 'bottom-up' contexts. Failure to control for the socio-interactive contexts underlying episodes of JA or the performance of JA behaviours contributes to the difficulties in comparing or replicating these studies.

Thirdly, the studies reported have a strong focus on visual or 'looking' skills, perhaps unsurprising given the significance that these have in the early development of JA. However, none of the studies report specifically on participant ability to perform the requisite skills of gaze fixation or gaze shifting. Although many give a broad description of visual skills (with reported visual impairments being common as would be expected in this population) the means used to assess these are either not reported (Hostyn et al., 2011; Neerinckx et al., 2014; Neerinckx & Maes, 2016; Olsson, 2005) or are based mainly on assessment of visual acuity (Arens, Cress, & Marvin,

2005). The potential link between visual abilities in respect of gaze fixation or gaze shifting is not discussed by any of the studies.

Finally, the conclusions drawn by these studies are based on relatively brief periods of interaction. None of the studies recorded interaction periods of more than twenty minutes, a fact which was sometimes justified by the characteristics of the participants. For example, Neerinx et al. suggest that ten minutes of engagement is 'a considerable length of time for most people with PIMD' (Neerinx et al., 2014, p724). However, it is also known that people with PIMD produce low frequencies of communicative behaviours (Cirrin & Rowland, 1985) and that their engagement can be affected by aspects such as fluctuating arousal levels, discomfort or seizures . This raises doubt about conclusions drawn on the basis of single, short periods of interaction. Increased accuracy might be obtained by repeating measurements several times and on separate occasions. Although one study (Neerinx and Maes, 2016) did base conclusions on more than one episode of interaction, each of the episodes was conducted under differing conditions. It is disappointing that studies focusing on participants with PIMD should fail to take such key aspects of their functioning into account.

#### **4.2.8. Conclusions**

In summary, recent decades of theoretical discussion and research findings have established JA as a critical aspect of communication development which evolves during the first months of life and is inextricably linked with the development of intentional communication. This provides a strong argument for its being considered as a significant area for assessment in prelinguistic communicators with PIMD who are believed to function, developmentally, within this phase. Assessing the extent to which an individual demonstrates behaviours on a trajectory or continuum of typical development would indicate their current capacity for engaging in episodes of JA which form a basis for intentional communication. Such an assessment could also be used to inform suitable approaches for support and intervention. Developing our understanding of how JA develops within this population would also enhance our knowledge of prelinguistic communication within this group and how it

may resemble or differ from that observed in the typically developing population. At the present time it may be concluded that;

“our knowledge about JA behaviours used by persons with PIMD in interactions remains limited and incomplete”

Neerinckx & Maes (2016, p 575)

Emerging research in the field has provided some useful information about contexts which may facilitate the occurrence of JA and initial evidence supporting a potential continuum of skills underpinning the occurrence of lean and then rich JA in typically developing infants. However, these studies have certain limitations. In particular they often fail to differentiate between behaviours facilitating JA and the state of JA itself, they do not describe or control for critical aspects of the socio-interactive context, they have not assessed pre-requisite functional vision skills and are based on brief, single episodes of interaction.

Therefore, the aim of this thesis was to contribute to our understanding of JA in people with PIMD through conducting some research into the JA behaviours they demonstrate, while avoiding some of the issues inherent in previous studies. By using structured sampling methods to measure these behaviours it was anticipated that findings would not only inform our understanding of JA in people with PIMD but could also be used to address some of the issues in current communication assessment approaches for people with PIMD.

#### **4.2.9. The proposed research**

The research carried out for this thesis aimed to answer the following research questions:

- 1) What patterns of JA behaviour are demonstrated by young people with PIMD?
- 2) Are these patterns of behaviour affected by the functional ability to fix and shift gaze?
- 3) Are structured probes an effective means of eliciting information about JA behaviours?

These questions were addressed through using structured probes to measure the JA behaviours demonstrated by a sample of young people with PIMD. The use of structured probes would enable the socio-interactive context to be controlled and would provide multiple opportunities for participants to demonstrate target behaviours. The structured probes would be administered on several occasions to address fluctuations in performance and both observable behaviours and the occurrence of the state of JA would be taken into consideration. The functional ability to fix and shift gaze would also be measured to explore the relationship between these skills and JA behaviours and additional background measures of cognition, communication and motor skills would be used to investigate whether abilities in these areas were predictive of performance on the structured probes.

The following chapter considers a range of methodological issues which were relevant to the design of this study including an account of structured sampling approaches and general methodological factors which need to be taken into account when carrying out research with people who have PIMD.

## 5. Methodological Issues

Although the numbers of people with PIMD are increasing (Emerson, 2009) the amount of research carried out with this population is relatively small and this is due, in part, to the methodological challenges they present to the researcher (Porter & Lacey, 2005). In this chapter I will consider a range of methodological issues in relation to a PIMD population, beginning with a discussion of structured sampling methods. Having presented evidence that such methods are underused with people with PIMD in clinical settings despite their potential benefits, I will go on to describe how they have been used in other relevant contexts such as the clinical assessment of pre-linguistic infants and children at risk of ASD and as part of research studies with people with PIMD. I will evaluate the existing methods used in these contexts, considering the extent to which they might inform development of a set of structured probes for eliciting JA behaviours in the current study.

Subsequently, I review broader methodological issues relating to research involving people with PIMD including challenges raised by the low incidence and heterogeneity of this population, and the difficulty in establishing cognitive/developmental level. Methodological decisions taken in the design of the current study will then be outlined with reference to these issues.

### 5.1. Current use of direct and standardised methods in the assessment of people with PIMD

Effective assessment is the bedrock of effective intervention in speech, language and communication disorders (Brinton & Fujiki, 2010). As discussed in section 3.4, this can involve multiple means of information gathering and it can be argued that robust assessment should be based on information taken from a range of sources. Brady and Halle (1997) propose that thorough assessment should consider an individual's functioning in natural contexts but should also include measures which can be reliably repeated over time. On this basis they recommend using the following three sources of information; observation, interview with family or caregivers and the use of 'analog probes' which they define as:

“systematic environmental manipulations, structured protocols, contrived communicative situations and simulations”

(Brady and Halle, 1997, p 96)

The findings reported in Chapter 3 suggested that, whilst SLTs currently make use of observation and interview data as part of their communication assessment of people with PIMD, they rarely implement such 'analog' or 'structured' probes (as they will be termed throughout this thesis). Some SLTs did report using direct methods for assessing clients which might signify the use of personally devised structured probes, for example, one reported “setting tasks in natural settings”. However, the precise format of these was unclear, likely varying across clinicians. In addition, none of the published assessment frameworks included the use of structured probes. As discussed, such a lack of shared approach means that measures cannot be reliably repeated over time, particularly where there are changes in clinician.

Structured probes have a number of advantages meaning they could make an effective contribution to communication assessment for people with PIMD. Firstly, they can be used to target specific skills which are of interest to the assessor. Secondly, by providing an environment specially designed to elicit these target skills, they offer multiple opportunities for them to occur

– particularly beneficial for a population whose rate of communicative opportunities in natural contexts may be low (Cirrin & Rowland, 1985). Thirdly, by providing a controlled context, they produce information about which aspects of the environment or types of scaffolding assist the individual to demonstrate target skills, thus providing a measure of their capacity to perform the skill even where this is not evidenced in natural contexts. Finally, they provide a clear and consistent procedure which can be replicated over time and by multiple SLTs ensuring that changes in performance reflect the individual's skills and not changes in assessment method.

The aim of the current study was to explore participants' capacity for engaging in JA behaviours. It was clear that, given their benefits, structured probes could provide an effective means of achieving this aim.

Although there is currently no clinical tool which uses structured probes to elicit JA behaviours in people with PIMD, such tools have been used in other relevant contexts. These include research studies where structured probes have been developed to measure a range of pre-linguistic skills in participants with PIMD and clinical contexts where they have been used to assess pre-linguistic skills in infants and early social communication skills in children at risk of ASD. An appraisal of the structured probes used in these contexts will now follow, alongside a discussion of whether and how they might inform the design of structured probes to be used in this study.

#### **5.1.1. The use of structured probes in relevant clinical contexts**

Two widely used clinical tools based on structured probes which will be considered here are the Communication and Symbolic Behaviours Scales (CSBS) (Prizant & Wetherby, 2003) and the Autism Diagnostic Observation Schedule (ADOS) (Lord et al., 2012).

The CSBS is described as an “informal procedure for sampling communication with preverbal children” (Prizant & Wetherby, 2003, p 6). This assessment tool is designed for use with children aged, developmentally, from 8-24 months, aiming to assess prelinguistic skills known to be associated with later communication and language development including intentional requesting (which is likely to include JA) and

engagement in joint attention, defined as drawing an adult's attention to a referent for social purposes. Information for the assessment is gathered both from a caregiver questionnaire and from a period of direct engagement with the child lasting up to one hour during which structured probes are administered in a standardised manner with guidance provided for the prompts and verbal behaviours which the adult might use. Activities include sharing a book, symbolic play and the use of eight 'communicative temptations' which are designed to provide multiple opportunities for the child to communicate for different functions and display target behaviours including gaze shifts and the sharing of positive affect. Examples of some of the communicative temptations are presented in Table 5.1.

*Table 5.1. Three examples of communicative temptations used in the CSBS*

| <b>Communicative Temptation</b> | <b>Description</b>   |
|---------------------------------|--|
| <b>Wind-up toy</b>              | Tester activates toy and allows it to deactivate. Allows child to pick it up or hand it to them, waiting for a signal to help re-start it.   |
| <b>Bubbles</b>                  | <p>Tester blows bubbles, catches on wand and encourages child to pop it.</p> <p>Closes jar and gives it to child to elicit communication for 'help'.</p> <p>Tester repeats activity multiple times, providing different cues each time e.g. holding wand up to mouth and waiting with an expectant look.</p> |
| <b>Balloons</b>                 | <p>Tester blows balloon up and lets air out gradually.</p> <p>Tester gives balloon to child and waits for signal.</p> <p>Tester repeats activity multiple times, providing different cues each time e.g. pausing during blowing up of balloon and giving child an expectant look.</p>                        |



The CSBS is of interest since it has been shown to be a reliable and valid clinical tool which aims to assess a developmental phase of communication relevant to some individuals with PIMD (Wetherby, Allen, Cleary, Kublin, & Goldstein, 2002). However, aspects of its protocol mean that its structured probes are not directly applicable to this population. Taking up to one hour to administer, it is likely to prove too lengthy for individuals with varying levels of engagement. In addition, it relies heavily on motor skills with children encouraged to blow bubbles, pick up and give objects. An attempt to use the communicative temptations from the CSBS with a group of children with PIMD, (Iacono, Carter, & Hook, 1998) concluded that the activities were not sufficiently engaging for this group, tending to be completed before participants showed any level of response.

The ADOS, while less relevant to a population with PIMD, is also of interest due to its success in the clinical field. It uses a series of structured probes to elicit social behaviours from children and adolescents who are potentially showing signs of ASD. For example, for one probe, the assessor will pause in the process of blowing up a balloon to see if the child will demonstrate requesting behaviours. Administered in conjunction with an interview schedule for parents (Rutter, Le Couteur, & Angeles, 1994), ADOS is widely used and has come to be considered the “gold standard” in methods for diagnosing autism (Akshoomoff, Corsello, & Schmidt, 2006, p689). Since it is designed for use with verbal individuals who have ASD, the ADOS procedures cannot be directly applied to people with PIMD. However, its contribution to assessment in the field of ASD provides a good example of how structured probes can be used to form an effective and widely shared assessment tool.

#### **5.1.2. The use of structured probes with people with PIMD in research studies**

Structured probes have been used to measure communication in participants with PIMD as part of research studies carried out by Cirrin and Rowland (1985), Iacono et al. (1998), McLean, McLean, Brady, and Etter (1991) and McLean and Snyder-McLean (1987). All of these studies report using standardised activities to elicit samples of intentionally communicative

acts in participants described as having severe or profound and multiple intellectual disabilities. The specific nature of the activities, or probes reported in this work, is of particular relevance to the current study and will now be evaluated.

Each of the aforementioned studies used a unique set of structured probes for eliciting intentional communication and all reported some success, particularly in eliciting communication for the imperative functions of requesting or rejecting. Communication for declarative functions was reported less frequently in all of these studies and this was interpreted as reflecting characteristics of the participants rather than issues with experimental methods (e.g. Cirrin & Rowland, 1985). However, closer examination of the methods used suggests that many studies used probes which were more likely to elicit imperative than declarative communication. For instance, probes frequently involved pausing during established routines or setting up situations in which participants needed to obtain help from the assessor, creating contexts in which any communicative signals were more likely to be construed as requests rather than commenting or sharing affect. This is reflected in examples of four of the seven probes used by Iacono et al. (1998) presented in Table 5.2 below.

*Table 5.2. Examples of four structured probes devised by Iacono et al., 1988*

(Reproduced from Iacono et al., 1988, p106)

| Situation/Probe    | Procedure   |
|--------------------|---|
| <b>Balloon</b>     | A balloon is slowly inflated, then deflated with the air directed onto the student's hand and then face. The activity is frequently interrupted (e.g. holding the inflated balloon close to the student's face, holding the deflated balloon in front of the student, ensuring she can see it). |
| <b>Social Game</b> | Involve student in song – switch activation of a tape recorder. The switch is initially placed within the student's reach with turns taken in activating and deactivating the tape recorder. The switch is then placed out of reach.  |
| <b>Book</b>        | The student is engaged in a book-reading activity with a preferred book. The teacher pauses frequently, waiting for the student to initiate turning of the page or some communicative behaviour.  |
| <b>Textures</b>    | The student's cheek is stroked with cotton wool. This is repeated with a cool, sticky substance. Both items are then held in front of the student.  |

In contrast, in their study, McLean and colleagues (1991) did include some structured probes which might be expected to elicit declarative communication since these were designed to “evoke a request for the interactor's attention to some unusual event or object” (McLean et al., 1991, p298) (see Table 5.3.. for examples).

Table 5.3. Examples of three structured probes devised by McLean et al., 1991

(Description reproduced from McLean et al., p307)

| Situation/Probe                 | Procedure  |
|---------------------------------|--|
| <b>Surprise Marker in a Box</b> | <p>E* puts a piece of paper in front of himself and one in front of S. E brings out a box of markers, takes one marker to draw with, and offers the box to S. S opens the box and takes out a marker. After drawing for about a minute, E says "I'm going to get a new color." E secretly places a plastic spider in the marker box, takes a new marker, and offers the box to S. S opens the box. If, after 5 seconds, S doesn't show E the spider, E will prompt:</p> <p>Level 1 = Takes box back, looks inside and gasps, then passes the box back to S.</p> <p>Level 2 = "Look at that" + points to spider</p> |
| <b>Moving Hand</b>              | <p>A rubber mechanical hand, covered by a cloth drape, is located on a shelf behind E's seat where it can be seen by S. During the course of interaction, E presses a hidden switch activating hand movement. E waits 5 s after S has noticed this movement. If S does not respond within 5 s, E prompts:</p> <p>Level 1 = "Do you see something?"</p> <p>Level 2 = E looks over shoulder, points, and says "Wow, look at that"</p>  |
| <b>Dangling Alligator</b>       | <p>At some point during the sample, a plastic alligator attached to an "invisible" fishline is dangled from the ceiling behind E's head. E waits 5 s after S has seen the alligator. If S does not respond within 5 s, E prompts:</p> <p>Level 1 = "Do you see something?"</p> <p>Level 2 = E turns, looks toward alligator, points and says, "Wow, look at that!"</p>   |

\*E = Experimenter, S = Subject

These probes are of particular interest in terms of the socio-interactive contexts they create (see section 4.2.5.). All three of the probes described in Table 5.3. create 'top-down' contexts in which JA may occur as part of declarative communication. In such contexts, the referent is only salient to the participant as it is out of the assessor's sight so the participant must direct the assessor's attention to it if JA is to occur. In contrast, probes such as the wind-up toys (used in the CSBS) and book sharing (used in the CSBS and by Iacono, Carter & Hook) all create 'bottom-up' contexts where the referent is visible and salient to both participant and assessor at the same time so that, potentially, JA can occur through a mutual sharing look without the need for the participant to draw the assessor's attention to the referent. None of the studies discussed explicitly differentiated between, 'top-down' and 'bottom-up' contexts or the potentially different communicative behaviours they might elicit, representing shortcomings in their methodology. However, the fact that the resulting range of probes they created elicited both 'top-down' and 'bottom-up' JA provides a source of information for the current study.

A potential risk of using structured probes with this population is that a single administration will not reflect participants' full capability, given the potential fluctuations in performance resulting from variable arousal levels and physical health (Guess, Roberts, & Rues, 2002). All of the studies discussed here showed some acknowledgement of this issue although they still only administered their probes on a maximum of two occasions. Data compared between both administrations of the probes in these studies indicated that variations in performance, were, indeed, observed (e.g. Iacono et al., 1998), highlighting the need to administer such structured sampling on more than one occasion in order to draw meaningful conclusions about an individual's ability.

The research studies considered here all demonstrated that structured probes can be used to sample the communicative behaviours of people with PIMD, with several authors commenting that they provided an effective means of observing a range of communicative acts in a population who are generally considered difficult to test (e.g. Cirrin & Rowland, 1985; McLean et al., 1991). However, it was notable that none of these studies compared the

data they obtained using the probes with that obtained from less structured contexts as caregiver interview or observation in natural contexts, meaning it is difficult to judge the extent to which behaviours elicited by the probes reflected 'everyday' performance. This provided a motivation for the current study to compare data about target behaviours from the use of both structured probes and less structured means.

The aim of the current study was to use structured probes to elicit JA behaviours. Although none of the studies considered here overtly focused on JA, all specified an element of coordinated attention as a criteria for the intentional communication they were aiming to elicit with, for example, Cirrin and Rowland only scoring behaviours as intentional communication if they included "alternation of gaze between listener and referent (joint focus)" (Cirrin & Rowland, 1985, p 54) and McLean et al. defining intentionally communicative acts as being:

"A motor or vocal act emitted in a state in which evidence of attention to another person is provided by eye gaze, body orientation, or directionality of motor/vocal act"

(McLean et al., 1991, p299)

It is clear, therefore, that many of the probes involved in these studies indirectly aimed to elicit JA behaviours in people with PIMD, further highlighting their relevance to the design of probes for the current study. However, it should be noted that none aimed to elicit behaviours indicative of responding to JA (RJA). There is one further set of probes which includes a specific focus on early JA behaviours including RJA. This set of probes, the Early Social Communication Scales, will now be evaluated.

### **5.1.3. The Early Social Communication Scales (ESCS).**

The Early Social Communication Scales (ESCS) are a measure designed to assess non-verbal social communication skills in children functioning within a developmental age range of 8 to 30 months) (Mundy et al., 2003; Seibert, Hogan, & Mundy, 1982). The ESCS gathers information using a set of structured probes which aim to elicit behavioural requesting, social interaction and joint attention and which are administered alongside a

questionnaire for caregivers. Although designed as a clinical tool, the ESCS would appear to have been used mainly within research contexts to date, particularly in the field of ASD (e.g. Bruinsma, Koegel, & Koegel, 2004). It was of particular relevance to the current study for two reasons.

Firstly, being designed as a clinical tool, the probes used in ESCS are accompanied by a manual describing the procedures and prompts used to administer them, a level of detail not provided by the probes reported in the research studies above. They have also been subject to testing for reliability and validity, again, unlike the probes used in the research studies (Mundy et al., 2003).

Secondly, the ESCS is especially designed to target behaviours relating to the typical development of JA covering a developmental range of 8-30 months which, at least partially, matches that of people with PIMD. Given the aims of the current study it was clearly relevant to evaluate the approach taken by the ESCS in some detail.

### **Structured Probes Used by the ESCS**

A set of nine probes is implemented by the ESCS, with three aiming to elicit what are described as 'JA behaviours' and one targeting 'behavioural requesting' which also involves engaging in JA, albeit in a lean form, since the attention of another person is directed to an object for the purposes of requesting it. A description of these four probes is given in Table 5.4. It should be noted that the Gaze Following probe requires four large, colourful posters to be hung on the walls. Two of the posters are hung to the left and right side of the child so that they are within their view and two further posters are hung behind and to either side of the child.

*Table 5.4. Structured Probes used by the ESCS for eliciting Joint Attention and Behavioural Requesting*

| Probe                   | Description   | Target behaviours   |
|-------------------------|---|---|
| <b>Object Spectacle</b> | <p>Three wind-up mechanical toys and three hand-held mechanical toys (balloon, squeeze toy, cone toy, bellows toy) are presented. In each presentation the tester activates the toy on the table in front of, but out of reach of the child.</p> <p>Toys should be wound up enough to remain active for at least 6 seconds but not so long that the child loses interest. The tester remains silent but attentive to the child while the toy is active to allow the child to initiate joint attention bids.</p> <p>Each item is presented three times in a row.</p> | <p>Initiating JA</p> <p>Initiating Behavioural Requests</p> |
| <b>Gaze Following</b>   | <p>Trials begin with the tester bringing the child's attention to her face.</p> <p>Tester looks and points to the posters on the wall while emphatically stating the child's name.</p> <p>Tester attempts to direct the child's attention to each poster in the following order: to the tester's Left, Left-Behind, Right, Right Behind. On all trials the tester obtains the child's attention, then turns her entire torso (not just her head)</p> <p>The Gaze Following task is administered twice during the session</p>  | <p>Responding to JA</p>                                     |



*Table 5.4. Structured probes used by the ESCS for eliciting Joint Attention and Behavioural Requesting (cont.)*

| <b>Probe</b>             | <b>Description</b>   | <b>Target behaviours</b>              |
|--------------------------|--|---------------------------------------|
| <b>Book Presentation</b> | A picture book is opened and presented on the table within the child's reach. Several distinct pictures should be displayed on the pages of the book. The tester asks, "What do you see?" The tester should allow the child to examine the book for approximately 20 seconds. If the child points to pictures spontaneously the tester should respond briefly, but naturally (e.g., "Yes, I see"). After a 20 second interval the tester should begin pointing to pictures in the book regardless of whether the child has pointed or not. | Initiating JA<br><br>Responding to JA |
| <b>Plastic Jar Task</b>  | Two wind-up toys are placed in a plastic jar in front of the child. The tester then gives the jar to the child and waits for approximately 10 seconds, or until the child gives the jar back to the tester. After one is taken out and played with the jar is returned to the child to request the second one.   | Initiating behavioural requests       |

### **Joint Attention Behaviours Measured by the ESCS**

Administration of the ESCS probes is video recorded. Each recording is analysed, and the frequency of all observed target behaviours is counted. The descriptors for target behaviours relating to initiating and responding to JA and to behavioural requesting are summarised in Table 5.5.

*Table 5.5. Descriptors for ESCS Target behaviours relating to Joint Attention and Behavioural Requesting. (Descriptors reproduced from Mundy et al., 2003)*

|                                    | <b>Initiating Joint Attention</b>   |
|------------------------------------|---|
| <b>1. Eye Contact</b>              | Child makes eye contact with the tester while manipulating or touching an inactive mechanical toy. Child must be touching the object.   |
| <b>2. Alternate [eye gaze]</b>     | Child alternates looking between an active object spectacle and the tester's eyes.  |
| <b>3. Point</b>                    | With a clear articulation of the index finger child points to an active toy, to pictures in the book, to wall posters or to any other unobtainable object or event  |
| <b>4. Show</b>                     | Child raises a toy upward toward the tester's face while looking at the tester.   |
|                                    | <b>Responding to Joint Attention</b>  |
| <b>1. Following Proximal Point</b> | During Book Presentation task, the tester points to 6 pictures in the book. The child gets credit if he/she clearly follows the tester's pointing gesture by immediately turning his/her head and eyes to the appropriate area of the book.                     |
| <b>2. Following Line of Regard</b> | On Left and Right look trials the child gets credit if he/she turns his/her eyes or head sufficiently to indicate that he/she is looking in the correct direction and beyond the end of the index finger of the tester, approximately 45-90 degrees off midline |
|                                    | <b>Initiating Behavioural Requests</b>  |
| <b>1. Eye Contact</b>              | Child makes eye contact with the tester when an object is inactive. The child must not be touching the object during the eye contact to receive this code.  |
| <b>2. Reach</b>                    | Child extends his/her arm toward an out of reach object   |
| <b>3. Appeal</b>                   | Child combines eye contact with the tester with reaching  |

Evaluation of the ESCS indicates several positive factors which could be used to inform the design of structured probes for the current study. Firstly, it is the only set of probes currently available which aims to elicit both initiating and responding to JA as well as differentiating between the use of JA for both imperative and declarative functions. It focuses on observable behaviours, providing clear descriptors for these and categorising them into different levels of complexity – eye contact and alternate gaze being described as ‘lower level’ behaviours and ‘pointing’ and ‘showing’ being described as ‘higher level’ behaviours. It has also been widely used and tested for reliability and validity (although not with people with PIMD).

Secondly, the ESCS procedure provides an example of how the socio-interactive context might be controlled, both in terms of assessor behaviour and aspects of the physical environment. For example, a score for ‘eye contact’ can only be given if this was motivated by the child and not if “the child’s behaviour may have been elicited by the tester’s movement or talking” (Mundy et al., 2003, p16). Specific guidance on the use of speech is provided for the tester as follows:

“During ESCS administration, the tester performs a variety of tasks with natural but minimized verbal interaction with the child. A reduction in tester verbalization allows for clearer differentiation of communicative bids that are initiated by the child. The tester should feel free to speak to and interact with the child during transitions in the testing procedure (e.g., while activating an object, while selecting a new object) but should keep verbal interaction to a minimum during actual task administration.”

(Mundy et al., 2003, p4)

Such guidance demonstrates how a balance may be struck between controlling the environment and allowing a degree of flexibility which can increase ecological validity and sustains the engagement of the child. For example, if the child points to pictures in the book the adult is encouraged to respond “briefly but naturally” (Mundy et al., 2003, p11). The order of presentation and duration of activities is also flexible depending on the engagement of the child.

Finally, the procedure used to implement the ESCS is efficient at providing multiple opportunities for target behaviours to occur. The ‘object spectacle probe’ is particularly strong in this regard since a number of stimuli are presented on three occasions each. This probe may also elicit JA for two different functions within one

activity, with JA behaviours occurring during activation of the toy being interpreted as initiating JA for social purposes and those occurring once the toy has finished being interpreted as requesting.

Despite its advantages, the ESCS has certain limitations which meant it was not directly suitable as a measure for this study in its current form. Most notably, both the probes and the target behaviours described in the ESCS assume a certain level of motor skill and visual ability. For example, the plastic jar probe requires that the jar be given to the child. The wind-up toys are small, the book involves multiple 2D images on a page and the posters are hung on the wall at a distance of at least two feet beyond the tester's extended arm and pointing index finger (Mundy et al, 2003, p3) all of which may make these probes challenging for individuals known to experience physical disabilities and visual impairment. The higher level target behaviours for initiating JA and behavioural requesting also involve physical dexterity in the form of pointing and reaching.

A further limitation of the ESCS is that, as discussed in relation to the research studies outlined above, it does not differentiate between 'top-down' and 'bottom-up' contexts with all probes creating 'bottom-up' contexts in which referents are simultaneously visible and salient to both tester and child.

Finally, the Gaze Following probe designed to elicit RJA behaviours in the ESCS has practical implications which limited the feasibility of its administration. In order for the probe to be carried out, posters need to be hung in specific locations and at specific angles in relation to the child. This is difficult, if not impossible, to achieve, when assessments are carried out in a variety of rooms which are not familiar to the tester since rooms may not have suitable walls on which to hang posters and, if they do, walls (and, therefore, posters) will be at varying distances from the participant, possibly affecting performance. This would be the case in the current study since participants were to be seen in rooms provided at their schools.

In summary, although the ESCS has limitations, meaning that it could not be used as a suitable measurement for the current study it does have many positive features which were used to inform the methodological design, including some aspects of the structured probes and the methods used to score them.

#### **5.1.4. Conclusions relating to the use structured sampling to measure pre-linguistic skills in people with PIMD**

This discussion and evaluation of structured probes has demonstrated how they have been used successfully to elicit and measure aspects of pre-linguistic communication, including behaviours relating to JA, in both clinical and research contexts. Although none of the existing tools using structured probes were directly suitable as measures of JA for the current study, it was possible to use their protocols and scoring systems as a foundation for developing a suitable measure.

## 5.2. Methodological challenges raised by research with people who have PIMD

Certain features of the PIMD population can raise challenges for traditional research methods and needed to be taken into consideration when designing the current study. Some of these challenges are outlined below alongside strategies which might be used to address them. This is followed by a discussion of how these strategies were adopted when making methodological decisions for the current piece of research.

### 5.2.1. Heterogeneity and low Incidence

Although people with PIMD are unified by the descriptors of profound intellectual impairment ability and multiple additional disabilities described in Chapter 2, members of this group demonstrate “hugely varied” profiles of ability in all areas (Porter, 2015, p 400) since their functioning is affected by so many cognitive, physical, sensory and medical factors.

From a research perspective, this degree of heterogeneity has a number of implications. Firstly, it affects the extent to which findings about a single case study or a sample group can be generalised to the whole population of people who have PIMD. Basing findings solely on quantitative analysis at a group level is particularly unreliable since results can be skewed by large intra-group variation as well as masking variability in performance between individuals (Porter & Lacey, 2005). This has potential implications for any practical outcomes resulting from research since, for example, a teaching strategy which yields positive results at a group level, may not be of benefit to all members of the group.

Taking a three-tier approach to data analysis as proposed by Light (1988) goes some way to addressing these issues. Under Light’s framework, data is analysed at molar, intermediate and molecular levels. At the molar level quantitative methods such as statistical analysis are used to explore relationships between independent and dependent variables and group patterns are identified. At the intermediate level raw data is analysed to investigate the frequency or proportion of participants whose results are congruent with or different from group patterns to evaluate its strength and the molecular level of analysis focuses on those individuals whose performance is typical or atypical in relation to the group patterns.

Secondly, the heterogeneity of this population means that experimental studies using matched groups to compare the effects of different independent variables are rarely feasible given the range of participant characteristics which need to be considered and the difficulty in measuring these with accuracy. Equally, there is no clear control group for people with PIMD. Although young typically developing children may be broadly matched with them in terms of cognitive age, they will have a very different physical and sensory profile as well as lacking the years of life experience which a chronologically older person with PIMD will have.

A further challenge to research is the low incidence of this group since, despite their growing numbers, people with PIMD represent a relatively small minority of the population (see section 2.4.). This means that large-scale recruitment of participants is difficult, resulting in much research in the field being small-scale (Porter & Lacey, 2005). Traditional research methods favour the use of large-scale studies since the larger and more representative a sample, the more likely that its results can be reliably generalised to the rest of the population (Robson & McCartan, 2015). Gersten, Baker, & Lloyd (2000) propose that, while small-scale studies are inevitable in the field of PIMD research, the issue can be addressed by synthesizing findings of a number of small-scale studies so that an overall contribution to knowledge can be made. In order to achieve this, they emphasize that procedures and participant characteristics for studies must be thoroughly described to enable replication and synthesis of findings.

A systematic approach to describing participant characteristics has been developed by Pennington, Marshall, & Goldbart (2007). Although designed to facilitate the synthesis of findings across small-scale studies in the field of Augmentative and Alternative Communication (AAC) this approach is also relevant to participants who have PIMD since AAC users may similarly demonstrate a range of motor, cognitive and sensory impairments.

The guidelines developed by Pennington and colleagues recommend that the following participant characteristics should be described; age; gender; medical diagnosis; vision; hearing; gross motor function (including mobility); fine motor function; cognitive and communication function. They also recommend that speech, literacy, memory and socio-economic status should be reported, but these are,



perhaps, less applicable to the population of children with PIMD being either absent, of little relevance or difficult to measure accurately.

### **5.2.2. Classifying cognitive functioning**

While the cognitive function of research participants with PIMD should, ideally, be described, classifying cognitive abilities in this population is notoriously difficult (see section 2.2.). The fact that people with PIMD are nonverbal and may give limited behavioural responses due to their physical and sensory deficits means that even informal methods approaches to estimating developmental age lack accuracy (DeVeney et al., 2012). Indeed, arguably, the careful assessment of joint attention abilities under exploration in the current study may provide a more robust, sensitive and measurable insight into cognitive functioning than existing measures. However, for the purposes of the current study, consideration would need to be given as to how cognitive functioning should best be classified.

### **5.2.3. Fluctuating performance**

Factors which affect the health and wellbeing of an individual with PIMD can lead to fluctuations in their level of alertness and ability to engage with the environment at any given time (see section 2.2.). Such factors include the effects of seizures, fatigue or discomfort caused by muscle spasms or digestive issues. Fluctuation in performance has implications for study designs where protocols are administered on a single occasion since such ‘snapshots’ may not reflect an individual’s full capacity and the reliability of findings from such studies may be called into question. Repeated administration of measures may help address this issue.

### **5.2.4. Ethical issues**

Over recent years efforts have been made to include people with learning disabilities in decision-making about research which concerns them, for example Mencap has recruited ‘research champions’ who have learning disabilities themselves (Mencap, 2019). However, there is a limit to the extent to which people with PIMD can be included in such developments since, whilst they are able to react to aspects of the immediate environment, they are unable to express a more complex ‘view’ about what types of research are valuable and whether they would like to take part (Ware, 2004). These limitations have raised questions about whether it is ethical to include them in research projects of which they have limited understanding (Mietola et al., 2017). In

response to this it has been argued that ceasing to include people with PIMD in research would be unethical in itself since it would prevent advances in understanding which could be of benefit to them (Tuffrey-Wijne, Bernal, & Hollins, 2008) but, to ensure that they have an ethical foundation, studies should aim to produce findings which have benefits for participants and should make these potential benefits clear (Mietola et al., 2017).

### **5.3. Methodological decision making for the current study**

Given the methodological issues described above, the following decisions were made concerning the research measures and procedures for the current study.

#### **5.3.1. The measurement of joint attention**

It was decided that JA behaviours would be examined through the use of structured probes, using methods developed from existing protocols as appropriate. The selected target behaviours would be based on the typical trajectory of JA development outlined in section 4.2.4., including behaviours which are pre-requisites to JA (attending separately to people and objects) as well as those which might indicate the occurrence of lean and rich forms of JA. The structured probes would aim to elicit both RJA and IJA behaviours including imperative and declarative JA. Tester behaviour would be controlled and both top-down and bottom-up contexts would be created.

#### **5.3.2. The measurement of functional vision**

A measurement of functional vision (specifically gaze fixation and gaze shifting) was required to answer the research questions for this study. Since there are no existing tools focusing on the measurement of these skills in people with PIMD, a decision was taken to trial a recently developed tool, the Rapid Assessment of Functional Vision or FunVis (Clarke et al., 2019). This measure is designed to test functional vision in the form of gaze fixation and gaze shifting in children with cerebral palsy aged, developmentally, from 9 months to 6 years. Although it was possible that some participants in the current study might be functioning outside this suggested developmental age range for this test, the lack of potential alternatives and the fact that it is suitable for individuals with complex physical impairments meant that the FunVis was considered a suitable option to trial (see Appendix C for FunVis target behaviours and procedure).

#### **5.3.3. The selection of participants**

Convenience sampling, in which participants are recruited on the basis of their accessibility is particularly suitable for low incidence populations (Polgar & Thomas, 2013) and was, therefore, selected as a sampling technique for this study. In terms of the age range for participants it was decided that the focus should be on young people rather than adults. This decision was taken since the aim was to improve

understanding about JA behaviours and the methods which might be used to assess them, factors which might improve approaches to intervention for this population . Since children and young people with PIMD are more likely to receive SLT intervention than adults (Mansell, 2010) it was reasoned that the study findings would be of most relevance to them.

Applying a convenience sampling approach to young people meant that participants would be most effectively accessed through special schools where the majority of children with PIMD in the UK are educated. Given the low incidence of population it was decided not to limit the potential for recruitment by specifying any particular age range for the children so that any school-aged child could be included. .

#### **5.3.4. The selection of appropriate measures of participant characteristics**

Following Gersten and colleagues' (2000) guidelines and drawing on recommendations by Pennington and colleagues (2007) core characteristics of the participants were measured and reported. Age, gender, and medical diagnosis were ascertained through liaison with teaching staff who were also asked whether participants were known to have issues with their hearing or vision.

##### **Motor function**

The Gross Motor Classification Scale (GMFCS) (Palisano, Rosenbaum, Bartlett, & Livingston, 2008) was used to classify the gross motor functioning of participants while fine motor function was classified using the Manual Ability Classification System (MACS) (Eliasson et al., 2006). Both of these tools are established and validated measures for children with motor impairments (cerebral palsy).

##### **Cognitive function**

As noted, accurate assessment of cognition in young people with PIMD is challenging. Initially, subscales of the Mullen Scales of Early Learning (Mullen, 1995), were considered as a potentially useful tool to assess cognitive ability. Early exploratory work however, identified that it did not offer an informative insight into participants' cognitive abilities, providing little differentiation at a developmental level of less than 12 months and including some items which were too dependent on physical abilities to be used with this population e.g. pulling on a cord to obtain a disc which is out of reach on the table (Mullen, 1995, p22). An alternative was, therefore, sought.

Since participants were being sampled from an educational setting it was decided that the P-Scales for English (Department for Education, 2014) might provide a suitable means of estimating cognitive and communicative abilities in this group. At the time of the study the P-Scales were a tool used UK-wide in educational settings to differentiate performance in pupils functioning below the first level of the national curriculum (although their use is now under review – see Standards and Testing Agency, 2016). The P-Scales are based on teacher judgements and categorise students into one of eight levels of functioning based on performance observed in the classroom. Although the fact that they are based solely on teacher judgements places some limitations on the strength of P-scales as a measure (see Chapter 3, p55) , it was felt that that they had certain advantages since they were well established in an educational setting, familiar to teachers and provided readily available information about participants since all were having their performance monitored regularly through the use of this system.

#### **5.3.5. The administration of measures**

In order to address the issue of fluctuating performance, it was decided that measures of JA and functional vision would be administered on three separate occasions with at least a week separating them. It was also decided that measures should be administered at a different time of day on each occasion in case participant level of engagement was, for example, higher in the morning than the afternoon.

#### **5.3.6. Data analysis**

In order to take heterogeneity into account, it was decided that data analysis for this research would take a three tier approach, as recommended by (Light, 1988), considering results at both a group and individual level.

#### **5.3.7. Identifying potential benefits of the study**

This study was considered to have a strong ethical foundation since it was anticipated that its findings would be of benefit to people with PIMD, ultimately improving the SLT input provided to support them. In order to make this clear to those giving consent on behalf of participants, it was decided that the aims and potential benefits of the study should be detailed on the information letter provided for schools and parents. To provide some more direct benefit to participants it was also decided that

feedback sessions including some information about each participant's profile of performance and overall study findings, would be offered to teaching staff and parents once the study had been completed.

#### **5.3.8. Summary of decisions**

Having considered a range of methodological issues it was decided that the research carried out as part of this thesis should focus on school-aged children with PIMD who would be accessed through special schools using convenience sampling. Structured probes would be used to elicit and measure JA behaviours and, where possible, these would be based on relevant existing methodology. Functional vision would be assessed through trialling a measure established as effective for children with cerebral palsy. A range of additional participant characteristics as outlined by Pennington et al. (2007) would be assessed and described to facilitate synthesis of findings with future research. Measures of JA and functional vision would be administered on three occasions to accommodate fluctuations in performance and results would take individual profiles of performance into account.

#### **5.4. The need for exploratory work**

The assessment of both JA behaviours and functional vision for this study would involve the use of measures which were either novel or had not previously been used with a population who have PIMD. It was, therefore, decided that an exploratory phase of work should be conducted before the main study was undertaken. Such exploratory work is generally carried out through the use of small-scale studies and is especially useful for field testing the feasibility and effectiveness of novel measures before they are used to draw conclusions in a larger-scale piece of research (Gorard, 2013; Swedberg, 2018). The following chapter describes this exploratory phase of work.

## 6. The Development and Testing of Measures: An Exploratory Phase of Study

This exploratory phase of study was undertaken to test the feasibility and effectiveness of two measures; firstly, a set of structured probes designed to elicit and measure JA behaviours in children with PIMD and, secondly, the FunVis assessment of functional vision (Clarke et al., 2019). The structured probes required testing in this manner since, although drawing on established approaches, they would constitute a novel measure, being specifically designed for this study. The feasibility and effectiveness of the FunVis needed to be tested in relation to children with PIMD since, although established as effective for children with cerebral palsy it had not been used with children with the degree of intellectual impairment experienced by those with PIMD.

In order to be considered feasible and effective, both the JA measure and the FunVis assessment would need to:

- 1) Elicit a range of target behaviours without floor or ceiling effects.
- 2) Reflect participants' performance of target behaviours in unstructured 'everyday' contexts, possibly highlighting greater capacity in structured contexts.
- 3) Be sufficiently interesting to engage participants with PIMD and practically feasible to administer

To test these areas the measures were administered to a small sample of seven children whose performance of JA behaviours were also measured through observation in everyday contexts and interviews with their teachers.

The following chapter will present this exploratory work, beginning by describing the development of the structured probes which were used to elicit and measure JA behaviour. The method used to administer this measure, additional measures of JA behaviour and the FunVis assessment will then be described. Finally, the results and conclusions of the exploratory work will

be presented along with a description of how these conclusions were used to inform methodology for the main body of research.



## **6.1. The development of structured probes to measure JA behaviours**

### **6.1.1. Target JA behaviours**

The target behaviours selected to be measured by the probes were based on the typical trajectory of JA development outlined in section 4.2.4. and included behaviours which might be considered pre-requisites to JA (attending separately to people and objects) as well as those indicating lean and rich forms of JA (initiating JA for imperative and declarative functions).

Target areas included both observable gaze behaviours (such as gaze shifting between person and object) as well as behaviours which were judged by raters to be intentionally communicative.

A list of the target behaviours and their descriptors is represented in Table 6.1

Table 6.1. Target JA behaviours measured during the exploratory phase of study.

|                           | Target behaviour                           | Code      | Descriptor   |
|---------------------------|--|-----------|--|
| Observable Gaze Behaviour | Attends to a single referent               | AO        | Fixes visual attention on an object  |
|                           |  | AP        | Fixes visual attention on a person   |
|                           |  | AO +AP    | Fixes visual attention separately on an object and on a person during the same trial |
|                           | Integrates attention to objects and people | AOP       | Shifts gaze between person and object  |
|                           | Follows gaze and head turn                 | RJA       | Follows the focus of another person's attention                                      |
| Rater Judgement           | Engages in JA                              | IJA (imp) | Engages in JA for <u>imperative</u> purposes (requesting or rejecting)               |
|                           |  | IJA (dec0 | Engages in JA for <u>declarative</u> purposes<br>(sharing affect or interest)        |

### **6.1.2. The structured probes**

The probes developed to elicit target JA behaviours are described below. Each probe consists of an activity which is repeated several times during every test session, with each repetition, or 'trial', providing an opportunity for the target behaviours to be demonstrated.

#### **6.1.2.1. Dynamic Object Probe**

**Aim of probe:** To elicit the initiation of JA for imperative and declarative purposes in a bottom-up context.

#### **Theoretical basis of probe**

This probe was based on the 'object spectacle' probe which has been used successfully as part of the ESCS to elicit the initiating of JA for imperative and declarative functions (Mundy et al., 2003). In the 'object spectacle' probe, objects and activities, such as wind-up toys and inflating balloons, are activated by the tester (providing an opportunity for the participant to engage in declarative JA). These items then either deactivate spontaneously or are deactivated by the tester, (providing an opportunity for the participant to engage in imperative JA). Since stimuli are simultaneously salient to tester and participant this constitutes a 'bottom-up' context.

#### **Equipment**

The stimuli used for the ESCS have been found to be insufficiently engaging for a PIMD population (Iacono et al., 1998). Therefore, a more appropriate set was selected for this probe. All chosen test items had a dynamic element so they could be activated and deactivated, were multisensory and, in the clinical experience of the author, frequently found to be engaging for people with PIMD. However, to address the possibility that participants would not find any of the test stimuli engaging, participants were also invited to bring an item of their own choosing which met the specified requirements.

The standard set of four test items was as follows:

**1. Light spinner**



When activated by a button the light spinner produces spinning light and optional sounds.

**2. Woodpecker**



When pulled to the top of the pole the woodpecker gradually 'pecks' its way down.

**3. Marble Tree**



When marbles are placed at the top of the tree they roll down, producing rewarding changes in sound as they go.

**4. Plane**



When the plane is activated by pressing a button, the foam propeller lights up and spins, producing a fan effect. Music may also be activated.

**Procedure for the Dynamic Object probe**

Test items are kept out of sight in a case under the table until needed. An item is brought out and placed on the table. The tester activates the toy and watches it for the initial part of its activation (approximately 30 seconds),

creating an opportunity for the participant to make active attempts at gaining her attention. She then turns to look at the participant, creating the chance for them to share attention through mutual gaze and additional signalling e.g. smiling.

As the test item deactivates, the tester looks back at it and continues looking at it for up to one minute while it is full deactivated, creating an opportunity for the participant to initiate JA for requesting a restart of the toy.

Throughout the procedure, the tester keeps any verbal comments or sounds to a minimum but, if the participant is judged to initiate declarative JA, the tester acknowledges this through appropriate facial expression and positive noises or brief comments e.g. “mmm, that’s a funny sound!”. If the participant is judged to engage in imperative JA for requesting or rejecting an item, the tester acknowledges this by reactivating the item one more time or by removing it.

### **Number of trials**

During each test session this procedure is administered once with each of the four standard test items and once with the familiar item that the participant brings to the session.

Total trials per test session = 5

### **Scoring of the Dynamic Object probe**

Scoring begins when the test item activates and ends one minute after it has deactivated. Further details of the scoring for all probes is given in section 6.1.3.

#### **6.1.2.2. Pencil Tin Probe**

**Aim of probe:** To elicit the initiation of JA for declarative purposes in a top-down context

#### **Theoretical basis of probe**

This probe aimed to create a ‘top-down’ context for eliciting JA (in contrast to the ‘bottom-up’ context created by the Dynamic Object probe) through providing a referent of which only the participant is aware. As discussed in

Chapter 5, some research studies have created ‘top-down’ probes for eliciting JA by introducing an unexpected event e.g. an alligator toy dangling down from the ceiling (McLean et al., 1991). However, these generally required a degree of setting-up within the physical context which was not practical for the present study as sessions were carried out in various rooms provided by schools and not necessarily accessible to the researcher in advance. Therefore, an effort was made to create a naturalistic context in which an unexpected event occurs, unnoticed by the tester.

### **Equipment**

A bright tin full of pencils sized approximately 15cm x 6cm.

### **Procedure for the Pencil Tin probe**

The tester engages in face-to-face singing, talking or play with the participant. An assistant, who is seated to the side of the tester holds the tin of pencils out towards her, gradually increasing cues such as shaking the tin and saying the tester’s name. The tester continues to engage with the participant, apparently oblivious to the assistant’s efforts to communicate with her. If the participant makes attempts to draw the tester’s attention to the pencil tin, she smiles, says “oh, thanks!” and takes the tin. If the participant does not draw the tester’s attention to the pencil tin, the assistant puts the tin away.

The side on which the assistant is sitting is alternated for each administration of the procedure.

### **Number of trials**

The procedure is administered five times during the test session interspersed with administration of other probes.

Total trials per test session = 5

### **Scoring of Pencil Tin probe**

Scoring begins when the assistant first holds out the tin of pencils and ends when the tester takes the pencils or the tin is put away.

### **6.1.2.3. Look at the Puppet Probe**

**Aim of Probe:** To elicit RJA

#### **Theoretical Basis of probe**

This probe was designed to elicit the following of gaze and a head turn as described in the research reported in section 4.2.4. In contrast to the poster-based probe used by the ESCS to assess RJA which was considered to be impractical to implement and unsuitable for those with potential visual impairments it employs a real, 3D object and a naturalistic context.

#### **Equipment**

An octopus shaped hand puppet approximately 10 x 30 cm in size.

#### **Procedure for the Look at the Puppet probe**

The assistant is seated to the side of the tester at a distance of approximately 1 metre. She is wearing the octopus hand puppet which is resting in her lap. The tester engages the participant's attention by calling his or her name. Once eye contact is established, the tester says "[name] look!" while turning her head to the side to look at the hand puppet in the assistant's lap. If the participant follows the head turn and looks toward the puppet it is wiggled slightly. The side on which the assistant sits is alternated for each administration of the probe.

#### **Number of trials**

Procedure is administered five times during the test session interspersed with administration of other probes.

Total trials per test session = 5

### **6.1.3. Scoring of the structured probes**

Five trials of every probe were administered during each test session making a total of fifteen trials. Since three test sessions were carried out, each participant completed an overall total of 45 trials. For the Dynamic Object and Pencil Tin probes a score of 0 or 1 was given for all target behaviours listed in Table 6.1 (with the exception of RJA) where:

0= target behaviour is not demonstrated during the trial.

1= target behaviour is demonstrated at least once during the trial.

Trials of the Look at the Puppet probe were given a score of 0 or 1 for RJA where:

0 = does not disengage gaze from the tester to look in the direction of her head turn.

1 = disengages gaze from the tester to look in the direction of her head turn.

Scores were combined across all three test sessions and a percentage score given for each target behaviour, representing the number of completed trials in which the behaviour was demonstrated. This allowed for the possibility that some trials might not be completed due to participants becoming distracted or tired.



## **6.2. Testing the structured probes and the FunVis for feasibility and effectiveness**

### **6.2.1. Method**

#### **6.2.1.1. Design**

For this exploratory study, measures of JA behaviour and of functional vision were tested for feasibility and effectiveness by administering them to participants on three separate occasions. Field notes were taken during the administration of the measures to gather observations about practical aspects of their use. Data about JA behaviours elicited by the structured probes was compared to those demonstrated in 'everyday' contexts by observing participants their school environment on three occasions and through conducting a survey interview with teachers.

#### **6.2.1.2. Ethical Approval**

Full ethical approval for the study was given by the UCL ethics committee, Project ID 7565/001. Since participants in the study were unable to give informed consent to take part, this was given by their parents on their behalf.

#### **6.2.1.3. Recruitment**

Recruitment was initiated through direct contact with five special schools catering for primary or secondary-aged children with PIMD within the Greater London and Surrey area. The headteacher of each school was sent a covering letter or email containing brief information about the project and an invitation to participate. Of the five schools contacted, two did not respond. The remaining schools expressed an interest in participating and were provided with further information about the requirements of the project including the need to:

- Provide a quiet room for administration of the testing sessions.
- Ensure that a member of staff familiar with each participating child would be able to accompany them to testing sessions.
- Allow researchers to carry out observations in everyday school environments agreed in advance with teaching staff.
- Allow time for a teacher familiar with each participant to complete the teacher interview.

All three schools agreed to meet these requirements. They were subsequently asked to identify participants who met the inclusion criteria for the study.

### **Inclusion Criteria**

Inclusion criteria were based on the definition of PIMD outlined in section 2.2. In addition, the P-Scales were used to provide further guidance for teachers with regard to the cognitive functioning of potential participants. Only children classified as being at P-Scale 3(ii) or below would be included in the study since P-Scale 4 and above represents verbal communicators who would not meet the criteria for having PIMD. Inclusion criteria also stipulated that participants should have sufficient vision and hearing for them to be able to engage with the activities included in the probes. The resulting inclusion criteria are represented in Table 6.2.

*Table 6.2. Inclusion criteria for the exploratory phase of study*

|  |
|--|
| <p><b>Participants should:</b></p> <ul style="list-style-type: none"><li>• be described as having a profound learning disability.</li><li>• have at least one additional sensory, motor or communication impairment e.g. physical disability, visual or hearing impairment, medical condition such as epilepsy.</li><li>• be pre-verbal i.e. should not use speech.</li><li>• have severely limited understanding of verbal language without additional cues.</li><li>• Be functioning at P-scale 3ii or below for English.</li><li>• have sufficient functional vision to respond to everyday items at school and at home.</li><li>• have sufficient functional hearing to be able to respond to another person speaking to them.</li></ul> <p><b>Participants should not:</b></p> <ul style="list-style-type: none"><li>• consistently use alternative formal means of communication e.g. signs/symbols.</li></ul> |
|--|

Ten potential participants were identified across the three schools and were sent information letters and consent forms by the schools themselves. No participants were contacted directly by the researcher. Parental consent was not obtained for three children, all of whom attended the same school. A total of seven children aged 4-17 years, from the two remaining schools, were given consent to participate and were recruited.

#### **6.2.1.4. Participants**

Participant characteristics are described in Table 6.3.

Table 6.3. Participant characteristics for the exploratory phase of study

|           | Age | Gender | Medical Diagnosis   | Hearing                                   | Vision  | P-Scale:<br>English | GMFCS<br>Level | MACS<br>Level                  |
|-----------|-----|--------|---|---|---|---------------------|----------------|--------------------------------|
| <b>P1</b> | 9   | M      | 1p36 Deletion Syndrome  | No known<br>issues                        | Impaired<br><br>(functional<br>with glasses)            | 3:ii                | 3              | 3<br><br>(left<br>hemiparesis) |
| <b>P2</b> | 11  | M      | Quadriplegic Cerebral Palsy<br><br>Learning disability<br><br>Epilepsy (uncontrolled)                 | No known<br>issues                        | No known<br>issues                                      | 3:i                 | 5              | 5                              |
| <b>P3</b> | 13  | M      | Allan-Herndon-Dudley<br>Syndrome  | No known<br>issues                        | No known<br>issues                                      | 3:i                 | 5              | 4                              |
| <b>P4</b> | 8   | M      | DOOR Syndrome<br><br>Kyphoscoliosis<br><br>Partial Agenesis of the<br>Corpus Callosum<br><br>Epilepsy | Impaired<br><br>(functional<br>with aids) | Impaired<br>(function<br>unknown –<br>wears<br>glasses) | 2:i                 | 5              | 3<br><br>Tactile<br>defensive  |

Table 6.3. Participant characteristics for the exploratory phase of study

|           | Age | Gender | Medical Diagnosis  | Hearing         | Vision  | P-Scale:<br>English | GMFCS<br>Level | MACS<br>Level |
|-----------|-----|--------|--|-----------------|---|---------------------|----------------|---------------|
| <b>P5</b> | 4   | M      | Angelman Syndrome  | No known issues | Impaired<br>(can track objects without glasses) | 2:i                 | 4              | 2             |
| <b>P6</b> | 17  | F      | Profound intellectual impairment (unknown aetiology)<br><br>Epilepsy | No known issues | Impaired<br>(functional with glasses)           | 2:i                 | 5              | 3             |
| <b>P7</b> | 7   | M      | Glut1 Deficiency Syndrome<br><br>Seizures (controlled)               | No known issues | No known issues                                 | 3:i                 | 4              | 3             |

#### **6.2.1.5. Researchers**

The principal researcher (the author) is a Speech and Language Therapist with more than twenty years' experience in working in school contexts and with people who have PIMD. Two further researchers were involved in supporting the principal researcher to conduct the testing activities. Both were final-year students on the MSc. Speech and Language Sciences programme at UCL and had previous experience of working with people who had PIMD.

#### **6.2.1.6. Measures**

Measures of JA, functional vision and background measures were administered to participants for this exploratory phase of study and are described below.

##### **Measures of JA**

The structured probes outlined in section 6.1. were administered to participants alongside two further measures of JA. The first was an observational measure designed to assess JA behaviours used in everyday contexts and the second was an interview-based questionnaire designed to explore teacher judgements of JA behaviour.

##### **Observational measure of JA**

This observational instrument was designed to measure whether participants engaged in target JA behaviours within their everyday school environment which they did not demonstrate during the structured probes. To achieve this, each participant was observed for half an hour on three separate occasions within their school setting. This period of time was chosen because it was similar to the length of time which participants spent engaged in the structured probes. An observation form was devised on which raters recorded whether the target behaviours described in Table 6.1. were seen to occur during each of three 30-minute long observation sessions (see Appendix B for an example of the observation form). For each observation session, participants were given a single score of 0 or 1 for each target JA behaviour dependent on whether or not it had been observed. Further details of the procedure used to administer the observational measure are outlined in section 6.2.1.7.

### Interview based questionnaire for teachers

An interview-based questionnaire was drawn up to gather teacher judgements about whether participants were able to demonstrate the target JA behaviours. Standardised questions were delivered in a face-to-face interview situation to ensure a high response rate (Polgar & Thomas, 2013) and so that teachers could clarify any questions if necessary.

The interview consisted of a set of six questions, each of which was designed to address one of the target JA behaviours. A binary yes/no answer was requested for each question although teachers could add qualitative information to clarify their answers if they wished to. Participants were given a score of 0 or 1 for each target behaviour dependent on whether or not teachers reported that they demonstrated it.

*Table 6.4. Interview-based survey questionnaire for teachers*

| Target JA behaviour                               | Question(s)   |
|---|---|
| Attention to objects (AO)                         | Does NAME look at objects in his/her environment?   |
| Attention to people (AP)                          | Does NAME look at people in his/her environment?  |
| Coordinated attention to objects and people (AOP) | Does NAME look back and forth between people and objects in his/her environment?  |
| Responding to Joint Attention (RJA)               | If you were to look over at something or somebody, would the child follow your gaze to see what you are looking at?   |
| Initiating Joint Attention (IJA(dec))             | Does NAME make active attempts to draw your attention to something to share it with you? If so, how?  |
| Requesting/Rejecting (IJA(imp))                   | Does NAME intentionally get you to continue an activity s/he enjoys or ask for something s/he would like? If so, how? (Please give an example)<br><br>Does NAME intentionally get you to stop an activity s/he dislikes or reject something s/he would like? If so, how? (Please give an example) |

The procedure used to administer the interview-based survey is outlined in section 6.2.1.7.

### **Functional vision measure**

Functional vision was measured using the FunVis assessment (Clarke et al., 2019). As discussed in section 5.3.8., this tool is designed for use with children who have cerebral palsy and are functioning, developmentally, between 9 months and 6 years.

For this assessment, circular cardboard targets which are 5cm in diameter, mounted on sticks and are either blank or have schematic smiley faces drawn on them (see Figure 6.1. Targets used for the Rapid Assessment of Functional Vision (FunVis)) are used to assess the ability to:

1. Fix gaze on a target at eye level or on a wheelchair tray/table-top.
2. Disengage gaze from a target and shift it to a second target when both targets are at eye level or on a wheelchair tray/table-top.
3. Track a target horizontally at eye level.



*Figure 6.1. Targets used for the Rapid Assessment of Functional Vision (FunVis)*



A score of 0 or 1 is given for the following six target behaviours dependent on whether the behaviour is demonstrated (see Appendix C for full descriptors of target behaviours):

- 1) Fixes gaze on target at eye level.
- 2) Fixes gaze on target on tray/table top.
- 3) Shifts gaze between targets at eye level.
- 4) Shifts gaze between targets on tray/table top.
- 5) Prefers schematic face target to blank.
- 6) Tracks target.

The detailed procedure for administering the FunVis may be seen in Appendix C.

### **Background measures**

In addition to the principal measures, a set of background measures was used to assess the cognitive, communication and motor skills of participants.

### **The P-Scales**

The P-Scales were used as a background measure of communicative and cognitive ability (see section 5.3.4.). Teachers were asked to provide the P-Scale level for English which had been ascribed to each participant at their most recent educational review. The descriptors for each level of the P-Scales may be seen in Table 6.5 with full performance descriptors to be found in Appendix D .

Table 6.5. *P-Scale descriptors for English (Department for Education, 2014).*

| P-Scale          | Descriptor   |
|------------------|--|
| P1(i)            | Pupils encounter activities and experiences.   |
| P1(ii)           | Pupils show emerging awareness of activities and experiences.  |
| P2(i)            | Pupils begin to respond consistently to familiar people, events and objects.   |
| P2(ii)           | Pupils begin to be proactive in their intentions.  |
| P3(i)            | Pupils begin to communicate intentionally.   |
| P3(ii)           | Pupils use emerging conventional communication.  |
| P4<br>(Speaking) | Pupils repeat, copy and imitate between 10 and 50 single words, signs or phrases or use a repertoire of objects of reference or symbols. |
| P4 (Listening)   | Pupils demonstrate an understanding of at least 50 words, including the names of familiar objects.                                       |
| P4 (Reading)     | Pupils listen and respond to familiar rhymes and stories.  |

### **The Gross Motor Classification Schedule (GMFCS)**

The GMFCS (Palisano et al., 2008) was used as a measure of gross motor ability. This tool provides descriptors for classifying a child's motor functioning into one of five categories (see Table 6.6.). A GMFCS level was assigned to each child on the basis of discussion with their teacher.

Table 6.6. Gross Motor Classification Scale (GMFCS) levels.

| Level | Descriptor   |
|-------|--|
| I     | <ul style="list-style-type: none"> <li>• Can walk indoors and outdoors and climb stairs without using hands for support</li> <li>• Can perform usual activities such as running and jumping</li> <li>• Has decreased speed, balance and coordination</li> </ul>  |
| II    | <ul style="list-style-type: none"> <li>• Can climb stairs with a railing</li> <li>• Has difficulty with uneven surfaces, inclines or in crowds</li> <li>• Has only minimal ability to run or jump</li> </ul>   |
| III   | <ul style="list-style-type: none"> <li>• Walks with assistive mobility devices indoors and outdoors on level surfaces</li> <li>• May be able to climb stairs using a railing</li> <li>• May propel a manual wheelchair and need assistance for long distances or uneven surfaces</li> </ul>  |
| IV    | <ul style="list-style-type: none"> <li>• Walking ability severely limited even with assistive devices</li> <li>• Uses wheelchairs most of the time and may propel own power wheelchair</li> <li>• Standing transfers, with or without assistance</li> </ul>  |
| V     | <ul style="list-style-type: none"> <li>• Has physical impairments that restrict voluntary control of movement</li> <li>• Ability to maintain head and neck position against gravity restricted</li> <li>• Impaired in all areas of motor function</li> <li>• Cannot sit or stand independently, even with adaptive equipment</li> <li>• Cannot independently walk but may be able to use powered mobility</li> </ul> |

### The Manual Ability Classification System (MACS)

Functional fine motor abilities were measured using the MACS. This tool classifies a child's functional manual abilities into one of five categories (see Table 6.7). A MACS level was assigned to each child on the basis of discussion with their teacher.

*Table 6.7. Manual Ability Classification System (MACS) levels (Eliasson et al., 2006)*

| <b>Level</b> | <b>Descriptor</b>   |
|--------------|---|
| Level 1      | Handles objects easily and successfully   |
| Level 2      | Handles most objects but with somewhat reduced quality and/or speed of achievement    |
| Level 3      | Handles objects with difficulty; needs help to prepare and/or modify activities       |
| Level 4      | Handles a limited selection of easily managed objects in adapted situations           |
| Level 5      | Does not hand objects and has severely limited ability to perform even simple actions |

#### **6.2.1.7. Procedures**

The structured probes and the FunVis assessment were administered during three test sessions. Observation of the participant in their school environment was also carried out on three occasions and the interview-based questionnaire for teachers was administered once, before any of the other measures were conducted. Procedures for administration of the structured probes, the observation sessions and the teacher interview are described below. As noted previously, the procedure for carrying out the FunVis can be seen in Appendix C.

##### **Procedure for administering the structured probes**

The structured probes described in section 6.1.2. above were administered during test sessions with their order defined by a protocol which may be seen in Appendix E. The session began with a warm-up period during which the tester and assistant engaged in informal interaction with both the participant and the member of teaching staff accompanying them. The member of teaching staff was shown an information letter about the project if they had not already seen it, and advised to sit quietly, to the side of the participant but to alert the tester if they observed any signs that the participant was unhappy or uncomfortable. Following the warm-up period, the FunVis assessment was administered and then the structured probes were carried out.

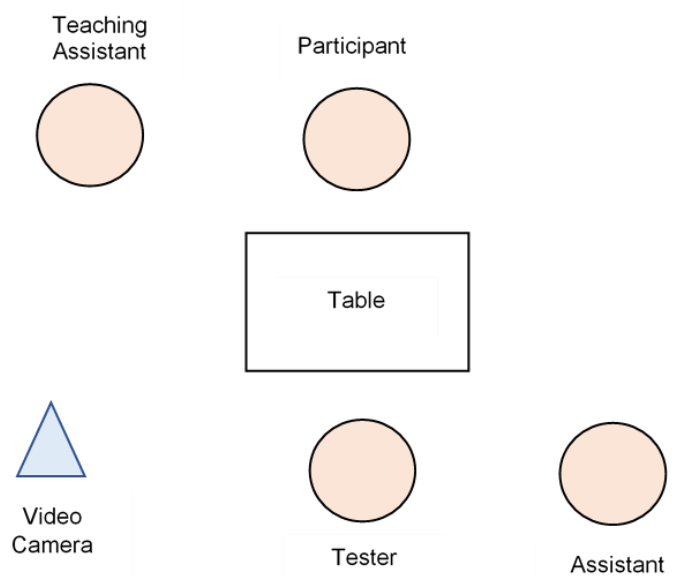
Test sessions were carried out on three separate occasions with at least a week in between each one. They were administered at a different time of day each time,

including at least one morning and one afternoon testing session. Test sessions lasted approximately 25-30 minutes in total.

All three researchers were fully trained in the administration and scoring procedures for the probes. For each participant the same two researchers would administer the structured probes during test sessions and the third would carry out observations. The two researchers administering the probes alternated their role as either tester or assistant for each test session.

### Room Set-Up

The room set-up is depicted in Figure 6.2.



*Figure 6.2. Room set-up for administration of the structured probes during the exploratory phase of study.*

### Video Recording

Test sessions were video recorded for subsequent analysis of data since it was challenging to record target behaviours 'live' during the session, partly because they could be fleeting and partly because both tester and assistant were involved in administering the measures.

A single digital camcorder was mounted on a tripod and situated so that, as far as possible, participant, target objects and the tester were all visible.

### **Procedure for conducting observations**

Three observations were carried out on three separate occasions for each participant and took place on the same day as the test sessions in which the structured probes were administered. Observations were conducted in the participant's everyday school context at a time convenient to teaching staff. As a result, different types of session were observed and the nature of each was noted on the observation recording form. Observed sessions included switch-work sessions, a whiteboard-based story time, 'hello' sessions, snack time and free-time after lunch.

Observations were carried out over a period of 30 minutes. This length of time was chosen as it equated, approximately, to the duration of the testing sessions in which the structured probes were administered

The two MSc. student researchers carried out all the observations, with each participant being observed by the same student researcher on all three occasions. This researcher was 'blind' to the participant's performance in test sessions.

### **Procedure for Teacher Interviews**

The teacher interview was administered by the principal researcher and all class teachers had known their students for at least nine months.

#### **6.2.1.8. Data analysis**

Participant percentage scores for each JA behaviour elicited by the structured probes were analysed to check for floor or ceiling effects. Results obtained from the structured probes and observational measure were compared by analysing the number of structured probe test sessions and observation sessions in which participants had demonstrated target behaviours. This data was compared with results from the teacher interview by calculating the number of agreements between teacher judgements about demonstration of target behaviours and their occurrence during structured probe test sessions and observation sessions.

## **6.2.2. Results**

Results relating to the feasibility and effectiveness of the structured probes will be presented first, followed by results relating to the FunVis assessment.

### **6.2.2.1. Results relating to the structured probes**

#### **Inter-rater agreement**

Video recordings of all 21 testing sessions were scored independently by two raters. Raters were the MSc. students assisting on the project, the first of whom had taken part in the session she scored and the second of whom had not.

Inter-rater agreement for JA behaviours scores was calculated using Cohen's kappa (see Table 6.8).

Table 6.8. Inter-rater agreement for JA behaviour scores on the structured probes

| JA Behaviour   | Code      | Kappa value                             | Level of Agreement (Altmann, 1991) |
|--|-----------|---|------------------------------------|
| Attention to objects   | AO        | .685<br>95% CI (.546 - .824)<br>p <.001 | Good                               |
| Attention to people  | AP        | .595<br>95% CI (.436 - .754)<br>p <.001 | Good                               |
| Integrates attention to objects and people using gaze shifts | AOP       | .354<br>95% CI (.225 - .483)<br>p <.005 | Fair                               |
| Follows the focus of another person's attention              | RJA       | .691<br>95%CI (.609 -.771)<br>p <.005   | Good                               |
| Initiating JA for <u>imperative</u> purposes                 | IJA (imp) | . 510<br>95%CI (.320 -.70)<br>p <.005   | Moderate                           |
| Initiating JA for <u>declarative</u> purposes                | IJA(dec)  | . 467<br>95%CI (.265 -.669)<br>p <.005  | Moderate                           |

Whilst judgements of AO, AP and RJA achieved good levels of inter-rater agreement, judgements about AOP and IJA achieved only fair or moderate levels of agreement.



### Target JA behaviours elicited by the structured probes

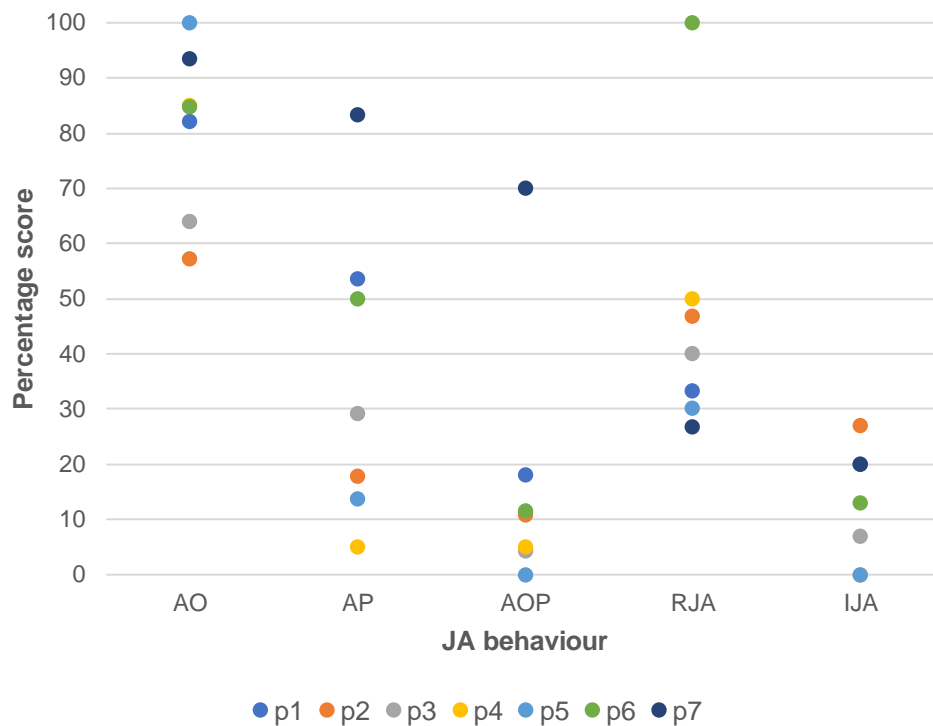
In order to explore whether the probes elicited a range of target behaviours without floor or ceiling effects, the overall percentage score for each target behaviour was analysed (see Table 6.9.). Since scores for IJA were generally low, measures of IJA(imp) and IJA(dec) were combined for this analysis.

*Table 6.9. Percentage scores for each target JA behaviour.*

| <b>JA Behaviour</b> | <b>Mean Score<br/>(%)</b> | <b>Range</b> | <b>S.D</b> |
|---------------------|---------------------------|--------------|------------|
| <b>AO</b>           | 81                        | 57-100       | 15         |
| <b>AP</b>           | 35                        | 0-83         | 29         |
| <b>AOP</b>          | 17                        | 0-70         | 24         |
| <b>RJA</b>          | 47                        | 27-100       | 25         |
| <b>IJA</b>          | 12                        | 0-27         | 11         |

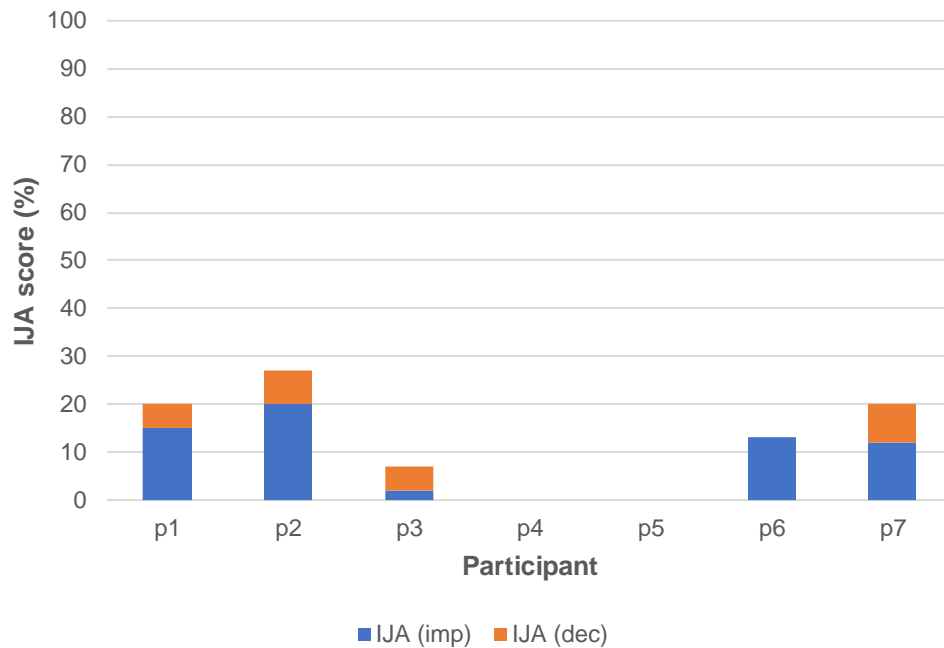
All target behaviours were elicited by the probes to at least some extent. As a group, participants demonstrated attention to objects (AO) more frequently than attention to people (AP) while the integration of attention to both people and objects through gaze shifting (AOP) was elicited in only 17% of trials. RJA behaviour in the form of following a head turn was elicited in 47% of trials and with IJA being the most infrequently observed behaviour, being judged to occur in only 12% of trials.

A range of target behaviours was also elicited across individuals without overall floor or ceiling effects (see Figure 6.3) .



*Figure 6.3. Individual Percentage Scores for each JA behaviour*

Detailed analysis of scores for IJA(imp) and IJA(dec) indicated that IJA was elicited in five participants with imperative IJA being scored more frequently than declarative IJA, mean score for IJA (imp) being 9% (range = 0-20%, S.D.= 8.1) and mean score for IJA (dec) being only 4% (range = 0-8%, S.D. = 4.2) (see Figure 6.5.).



*Figure 6.4. Imperative and declarative IJA demonstrated by individual participants*

Both the Dynamic Object and Pencil Tin probes had been devised to elicit declarative IJA behaviours. However, further analysis of the scores obtained from each probe indicated that all IJA(dec) behaviours were demonstrated during the Dynamic Object probe and none during the Pencil Tin probe, representing a floor effect for the latter measure. Field notes reported that, completion rates for this probe were relatively low. When participants did engage with it, they would often show awareness of the assistant proffering the pencil tin by looking at the tin and, sometimes, smiling. However, they did not make active attempts to direct the tester's attention to it.

#### **Comparison between JA behaviours elicited by the structured probes and those demonstrated during observation**

In order to investigate the relationship between JA behaviours elicited by the structured probes and those observed in less structured, everyday, contexts, the number of structured probe test sessions and in which each participants demonstrated the target behaviours was compared with the number of observation sessions in which they had demonstrated them (see Table 6.10. Number of structured probe test sessions and observation sessions in which target behaviours were demonstrated Table 6.10).

*Table 6.10. Number of structured probe test sessions and observation sessions in which target behaviours were demonstrated*

(maximum = 3)

|                      | AO |    | AP |    | AOP |    | IJA |    | RJA |    |
|----------------------|----|----|----|----|-----|----|-----|----|-----|----|
|                      | O* | P  | O  | P  | O   | P  | O   | P  | O   | P  |
| P1                   | 3  | 3  | 3  | 3  | 3   | 3  | 3   | 3  | 0   | 2  |
| P2                   | 2  | 3  | 2  | 2  | 0   | 2  | 0   | 2  | 0   | 2  |
| P3                   | 3  | 3  | 3  | 2  | 0   | 1  | 1   | 1  | 0   | 2  |
| P4                   | 3  | 3  | 3  | 1  | 0   | 1  | 0   | 0  | 0   | 2  |
| P5                   | 2  | 3  | 3  | 3  | 0   | 0  | 0   | 0  | 0   | 2  |
| P6                   | 2  | 3  | 3  | 3  | 0   | 3  | 0   | 1  | 0   | 1  |
| P7                   | 3  | 3  | 3  | 3  | 3   | 3  | 3   | 3  | 0   | 3  |
| Total<br>(out of 21) | 18 | 21 | 20 | 17 | 6   | 13 | 7   | 10 | 0   | 14 |

\* O= observational measure P = structured probes



= behaviour demonstrated in more structured probe sessions



= behaviour demonstrated in more observation sessions



= behaviour demonstrated with equal frequency on both measures

Results indicated that all target behaviours except attending to a person (AP), were demonstrated in as many or more structured probe test sessions as observation sessions. The exception was attributed to two participants who demonstrated AP in more observation sessions than structured probe test sessions. Overall, gaze shifting between object and person (AOP) and following a head turn and gaze (RJA) were especially more likely to be demonstrated during structured probe test sessions. In the case of RJA, field notes indicated that this was due to lack of opportunity with no relevant situations arising in which this behaviour could be demonstrated.

**Comparison between JA behaviours reported by teacher interview with those demonstrated during the structured probe test sessions and observation sessions.**

To explore whether teacher judgements reflected the JA behaviours demonstrated during administration of the structured probes and those seen during observation, the number of agreements between teacher judgements and scores for target behaviours on these measures was calculated for each participant. Agreement was judged to be reached if the teacher judged that a participant demonstrated the target behaviour and it was also seen during administration of the probes or during observation or if the teacher judged that a participant never demonstrated the target behaviour and it was not seen during administration of the probes or observation. The number of agreements between the measures is represented in Table 6.11. Since there were 7 participants, the total possible number of agreements for each target behaviour was 7.

*Table 6.11. Number of agreements between teacher judgements and demonstration of the target behaviours during administration of the probes and observation.*

(maximum = 7)

| <b>JA behaviour</b>      | <b>Observational Measure</b> | <b>Structured Probes</b> |
|--------------------------|------------------------------|--------------------------|
| <b>AO</b>                | 7                            | 7                        |
| <b>AP</b>                | 7                            | 7                        |
| <b>AOP</b>               | 5                            | 4                        |
| <b>IJA</b>               | 6                            | 4                        |
| <b>RJA</b>               | 7                            | 0                        |
| <b>Total (out of 35)</b> | <b>32</b>                    | <b>22</b>                |

Overall there was a higher level of agreement between teacher judgements and the observational measure than between teacher agreements and data obtained from the structured probes although there was complete agreement across all measures in relation to AO and AP behaviours which were demonstrated or reported in all contexts.

There were thirteen disagreements between teacher judgements and data obtained from the structured probes. In every case the disagreement was the result of the teacher judging that participants did not demonstrate the behaviour whereas it was seen to occur during the structured probes. This was most markedly the case for RJA where teachers judged no participants to demonstrate this skill, but all demonstrated it on at least some trials of the structured probes.

There were only three disagreements between teacher judgements and the observational measure. In the case of two of these, the teacher did not judge a behaviour to be demonstrated but it was seen during observation.

### **Engagement with the structured probes**

In order to judge whether the structured probes were sufficiently engaging for participants with PIMD, completion rates for trials of each probe were analysed. A trial was classified as incomplete if the participant became disengaged during its administration, for example, looking around the room, looking down or closing their eyes.

None of the participants completed all trials for each probe, with field notes indicating that incomplete trials tended to happen in clusters, sometimes occurring with a higher frequency for a participant's whole test session or towards the end of a session if a participant became tired or lost interest. However, there was also some variation according to probe type (see Table 6.12).

Table 6.12. Percentage of completed trials by probe and participant

| Participant  | Dynamic Object | Pencil Tin | Look at the Puppet | % of completed trials |
|--------------|----------------|------------|--------------------|-----------------------|
| <b>P1</b>    | 100%           | 13%        | 80%                | <b>77%</b>            |
| <b>P2</b>    | 100%           | 100%       | 100%               | <b>93%</b>            |
| <b>P3</b>    | 100%           | 73%        | 100%               | <b>84%</b>            |
| <b>P4</b>    | 67%            | 67%        | 67%                | <b>69%</b>            |
| <b>P5</b>    | 67%            | 33%        | 67%                | <b>71%</b>            |
| <b>P6</b>    | 53%            | 0          | 20%                | <b>48%</b>            |
| <b>P7</b>    | 100%           | 47%        | 100%               | <b>84%</b>            |
| <b>Total</b> | <b>84%</b>     | <b>48%</b> | <b>76%</b>         | <b>69%</b>            |

The Dynamic Object probe had the highest completion rate with 84% trials completed, while the Pencil Tin probe had a relatively low completion rate with only 48% trials completed. There was also some inter-participant variation with P6 being particularly prone to low completion, failing to complete any Pencil Tin trials and only 20% of Look at the Puppet trials.

### **Feasibility issues with the structured probes identified by field notes**

Field notes confirmed that participants seemed to show the highest levels of engagement during the Dynamic Object probe, perhaps due to the novelty of the items which were being presented, with all participants showing interest in at least some of the standard test items. Their level of engagement with these items meant that there was, in fact, limited need for them to bring an item of their own for the assessment.

In contrast with the Dynamic Object probe, practical issues were identified with the Look at the Puppet probe. Having the assistant seated next to the tester throughout the testing session proved to be somewhat distracting for participants and the fact that she occasionally provided an interesting stimulus by waving the hand puppet

meant that participants would sometimes disengage during trials to look at her – perhaps expecting that she might wave the puppet for them.

The system used to video sessions also led to some issues, with notes taken during scoring indicating that it was particularly difficult to score the occurrence of gaze shifts. The slightly side-long angle of the camera (see Figure 6.2) meant that the participant, test item and tester were not always in the frame at the same time so that it was difficult to judge the focus of gaze shifts. Additionally, the quality of the video and the fact that the footage could not be enlarged during playback contributed to the difficulty in making accurate judgements. Improving both the angle of the camera and the quality of video obtained could be used to address this issue.

#### 6.2.2.2. Results relating to the feasibility and effectiveness of the FunVis

For operational reasons, the FunVis assessment was administered only once (see further discussion below) and the scores obtained are represented in Table 6.13.

*Table 6.13. FunVis assessment scores*

|              | Fix:<br>Target<br>at eye<br>level | Fix:<br>Target on<br>tray/table-<br>top | Shift:<br>Between<br>targets eye<br>level | Shift:<br>Between<br>targets<br>tray/table-<br>top | Prefers<br>schematic<br>face target to<br>blank | Tracks<br>target | Total    |
|--------------|-----------------------------------|---|---|--|---|------------------|----------|
| <b>P1</b>    | 1                                 | 0                                       | 0   | 0  | 0   | 0                | <b>1</b> |
| <b>P2</b>    | 0                                 | 0                                       | 0   | 0  | 0   | 0                | <b>0</b> |
| <b>P3</b>    | 1                                 | 0                                       | 0   | 0  | 0   | 0                | <b>1</b> |
| <b>P4</b>    | 0                                 | 0                                       | 0   | 0  | 0   | 0                | <b>0</b> |
| <b>P5</b>    | 1                                 | 1                                       | 1   | 0  | 0   | 0                | <b>3</b> |
| <b>P6</b>    | 0                                 | 0                                       | 0   | 0  | 0   | 0                | <b>0</b> |
| <b>P7</b>    | 1                                 | 1                                       | 1   | 0  | 0   | 1                | <b>4</b> |
| <b>total</b> | <b>4</b>                          | <b>2</b>                                | <b>2</b>                                  | <b>0</b>   | <b>0</b>  | <b>1</b>         |          |

Scores on this measure were generally very low. Four participants fixed gaze on the targets at eye level, two additionally fixed gaze on targets on the table-top and only two participants demonstrated gaze shifting between targets. These scores contrasted with the gaze behaviours demonstrated by participants both during



observation and sessions and administration of the structured probes during which all participants were able to fix their gaze on objects and people at eye level and all demonstrated at least some ability to shift gaze as part of AOP, IJA or RJA behaviour.

Field notes suggested that participants were not sufficiently engaged by the targets used in the FunVis to pay attention to them. Indeed, they became so disinterested during administration of the protocol, which was carried out just after the warm-up period of the testing session, that it was hard to re-engage them for administration of the probes. It was decided not to change the order of administration to conduct the FunVis later in the session since this risked losing participant engagement at a time when some began to tire. Due to these operational challenges and the low scores it yielded, administration of the FunVis was abandoned after the first test session.

### **6.2.3. Discussion**

The aims of this exploratory study were to explore the feasibility and effectiveness of a set of structured probes designed to measure JA behaviours and of the FunVis assessment of functional vision, evaluating whether they were able to:

- 1) Elicit a range of target behaviours without floor or ceiling effects.
- 2) Reflect participants' performance of target behaviours in unstructured 'everyday' contexts, possibly highlighting greater capacity in structured contexts.
- 3) Be sufficiently interesting to engage participants with PIMD and practically feasible to administer

The effectiveness of the structured probes and the FunVis assessment will now be evaluated with reference to each of these areas alongside a discussion of changes which needed to be made in order to create effective measures of JA behaviours and functional vision for the main phase of the study.

#### **Feasibility and effectiveness of the structured probes**

The structured probes were effective in eliciting all target JA behaviours without floor or ceiling effects although there was variation between the probes in this respect which will be further discussed below.

Scoring of judgements about target behaviours using the criteria devised for the probes led to a good level of inter-rater agreement for judgements of AO, AP and RJA. However, agreement for judgements about gaze shifting (AOP) and IJA were lower. In the case of gaze shifting this may have been a consequence of the video recording method used, since it was found that gaze shifts were not always easy to see. This could be addressed by using improved camera angles and gathering footage which allows for slowing-down and zooming-in.

Lack of agreement for judgements about whether IJA had occurred may have resulted from the subjectivity of such judgements. As judgements relating to observable behaviours generally achieved higher levels of agreement, adding behavioural criteria for IJA might improve consistency of rating in this area.

The structured probes appeared to be effective in eliciting all the JA behaviours which each participant demonstrated in less structured, everyday contexts. There were only two incidences of participants demonstrating target JA behaviours more frequently during observation settings than structured probe test sessions and no incidences of teachers reporting behaviours which were not subsequently elicited by the probes. In contrast, there were many incidences where the structured probes elicited behaviours which were seen rarely or not at all during observation sessions. For example, two participants only demonstrated IJA during the probes, both of whom were judged not to do this by teachers. The probes were particularly useful for eliciting information about RJA behaviours since no opportunities for these to occur arose during observation.

The high degree of agreement between teacher judgements and data obtained from observation suggested that teacher judgements were an accurate reflection of performance demonstrated within everyday contexts but did not reflect a child's full capacity to engage in behaviours when structured opportunities were provided.

Of the three structured probes used, the Dynamic Object probe was found to be the most effective since it elicited both imperative and declarative JA in some participants. Its high completion rate also suggested that it was engaging for participants, all of whom responded to at least some of the standard test items used in its administration.

The Look at the Puppet probe had a relatively high completion rate of 76% and did elicit RJA from all participants on at least some trials although it was sometimes hard to gain initial eye contact in order for the probe to be administered. Field notes also highlighted practical issues with this probe since the presence of the assistant seated to the side of the tester was distracting, an issue which could be overcome by providing a wholly non-social stimulus as a target for this probe in the main phase of study.

The Pencil Tin probe, designed to elicit declarative JA in top-down context, was the least effective of the three, having a completion rate of less than 50% and failing to elicit the target behaviour in any participants despite some of them demonstrating it in response to the Dynamic Object probe. Indeed, the issue of whether the probe was truly a 'top-down' context could be debatable since, although the tester behaved as if she had not noticed the tin, participants may have inferred that she could see it and, therefore, not felt the need to draw her attention to it. Issues with the Pencil Tin

probe would need to be addressed by devising a more engaging and genuinely top-down context in which declarative JA might occur.

Overall, declarative IJA was the least commonly demonstrated target behaviour, a finding which may reflect the developmental stage of participants since it is consistent with results from other studies investigating declarative IJA in participants with PIMD (e.g. Neerinx & Maes, 2016). The low occurrence of this target behaviour highlights the need for any set of probes to include multiple contexts and opportunities to elicit it so that an individual's full capacity in this area may be measured.

The experience of testing probes for this exploratory study highlighted the complexities of judging whether failure to demonstrate target behaviours is due to aspects of the probes or participant ability. For example, if only the Pencil Tin probe had been administered it might have been concluded that none of the participants were able to engage in declarative JA. Therefore, it was decided that the modified set of probes to be used in the main phase of research should be tested on a reference group of typically developing children to ensure that they were effective in eliciting target behaviours before they were used as a basis for conclusions about JA abilities in the sample of participants with PIMD. The challenges raised by comparing the performance of typically developing children matched for developmental age with those with PIMD were raised in Chapter 5 and include issues with conducting accurate age matching and the fact that typically developing children do not share the varied profiles of physical and sensory abilities which children with PIMD demonstrate. However, given the absence of a more suitable alternative, in this case it was judged appropriate to use such a group as a means of testing the effectiveness of measures.

### **Feasibility and effectiveness of the FunVis assessment**

The FunVis assessment was not found to be a feasible or effective measure of functional vision for participants with PIMD. It was not sufficiently engaging and did not reflect the gaze fixation and shifting abilities which participants were able to demonstrate in other contexts. It seems likely that this was due to the relatively small and static 2D stimuli which are used for this assessment. Using real, 3D stimuli with a dynamic element would address this issue. However, since there is no existing measure of functional vision which uses such stimuli, one would need to be devised for the purposes of this research.

### 6.3. Conclusions and decisions relating to the main phase of study

Results of the exploratory phase of the study indicated that structured probes could be used effectively to elicit and measure JA behaviours in a sample of children with PIMD. However, findings also indicated that the following adaptations to measures and procedures would be required before further research was carried out on a larger scale:

- New probes should be developed to elicit declarative JA including one involving a top-down context. The effectiveness of these probes should first be verified using a reference group of typically developing children functioning within a similar developmental range to children with PIMD.
- Behavioural criteria should be developed to guide judgements about the initiation of JA for communicative purposes.
- Improved video recording techniques should be used to ensure gaze shifts are captured and can be analysed in detail.
- A new measure of functional vision should be developed which involves 3D targets with a dynamic aspect to ensure participants are engaged. This should also be tested for effectiveness on a reference group of typically developing children.

The work carried out during this exploratory phase of study was used to inform the measures and design for the main phase of work in which a measure of functional vision and a measure of JA behaviour were administered to a sample of children with PIMD for the purposes of answering the research questions set out in section 4.2.9. The following chapters will describe this work.

## **7. Development and Administration of a Functional Gaze Behaviour Measure**

This chapter describes the development of an alternative measure of functional vision to the FunVis assessment and its administration

### **7.1 Background**

As discussed in section 4.2.6., the functional vision skills of gaze fixation and gaze shifting constitute pre-requisites for JA behaviours which include the shifting of visual attention between objects and people. While typically developing infants are able to fix and shift their gaze between stimuli by the age of around four months (Atkinson & Braddick, 2012), evidence suggests that these skills are frequently impaired in atypically developing populations where, for example, phenomena such as the ‘competition effect’ which are seen in young infants, persist throughout childhood and, possibly, beyond (Atkinson et al., 2003; Cornish, Scerif, & Karmiloff-Smith, 2007). A persisting competition effect is likely to have implications for an individual's ability to engage in gaze shifting as part of JA since referents (the object and the person) are simultaneously present and, therefore, in competition.

To date such impairments of gaze fixation and gaze shifting have not specifically been researched and reported in a population described as having PIMD. However, given that they are an atypically developing group known to experience a high prevalence of visual dysfunction (Evenhuis et al., 2001) it was hypothesized that they might experience varying levels of difficulty with gaze fixation and shifting and that these difficulties might impact on their ability to demonstrate JA behaviours. To explore this hypothesis further it was necessary to develop a suitable measure of functional vision which would focus on gaze fixation and gaze shifting and which would be suitable for administration to participants with PIMD.

An attempt to measure these skills using the FunVis assessment designed for children with cerebral palsy (Clarke et al., 2019) had elicited limited responses from participants with PIMD in the exploratory work described in Chapter 6. While this finding may have been a reflection of the functional vision problems experienced by participants, it was noted that they were able to demonstrate better ability to fix and shift gaze when relating to the test items used as part of the experimental protocol. Unlike the target stimuli used as part of the FunVis, which were relatively small and 2D, these test items were larger, real objects which included movement and, sometimes, sound. These qualities may have meant that they were more engaging as well as being easier to perceive than the FunVis targets.

Existing studies of gaze shifting in atypically developing children have typically used a screen-based version of the fixation shift paradigm initially described by Tronick (1975) (e.g. Atkinson, Hood, Wattam-Bell, & Braddick, 1992; Cornish et al., 2007; Mercuri et al., 1999). During this procedure the participant is seated in front of a screen upon which a shape or a schematic face appears centrally. A second stimulus then appears to the side of the original stimulus. The original stimulus either remains in place and in competition with the second target ('competition condition') or disappears once the second stimulus has appeared leaving only the second target visible ('no-competition' condition). An observer then records if and when the child demonstrates a lateral gaze shift from the first to the second stimulus under both conditions.

Since it is an established method of measuring gaze behaviour, including the effect of competition, it was decided that the core aspects of the fixation shift paradigm could prove useful for the current study. However, previously used methods would require adaptations for use with participants with PIMD.

An issue with the commonly used protocol for the fixation shift paradigm described above is its dependence on screen-based, two-dimensional stimuli since the extent to which people with PIMD can engage with such stimuli is unclear. In the clinical experience of the author, many people with PIMD fail to engage with screen-based activities involving TV, tablets or PCs. They may also show limited interest in 2D, paper-based images. This is consistent

with the limited responses they demonstrated in response to the 2D schematic faces used as target stimuli by the FunVis assessment described in Chapter 5. Although, to the author's knowledge, no research has been carried out into the ability of people with PIMD to respond to screen-based stimuli, the lack of evidence means that such an approach not been established as a reliable means of exploring the skills of individuals with PIMD. Furthermore, the potential for generalising information gained from screen-based stimuli to functional performance in the 'real world' is unknown (Atkinson & Braddick, 2012). Therefore, to adapt the fixation shift paradigm for use with participants with PIMD and to improve the ecological validity of results, a procedure involving real rather than 2D or screen-based stimuli was devised. Since the procedure aimed to measure the functional behaviours of gaze fixation and gaze shifting, it was labelled the Functional Gaze Behaviour Measure (FGBM).



## **7.2. Development of the Functional Gaze Behaviour Measure (FGBM)**

The FGBM then, was a novel measure developed to assess participants' ability to fix and shift gaze between stimuli so that the impact of impairment in these areas on gaze behaviour used for JA could be measured.

### **7.2.1. Target behaviours to be measured by the FGBM**

Since JA behaviours include fixing gaze on both objects and people as well as shifting gaze between a person and an object which are simultaneously present, the following functional gaze behaviours were measured.

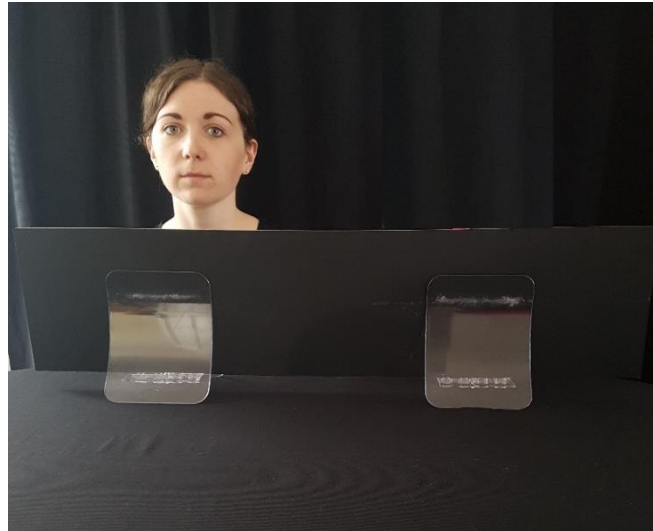
- 1) Fixation on faces.
- 2) Fixation on objects.
- 3) Gaze shifting under competition conditions.
- 4) Gaze shifting under no-competition conditions.

### **7.2.2. Test conditions**

Four different test conditions were devised to measure the target behaviours listed above. A system of dynamic and auditory cues was incorporated into each test condition.

### **Test condition 1: Fixation on faces**

For this test condition the tester's face was presented above a screen in front of the participant and slightly to one side of the midpoint (see Figure 7.1).

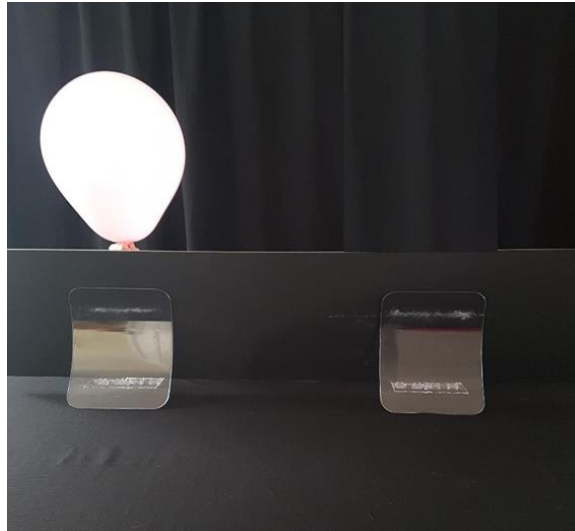


*Figure 7.1. FGBM Test condition 1: Fixation on a face*

Facial expression was kept neutral but friendly. If the participant failed to fix gaze on the face after three seconds a dynamic cue was added through the tester turning her head slowly from side to side. If fixation was still not achieved after a further three seconds, she added an auditory cue by saying “hi” as she continued to turn her head.

### **Test condition 2: Fixation on an object**

For this test condition a pink balloon, blown up to match the approximate size of the face stimulus, was presented above the screen, again, in front of the participant and slightly to one side of the midpoint (see Figure 7.2).



*Figure 7.2. FGBM Test condition 2: Fixation on an object*

If the participant failed to fix their gaze on the balloon after three seconds a dynamic cue was added by waving it from side to side. If fixation was still not achieved after a further three seconds, an auditory cue was added through the tester (who was holding the balloon but was not visible) saying “balloon!”.

### **Test condition 3: Gaze shifting under competition conditions**

For this test condition a second stimulus was presented as soon as gaze fixation on the first stimulus had been obtained, with both stimuli remaining visible and in competition. A number of trials were carried out for this condition involving a balanced combination of face and object stimuli (see Figure 7.3 for an example of a face-object gaze shift trial).



*Figure 7.3.FGBM Test condition 3: Gaze shifting under competition conditions*

If a participant did not disengage from the first to the second stimulus after three seconds, the same system of cuing as described for test conditions 1 and 2 was applied to the second stimulus.

#### **Test condition 4: Gaze shifting under no-competition conditions**

Under this condition a second stimulus was presented as soon as gaze fixation on the first stimulus had been obtained as in test condition 3. However, in this condition the first stimulus was withdrawn as soon as the second stimulus was fully visible.

#### **8.2.3. FGBM Protocol**

The FGBM protocol consisted of 16 counterbalanced trials incorporating an equal number of competition and no-competition presentations of stimuli and in which an equal number of faces and objects were presented. To ensure that the side upon which stimuli were presented did not have a significant effect on results, an equal number of faces and objects were presented on the left and right side. The resulting sequence of trials is represented in Table 7.1.

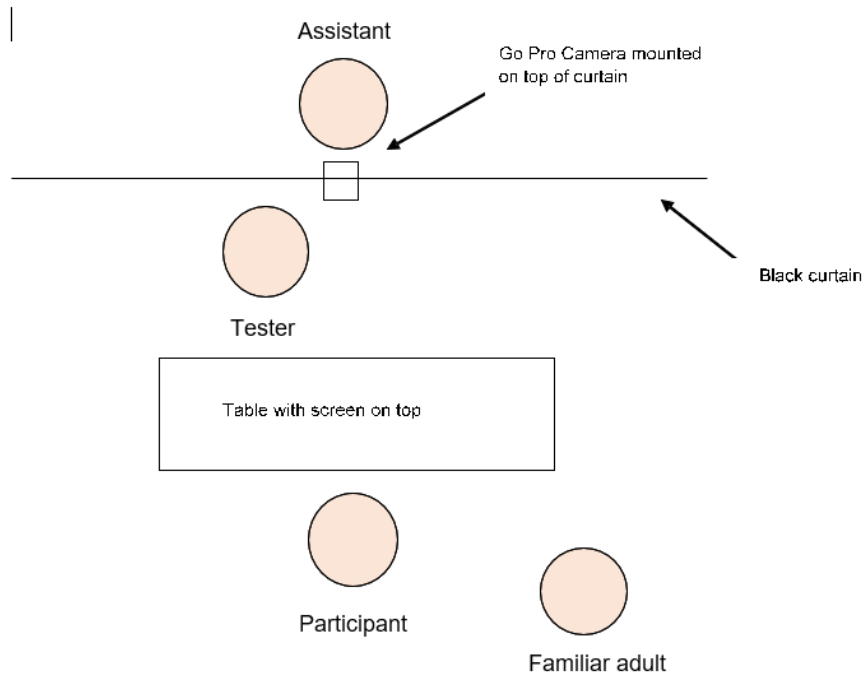
Table 7.1. Sequence of FGBM trials

| Trial number | Presentation   | Type of Stimulus and Position |                 |
|--------------|----------------|-------------------------------|-----------------|
|              |                | First stimulus                | Second stimulus |
| 1            | No Competition | Face (left)                   | Face (right)    |
| 2            | Competition    | Face (right)                  | Face (left)     |
| 3            | No Competition | Balloon (left)                | Balloon(right)  |
| 4            | No Competition | Face (right)                  | Balloon (left)  |
| 5            | Competition    | Balloon (right)               | Balloon (left)  |
| 6            | Competition    | Face (left)                   | Balloon (right) |
| 7            | No Competition | Balloon (right)               | Balloon (left)  |
| 8            | No Competition | Face (right)                  | Face (left)     |
| 9            | Competition    | Face (left)                   | Face (right)    |
| 10           | No Competition | Face (left)                   | Balloon (right) |
| 11           | Competition    | Balloon (left)                | Balloon (right) |
| 12           | Competition    | Balloon (left)                | Face (right)    |
| 13           | No Competition | Balloon (right)               | Face (left)     |
| 14           | No Competition | Balloon (left)                | Face (right)    |
| 15           | Competition    | Face (right)                  | Balloon (left)  |
| 16           | Competition    | Balloon (right)               | Face (left)     |

### 7.2.3. FGBM Procedure

The stimuli for the four test conditions of the FGBM were presented above a black screen which was situated on a table approximately 1.5 metres in front of the participant. The height of the cardboard screen could be adapted to ensure that the stimuli were presented at the participant's eye line. Stimuli were presented 60 cm apart from each other with the distance measured using the central point of each stimulus. The procedure was conducted in front of a black curtain hung from a garment rail in order to minimise visual distractions. A hole in the curtain, disguised with black mesh, allowed the assistant to view the participant during the procedure and carry out some live scoring.

The resulting room set-up can be seen in Figure 7.4.



*Figure 7.4. Room set-up for the FGBM.*

**Presentation of face stimuli**

For the presentation of face stimuli, the tester, who was crouching unseen behind the screen, would kneel up so her face appeared above it. Where two faces were presented, the assistant would move from behind the curtain to crouch behind the screen with the tester. Both tester and assistant would then kneel up to present their faces at the appropriate time.

**Presentation of object stimuli**

For the presentation of object stimuli, the tester behind the screen would raise a balloon until it appeared above the screen at the participant's eye level.

**Presentation of the second stimulus**

As soon as the participant fixed gaze on the first stimulus (face or object) the second stimulus was presented.

When the first stimulus was a face, the tester who was providing the face stimulus was aware when the participant had fixed gaze on her and either presented the second stimulus above the screen if it was a balloon or tapped the assistant who was also behind the table, signalling for her to also kneel up and present her face as second stimulus.

When the first stimulus was a balloon, the tester presented it from behind the screen so was unable to see when the child fixed gaze on it. Therefore, the assistant, who was observing from behind the curtain, would signal the appropriate moment for the presentation of the second stimulus using a quiet click sound produced by a clicking pen.

#### 7.2.4. Scoring the FGBM

Data obtained from the FGBM was scored from video recordings of the test sessions, with live scores taken during the sessions used to clarify any instances where the video data was unclear.

Each trial of the FGBM was given a score for gaze fixation, gaze shifting and the use of multiple gaze shifts dependent on the level of cuing which was required (see Table 7.2).

*Table 7.2. Scoring system for the FGBM*

| Gaze Fixation Scores         |   | Gaze Shift Scores                                 |    | Multiple Gaze Shift Scores                                      |   |
|------------------------------|---|---|----|---|---|
| No gaze fixation             | 0 | No gaze shift (continues to fixate on stimulus 1) | 0  | Single gaze shift from stimulus 1 to 2                          | 0 |
| Gaze fixation after DC + AC* | 1 | Gaze shift from stimulus 1 to 2 following DC + AC | 1  | Further spontaneous gaze shift(s) occur following initial shift | 1 |
| Gaze fixation after DC       | 2 | Gaze shift from stimulus 1 to 2 following DC      | 2  |   |   |
| Gaze fixation with no cue    | 3 | Gaze shift with no cue                            | 3  |   |   |
|                              |   | No fixation on stimulus 1                         | NF |   |   |

\*DC = Dynamic cue

AC = Auditory cue

Gaze fixation was defined as a look of more than 1 second at the first stimulus presented in each trial. Gaze fixation scores ranged from 0 to 3 and reflected the degree of dynamic or auditory cuing required to elicit fixation.

Gaze shift scores also ranged from 0-3 depending on the degree of cuing required to elicit a gaze shift from Stimulus 1 to Stimulus 2. A code of NF



(No Fixation) was applied to trials where gaze shifting could not be scored because the participant failed to fix gaze on stimulus 1.

A score for multiple gaze shifting was given for competition trials where both stimuli remained simultaneously visible. For these trials, participants would be given a score if they spontaneously demonstrated more than a single gaze shift between the two stimuli e.g. shifting their gaze from Stimulus 1 to Stimulus 2 and then back to Stimulus 1.

Finally, a code of NT (null trial) was applied to trials where participants became distracted or disengaged. For example, becoming preoccupied by something else in the room, closing their eyes or looking at their own body, so that they were unaware of the stimuli being presented over the screen.

### 7.3. Testing the FGBM on a typically developing reference group

Since the FGBM was a novel measure designed for this study, its effectiveness for eliciting the target behaviours of gaze fixation and gaze shifting was tested on a reference group of typically developing children. As a measure it was designed to assess early gaze behaviours and, thus, would not be appropriate for use with children who were matched to the PIMD sample in terms of chronological age. Therefore, the decision was taken to test it on a sample matched broadly in terms of developmental age to the PIMD sample.

#### **Design**

A cross-sectional design was adopted to test the FGBM on a small reference group of typically developing children.

#### **Ethical approval**

This part of the study was approved by the UCL ethics committee. Project number 7565/001 (ethics amendment).

#### **Recruitment**

Since people with PIMD are likely to function at a developmental level of around 18 months or younger (see section 2.1), the upper age limit for the pilot sample was set at 18 months, and the lower age limit at 6 months. The inclusion criteria specified this age range and required that children who took part were not known to have any developmental issues or sensory impairments.

Participants were recruited using convenience sampling. A day nursery was approached, as were contacts of the principal researcher known to have young infants. An information letter containing information about the purpose and nature of the study was given to the nursery who circulated it to parents of infants who met the inclusion criteria. The information letter was also given to contacts of the principal researcher who were asked to pass it on to any other parents who might be interested in their infants taking part. Any parents who expressed an interest in participating were asked to sign a

consent form and their infant was recruited to the study. Six participants were recruited overall, one through the day nursery and the remainder through contacts of the researcher.

### Participants

Six participants were recruited to take part in the testing of the FGBM (see Table 7.3).

*Table 7.3. Characteristics of participants taking part in the pilot study*

| Participant | Age in months | Gender | Location of testing |
|-------------|---------------|--------|---------------------|
| 1           | 12            | F      | University          |
| 2           | 10            | F      | Home                |
| 3           | 8             | F      | Home                |
| 4           | 7             | F      | Home                |
| 5           | 18            | F      | Nursery             |
| 6           | 15            | M      | Home                |

### Procedure

Participants attended one test session in which both the FGBM and the JA behaviour measure (to be reported in Chapter 8) were administered. Testing was carried out in a location which was chosen by parents as being most convenient to them. Four participants were tested in their own home, one was tested in a quiet room at the principal researcher's university and one was tested at her day nursery. Infants were seated on their parent's lap (or the lap of a familiar member of staff at the day nursery) during administration of the FGBM with the adult being instructed to remain quiet and neutral.

## **Results**

Five participants completed all the trials of the FGBM. One participant (P6) failed to complete the final four trials due to becoming distracted and leaving his mother's lap. These trials were omitted from scoring.

### **Gaze fixation**

Participants achieved the maximum score of 3 for gaze fixation on 91 of the 92 trials administered, fixing their gaze on all stimuli without the need for cuing. This represented near ceiling performance. One participant (P6) failed to demonstrate fixation on the final trial he completed before becoming distracted.

### **Gaze shifting**

Participants achieved the maximum score of 3 for gaze shifting on 88 (96%) of the 92 trials administered, shifting their gaze from Stimulus 1 to Stimulus 2 without the need for cuing. P1 scored 0 for gaze shifting on two trials and P4 scored 0 for gaze shifting on one trial. P6 was unable to achieve a gaze shifting score for his final trial since he had failed to fixate on Stimulus 1.

### **Multiple gaze shifts**

Eight trials which involved stimuli being presented in competition could be given a score for multiple gaze shifting. All participants demonstrated multiple gaze shifting on some of these trials with the mean score being 4.5. (range = 3-7). There was no apparent association between age and use of multiple gaze shifts with the youngest participant (P4) achieving a score of 7 and the oldest participant (P5) achieving a score of 4.

## **Conclusions**

Score for gaze fixation and gaze shifting were near ceiling for this reference group of typically developing infants confirming that the FGBM was an effective means of eliciting these visual behaviours in a typically developing sample with a developmental age of between 7 and 18 months.

## **7.4. Administration of the FGBM to participants with PIMD**

### **7.4.1. Introduction**

Testing on a reference group had established the FGBM to be an effective means of measuring gaze fixation and gaze shifting in a sample of typically developing children functioning within a similar developmental range to people with PIMD. For the next phase of the study it was administered to a sample of children with PIMD so that the relationship between their functional vision abilities and their use of gaze behaviour for JA could be explored.

### **7.4.2. Method**

#### **7.4.2.1. Design**

A cross-sectional design was used to measure gaze fixation and gaze shifting abilities demonstrated by a sample of children with PIMD using a novel measure, the FGBM, which was designed for the purposes of the study. Quantitative data was obtained from the measure and analysed using descriptive and statistical methods.

#### **7.4.2.2. Ethical approval**

The study was approved by the UCL ethics committee, project number 7565/001.

#### **7.4.2.3. Recruitment**

Recruitment was initiated through contacting two specialist residential schools for children aged 5-18 with PIMD and three community special schools for children with a range of learning disabilities in the south of England. These included one primary school, one secondary school and one school for children aged 5-18 years. Headteachers of the schools were sent a covering letter or email containing brief information about the study and an invitation to participate. All expressed an interest in participating and were provided with further information about the requirements of the project and inclusion criteria for participants. Inclusion criteria replicated those used in the exploratory work described in Chapter 5 and are reproduced in Table 7.4.

*Table 7.4. Inclusion Criteria for the main phase of study*

**Participants should:**

- be described as having a profound learning disability.
- have at least one additional sensory, motor or communication impairment e.g. physical disability, visual or hearing impairment, medical condition such as epilepsy.
- be pre-verbal i.e. should not use speech.
- have severely limited understanding of verbal language without additional cues.
- Be functioning at P-scale 3ii or below for English.
- have sufficient functional vision to respond to everyday items at school and at home.
- have sufficient functional hearing to be able to respond to another person speaking to them.

**Participants should not:**

- consistently use alternative formal means of communication e.g. signs/symbols.

Information letters and consent forms were then sent by schools to parents of students who met the inclusion criteria. No parents were contacted directly by the research team.

Twenty-one participants were initially recruited to the study, six from specialist schools and fifteen from community special schools. However, upon meeting two of the participants and talking to their teachers, it became clear that they did not, in fact, meet the inclusion criteria due to insufficient levels of vision. Both were only able to respond to changes in light and showed no awareness of objects. They were, therefore, excluded from the study. A further two participants attended one test session each but were unable to attend any further sessions due to illness and/or medical appointments. Given the known fluctuating levels of attention in this group

one test session would provide insufficiently reliable data for the purposes of the study, so these participants were also excluded. Seventeen participants were finally included in the study, six from specialist schools and eleven from community special schools.

#### **7.4.2.4. Participants**

Participant characteristics are represented in Table 7.5. To ensure that participants had sufficient visual acuity to engage with the FGBM, they were assessed by a specialist orthoptist using Cardiff or Keeler cards. Two participants (P16 and P17) could not be assessed by the orthoptist for operational reasons (the orthoptist being unable to travel to their school within the time frame of the study). Orthoptist assessment confirmed that the fifteen participants seen all had sufficient vision to engage with the FGBM. While some vision related issues were reported such as strabismus (misaligned eyes) and the tracking of objects these were not considered significant in relation to the demands of the FGBM task.

Table 7.5. Participant characteristics for the main phase of study

|           | Age   | Gender | Medical Diagnosis                                     | School type | GMFCS Level | MACS level | P-scale | Hearing  | Vison                                | Visual Acuity           |
|-----------|-------|--------|---|-------------|-------------|------------|---------|--|--------------------------------------|-------------------------|
| <b>P1</b> | 7:9   | F      | Rett Syndrome   | CSS*        | 4           | 5          | P3i     | No known issues  | No known issues                      | 50cm 0.8 logMAR         |
| <b>P2</b> | 12:1  | M      | Unspecified chromosomal abnormality                   | CSS         | 4           | 4          | P3ii    | No known issues  | No known issues                      | Keeler cards 0.9 logMAR |
| <b>P3</b> | 5:5   | F      | Aicardi-Goutieres syndrome                            | CSS         | 4           | 5          | P2ii    | No known issues  | Estropia (right eye)<br>Astigmatism. | 0.2 logMAR              |
| <b>P4</b> | 6:4   | M      | Wolf-Hirschorn Syndrome                               | CSS         | 4           | 4          | P2ii    | No known issues  | Long-sighted.<br>Wears glasses.      | 0.6 logMAR (corrected)  |
| <b>P5</b> | 13:0  | M      | Cerebral Palsy, microcephaly<br><br>Unknown aetiology | CSS         | 4           | 4          | P2ii    | Severe hearing deficit. Functional hearing with cochlear implant | No known issues                      | 1.0 logMAR              |
| <b>P6</b> | 14:10 | F      | Cerebral palsy.<br><br>Unknown aetiology.             | CSS         | 5           | 5          | P3      | No known issues  | No known issues                      | No response             |



Table 7.5. Participant characteristics for the main phase of study

|            | Age  | Gender | Medical Diagnosis   | School type | GMFCS Level | MACS level | P-scale | Hearing         | Vison                                | Visual Acuity           |
|------------|------|--------|---|-------------|-------------|------------|---------|-----------------|--------------------------------------|-------------------------|
| <b>P7</b>  | 8:7  | M      | Epilepsy.<br>Cerebral Palsy.                                | CSS         | 5           | 5          | P2ii    | No known issues | Has glasses,<br>purpose unknown.     | No response             |
| <b>P8</b>  | 3:5  | M      | Allan-Herndon-Dudley syndrome.                              | CSS         | 5           | 5          | EYFS    | No known issues | No known issues                      | No response             |
| <b>P9</b>  | 6:1  | M      | Epilepsy.<br>Microcephaly.                                  | CSS         | 3           | 2          | P3ii    | No known issues | No known issues                      | 0.4 logMAR              |
| <b>P10</b> | 15:4 | F      | PIMD Aetiology unknown.                                     | CSS         | 4           | 4          | P2i     | No known issues | Issues reported but pattern unknown. | No response             |
| <b>P11</b> | 15:9 | M      | PIMD.<br>Aetiology unknown                                  | SS          | 5           | 5          | P1ii    | No known issues | Short-sighted.<br>Wears glasses.     | 0.6logMAR               |
| <b>P12</b> | 17:0 | M      | Epilepsy.<br>Cerebral Palsy.<br><br>PIMD aetiology unknown. | SS          | 5           | 4          | P1ii    | No known issues | Short-sighted.<br>Wears glasses.     | Keeler cards 2.0 logMAR |

Table 7.5. Participant characteristics for the main phase of study

|            | Age   | Gender | Medical Diagnosis                       | School type | GMFCS Level | MACS level | P-scale | Hearing                                | Vison   | Visual Acuity          |
|------------|-------|--------|---|-------------|-------------|------------|---------|--|---|------------------------|
| <b>P13</b> | 10:3  | M      | Brain injury.<br>Epilepsy.              | SS          | 4           | 3          | P2i     | No known issues                        | Cortical visual impairment showing improvement. | No response            |
| <b>P14</b> | 16:4  | F      | Angelman syndrome.<br>Epilepsy.         | SS          | 5           | 4          | P2ii    | No known issues                        | No known issues                                 | 0.4logMAR              |
| <b>P15</b> | 15:11 | F      | Rett syndrome                           | SS          | 4           | 4          | P3      | No known issues                        | Wears glasses. Purpose unknown.                 | 0.9 logMAR (corrected) |
| <b>P16</b> | 16:5  | F      | Rett syndrome                           | SS          | 4           | 4          | P3ii    | No known issues                        | No known issues                                 | Not tested             |
| <b>P17</b> | 16:7  | F      | Pallister-Killian syndrome.<br>Epilepsy | SS          | 5           | 4          | P2i     | Moderate hearing loss, no hearing aids | Hypermetropia. Exotropia. Wears glasses         | Not tested             |

\*CSS = community special school    SS = specialist residential school for PIMD

#### **7.4.2.5. Researchers**

The FGBM was administered by the principal researcher assisted by two final-year students on the MSc. Speech and Language Sciences programme at UCL. These students were different to those who had assisted in the exploratory phase of the study. Both students had prior experience of working with children with PIMD.

#### **7.4.2.6. Procedure**

The FGBM was administered three times to each of the 17 participants meaning that 51 administrations of the measure were carried out overall. Each administration took place in a separate test session with sessions taking place at least one week apart. Each test session was carried out at a different time of day and took place in a suitable room at the participants' schools.

Administration of the FGBM took approximately 10-15 minutes. Participants were seated in their wheelchairs or supportive seating with the exception of P9 who was able to sit in a non-adapted chair. A familiar teaching assistant accompanied participants throughout the procedure. This teaching assistant was provided with information letter about the project if they had not previously seen one and advised to sit quietly to the side of the participant but to alert the tester if they observed any signs that the participant was unhappy or uncomfortable. If the participant did become distressed or disengaged, the session was temporarily discontinued while attempts were made to re-engage or settle them. On rare occasions when this was not possible, sessions were discontinued and administered at another time.

#### **Video recording**

Video recordings were made using a GoPro Hero4 camera mounted centrally on the top of the garment rail which held the curtain. The GoPro camera in this position provided an overhead 'fish-eye view', enabling both person and object to be seen clearly. Footage could be viewed on an iPad tablet which allowed for zooming in and frame-by-frame viewing so that gaze shifts and gaze direction could be analysed in detail. During the procedure the video was live-screened to an iPad situated under the table so that, during trials

where both researchers were behind the cardboard screen, they could check that the participant remained engaged.

#### **7.4.2.7. Scoring and Analysis**

Data obtained from the FGBM was scored from video with data taken from live scoring used to clarify scoring in instances where the video was unclear. The scoring system described in section 7.2.4. was used, with each participant being given a score between 0 and 3 for gaze fixation and for gaze shifting and a score of 0 or 1 for the use of multiple gaze shifts. Scores from all three testing sessions were combined and, as the number of trials completed by each participant varied (see section 7.4.3.2.), overall scores were expressed as a percentage of the total number of trials completed.

The resulting scores were analysed to identify patterns at a group and individual level.

### **7.4.3. Results**

As discussed in section 7.3.5. the FGBM assessed the ability of this sample of children with PIMD to fix and shift gaze between stimuli so that the impact of impairment in these areas on gaze behaviour used for JA could be measured. In reporting the results, inter-rater agreement will first be reported, followed by a consideration of trial completion rates. Group and individual level scores for gaze fixation and gaze shifting will then be presented followed by an analysis of the relationship between these functional vision skills and background measures of cognition and motor skills.

#### **7.4.3.1. Inter-rater agreement**

Data from 20% (10) of the 51 test sessions was scored by a second rater, one of the MSc. students, who had not been involved in administering any of the sessions she scored but was familiar with the scoring system. Inter-rater reliability for judgements about gaze fixation, gaze shifting and use of multiple gaze shifts was calculated using Cohen's kappa (see Table 7.6). Good levels of agreement were obtained for judgements in all three areas.

*Table 7.6. Inter-rater agreement for gaze fixation, gaze shifting and multiple gaze shifts scores on the FGBM*

| <b>Gaze Behaviour</b>              | <b>Kappa value</b>                             | <b>Level of Agreement</b><br>(Altmann, 1999) |
|------------------------------------|--|--|
| <b>Gaze fixation</b>               | <b>.797</b><br>95% CI (.751 - .848)<br>p <.001 | Good   |
| <b>Gaze shifting</b>               | <b>.662</b><br>95% CI (.60-.722),<br>p <.001   | Good   |
| <b>Use of multiple gaze shifts</b> | <b>.589</b><br>95% CI (.436 - .754)<br>p <.001 | Good   |

#### **7.4.3.2. Trial completion**

Each administration of the FGBM comprised 16 trials. Overall, 48 trials were administered to each participant (16 during each of the three test sessions). Of the total 816 trials thus administered, 105 (13%) were not completed – being coded as a null trial (NT) (mean per participant = 6.2, range = 0-31, S.D. = 8.1). There was a degree of individual variation with one participant, P17, completing only 17 (35%) of the trials administered. All other participants completed at least 31 (65%) of the trials administered to them. Null trials were excluded from subsequent analysis.

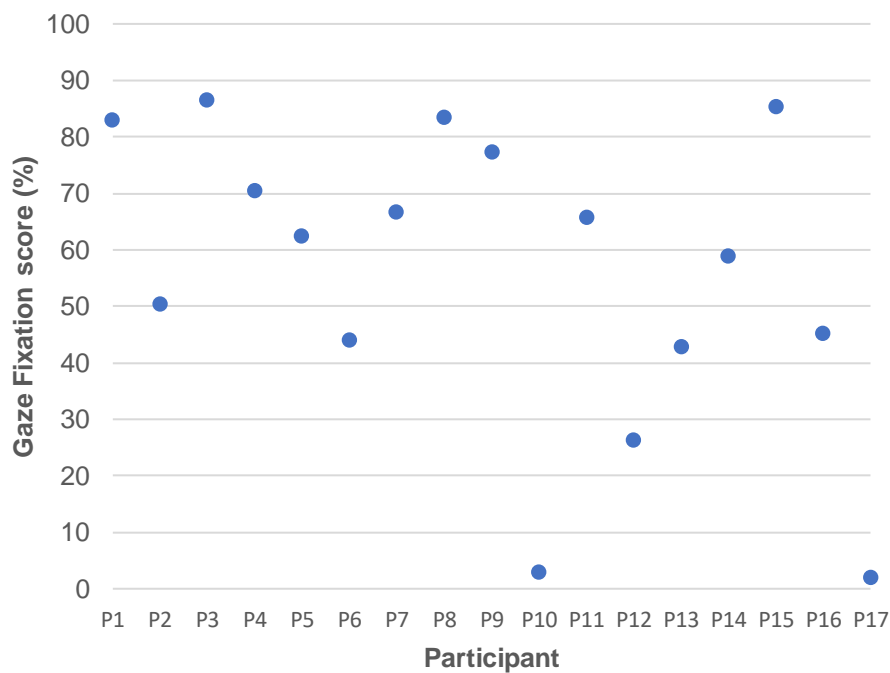
#### **7.4.3.3. Gaze fixation**

Overall, gaze fixation was demonstrated in 473 (67%) of the 711 completed trials.

As described in section 7.3.5., participants each received a score for gaze fixation which reflected the degree of cuing they had required. Scores were

calculated as a percentage of the total score they could have achieved given the number of trials they completed.

A mean gaze fixation score of 56% was achieved at group level (range = 2-87, S.D. = 26.5) with individual fixation scores represented in Figure 7.5.



*Figure 7.5. Participant scores for gaze fixation on the FGBM*

None of the participants achieved a score of 100% which would have indicated spontaneous fixation on all stimuli. However, four participants (P1, P2, P8 and P15) did achieve a score of over 80%. Two participants (P10 and P17) achieved particularly low scores of less than 5% which, in the case of P17, represented only one incidence of fixation.

### The effect of cuing on gaze fixation

Of the 473 trials in which gaze fixation was demonstrated, 337 (71%) involved spontaneous fixation on a static stimulus. In 79 (17%) trials, participants only fixed gaze following a dynamic cue and in a further 57 (12%) trials a dynamic and auditory cue was required before fixation occurred. The proportion of cued vs spontaneous fixation demonstrated by each participant can be seen in Figure 7.6.

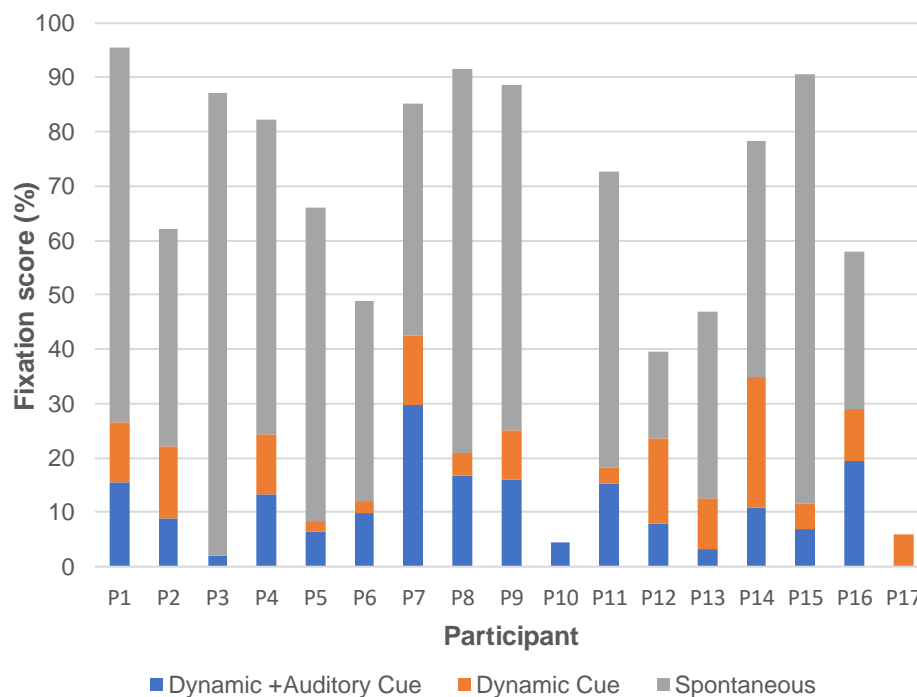


Figure 7.6. Fixation on static vs dynamic stimuli demonstrated by each participant on the FGBM

Whilst participants most frequently fixed gaze spontaneously, fixation rates did increase in response to cuing and it was notable that the two participants who achieved very low fixation scores (P10 and P17) only fixed their gaze on stimuli following a cue. Performance here contrasted with the reference



group of typically developing children who fixed their gaze on all stimuli and did not require any cues to do so.

### **Gaze fixation on faces and objects**

To investigate whether participants were more likely to fix their gaze on either faces or objects, mean fixation scores for both types of stimuli were compared. The mean fixation score for faces was 60.8% (range = 0-92.8, S.D. = 28.5) and the mean fixation score for objects was 50.5% (range = 2.9-85.5, S.D. = 28.6). Since scores for fixation on faces did not meet the assumption of normality (Shapiro-Wilk,  $p < .05$ ), a Wilcoxon signed ranks test was carried out to compare performance. Data from P17 was omitted from this calculation as it was based on only one incident of fixation and may have skewed results. The outcome of the Wilcoxon signed ranks test indicated that, at a group level, there was no significant difference in fixation scores for faces or objects ( $z = 1.647$ ,  $p = .10$ ).

Despite the lack of group difference in this area, descriptive analysis of individual participant responses suggests that ten were more likely to fixate on faces than objects with P2 and P13 showing a particularly strong tendency to do so (see Figure 7.7.).

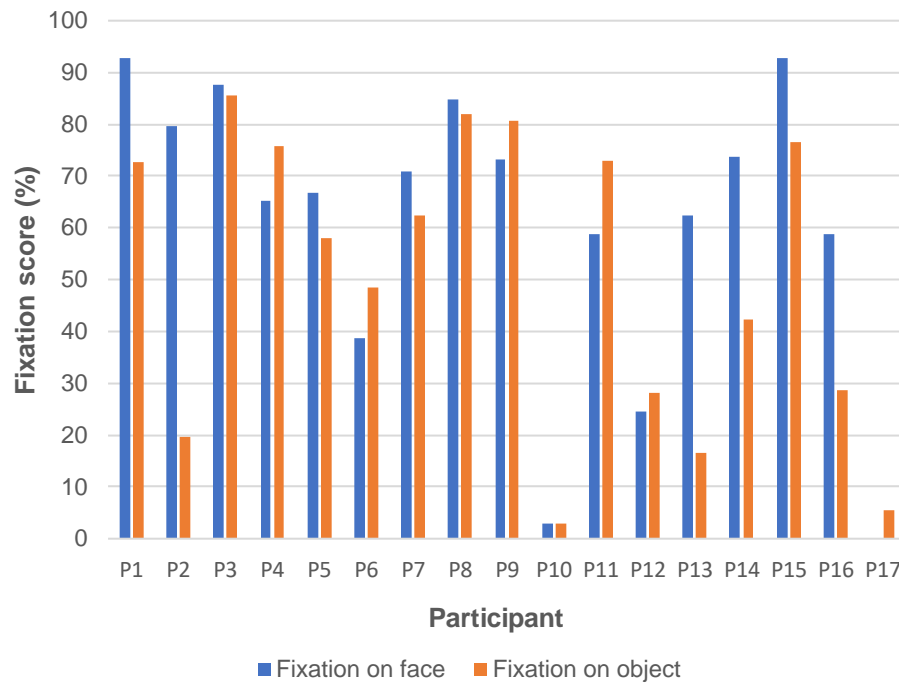


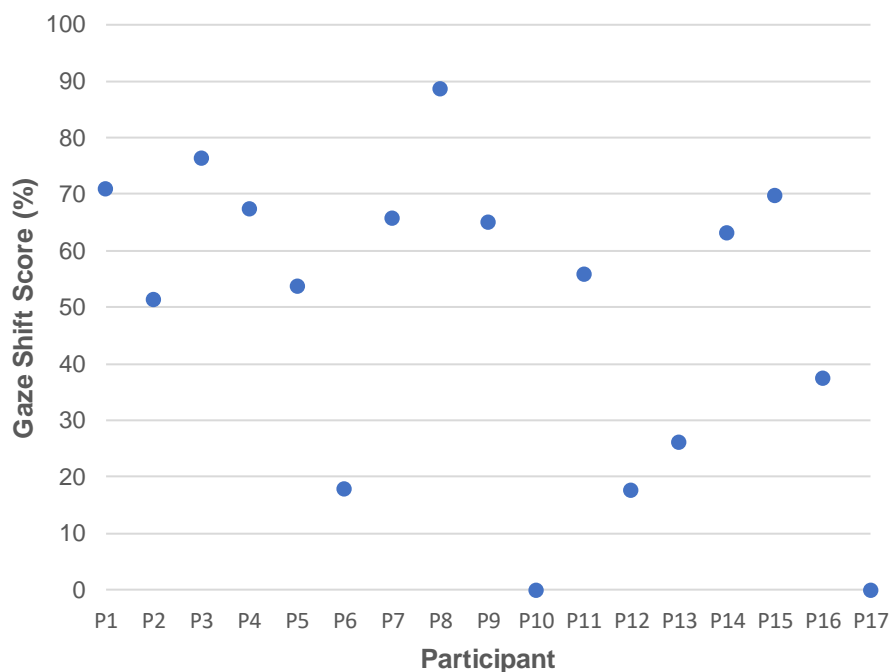
Figure 7.7. Proportion of gaze fixation directed towards faces vs objects

### The effect of left/right presentation on gaze fixation scores

FGBM trials had been counterbalanced to ensure that an equal number of targets were presented to the left and right hand side of the participant's midline. An independent samples t-test confirmed that the side on which the target was presented did not have a significant effect on fixation scores  $M = 1.4$ , 95% CI [-17.7, 20.6],  $t(32) = 0.154$ ,  $p = .94$ .

#### 7.4.3.4. Gaze shifting

Overall, participants demonstrated a gaze shift in 328 (46%) of the 473 trials in which gaze fixation on the first stimulus had occurred (competition and no-competition conditions combined). A mean gaze shifting score of 48.7% was achieved by the group (range = 0-89, S.D. = 27.1 with individual scores represented in Figure 7.8.

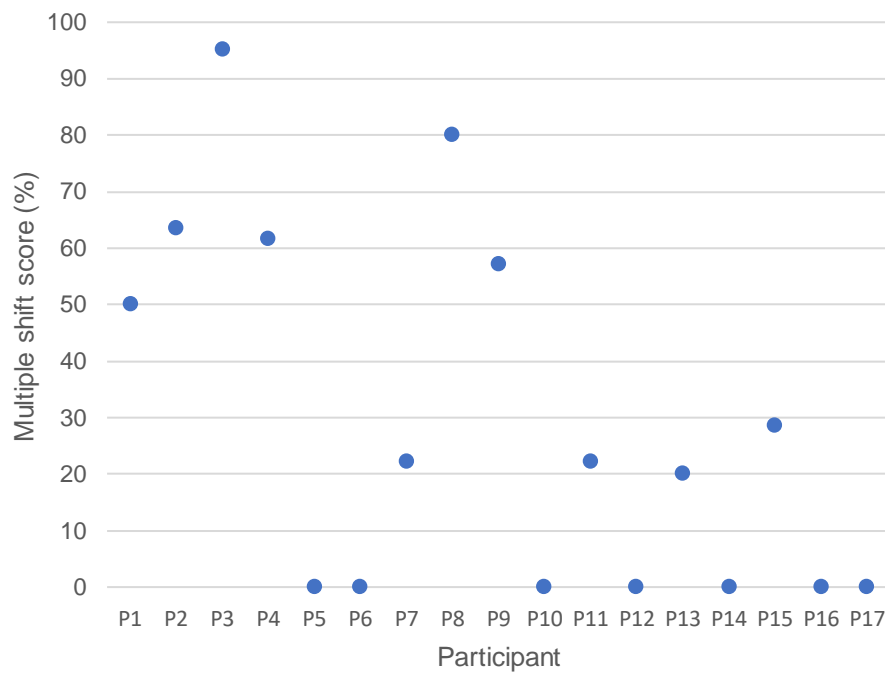


*Figure 7.8. Participant scores for gaze shifting on the FGBM*

Once again, none of the participants achieved a score of 100% which would have indicated spontaneous gaze shifting between stimuli on all trials. One participant (P8) achieved a score of over 80% but two participants (P10 and P17) failed to shift gaze on any trials. Again, this contrasted with the performance of the reference group of typically developing children who were able to shift their gaze between stimuli in 96% of trials without the need for cuing.

### Multiple gaze shift scores

The mean multiple gaze shift score for the group was 29.4 (range = 0-80, S.D. = 32.1) with individual participant scores presented in Figure 7.9.



*Figure 7.9. Participant scores for use of multiple gaze shifts on the FGBM*

While two participants (P3 and P8) achieved scores of over 80% for the use of multiple gaze shifts, seven did not demonstrate this skill at all.

### **The effect of competition on gaze shifting**

To test whether participants were more likely to shift gaze when stimuli were not in competition, mean gaze shift scores for 'competition' and 'no-competition' conditions were compared. The mean score for gaze shifting under the competition condition was 51% (range = 0-91, S.D. = 28.9) while the mean score for gaze shifting under the no-competition condition was 46.2% (range = 0-96, S.D. = 28.5). Since data met the assumption of normality (Shapiro-Wilk,  $p > .05$ ) a related samples t-test was used to compare these means. This indicated that there no statistically significant difference between them,  $t(16) = 1.058$ ,  $p = .31$  providing no evidence for an ongoing 'competition effect' in this group. Descriptive analysis of individual participant scores confirmed that the scores of the majority reflected the group level finding with only one (P5) demonstrating more gaze shifts when targets were not in competition.

#### 7.4.3.5. The relationship between scores for gaze fixation and gaze shifting

A correlation matrix was used to investigate whether there was a statistically significant relationship between the three measures of functional gaze behaviour scored by the FGBM (see Table 7.7). Since multiple gaze shift scores were not normally distributed (Shapiro-Wilk,  $p < .05$ ), a Spearman's correlation was used.

*Table 7.7. Correlation matrix comparing scores for gaze fixation, gaze shifting and the use of multiple gaze shifts on the FGBM*

|               | Gaze fixation | Gaze shifting     | Multiple gaze shifts |
|---------------|---------------|-------------------|----------------------|
| Gaze fixation |               | .970**<br>P=.0001 | .776**<br>p=.0001    |
| Gaze shifting |               |                   | .779**<br>P=0001     |

*Each cell shows the correlation coefficient (Spearman's rho) and the significance level.*

*\*\* = significant correlation at the .01 level*

Results indicated that scores for all three types of gaze behaviour were significantly positively correlated so that participants who were most likely to fix their gaze on stimuli were also more likely to shift gaze from one to the other and to spontaneously engage in multiple shifts back and forth between them when they remained present at the same time.

#### 7.4.3.6. Consistency of gaze behaviour across test sessions

The data analysis reported above was based on combined scores from all three test sessions, so further analysis was carried out to explore the extent to which participant performance varied between sessions. Since scores for gaze fixation and gaze shifting were highly correlated, these two variables were combined to provide a mean overall FGBM score for each of the three test sessions (see Table 7.8). Table 7.8. Mean FGBM score for each test session

| Test session | mean FGBM | range | S.D. | 95% confidence interval |
|--------------|-----------|-------|------|-------------------------|
| 1            | 49.2      | 0-93  | 28.1 | 34.7 - 63.6             |
| 2            | 54.7      | 3-96  | 27.5 | 40.6 - 68.8             |
| 3            | 50.9      | 0-95  | 29.8 | 35.6 - 66.1             |

A one-way ANOVA was conducted to determine if there was a significant difference between the mean FGBM scores for each test session. Data was normally distributed for each test session (Shapiro-Wilk,  $p > .05$ ), there were no outliers as assessed by boxplot and there was homogeneity of variance as assessed by Levene's test of homogeneity of variance ( $p = .842$ ). Results indicated no significant difference between mean FGBM scores for each of the test sessions  $F(2, 48) = 0.168$ ,  $p = .85$ ).

Although there was no significant group level difference, some participants did demonstrate marked variability in performance between sessions (see Figure 7.10) with, for example, P11 scoring 17% in test session 1 but 83% in test session 2.

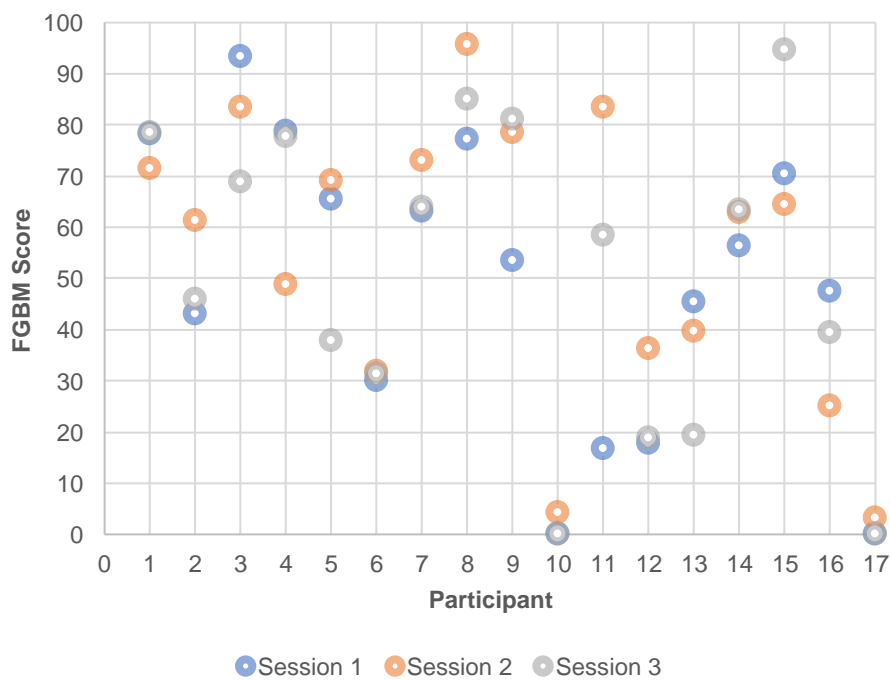


Figure 7.10. Participant FGBM scores for each test session

#### 7.4.3.7. The relationship between FGBM scores and gross motor skills

To explore the relationship between gross motor skills and JFGBM scores, participants were divided into two groups depending on their GMFCS level. The six participants with a GMFCS level of 5 were grouped together, as were the eleven participants with a GMFCS level of 3 or 4 (only one participant had a GMFCS level of 3 hence the merging of scores for these two GMFCS levels). An overall FGBM score was calculated by combining the percentage scores for gaze fixation and gaze shifting and dividing these by two.

An independent samples t-test was used to test the difference in FGBM scores between the two GMFCS groups since data was normally distributed



(Shapiro-Wilk,  $p > .05$ ) and there was homogeneity of variance, as assessed by Levene's test for equality of variance ( $p = .552$ ). Results indicated no significant difference in FGBM scores between the GMFCS level 3/4 group (mean = 51, range = 1.0– 1.6, S.D.= 29.1) and the GMFCS level 5 group (mean = 54.5, range = 22.1–86.0. S.D. = 23.8)  $t(15) = -0.25$ ,  $p = .81$ ).

#### **7.4.3.8. The relationship between FGBM scores and cognitive skills**

P-scales had been used as a measure of cognitive ability for this sample. Participants were functioning between p1ii and p3ii (see Table 7.5) for each participant's P-scale score) with two functioning at 1ii, three at p2i, five at p3ii, two at p3i and four at p3ii. P8 did not have a P-scale score since, due to his age (3:5), he had not yet begun to be assessed using this framework. He was, therefore, excluded from this analysis.

Participants were divided into a low P-scale and high P-scale group since the numbers of participants in individual p-scale groups was too small for robust statistical comparison to be carried out. The low P-scale group included participants at P-scales 1ii, 2i and 2ii ( $n = 10$ ) and the high P-scale group included participants at P-scales 3i and 3ii ( $n = 6$ ).

An independent samples t-test was used to test the difference in FGBM scores between the two P-scale groups since data was normally distributed (Shapiro-Wilk,  $p > .05$ ) and there was homogeneity of variance, as assessed by Levene's test for equality of variance ( $p = .448$ ). Results indicated no significant difference in FGBM scores between the low P-scale group (mean = 48.2, range = 1.0–81.6, S.D. = 29.0) and the high P-scale group (mean = 57.3, range = 30.1– 86.0, S.D. = 23.6) ( $t(14) = -6.10$ ,  $p = .551$ ).

#### **7.4.4. Discussion**

The FGBM provided a measure of the functional gaze behaviour in this sample of children with PIMD. Results indicated that, as might be expected, they showed deficits in their ability to fix and shift their gaze with none demonstrating these skills in all trials. This contrasted with the performance of a reference group of typically developing children, even the youngest of whom spontaneously fixed their gaze on all the stimuli presented and shifted their gaze between stimuli at near ceiling level, frequently engaging in multiple gaze shifts between stimuli.

The extent of observed deficits varied across the range of participants with some (P1, P3, P8 and P15) achieving both gaze fixation and gaze shifting scores of 70% or more, while others (P10 and P17) achieved a score of less than 5% for gaze fixation and did not demonstrate any gaze shifting at all. The spontaneous use of multiple gaze shifts, looking back and forth between stimuli, was particularly varied, with P1 and P3 doing so on more than 80% of occasions when the opportunity was presented whereas seven of the participants never engaged in this behaviour.

Although most instances of gaze fixation occurred spontaneously, all participants achieved higher scores when cues were used, in contrast to the reference group who did not required any cuing. The two participants who demonstrated the lowest FGBM scores (P10 and P17) were wholly dependent on cues. This may have implications for the use of gaze as a component of JA behaviour where gaze fixation and shifting occurs in response to an internal motivation (a desire to focus or share attention) rather than in response to extrinsic cues.

All participants, (with the exception of P17 who was virtually unable to fix gaze at all) fixed their gaze on both faces and objects, several being more likely to fix on faces than objects although this was not a statistically significant trend. This suggests that all should, functionally, be able to fix their attention on single referents regardless of whether they are people or objects – a key pre-requisite for JA although the extent of this ability varied between participants.

While existing research has suggested that some atypically developing populations, such as those with William's syndrome (Atkinson et al., 2003) demonstrate a persisting 'competition effect', finding it difficult to shift gaze between two stimuli which are simultaneously present, no persisting competition effect was found for the current sample of children with PIMD, all of whom were able to shift gaze between stimuli in competition (with the exception of P10 and P17 who did not demonstrate any gaze shifting). This is significant since gaze shifting, when used as a JA behaviour involves shifting attention between two, simultaneously present, referents. Therefore, the absence of a competition effect suggests that those participants who were able to shift gaze should have the functional ability to do so as part of JA behaviour and that, therefore, any deficits in their JA behaviours could not be attributed to functional vision impairment.

As noted, a wide range of functional gaze abilities was demonstrated by participants and it might be expected that this would be explained by their underlying degree of impairment as reflected by scores on measures of cognitive and motor ability. However, this was not supported by the evidence since no significant link was identified between P-scale or GMFCS scores and functional vision scores on the FGBM. This suggests that functional vision ability cannot be predicted on the basis of cognitive or motor ability and highlights the need for these skills to be independently assessed.

As a tool, the FGBM appeared to be an effective means of measuring of gaze fixation and shifting in children with PIMD. Scores for gaze fixation, gaze shifting, and the use of multiple gaze shifts were all significantly correlated suggesting that the measure was successful at assessing a single underlying competence in functional vision.

For the most part, participants seemed happy to engage in the procedure and appeared interested. However, the lack of social interaction involved in administration of the measure did mean that, if participants did lose interest at any point, the procedure had to be interrupted in order to attempt to re-engage them and re-engaging was not always possible. This led to a relatively high number of null trials compared to the typically developing reference group. As outlined in Chapter 2, fluctuating levels of arousal and

interest are common in people with PIMD and it can be challenging to engage them in any formal procedure so, in this context, the fact that 87% of the FGBM trials were successfully administered should be considered positive. The benefits of administering the measure on more than one occasion were clear from analysing performance across test sessions. While there was no group level evidence of significant variation which might indicate a possible learning effect across administrations, the performance of several participants did vary between the three testing sessions emphasizing the fact that single administrations of measures may not accurately reflect capacity in individuals with PIMD.

## **7.5. Conclusions**

On the basis of findings from the FGBM it could be concluded that the participants in this study demonstrated varying degrees of impairment in gaze fixation and gaze shifting which appeared to be idiosyncratic and unrelated to other abilities such as cognition and motor skills. Given that the typical developmental progression of JA involves gaze fixation on objects and people followed by the integration of this attention through gaze shifting it would be expected that impairments in functional gaze experienced by these participants would have an impact on their ability to engage in JA. Therefore, it was hypothesized that performance on the FGBM would be associated with performance on the structured probes designed to elicit JA.

## **8. Measuring JA behaviours in a sample of children with PIMD**

This chapter provides an account of how structured probes were used to measure JA behaviours in a sample of young people with PIMD. I begin by describing the development of these probes which was based upon the exploratory work described in Chapter 6 and the process of testing them on a sample of typically developing children. Next, I describe the development of a measure of JA in unstructured play which was used to compare performance in structured and less structure contexts. The administration of both measures to a sample of young people with PIMD is then described. Results are presented, firstly with reference to performance on the JABM, followed by the relationship between performance on this measure with performance on the measure of unstructured play and background measures. The impact of probe type on performance is then reported. Finally, the results will be discussed in relation to the research questions which were posed by this study.

## **8.1. The Joint Attention Behaviour Measure (JABM) – using structured probes to measure Joint Attention.**

Since the set of structured probes and their associated scoring system would constitute a measure of behaviours relating to JA this measure was labelled the Joint Attention Behaviour Measure (JABM).

### **8.1.1. Target behaviours to be measured by the JABM**

The JABM aimed to measure the target behaviours identified during the exploratory phase of the study as representing the developmental trajectory of JA (see section 6.1.1.). However, the exploratory phase of work had identified that subjective rater judgements of whether IJA had occurred achieved only moderate levels of inter-rater agreement. Therefore, a behavioural descriptor for IJA was included.

According to the accounts of typical pre-linguistic communication development discussed in Chapter 4, early attempts at initiating JA are signified by the combination of integrated attention (usually in the form of a gaze shift), with additional signals such as vocalisations, body movements or gestures. For example, an infant might look back and forth between a desired item and a caregiver whilst holding a hand out towards the item and vocalising (e.g. Bates et al., 1975). Therefore, the combination of a gaze shift alongside behaviours such as smiles, vocalisations or body movements may be considered a behavioural indicator of IJA. Since such behaviours may be idiosyncratic and depend partly on the physical abilities of individuals with PIMD it was not possible to list them definitively. However, following the example of Sigafos and colleagues (e.g. Sigafos et al., 2011) and Brady and colleagues (Brady et al., 2012), any behaviour which could potentially be used as a communicative signal was considered. For the purposes of this study the term ‘potentially communicative behaviour’ (PCB) was used to describe these behaviours.

According to Hobson and Hobson (2007), the presence of a ‘sharing look’ is another behavioural indicator of JA, engagement in sharing looks signifying a rich form of JA where both partners are aware of their mutual attention and

use it intentionally to share affect for social purposes. In a PIMD population where physical disabilities may limit the number of potentially communicative behaviours that can be produced, such sharing looks may be a particularly important indicator of declarative JA. Hobson & Hobson (2007) explored the use of sharing looks in a sample of children with autism, defining them as follows:

“...those looks directed to the tester that could be seen to express a participant sharing experience through interpersonal contact with the tester. They involved a deep gaze that conveyed personal involvement”

(Hobson & Hobson, 2007, p 418)

Clearly there is a subjective aspect to identifying such looks, with Hobson and Hobson's study identifying no measurable difference in terms of duration or other qualities, between sharing looks and other types of look, such as those they classified as checking looks. However, they did achieve high levels of agreement between the raters who scored the looks from videos of a structured interaction between participant and researcher, reporting that, in practice, this was “straightforward” (Hobson & Hobson, p 418).

With these potential behavioural indicators of IJA and intentional communication in mind, the additional target behaviour which was added to the JABM, was the behavioural descriptor of ‘gaze shifts accompanied by ‘additional signals’ which could be a PCB, a sharing look or a combination of both. To meet the criterion for this target behaviour, the PCB or sharing look had to be clearly produced in conjunction with the gaze shift rather than something which had been previously occurring.

For the purposes of the current study, a sharing look was defined as a look during which the child seemed to be sharing some enjoyment or other emotion with the communication partner. Gaze would be mutual, possibly accompanied by a PCB such as smiling and probably prolonged rather than brief but this was not a necessary criterion.

More subjective scoring categories of IJA(imp) and IJA(dec), as described during the exploratory phase, would be retained alongside the behavioural

criteria which were represented by the code AOP+ so that the relationship between these might be compared.

A summary of the final target behaviours to be measured by the JABM is represented in Table 8.1.

*Table 8.1. Target behaviours measured by the JABM*

|                         |  | <b>Target Behaviour</b>  | <b>Code</b> |
|-------------------------|--|--|-------------|
| <b>Gaze Behaviour</b>   | <b>Attends to a single referent</b>                    | Fixes visual attention on an object  | AO          |
|                         |  | Fixes visual attention on a person   | AP          |
|                         |  | Fixes visual attention separately on an object and on a person during the same trial   | AO + AP     |
|                         | <b>Integrates attention between objects and people</b> | Shifts gaze between an object and a person with no additional behaviours   | AOP         |
|                         |  | Shifts gaze between an object and a person whilst demonstrating a PCB and/or sharing look which was not previously occurring | AOP+        |
|                         | <b>Follows gaze</b>                                    | Follows the focus of another person's attention  | RJA         |
| <b>Rater judgements</b> | <b>Engages in JA</b>                                   | Engages in JA for <u>imperative</u> purposes (requesting or rejecting)   | IJA (imp)   |
|                         |  | Engages in JA for <u>declarative</u> purposes (sharing affect or interest)   | IJA (dec)   |



### **8.1.2. Communication partner behaviour during the JABM**

As discussed in section 4.2.5., the gaze behaviour of the communication partner can influence whether or not an individual engages in JA behaviours, with the direction of gaze being particularly important. It was reasoned that, if a participant shifted their gaze from an object to the tester who was already looking at them, they might be more likely to engage in JA than if they had to make active attempts to gain the tester's attention first. Therefore, tester gaze behaviour was manipulated during the procedure for each probe so that, in the initial phase of its administration, the participant would need to gain the attention of the tester to engage in JA, whereas, in the second phase the tester was looking at them, providing an optimal social context for JA to occur. Including both types of communication partner behaviour as part of the protocol for each probe ensured that there was maximum opportunity for JA to occur.

### **8.1.3. A description of the structured probes making up the JABM**

Five probes made up the final version of the JABM. The first, the Dynamic Object probe, which had been used successfully as part of the exploratory phase of study, was refined slightly for use in the JABM. The remaining probes were devised on the basis of information gained during the exploratory phase. Since the initiation of JA for declarative purposes had been the least observed behaviour during exploratory work, three probes were designed to elicit it for the main phase of the study. This would give participants as many opportunities as possible to demonstrate this behaviour in different contexts. In addition, a probe designed to provide a truly 'top-down' context for JA was included. A description of the development and nature of each probe is now given, followed by an account of the scoring system used for the JABM.

#### **8.1.3.1. Probe 1: Dynamic Object (1)**

**Aim of probe:** To elicit declarative JA in a 'bottom-up' context.

##### **Theoretical background to the probe**

The theoretical background to this probe was described in section 6.1.2.1. During exploratory work the Dynamic Object probe had been considered to have two phases – during the first phase an engaging stimulus was activated, creating an opportunity for declarative JA (sharing attention about an engaging object) and, during the second phase, the stimulus was left deactivated, creating an opportunity for imperative JA (requesting or rejecting the object). For the main phase of the study this was simplified by treating the two phases as two separate probes, termed Dynamic Object (1) and Dynamic Object (2), with the first designed to elicit declarative IJA and the second designed to elicit imperative IJA. This facilitated judgements about the function of any episodes of IJA (i.e. whether they were likely to have been for the purposes of sharing attention or requesting/rejecting) thus improving inter-rater agreement.

##### **Equipment**

The light Spinner, woodpecker, marble tree and plane described in section 6.1.2.1. were used as a standard set of test items for the probe.

##### **Procedure for the Dynamic Object (1) Probe**

The child is seated in their typical supportive seating (e.g. wheelchair) at an adjustable table opposite the tester. Test items are kept out of sight in a case under the table until needed. A test item is brought out and placed on the table slightly to the side of the participant and tester so that gaze shifting between the item and the tester can be observed. The tester activates the test item, focusing her gaze on it for the initial period of its activation (approximately 30 seconds). She then turns to look at the participant for the remaining period during which the item is activating.

During the procedure the tester avoids smiles or animated facial expression, appearing mildly interested but neutral, also avoiding verbal comments or sounds. If the participant initiates declarative JA, this is acknowledged

through appropriate facial expression and positive noises or brief comments e.g “mm, that’s a funny sound!”.

Since the probe is administered four times during each test session (see section 7.1.4. below), each test item is presented once. However, flexibility is allowed to reflect the preference of the participant i.e. if they seem to have a strong preference for one item over the others (as established during the session of ‘unstructured play’ preceding administration of the JABM) this item may be used in all trials.

### **Scoring of the Dynamic Object (1) probe**

Scoring begins when the test item is activated and ends when it deactivates. The scoring system used for all the probes is described in section 7.1.5., below.

#### **8.1.3.2. Probe 2: Dynamic Object (2)**

**Aim of probe:** To elicit imperative IJA in a bottom-up context

#### **Equipment**

Equipment is as listed for the Dynamic Object (1) probe.

#### **Procedure for the Dynamic Object (2) Probe**

As the test item deactivates, the tester looks back at it and continues looking at it while it remains deactivated, creating an opportunity for the participant to initiate JA for the purposes of requesting. After 30 seconds, she turns to look at the participant to facilitate any attempts to engage in imperative JA.

The test item should remain in place for approximately one minute then be put away. However, if the participant appears to request the item, the tester should acknowledge this by reactivating it one more time.

### **Scoring of the Dynamic Object (2) probe**

Scoring begins when the test item deactivates and ends when the tester reaches to pick it up and put it away.

### **8.1.3.3. Probe 3: Surprise Puppy Probe**

**Aim of probe:** To elicit declarative IJA in a top-down context.

#### **Theoretical background to the probe**

The 'pencil tin' probe which was designed to create a top-down context for eliciting JA in the exploratory phase of study had not proved to be effective and it was possible that this probe had not created a true top-down situation with participants assuming that the tester could see the tin, even though she ignored it.

The Surprise Puppy probe was devised as an alternative and was based on an activity previously described by (McLean, McLean, Brady, & Etter (1991) (see section 5.1.3.). In this activity, an alligator toy is lowered from the ceiling behind the tester's back, providing an interesting and unexpected event which can only be seen by the participant and not the tester thus creating an opportunity for the participant to direct the tester's attention to it and share enjoyment. It had not been feasible to implement such an activity during the exploratory phase of the study due to the difficulty in setting it up in rooms provided by schools. However, the protocol for the FGBM (which was administered at the same time as the JABM) necessitated the use of a curtain hanging on a garment rail to provide a plain, dark background to stimuli. The presence of this curtain allowed a similar activity to the 'alligator' activity to be created.

#### **Equipment**

A 'puppy on a lead' toy was used.

#### **Procedure for the 'Surprise Puppy' probe**

The tester engages the participant's attention through talking or singing to them. Once the participant's attention is engaged, the assistant (who is behind the curtain) dangles the puppy over the top of the curtain until it is hanging slightly behind the tester and over her shoulder. A participant's view of the probe can be seen in Figure 8.1.



*Figure 8.1. Participant view of the Surprise Puppy probe*

The tester continues to look at the participant while talking or singing, to appear as if she is unaware of the puppy. After 30 seconds she turns her head, looks at the puppy, smiles and says “oh, it’s a puppy!”, then looks back at the participant. The puppy is withdrawn after approximately one minute. If the participant makes attempts to direct the tester’s attention to the puppy, the tester turns her head to look at it, makes a surprised face, and then looks back at the participant saying “oh, look, it’s a puppy!”.

The side on which the puppy is presented is alternated for each trial.

### **Scoring of the Surprise Puppy probe**

Scoring begins as soon as the puppy becomes visible over the curtain and ends when it is withdrawn.

#### **8.1.3.4. Probe 5: The Phone probe**

**Aim of probe:** To elicit declarative IJA in a familiar, bottom-up context.

#### **Theoretical background to the probe**

This probe was based on two previously reported activities which have been used to elicit declarative IJA in participants with ASD or PIMD. In a 'remote control toy' activity used by Summers & Impey (2011), a static toy on the floor is suddenly activated by remote-control and, in a 'moving hand' probe described by McLean et al. (1991), a mechanical hand on a shelf behind the tester is activated by a hidden switch (see section 5.1.3.). For the current probe, the stimulus used was a mobile phone situated on the table which begins to ring during the test session. It was reasoned that participants might be familiar with such a context and could use their previous experience to understand that the adult might want to look at the phone and pick it up, thereby creating a naturalistic situation for them to direct the attention of the tester to the phone.

#### **Equipment**

A mobile phone which was placed on the table at the beginning of the test session.

#### **Procedure for the Phone probe**

Following the end of the previous trial, the tester looks down at a pad on her knee and begins to write on it. As she is writing, the assistant, who is behind the curtain, rings the phone. The tester ignores the phone for the first four rings and continues writing, she then looks up at the participant for the final four rings of the phone.

After eight rings the tester answers the phone, shrugs and says, "nobody there". If the participant makes attempts to direct the tester's attention to the phone, she responds to these by saying "oh, my phone" and answering it.

At the end of the trial the tester places the phone back on the table, alternating the side on which it is situated.

### **Scoring for the Phone probe**

Scoring begins when the phone rings and ends when the tester turns to reach for it.

#### **8.1.3.5. Probe 5: The Look at the Ball probe**

**Aim of probe:** To elicit RJA

#### **Theoretical basis of the probe**

This probe was based on a procedure described by Senju & Csibra (2008) who used it successfully to elicit gaze following in typically developing infants from the age of six months. In their study, infants were shown a video of an adult who looked straight ahead, then raised their eyebrow and said “look” before turning their head and looking down at one of two objects on a table. For the current study the procedure was adapted to be carried out in real life, rather than on a video since the ability of people with PIMD to process images on a screen is uncertain. This procedure was chosen, firstly, because stimuli were presented within a relatively close visual field to the participant and one to which they had already been attending since stimuli for the Dynamic Object and Phone probes were also presented on the table top. Secondly, the procedure was operationally feasible since it did not require stimuli to be placed on walls (as in the ESCS) or held by another person (as in the Look at the Puppet probe used during the exploratory phase of this study).

#### **Equipment**

Two transparent balls filled with liquid and glitter were used. The balls were an equal size of approximately 10 cm in diameter although the colour of the glitter inside them was different.

#### **Procedure for the Look at the Ball probe**

The tester engages the participant's attention using strategies such as calling their name and/or wagging her finger in front of her own face. Once eye contact is established, she says “look” then turns her head and looks down at one of the balls. After looking at the ball for a few seconds she picks it up, and shows it to the participant, shaking it slightly to show the glitter swirling.

### **Scoring for the Look at the Ball probe**

Scoring began when eye contact was established with the participant and ended when the tester reached to pick up the ball.

#### **8.1.4. Protocol for the JABM**

In summary, the JABM consisted of the following five probes:

- 1) Dynamic Object (1)
- 2) Dynamic Object (2)
- 3) Surprise Puppy
- 4) Phone
- 5) Look at the Ball

All five probes were administered four times during each test session constituting twenty trials overall. Probes were interspersed with each other with none being administered twice in a row and trials were balanced so that stimuli were presented an equal number of times to the left and right. The resulting protocol and score sheet for the JABM can be seen in Appendix F.



### **8.1.5. Scoring and analysis of the JABM**

#### **Scoring of RJA behaviours**

RJA behaviours were elicited and measured by one probe – the Look at the Ball probe. Four trials of this probe were administered during each of the three test sessions, meaning that twelve trials of the probe were administered to each participant overall. Each trial was given a score of either NF, 0 or 1 for RJA as follows:

**No fixation (NF)** = the participant does not make eye contact with the tester for long enough for the trial to be effectively administered. i.e. the participant is not looking at the tester's eyes when she shifts her gaze.

**0** = the participant is making eye contact with the tester when she shifts her gaze but does not look towards the same object as the tester.

**1** = the participant is making eye contact with the tester when she shifts her gaze and follows this focus of gaze by looking towards the same object as the tester.

#### **Scoring of IJA behaviours**

The term 'IJA behaviours' is used here to include all target behaviours except RJA. The presence of these behaviours was measured using data from all probes except the Look at the Ball probe i.e. the Dynamic Object (1), the Dynamic Object (2), the Phone and the Surprise Puppy probes. Four trials of each of these four probes were administered in each of the three testing sessions completed by participants meaning that each participant completed 48 trials which were scored for target IJA behaviours. Scoring and analysis of these trials was carried out in two phases.

In the first phase, each trial was placed in one of the three following categories:

**1) No fixation**

The participant does not fix their gaze on any particular stimulus (i.e. their gaze may shift around in a fleeting manner, perhaps generally looking around the environment).

**2) Fixation on single referent**

The participant fixes their gaze on an object and/or person at least once during the trial but does not integrate their attention by shifting their gaze between the two i.e. there will be a period of disengagement from one referent before gaze is fixed on another.

**3) Gaze shifting**

The participant shifts their gaze from an object to a person or vice versa at least once during the trial. There must be at least one 'object-person' or 'person-object' shift and there may be more (e.g. the participant may demonstrate an 'object-person-object' shift or continue looking back and forth between object and person).

Each trial was placed in the category representing the most advanced type of gaze behaviour observed during its administration. Therefore, if a participant demonstrated several instances of fixation on an object during a trial but also shifted their gaze between that object and a person on one occasion, the trial would be categorised as 'gaze shifting'.

For the second phase of scoring, each trial was given a score for JA behaviour dependent on the category into which it had been placed as follows:

**No fixation trials** – these were not given a score for JA behaviours.

**Fixation on single referent trials ('fixation trials')** – for trials in this category, participants were given a score of 1 for one of the following behaviour patterns:

**a) Attention to object (AO)**

Participant only fixes gaze on object during the trial.

**b) Attention to person (AP)**

Participant only fixes gaze on a person during the trial.

**c) Attention to object and to person (AO + AP)**

Participant fixes their gaze on both an object and a person at different times during the trial but disengages their gaze between these separate instances of fixation.

**Gaze shifting trials** – for each these trials, participants were given a score of 1 for one of the following behaviours:

**a) Integrated Attention to Object and Person (AOP)**

Gaze shifting occurs (as described under the 'gaze shifting' category above) and is not accompanied by any communicative signals.

**b) Integrated Attention to Object and Person plus Communicative signals (AOP+)**

Gaze shifting occurs and is accompanied by a communicative signal in the form of a PCB, a sharing look or a combination of the two. The signal must clearly be produced in conjunction with a gaze shift rather than occurring continuously.

The most complex gaze behaviour was scored, with AOP+ considered to be more complex than AOP.

### **Further analysis of AOP+ gaze shifts**

Trials scored as AOP+ were subject to further analysis being given a score of 0 or 1 for the occurrence of IJA where:

**0** = Participant demonstrates a gaze shift between person and object but this is *not* judged to have a communicative function.

**1** = Participant demonstrates a gaze shift between person and object and this *is* judged to have a communicative function.

Trials coded as IJA were then given an additional score for either IJA(imp) or IJA(dec) defined as follows.

#### **IJA(imp) = IJA for imperative functions**

An AOP+ gaze shift is used to request a desired item or reject a disliked item.

#### **IJA(dec) – IJA for declarative functions**

An AOP+ gaze shift is used to draw an adult's attention to an engaging item and share affect about it for social purposes.

### **Scoring two and three-point gaze shifts**

All gaze shifting trials (scored as AOP or AOP+) were also given a score for being either a two-point or three-point shift, defined as follows:

**Two-point shift** – the participant shifts their gaze from one referent to another but does not shift their gaze back to the first referent.

**Three-point shift** – the participant shifts their gaze from one referent to another and then back to the first referent. They may continue looking back and forth between the referents multiple times.

Trials were given a score for the most complex gaze shift pattern seen so that, for example, if a participant demonstrated three two-point gaze shifts and one three-point gaze shift during a trial, it would be given a score for three-point gaze shifting.

## 8.2. Testing the JABM on a typically developing reference group

### Introduction

As with the FGBM, the JABM was designed to measure behaviours occurring during the first months of typical development and so it would not have been appropriate to test it on children matched with the PIMD sample in terms of chronological age. It was, therefore, tested on the same reference group of typically developing children as the FGBM. The aim of this work was to ensure that the JABM was effective in eliciting the target behaviours in a typically developing sample of participants.

### Design

This work used a cross sectional design to test performance on the JABM in a small group of neurotypical children.

### Ethical

The study was approved by the UCL ethics committee. Project number 7565/001 (ethics amendment).

### Recruitment

Participants were recruited using convenience sampling from day nursery in southern England (see section 7.3 for details).

### Participants

Six participants were recruited to the reference group (see Table 7.3 for details).

### Procedure

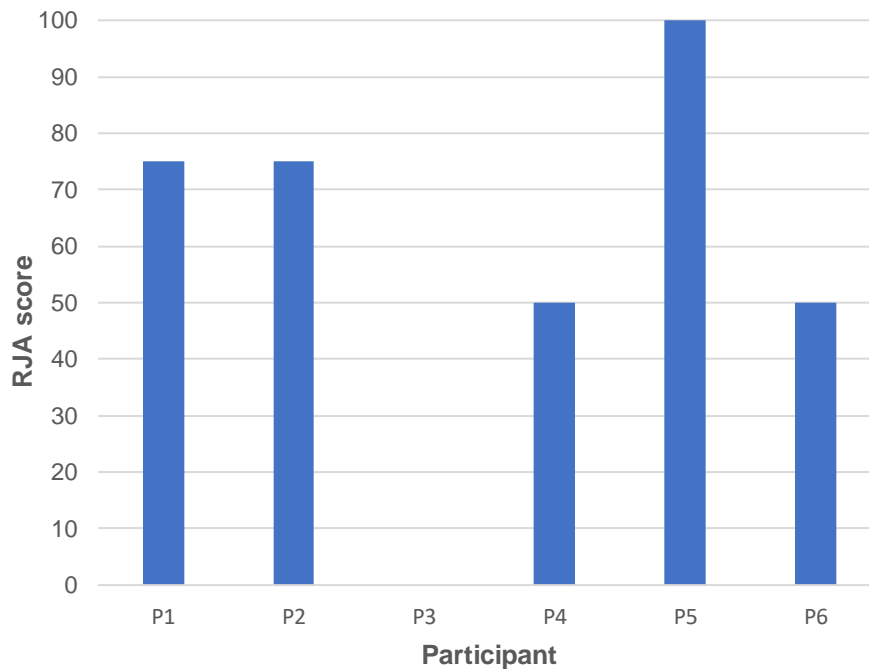
The JABM was administered during one test session carried out at the university or at the participant's home or nursery (see section 7.3). It was administered after the FGBM since it was considered to be a more engaging procedure and more likely to retain participant interest if they were tiring or becoming distracted. As with the FGBM, infants were seated on their parent's lap (or the lap of a familiar member of staff at the day nursery) with the adult being instructed to remain quiet and neutral.

## **Results**

All six participants engaged with all the trials of the JABM which were administered to them. However, practical issues (lack of phone signal) meant that it was not possible to administer the Phone trial to P6. These four trials were omitted from scoring meaning that an equal number of trials was not administered for each probe. To address this, a percentage score was calculated to reflect the proportion of trials for each probe in which target behaviours were demonstrated. For the purposes of testing on the reference group, scores for two and three-point gaze shifts were combined. Scores for IJA(imp) and IJA(dec) were also combined into a single score for IJA.

## RJA behaviours

One probe – the Look at the Ball probe – was designed to elicit RJA, with a total of 24 trials of this probe being administered (four to each participant). RJA in the form of gaze following was demonstrated in a total of 67% of these trials (mean = 58.3, range = 0-100, S.D. = 34.2). There was a possible association between age and performance with the two oldest participants P5 (who was aged 18 months) and P6 (who was aged 15 months) achieving ceiling scores, P4 (who was aged 7 months) achieved a score of 50% and P3 (who was aged 8 months) did not demonstrated any gaze following (see Figure 8.2).



*Figure 8.2. JABM RJA scores for the typically developing reference group*

## IJA behaviours

Out of the 92 trials administered which were designed to elicit IJA behaviours, 35% were scored as AOP+ (gaze shifts accompanied by communicative signals) (mean = 35.8%, range = 12.5 – 62.5, S.D. = 22.3) and IJA was judged to occur in 27% (mean = 28.1, range = 6.3-50, S.D. = 16.6).

Of the remaining trials 38% were scored as AOP (unaccompanied gaze shifts) (mean = 37.2, range = 12.5 – 43.8, S.D. = 17.7) and 25% were scored as attention to a single referent (mean = 25.0, range = 12.5-37.5, S.D. = 8.8) with 12% being scored as AO (attention to object only) (mean = 12.5, range = 6.3-18.8, S.D. = 7.9) and 13% being scored as AP (attention to person only) (mean = 12.50, range = 0-18.8, S.D. = 7.91). 'No fixation', in which the participant was generally looking around the room, was scored in 2% of trials (mean = 2.08, range = 0-6.3, S.D. = 3.2). Individual profiles of performance are represented in Table 9.2.

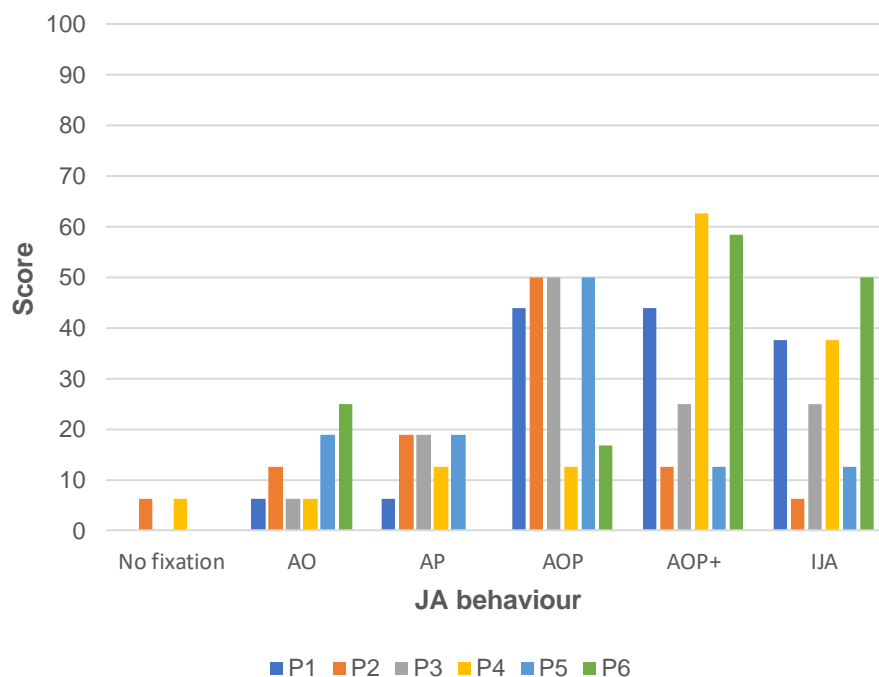


Figure 8.3. JABM IJA scores for the typically developing reference group



Results indicated that all four probes were successful in eliciting gaze shifts which were accompanied by communicative signals and represented communicative IJA although these did not occur in all trials

Probes varied in the extent to which they elicited IJA behaviours, with the Surprise Puppy trial eliciting the highest frequency of all types of IJA behaviour and the Phone trial eliciting the least. There was some variation in performance between participants, however, with P4 demonstrating AOP+ in all trials of the Phone probe.

There was no apparent association between age and demonstration of IJA behaviours, with the youngest participant (P4) who was aged 7 months, achieving higher scores than the oldest participant (P5) who was aged 18 months.

## **Conclusions**

Testing of the JABM on a typically developing reference group indicated that all probes were effective in eliciting JA behaviours. Patterns of JA behaviour similar to those described in the observational studies of Bates et al. (1975) were commonly seen in response to the probes. For example, P6 (aged 15 months) looked back and forth between the surprise puppy and the tester whilst pointing and vocalising persistently. Although JA behaviours were not elicited in all trials, with the most advanced (AOP+ and IJA) being least commonly demonstrated, this may reflect individual differences in capacity for these behaviours, given the age of participants since the youngest would not be expected to have developed these skills fully. It is also questionable whether any structured sampling procedure could elicit target JA behaviours on all trials since the aim is to create opportunity rather than demand responses. The Phone trial elicited the lowest number of JA behaviours, but a decision was made to retain it as part of the JABM since it did elicit some JA behaviour and was particularly effective in doing so for one participant.

### **8.3. A measure of Joint Attention behaviours in unstructured play**

#### **8.3.1. Aims of the unstructured play measure**

The unstructured play measure was developed to investigate the JA behaviours demonstrated by participants in a less structured and, therefore, more naturalistic context which included a familiar communication partner. Data elicited by this measure would be used to explore the relationship between performance in the structured context of the JABM and performance in this more naturalistic context. The unstructured play measure focused on measures of IJA behaviours since exploratory work had indicated that contexts eliciting RJA were an infrequent occurrence in everyday situations and trying to introduce opportunities for it would risk imposing a structure on a session which was designed to be participant-led.

Since the unstructured play measure was administered immediately before the JABM it also served as a 'warm-up' activity during which participants could become accustomed to the situation and the tester. The test items for the Dynamic Object (1) probe were used as part of the unstructured play measure, this also allowed the tester to gauge which proved most motivating for each participant.

#### **8.3.2. Development of the unstructured play measure**

The nature of the physical disabilities experienced by the sample of children with PIMD in this study meant that they were unable to engage in truly 'free' play since only one participant was independently able to reach and grasp objects or engage physically with the environment. Therefore, this measure involved play which was initiated by an adult but continued in a minimally structured, flexible manner which was led by the child's behaviours. For example, if a child showed a positive response to an object and tried to move their hand towards it, the adult could move the object closer to them so that they could feel it or could help them explore it with hand-over-hand assistance. In one case, a participant enjoyed moving her head close to the soft, foam propeller of the plane and stopping it with her nose so this was

continued as a game with the adult repeatedly activating the plane and responding positively with smiles and laughing each time the participant stopped the propeller.

The unstructured play session included the familiar adult who accompanied each participant to test sessions. Due to time limitations of the study it was not feasible to train all the familiar adults on the requirements of the session in order for them to carry it out independently and so it was led by the tester who would encourage the familiar adult to take a turn at engaging the participant in play with each test item.

### **8.3.3. Protocol for the unstructured play measure**

#### **Equipment**

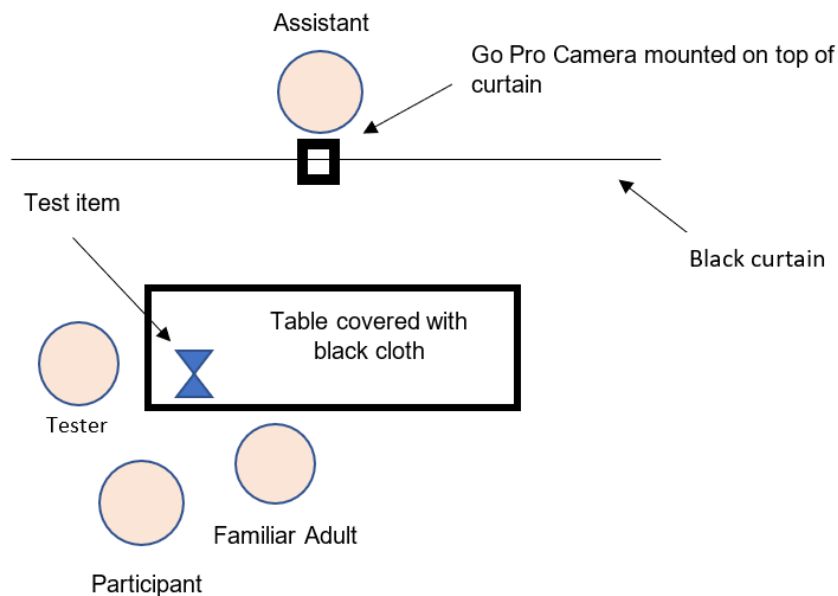
The four standard test items used in the JABM (the light spinner, woodpecker, marble tree and plan) were used to engage participants in play.

#### **Communication partners**

Both the assessor and a familiar adult (FA) were potential communication partners during this measure. The FA was the person who had accompanied the participant to their session – usually a member of teaching staff or, in one case, a family member.

## Room set-up

The room set-up for the unstructured play measure can be seen in Figure 8.4. The environment was arranged so that each test item was presented to the participant on the table in front of them. Both the tester and the familiar adult were seated close to the child so that the participant was able to look at either of them. The assistant viewed the procedure from behind the curtain and carried out some 'live-scoring' of the session.



*Figure 8.4. Room set-up for the unstructured play measure*

## Recording

Video recordings of all test sessions were made using a GoPro Hero4 camera mounted centrally on the top of the garment rail which held the curtain.

### **Procedure for the unstructured play measure**

At the beginning of the session, the familiar adult was asked if they would be happy to 'join in' with this part of the session. They were reminded that the session was being videoed and gave written consent for this to occur. Familiar adults were instructed that this part of the session was an informal time to 'have a play' and to engage with the participant as they would normally do.

Test items were then introduced one at a time. Each test item was initially introduced and demonstrated by the assessor, who then passed it on to the familiar adult so that they could operate it and engage the participant.

The session was continued until all of the test items had been engaged with or sooner if participants lost interest.

#### **8.3.4. Scoring and analysis of the unstructured play measure**

The unstructured play measure was scored from video recordings with live scores taken by the assistant during the session used to clarify scoring in instances where the video was unclear.

The length of each session varied between 4:02 minutes and 6:35 minutes, depending on how long it took to present all the test items. To ensure equitable comparison, only the first 4 minutes of each session, starting from presentation of the first test item, were scored. All participants had engaged with at least three of the test stimuli within this period of time.

Since the purpose of the unstructured play measure was to explore the relationship between IJA behaviours demonstrated during the JABM with those demonstrated in a less structured context, the same target behaviours were scored and analysed as follows (see Appendix G for the unstructured play measure score sheet).

### **Scoring gaze fixation on the unstructured play measure**

Scoring every incidence of fixation on people and objects during unstructured play sessions would have been highly time intensive so, given that gaze fixation was not the prime focus area for this study, a broad measure of gaze fixation was taken, with each participant being given a score of 0-2 for AO and AP for each administration of the unstructured play measure, as follows:

#### **AO**

0= no gaze fixation

1 = fixes on an object less than five times during the session

2 = fixes on an object more than five times during the session

#### **AP**

0= no gaze fixation

1 = fixes on a person less than five times during the session

2 = fixes on a person more than five times during the session

### **Scoring gaze shifting and IJA on the unstructured play measure**

Every incidence of gaze shifting which occurred during the four-minute unstructured play sessions was analysed and subject to the same scoring system as gaze shifts occurring in trials of the JABM so that each shift was categorised as either AOP or AOP+. All shifts were also given a score for being either two-point or three-point in nature. All AOP+ shifts were given a further score for 0 or 1 to reflect whether they were judged to signify JA for a communicative function. Those that were given a score of 1 were further categorised as either IJA(imp) or IJA(dec).

## **8.4. Administration of the JABM and the unstructured play measure to participants with PIMD**

The JABM and the unstructured play measure were administered to the same participants who had completed the FGBM (see Chapter 7).

### **8.4.1. Method**

#### **8.4.1.1. Design**

A cross sectional approach was used to examine JA abilities in children with PIMD. Two measures were administered on three occasions each. The first measure was the JABM (a set of structured probes designed to elicit JA behaviours) and the second was a measure of JA in unstructured play. Scores on the JABM were compared to performance on the measure of JA in unstructured play, FGBM scores and background measures of cognition, motor skills and communication.

#### **8.4.1.2. Ethical approval**

The study was approved by the UCL ethics committee, project number 7565/001.

#### **8.4.1.2. Recruitment**

Participants were those who had completed the FGBM, see section 7.4.2.3. for details of recruitment.

#### **8.4.1.3. Participants**

See section 7.4.2.4. for participant characteristics.

#### **8.4.1.4. Procedure**

The JABM and the unstructured play measure were administered to participants in three test sessions, at least one week apart. Each test session was carried out at a different time of day and took place in a suitable room at the participants' schools. The unstructured play measure was administered before the JABM and the FGBM was administered in the same session (see section 7.4.2.5.).

Administration of the unstructured play measure took approximately 4-7 minutes and administration of the JABM took approximately 15 minutes. Participants were accompanied by a familiar adult throughout the procedure.



## 8.5. Results

I will begin this section by presenting results obtained from the JABM, describing the profiles of JA behaviour demonstrated by participants at a group and individual level. This will include analysis of scores for gaze behaviour, IJA and RJA, and the use of two-point and three-point gaze shifts. I will then explore the relationship between performance on the JABM and other measures including performance on the FGBM and background measures of cognition, communication and motor skills. Finally, I will consider aspects of the data relevant to the methodology used in this study, comparing the frequencies of target behaviours elicited by each probe, reporting on consistency of performance across test sessions and comparing performance on the JABM with performance on the unstructured play measure.

### 8.5.1. Performance on the JABM

#### 8.5.1.1. Imputation of missing data

The JABM consisted of 20 trials (representing four trials each of five probes) which were administered to 17 participants on three occasions. This meant that a potential 1,020 trials could be carried out overall. However, 39 (4%) of these potential trials could not be administered and were classified as null trials (see Table 8.2 for a breakdown of null trials by participant).

*Table 8.2. Number of null trials on the JABM by participant*

| <b>Participant</b> | <b>Number of null trials</b> | <b>Null trials as a percentage of possible trials.</b> |
|--------------------|------------------------------|--|
| <b>P1</b>          | 1                            | 2%   |
| <b>P2</b>          | 8                            | 13%  |
| <b>P3</b>          | 0                            | 0%   |
| <b>P4</b>          | 0                            | 0%   |
| <b>P5</b>          | 1                            | 2%   |
| <b>P6</b>          | 0                            | 0%   |
| <b>P7</b>          | 0                            | 0%   |
| <b>P8</b>          | 0                            | 0%   |
| <b>P9</b>          | 0                            | 0%   |
| <b>P10</b>         | 0                            | 0%   |
| <b>P11</b>         | 0                            | 0%   |
| <b>P12</b>         | 10                           | 17%  |
| <b>P13</b>         | 0                            | 0%   |
| <b>P14</b>         | 0                            | 0%   |
| <b>P15</b>         | 0                            | 0%   |
| <b>P16</b>         | 18                           | 30%  |
| <b>P17</b>         | 1                            | 2%   |
| <b>Total</b>       | <b>39</b>                    | <b>4%</b>  |

The two principal causes of null trials were as follows:

- 1) **Practical issues** – these included equipment failure. For example, 10 out of the 39 null trials were caused by failure of the mobile phone to ring during the trial due to loss of charge or poor signal. Where these issues could not be resolved, the Phone trials were not administered for the remainder of that session.
- 2) **Discontinuation** – the missing data for P16 and P12 (a total of 28 out of the 39 null trials) was caused by the need to discontinue a session part-way through due to the participant being tired and no longer able to engage. Wherever possible, discontinued trials were repeated on another occasion but, for practical reasons, this was not possible for these participants.

Missing data is a common feature of quantitative research and, although it is felt that having 5% or less missing data is unlikely to lead to statistical bias (Bennett, 2001; Schafer & Graham, 2002), it needs to be dealt with in a systematic manner since most statistical analyses are designed to be carried out on a full data set (Dong & Peng, 2013). Given the relatively small amount of missing data here, the decision was made to impute the missing values using a systematic process based on the participant's scores on completed trials.

### **Imputation Method**

It was reasoned that each participant might show a similar response pattern to each probe. Therefore, missing data for each probe was imputed based on their existing scores for performance on that measure using the following process.

For each probe containing missing data, the participant's average score for completed trials of that probe was calculated. This was then multiplied by the number of null trials for that probe and the total added to their overall score for performance on the probe.

#### **8.5.1.2. Inter-rater agreement**

Video recordings from 20% (10) of the test sessions in which the JABM was administered were selected at random and scored by a second rater (= 200 trials). The second rater was one of the MSc students assisting with the project who was familiar with the scoring system but had not participated in any of the sessions she was scoring. Inter-rater agreement for judgements about the occurrence of target behaviours was calculated using Cohen's kappa (see Table 8.3).

Table 8.3. Inter-rater agreement for scoring of target behaviours on the JABM.

| Type of Gaze behaviour                         | Kappa Value                                       | Level of agreement<br>(Altmann, 1999) |
|--|---|---------------------------------------|
| <b>AO</b>                                      | <b>.708</b><br>95% CI (.615 -.801)<br>$p < .0005$ | Good                                  |
| <b>AP</b>                                      | <b>.659</b><br>95% CI (.567 -.751)<br>$p < .0005$ | Good                                  |
| <b>AOP</b>                                     | <b>.68</b><br>95% CI (.566 -.794)<br>$p < .0005$  | Good                                  |
| <b>Gaze shift category</b><br>AOP(2) or AOP(3) | <b>.646</b><br>95% CI (.545 -.747)<br>$p < .0005$ | Good                                  |
| <b>AOP+</b>                                    | <b>.704</b><br>95% CI (.596 -.812)<br>$p < .0005$ | Good                                  |
| <b>Presence of PCB</b>                         | <b>.69</b><br>95% CI (.572 -.808)<br>$p < .0005$  | Good                                  |
| <b>Presence of Sharing Look</b>                | <b>.287</b><br>95% CI (.154 -.420)<br>$p < .0005$ | Fair                                  |
| <b>Presence of communicative IJA</b>           | <b>.29</b><br>95% CI (.157 -.423)<br>$p < .0005$  | Fair                                  |
| <b>IJA Category</b><br>(Req or Dec)            | <b>.33</b><br>95% CI (.195-.465)<br>$p < .0005$   | Fair                                  |
| <b>RJA</b>                                     | <b>.68</b><br>95% CI (.533-.827)<br>$p < .0005$   | Good                                  |

Agreement was considered good for most target behaviours but only fair for judgements about the presence of sharing looks and IJA.

#### **8.5.1.3. Results relating to RJA behaviour**

The seventeen participants each completed a total of 12 trials of the Look at the Ball probe which was designed to elicit RJA in the form of gaze following. As a group, participants followed the direction of the tester's gaze in 45 (22%) of these 204 trials, with mean score per participant being 2.7 out of a possible 12 (range = 0-8, S.D. = 2.1). Participants failed to follow the tester's gaze in 96 (47%) trials with a further 63 (31%) trials being scored NFP since participants did not engage in sufficient eye contact with the tester for the trial to be administered.

At an individual level, fourteen out of the seventeen participants demonstrated some gaze following in response to this probe (see Figure 8.5). However, none demonstrated gaze following in all of the trials with only two (P3 and P15) doing so in six or more of the twelve trials administered to them. Three participants (P7, P8 and P14) failed to demonstrate any gaze following and a further four (P1, P10, P16 and P17) only did so in only one trial. The incidence of NFP varied across participants and was particularly frequent for P10, P16 and P17, occurring in over half of the trials administered to these participants.

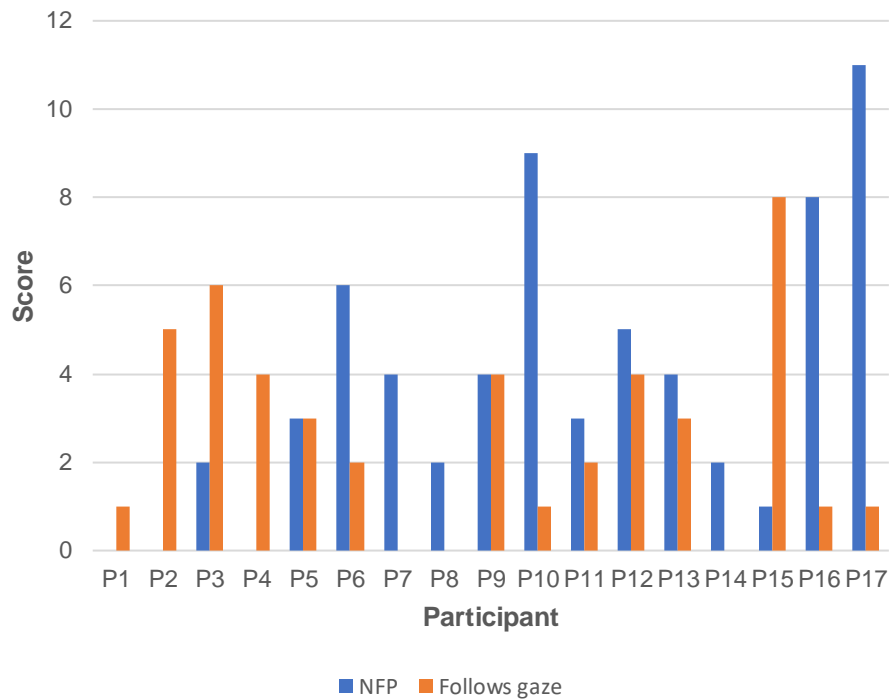


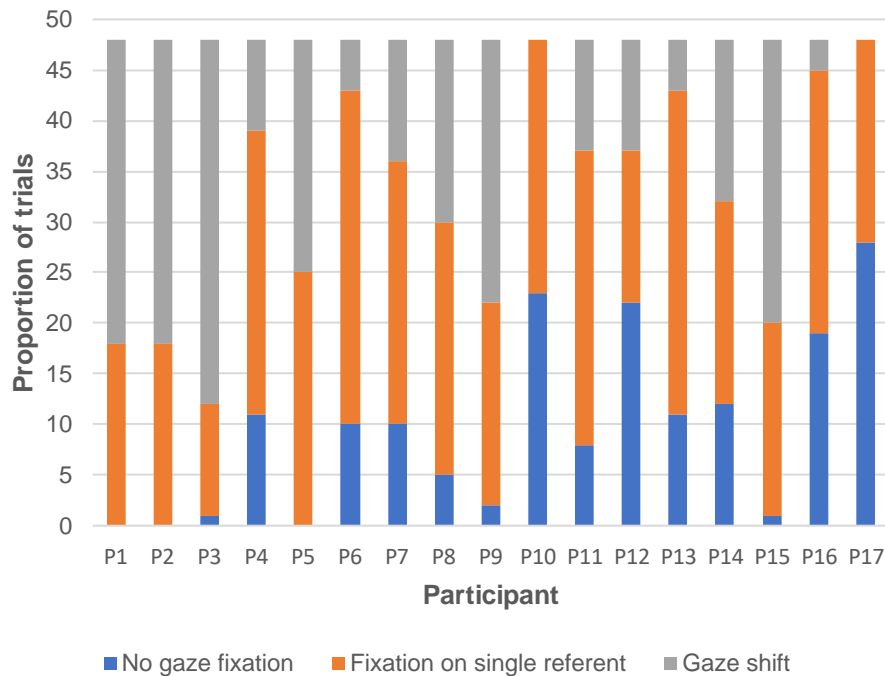
Figure 8.5. Participant scores for RJA on the JABM.

#### 8.5.1.4. Results relating to IJA Behaviour

The JABM included four probes designed to elicit behaviours relating to IJA. Four trials of each probe were administered during each of the three test sessions completed by participants. Therefore, the total number of trials during which IJA behaviours could be demonstrated totalled 816.

As outlined in section 8.1.5., each trial was placed into one of three categories according to the most complex gaze behaviour demonstrated – no fixation, fixation on a single referent or gaze shifting. The most commonly demonstrated trial type was fixation on a single referent, with 391 (48%) of trials being placed in this category (mean = 23, range = 11-33, S.D. = 6.0). Gaze shifts were demonstrated in 263 (32%) trials (mean = 15.5, range = 0-36, S.D. = 11.5) and 162 (20%) trials (mean = 9.6, range = 0-28, S.D. = 8.9) were categorised as no fixation.

Individual patterns of participant gaze behaviour are represented in Figure 8.6.



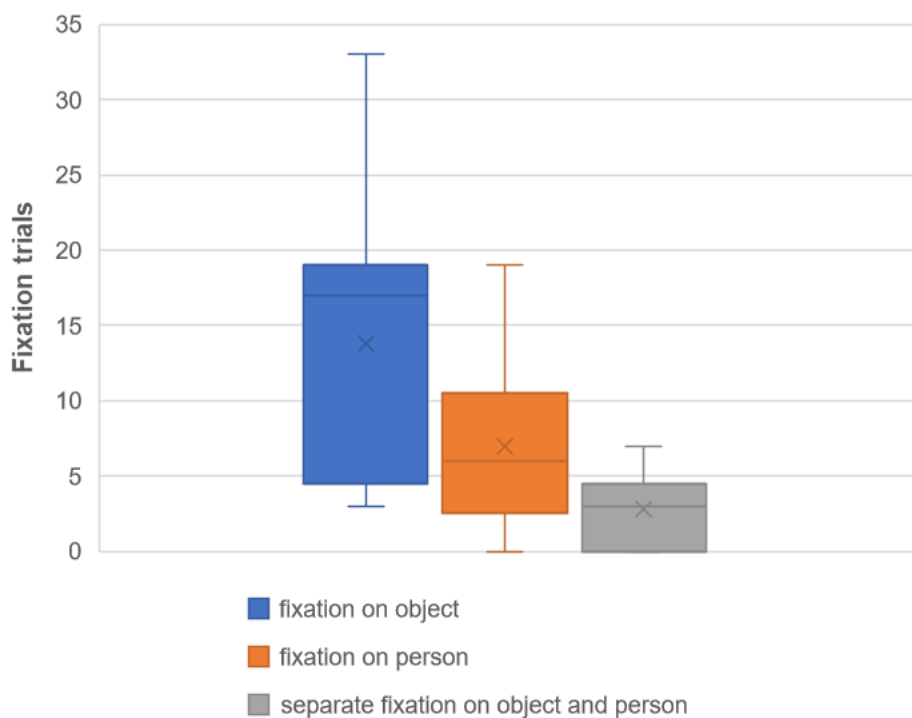
*Figure 8.6. Proportion of trials in which participants demonstrated each type of gaze behaviour (no gaze fixation, fixation on a single referent or gaze shifting).*

Not all participants followed the group pattern, with some (P1, P2, and P3) demonstrating a higher proportion of gaze shifting than either attention to a single referent or no fixation.



## Fixation Trials

The 391 trials in which participants demonstrated only gaze fixation on single referents were further analysed to explore whether this fixation was more likely to be focused on an object (AO), on a person (AP) or whether participants demonstrated separate incidences of attention to both object and person during the trial (AO+AP) (see Figure 8.7).

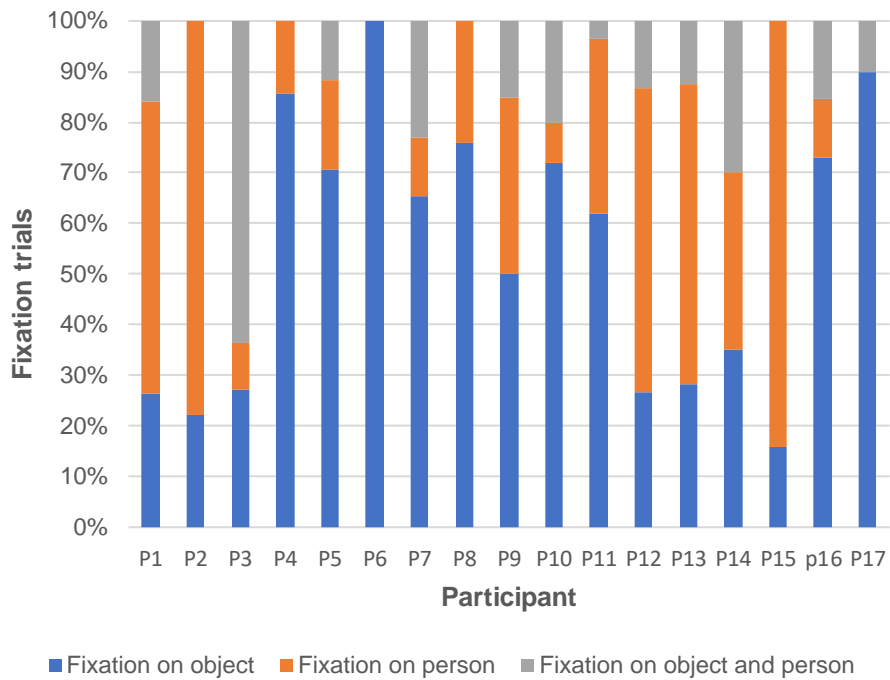


*Figure 8.7. Focus of fixation in trials in which only fixation on single referents occurred.*

As a group, participants were most likely to fixate on objects during fixation trials (total = 235, mean = 13.8, range = 3-33, S.D. = 9.0) followed by fixing on a person (total = 118, mean = 6.9, range = 0-19, S.D. = 5.6) with separate fixation on both objects and people being least frequently demonstrated (total = 47, mean = 2.8, range = 0-7, S.D. = 2.4).

Not all participants followed this group pattern, with six participants (P1, P2, P3, P9, P12 and P15) fixing on a person in a higher proportion of fixation trials (see Figure 8.8). It was notable that five of these participants who spent

a higher proportion of fixation trials attending to a person, also demonstrated the highest scores for gaze shifting (AOP and AOP+ combined) and the highest scores for IJA.



*Figure 8.8. Percentage of fixation trials in which participants fixed attention on a person, on an object or on both an object and a person.*

### Gaze shifting trials

Of the 263 trials categorised as 'gaze shifting', 145 (18% of all trials) were categorised as AOP (mean = 8.5, range = 0-22, S.D. = 6.3) and 118 (15% of all trials) were categorised as AOP+ (mean = 7.9, range = 0-25, S.D. = 8.5).

Thirteen participants demonstrated at least some AOP+ gaze shifts, with P2, P3 and P9 achieving the highest AOP+ scores. Two participants (P14 and P16) only demonstrated AOP gaze shifts and a further two participants (P10 and P17) did not demonstrate any gaze shifting at all (see Figure 8.9).

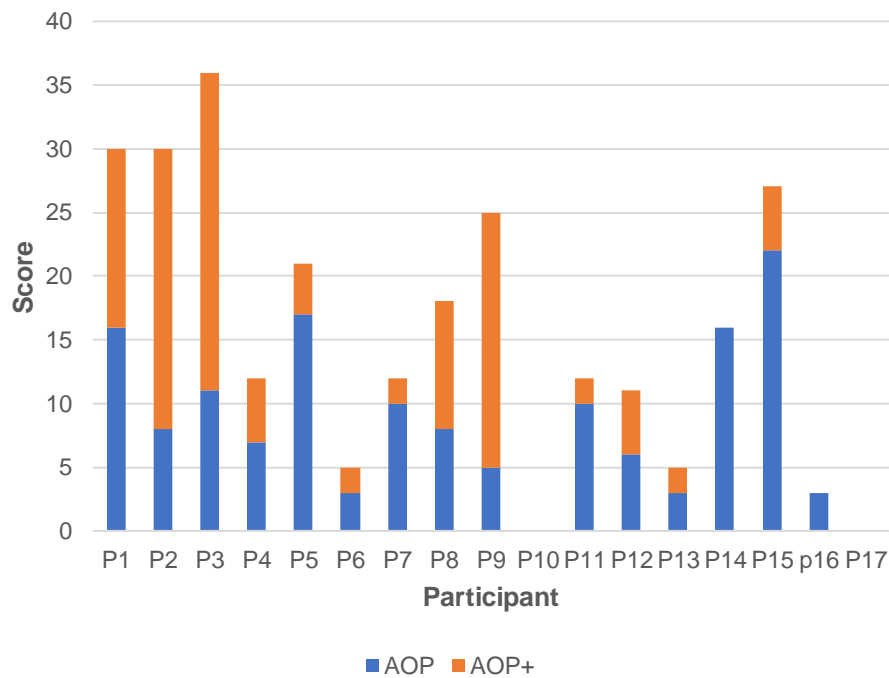
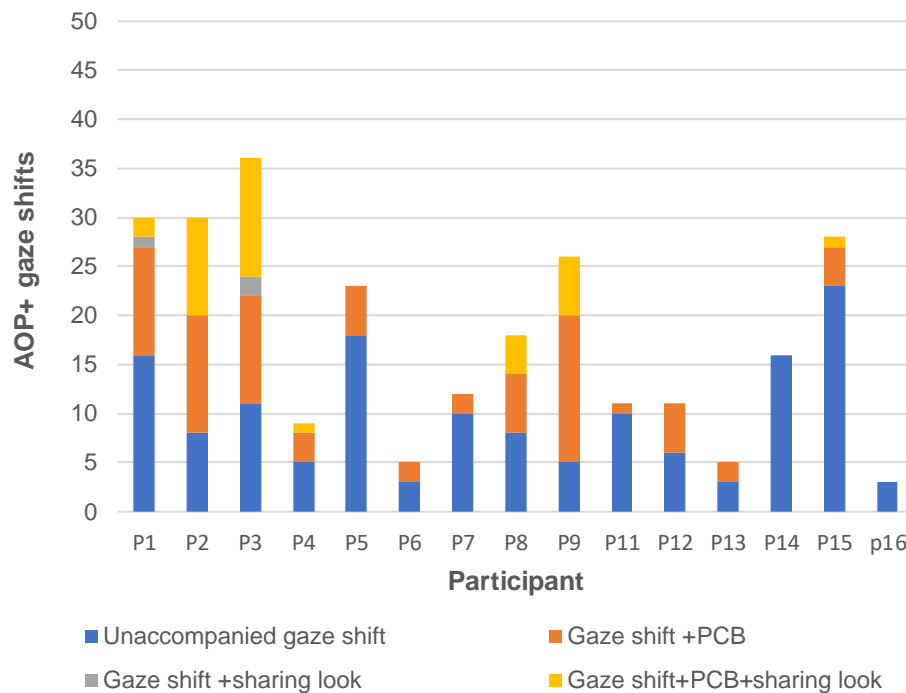


Figure 8.9. AOP and AOP+ gaze shift scores for each participant.

In order to explore the type of additional signals participants used to accompany their AOP+ gaze shifts, the frequency of AOP+ shifts which incorporated a PCB, a sharing look or a combination of the two was compared. Since P10 and P17 had not demonstrated any gaze shifts their data was excluded from this analysis (see Figure 8.10).



*Figure 8.10. Number of AOP+ gaze shifts accompanied by either a PCB, a sharing look or a combination of the two demonstrated by each participant.*

The most commonly used communication signals were PCBs which accompanied gaze shifts in 79 (66%) of the 119 AOP+ trials (mean = 5.3, range = 0-15, S.D. = 4.8). Thirty-six (30%) AOP+ trials were accompanied by a combination of both a PCB and a sharing look (mean = 17.5, range = 0-12, S.D. = 3.9) whilst only 3 were accompanied by a sharing look alone (mean = 0.2, range = 0-2, S.D. = 0.6). There was a significantly positive association between the frequency of gaze shift trials demonstrated overall (AOP and AOP+ combined) and the percentage of each participant's gaze shifts which were scored AOP+ ( $r = .64$ ,  $p = .005$ ), suggesting that participants

who more likely to demonstrate any type of gaze shifting were the most likely to accompany these with additional communicative signals.

### Frequency of gaze shifts judged to signify IJA

Sixty-eight (26%) of all AOP+ trials were also coded as signifying IJA (mean = 4.5, range = 0-14, S.D. = 5.4). Of these shifts, 41 (60%) were coded as declarative IJA (mean = 2.7, range = 0-12, S.D. = 4.4) and 27 (40%) were coded as imperative IJA (mean = 1.8, range = 0-7, S.D. = 2.2).

Marked variation between individual participants was observed with six of the fifteen who demonstrated gaze shifts, failing to produce any coded as IJA (see Figure 8.11).

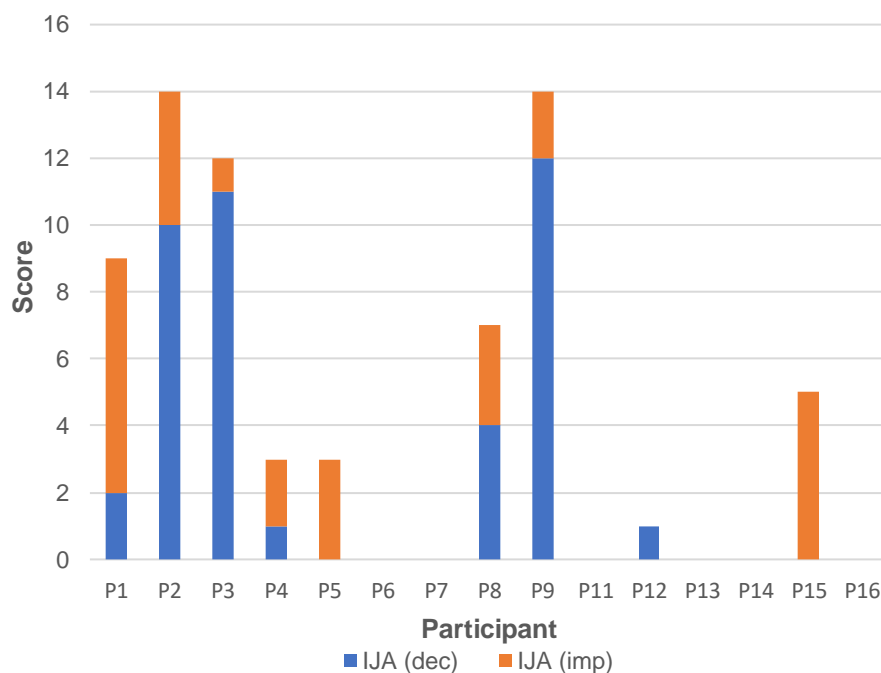


Figure 8.11. Participant use of gaze shifts judged to reflect imperative or declarative IJA.

### **The relationship between RJA and IJA behaviours**

Performance on the RJA trials of the JABM was compared to scores for AOP, AOP+ and IJA using Spearman's correlation since none of variables involved were normally distributed (Shapiro-Wilk,  $p < .05$ ).

There was no significant relationship between RJA and AOP scores ( $r = .60$ ,  $p = .083$ ). However, there was a small but significantly positive relationship between RJA and AOP+ scores ( $r = .555$ ,  $p = .032$ ) and between RJA and IJA scores ( $r = .537$ ,  $p = .039$ ) indicating that participants who demonstrated high scores for gaze following were more likely to demonstrate AOP+ gaze shifts including those judged to signify IJA. There were, however, some individual differences in this pattern since, for example, P1, who demonstrated one of the highest scores for the use of AOP+ gaze shifts only demonstrated RJA on one trial.

### **Frequency of two-point and three-point gaze shifts**

Of the 263 trials in which gaze shifting was demonstrated, 127 (48%) were scored as two-point gaze shifts (mean = 7.4, range = 0-21, S.D. = 5.6) and 136 (52%) were scored as three-point gaze shifts (mean = 8.1, range = 0-26, S.D. = 8.8).

All participants demonstrated a combination of both two-point and three-point shifts with nine demonstrating more two-point than three-point shifts including two (P14 and P16) who only used two-point shifts (see Figure 8.12).

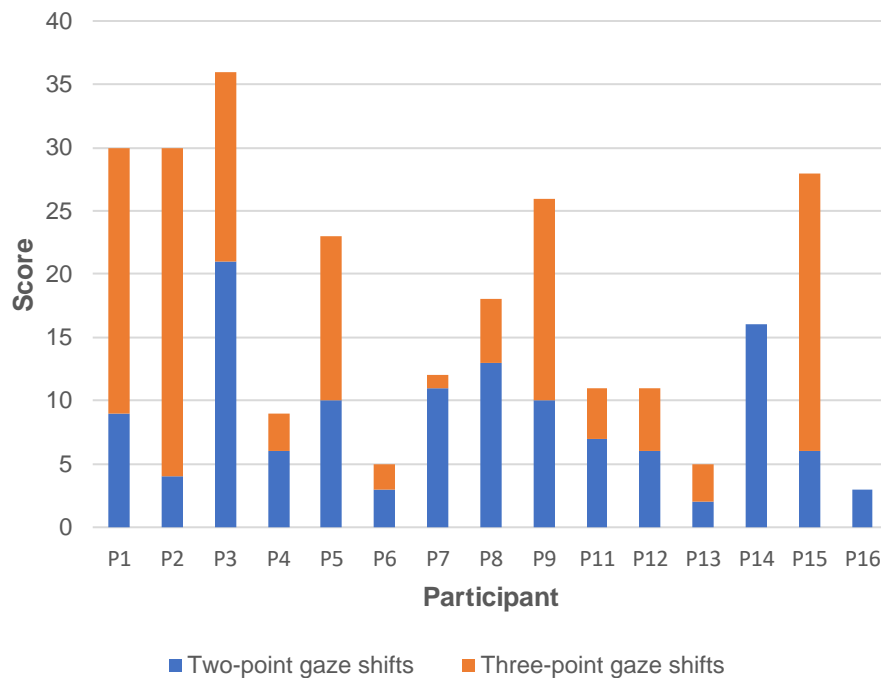


Figure 8.12. Participant use of two-point and three-point gaze shifts.

Further analysis using Pearson's correlation, since data for the use of AOP+ shifts was not normally distributed (Shapiro-Wilk,  $p < .05$ ), indicated that the use of three-point gaze shifts was significantly positively correlated with the use of AOP+ gaze shifts ( $r = .865$ ,  $p < .001$ ) whilst the use of two-point gaze shifts was not ( $r = .287$ ,  $p = .30$ ). Demonstration of three-point gaze shifts was also significantly positively correlated with scores for IJA ( $r = .67$ ,  $p < .001$ ) while the occurrence of two-point gaze shifts was not ( $r = .292$ ,  $p = .282$ ). This suggested that three-point gaze shifts were significantly more likely to be accompanied by communicative signals and to be judged to signify IJA than two-point shifts.

#### 8.5.1.5. Consistency of JA behaviour across sessions

The data analysis reported above was based on combined performance across sessions. Although fluctuations in performance were not the subject of this study, some investigation of consistency was carried out by comparing scores for the demonstration of AOP+ gaze shifts across sessions (see Table 8.4). AOP+ was selected to be the comparison score here because it represents the gaze shifting behaviour most likely to be associated with IJA.

*Table 8.4. Mean AOP+ scores for each test session*

| Test session | Mean AOP+ score | Range | S.D. | 95% confidence interval |
|--------------|-----------------|-------|------|-------------------------|
| 1            | 1.8             | 0 - 9 | 2.9  | 0.3 – 3.3               |
| 2            | 2.7             | 0 - 9 | 3.1  | 1.0 – 4.3               |
| 3            | 2.5             | 0 - 9 | 2.8  | 1.0 – 3.9               |

A one-way ANOVA was conducted to determine if there was a significant difference between the mean AOP+ scores for each test session. The ANOVA was carried out despite data not being normally distributed (Shapiro-Wilk,  $p < .05$ ) as the risk of Type 1 error is not high provided sample sizes in each group are the same (Maxwell, Delaney, Kelley, Delaney, & Kelley, 2017). There were no outliers as assessed by boxplot and there was homogeneity of variance as assessed by Levene's test of homogeneity of variance ( $p = .705$ ). Results indicated no significant difference between mean AOP+ scores for each of the test sessions  $F(2, 48) = 0.366$ ,  $p = .70$ .



Analysis of individual scores does not indicate marked variation for the majority of participants although there were some exceptions. For example, P8 achieved an AOP+ score of 1 in test session 1 but 6 in test session 2 (see Figure 8.13).

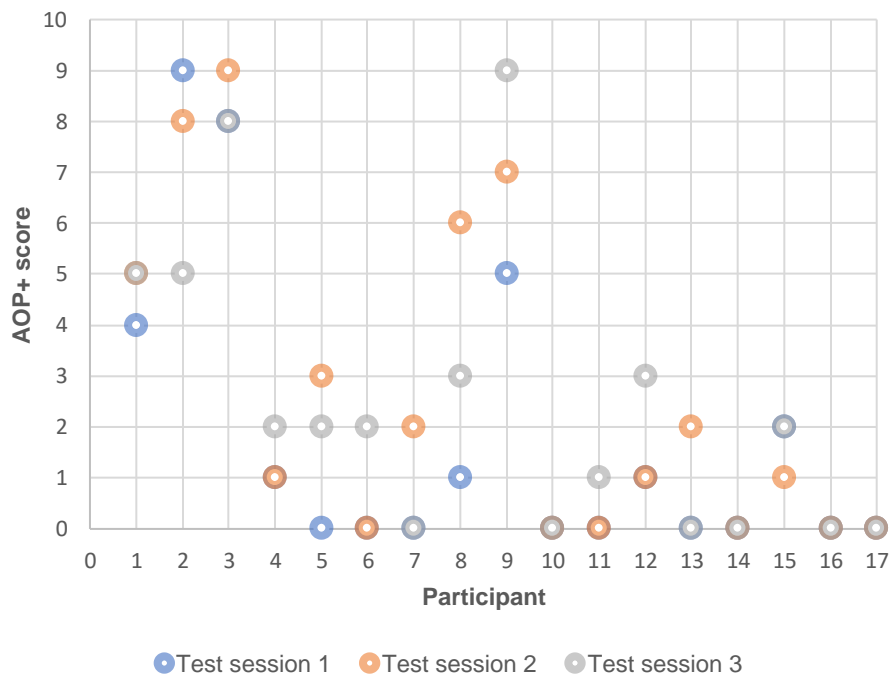


Figure 8.13. Participant AOP+ scores for each test session

#### 8.5.1.6. Identification of performance profiles on the JABM

On the basis of the data reported above, it is possible to identify distinct profiles of performance on the JABM which can be illustrated by focusing on some of the individual participants. P1 and P2 demonstrated a relatively high number of gaze shifts, the majority of which were three-point rather than two-point and were more likely than other participants to use gaze shifts coded as AOP+ and judged to signify IJA. During trials where they did not demonstrate gaze shifting, they always fixed their gaze on a referent and were more likely to fix their gaze on a person than an object. In contrast, P6 and P16 achieved low scores for gaze shifting and their gaze shifts were more likely to be two-point than three-point. They used few or no PCBs and

no sharing looks, with none of their gaze shifts judged to signify IJA. In trials where they did not demonstrate gaze shifting they sometimes failed to fix gaze on a referent and were more likely to fix their gaze on an object than a person.

## 8.5.2. The relationship between performance on the JABM and other measures

### 8.5.2.1. The unstructured play measure

As discussed in section 8.3., the unstructured play measure was designed as a comparison measure to explore the relationship between the demonstration of IJA behaviours on the JABM with performance in a less structured setting

#### Inter-rater agreement

Twenty percent of video recordings of the unstructured play measure test sessions ( $n=10$ ) were scored by a second rater. This rater was one of the MSc students assisting on the project and so was familiar with the scoring protocol but had not been present during the test session she scored. Scores for the different types of gaze shift (AOP and AOP+) and for judgements of IJA were tested for agreement using Cohen's kappa (see Table 8.5.). Inter-rater agreement ranged from Moderate for judgements of AOP to Very Good for judgments of IJA (Altmann, 1999).

*Table 8.5. Inter-rater agreement for scores on the unstructured play measure*

| Gaze behaviour | Kappa Value                                | Level of agreement (Altmann, 1999) |
|----------------|--|------------------------------------|
| AOP            | .509<br>95% CI (.290 -.732)<br>$p < .05$   | Moderate                           |
| AOP+           | .833<br>95% CI (.684 -.981)<br>$p < .0005$ | Very Good                          |
| IJA            | .609<br>95% CI (.390-.834)<br>$p < .05$    | Good                               |

### Performance on the unstructured play measure

All incidences of gaze shifting occurring during the 51 administrations of the unstructured play measure ( three for each of the seventeen participants) were given a score for either AOP, AOP+ and IJA, with a broad score being given for the occurrence of AO and AP (see section 8.3.4).

Overall, 193 gaze shifts were demonstrated (mean = 11.4, range = 0-46, S.D. = 12.3). Only two participants (P2 and P3) demonstrated more than 15 gaze shifts across the three administrations of the measure with three (P6, P16 and P17) demonstrating no gaze shifts at all (see Figure 8.14).

Of the 193 gaze shifts, 85 (44%) were scored as AOP (mean = 5.0, range = 0-17, S.D. = 4.8) and 108 (56%) were scored as AOP+ (mean = 6.4, range = 0-34, S.D. = 9.8). Of the 108 AOP+ gaze shifts, 52 (48%) were judged to signify IJA , with 29 being scored as IJA(imp) and 23 (being scored as IJA(dec).

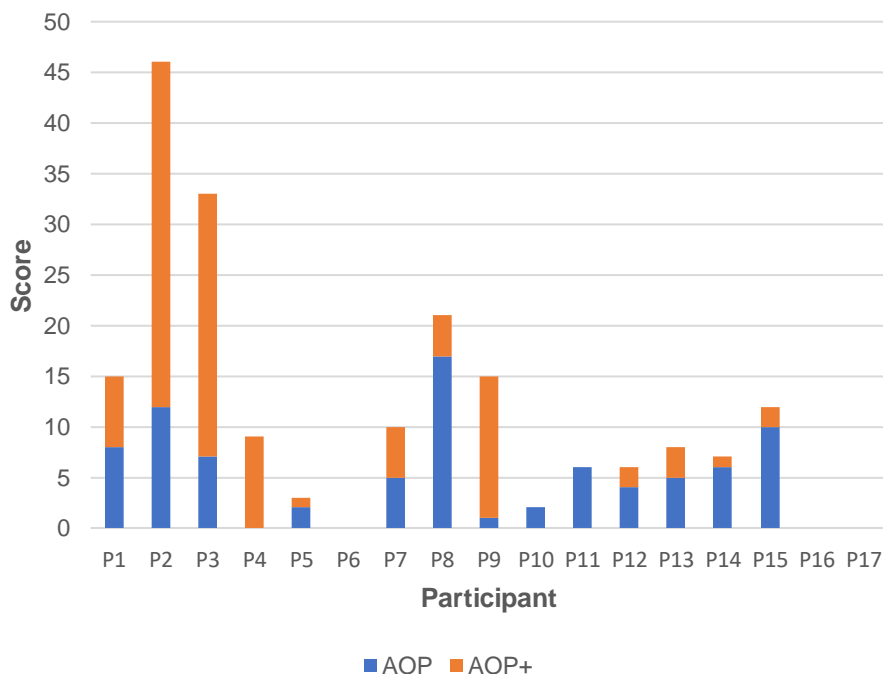
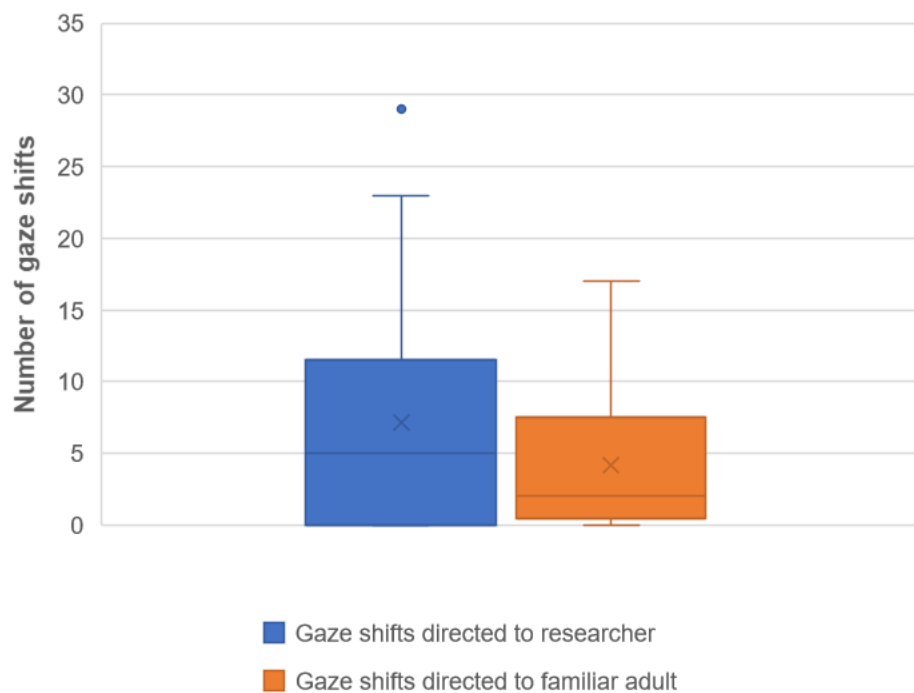


Figure 8.14. Number of AOP and AOP+ gaze shifts demonstrated by each participant during the unstructured play measure.

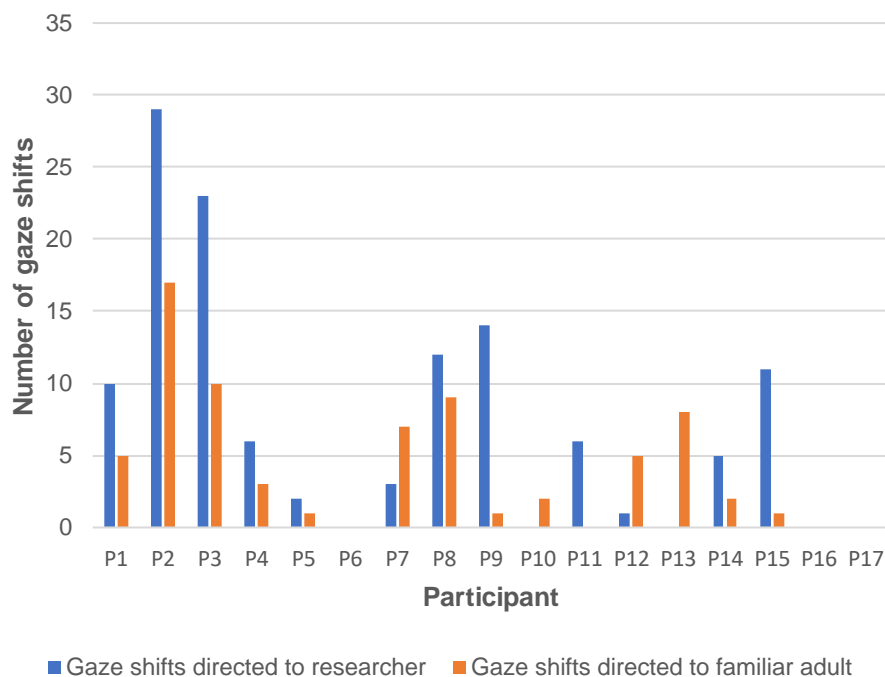
For the participants who did not demonstrate gaze shifting, (P6, P16 and P17), broad scores for AO and AP were analysed to discover the predominant focus of their gaze fixation during the unstructured play measure. All three scored higher for AO than AP suggesting that the pattern observed in the JABM, in which those who demonstrated fewer gaze shifts were more likely to fix gaze on objects than people, was also present during the unstructured play measure.

Unlike the JABM, the unstructured play measure involved a familiar adult as a potential communication partner. To explore whether JA behaviours were more likely to involve a familiar adult than the researcher, the number of gaze shifts directed to each person was compared (see Figure 8.15).



*Figure 8.15. Mean number of gaze shifts directed to the researcher and to the familiar adult during the unstructured play measure.*

Overall, 122 (63%) of observed gaze shifts were directed to the tester (mean = 7.2, range = 0-29, S.D. = 8.5) and 71 (37%) were directed to the familiar adult (mean = 4.2, range = 0-17, S.D. = 8.5.) Three participants (P10, P12 and P13) were more likely to involve the familiar adult than the researcher in their gaze shifts (see Figure 8.16). P10 only demonstrated gaze shifting during the unstructured play measure, having failed to do so during the JABM, and, although only two shifts were seen, both of these involved a familiar adult.



*Figure 8.16. Number of gaze shifts directed by each participant to the familiar adult or the researcher.*

#### **8.5.2.2. The relationship between performance on the JABM and performance on the unstructured play measure**

The nature of scoring for the JABM and the unstructured play measure differed in that all gaze shifts were scored during unstructured play sessions whereas a maximum of one gaze shift per trial was taken into account in scoring the JABM (see section 8.1.5. and section 8.3.4.). As a result, a direct comparison of target behaviour frequency was not feasible. However, it was possible to explore the relationship between scores across both measures using a Spearman's test for correlation since data was not normally distributed (Shapiro-Wilk,  $p < .05$ ) (see Figure 8.17).

Scores on all target behaviours, with the exception of IJA(imp) were significantly positively correlated across the two measures suggesting that performance on the JABM generally reflected performance in a less structured setting.

| Unstructured play measure |                     | JABM               |                    |                  |                  |                     |                     |                |                   |
|---------------------------|---------------------|--------------------|--------------------|------------------|------------------|---------------------|---------------------|----------------|-------------------|
|                           |                     | AO                 | AP                 | AOP              | AOP+             | 2-point gaze shifts | 3-point gaze shifts | IJA(imp)       | IJA(dec)          |
|                           | AO                  | .660**<br>P = .004 |                    |                  |                  |                     |                     |                |                   |
|                           | AP                  |                    | .649**<br>P = .005 |                  |                  |                     |                     |                |                   |
|                           | AOP                 |                    |                    | .831**<br>p<.001 |                  |                     |                     |                |                   |
|                           | AOP+                |                    |                    |                  | .830**<br>p<.001 |                     |                     |                |                   |
|                           | 2-point gaze shifts |                    |                    |                  |                  | .639**<br>P = .006  |                     |                |                   |
|                           | 3-point gaze shifts |                    |                    |                  |                  |                     | .717**<br>P <.001   |                |                   |
|                           | IJA (imp)           |                    |                    |                  |                  |                     |                     | .46<br>P = .47 |                   |
|                           | IJA (dec)           |                    |                    |                  |                  |                     |                     |                | .70**<br>P = .002 |

Each cell shows the correlation coefficient (Spearman's rho) and the significance level. \*\* = significant correlation at the .01 level

Figure 8.17. Correlation matrix comparing scores for target behaviours on the JABM and the unstructured play measure.

### 8.5.2.3. The relationship between performance on the JABM and performance on the FGBM

As described in Chapter 7, The FGBM measured participants' functional ability to fix and shift their gaze. To investigate whether these functional visual abilities were related to performance on the JABM, a correlation matrix was conducted to compare scores across the two measures (using Spearman's correlation since some data sets were not normally distributed (Shapiro-Wilk,  $p < .05$ ). FGBM scores for gaze fixation, gaze shifting and the use of multiple gaze shifts were compared with JABM scores for RJA, AOP, AOP+ and IJA as well as with trials coded as 'no fixation' in order to investigate whether failure to fixate was associated with poor functional vision skills (see Figure 8.18).

|             |                            | JABM scores            |                     |                                   |                       |                       |                  |
|-------------|----------------------------|------------------------|---------------------|-----------------------------------|-----------------------|-----------------------|------------------|
|             |                            | NFP<br>(RJA<br>trials) | RJA                 | No<br>fixation<br>(IJA<br>trials) | AOP                   | AOP+                  | IJA              |
| FGBM scores | Gaze<br>fixation           | -.721**<br>P=.001      | .239<br>P =<br>.356 | -.768**<br>P <.001                | .698**<br>p =<br>.002 | .642**<br>P =<br>.005 | .650**<br>P=.005 |
|             | Gaze<br>shifting           | -.757**<br>P <.001     | .740<br>P=.776      | -.712**<br>P =.001                | .674**<br>P=.003      | .582*<br>P=.014       | .589*<br>P=.013  |
|             | Multiple<br>gaze<br>shifts | -.714**<br>P=.001      | .339<br>P=.216      | -.669**<br>P=.006                 | .128<br>P=.649        | .723**<br>P=.002      | .755**<br>P=.001 |

Each cell shows the correlation coefficient (Spearman's rho) and the significance level.

\*\* = significant correlation at the .01 level

\* = significant correlation at the .05 level.

Figure 8.18. Correlation matrix comparing scores on the FGBM and the JABM.



No significant relationship was found between RJA scores on the JABM and performance on the FGBM. There was also no significant relationship between the use of multiple gaze shifts on the FGBM and the use of unaccompanied gaze shifts (AOP) during the JABM. However, all other JABM scores were significantly associated with performance in all three areas of the FGBM. There was an inverse relationship between performance on the FGBM and the number of 'no fixation' trials on the JABM trials confirming a relationship between poor functional vision skills and failure to fixate during the JABM. In contrast, performance on the FGBM was positively associated with the use of AOP and AOP+ gaze shifts as well as with the demonstration of gaze shifts judged to signify IJA suggesting that participants with better functional vision skills were more likely to engage in referential gaze shifting, including those used for communicative functions.

Further analysis of the relationship between the use of two or three-point gaze shifts (both AOP and AOP+) and scores on the FGBM indicated that there was a positive and significant relationship between the use of multiple gaze shifts during the FGBM and the use of three-point shifts during the JABM ( $r = .622$ ,  $p = .013$ ) but not between the use of multiple gaze shifts and the use of two-point shifts ( $r = .352$ ,  $p = .199$ ) suggesting that the use of three-point shifts may be related to functional visual ability.

#### **8.5.2.4. The relationship between performance on the JABM and performance on background measures of motor skills, cognition and communication.**

##### **Motor skills**

To explore the relationship between gross motor skills and JABM scores, participants were divided into two groups depending on their GMFCS level. The six participants with a GMFCS level of 5 were grouped together, as were the eleven participants with a GMFCS level of 3 or 4 (only one participant had a GMFCS level of 3 hence the merging of scores for these two GMFCS levels).

The difference between each group's scores for RJA and for AOP+ were then compared. AOP+ was selected to be the IJA score upon which they would be compared because it represents the gaze shifting behaviour most likely to be associated with IJA and had been found to be significantly related to both AOP and communicative IJA.

An independent samples t-test was used to test the difference in RJA scores between the two GMFCS groups since data was normally distributed (Shapiro-Wilk,  $p > .05$ ) and there was homogeneity of variance, as assessed by Levene's test for equality of variances ( $p = .221$ ). Results indicated no significant difference in RJA scores between the GMFCS level 3/4 group (mean = 3.3) and the GMFCS level 5 group (mean = 1.5)  $t(15) = 1.91$ ,  $p = .075$ ).

A Mann-Whitney U test was used to determine whether there was a significant difference between AOP+ scores for the two GMFCS groups since data was not normally distributed (Shapiro-Wilk,  $p < .05$ ). Distributions of the AOP+ scores for both groups were similar, as assessed by visual inspection. There was found to be no statistically significant difference between AOP+ scores for the GMFCS level 3/4 group (median = 5.0) and the GMFCS level 5 group (median = 2.0),  $U = 24.5$ ,  $z = -.864$ ,  $p = .404$ ).

In summary, statistical analysis found no relationship between motor skills as measured by the GMFCS and JA behaviours measured using JABM scores for RJA and AOP+.

## **Cognitive Skills**

P-scales had been used as a measure of cognitive ability for this sample. Participants were functioning between p1ii and p3ii (see Table 7.5 for each participant's P-scale score) with two functioning at 1ii, three at p2i, five at p3ii, two at p3i and four at p3ii. P8 did not have a P-scale score since, due to his age (3:5), he had not yet begun to be assessed using this framework. He was, therefore, excluded from this analysis.

Participants were divided into a low P-scale and high P-scale group since the numbers of participants in individual P-scale groups was too small for robust statistical comparison to be carried out. The low P-scale group included participants at P-scales 1ii, 2i and 2ii ( $n = 10$ ) and the high P-scale group included participants at P-scales 3i and 3ii ( $n = 6$ ). Mean scores for RJA were compared between the two groups as were scores for AOP+.

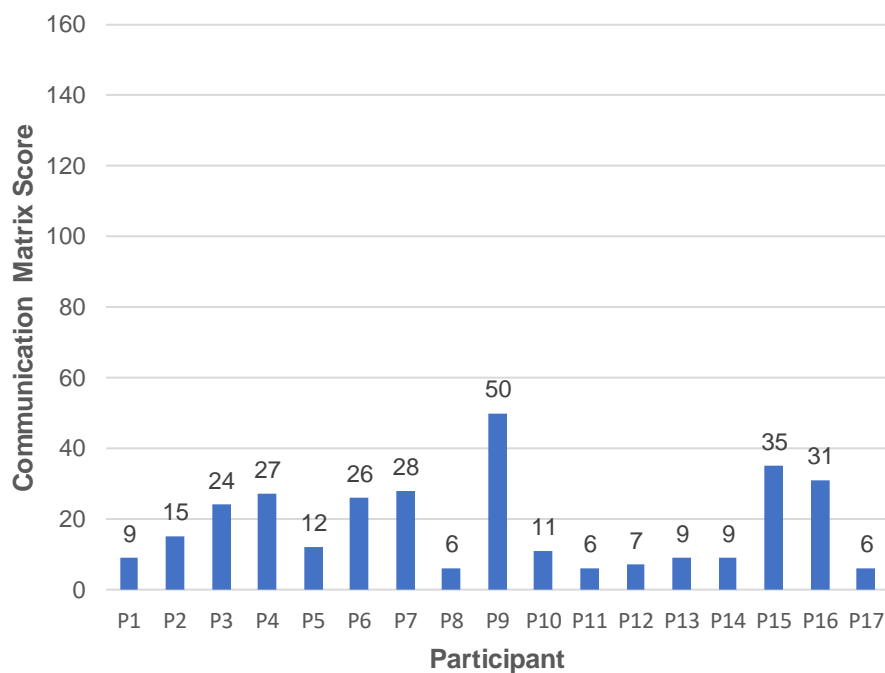
An independent samples t-test was used to test the difference in RJA scores between the two P-scale groups since data was normally distributed (Shapiro-Wilk,  $p > .05$ ) and there was homogeneity of variance, as assessed by Levene's test for equality of variances ( $p = .538$ ). Results indicated no significant difference between RJA scores for the low P-scale group (mean = 2.4) and the high P-scale group (mean = 3.4) ( $t(14) = -1.02$ ,  $p = .325$ ).

A Mann-Whitney U test was used to determine whether there was a significant difference between AOP+ scores for the two P-scale groups since data was not normally distributed (Shapiro-Wilk,  $p < .05$ ). Distributions of the AOP+ scores for both groups were similar, as assessed by visual inspection. There was found to be no statistically significant difference between AOP+ scores for the low P-scale group (median = 2.0) and the high P-scale group (median = 5.0),  $U = 44.5$ ,  $z = .804$ ,  $p = .441$ ).

In summary, statistical analysis found no relationship between cognitive skills as measured by the P-scales and JA behaviours measured using JABM scores for RJA and AOP+.

## Communication

To explore the relationship between scores on the JABM and scores obtained from an existing measure of communication, participant scores on the Communication Matrix were compared with their JABM scores for RJA and AOP+. Participants had achieved a mean score of 18.3 (out of a possible 160) on the Communication Matrix (range = 0-50, S.D. = 12.9), with individual scores being represented in Figure 8.19.



*Figure 8.19. Participant scores on the Communication Matrix*

Descriptive analysis suggested little association between scores on the Communication Matrix and performance on the JABM with, for example, P1, who had demonstrated some of the highest scores for AOP+ and IJA on the JABM, achieving a relatively low score on the Communication Matrix while P16, who had performed poorly on JABM measures of JA, achieved one of the higher scores on the Communication Matrix. This relationship was tested using a Spearman's correlation since data was not normally distributed, (Shapiro-Wilk < .05) which confirmed no significant association between

Communication Matrix scores and either JABM RJA scores ( $r = .313$ ,  $p = .221$ ) or JABM AOP+ scores ( $r = .205$ ,  $p = .429$ ).

### 8.5.3. The Impact of probe type on JA behaviours

To investigate whether the contexts provided by different probes had an effect on the frequency and type of JA behaviours elicited, JABM scores for the four probes designed to elicit IJA behaviours were compared. Since the Look at the Ball probe was only designed to elicit RJA it was excluded from this analysis.

#### 8.5.3.1. The impact of probe type on gaze behaviour

Each probe was administered a total of 204 times (four times in each of the three test sessions completed by the seventeen participants). The pattern of gaze behaviours elicited by each probe can be seen in Figure 8.20.

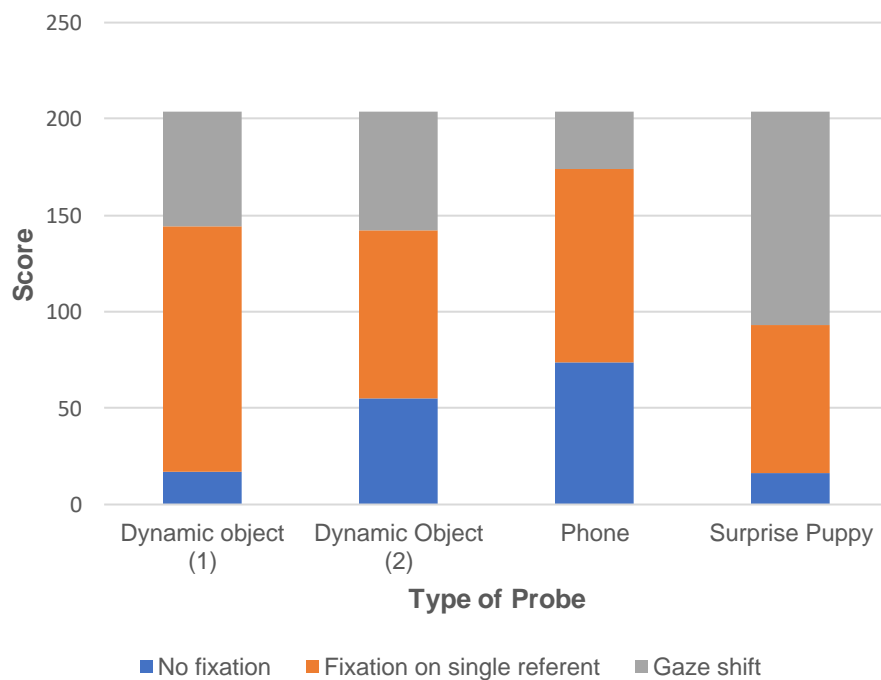


Figure 8.20. Gaze behaviour elicited in response to each probe

Of the four probes, the Phone probe resulted the greatest number of no fixation trials (total = 73, mean = 4.3, range = 0-10, S.D. = 3.5) and the smallest number of gaze shifts (total = 30, mean = 2.2, range = 0-11, S.D. = 2.9). In contrast, the Surprise Puppy trial resulted in few no fixation trials

(total = 16, mean = 0.94, range = 0-4, S.D. = 2.08) and the highest number of gaze shifts (total = 111, mean = 7.4, range = 3-13, S.D. = 2.82).

### 8.5.3.2. The impact of probe type on the use of AOP and AOP + gaze shifts

The number of AOP and AOP+ gaze shifts elicited by each probe was compared to investigate whether any of the probes elicited a greater proportion of AOP+ gaze shifts (see Figure 8.21).

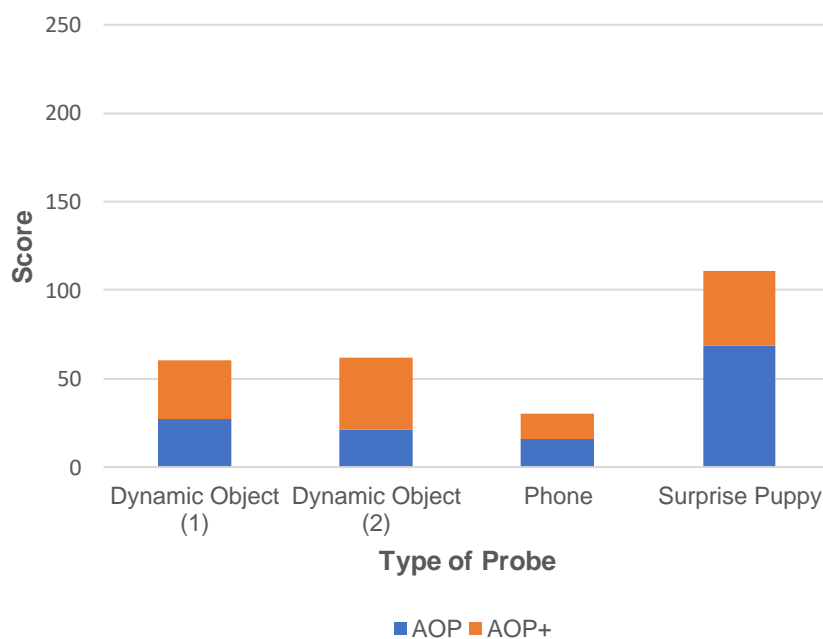


Figure 8.21. AOP and AOP+ scores for each type of probe.

All probes elicited both AOP and AOP+ gaze shifts. Although the Surprise Puppy trial had elicited the highest number of gaze shifts overall, only 38% of these were AOP+ shifts (total = 42, mean = 2.8, range = 0-7, S.D. = 2.7). The Dynamic Object (2) probe elicited a similar number of AOP+ shifts to the Surprise Puppy trial (total = 41, mean = 2.8, range = 0-7, S.D. = 2.4) despite eliciting fewer shifts overall. The smallest number of AOP+ shifts was elicited by the Phone probe (total = 14, mean = 1.0, range = 0-10, S.D. = 2.6) although it did result in a high number of AOP+ shifts (10) from one participant (P3).

### 8.5.3.3. The effect of probe type on the use of communicative IJA

The frequency of trials coded for IJA was compared across probe types to see if any were more likely to result in communicative IJA as judged by raters (see Figure 8.22).

Of the 68 trials judged to signify IJA, the greatest number were elicited by Dynamic Object (2) trials (total = 29, mean = 1.9, range = 0-6, S.D. = 2.2), followed by the Surprise Puppy trial (total = 15, mean = 1.0, range = 0-5, S.D. = 1.5) and the Dynamic Object (1) trial (total = 13, mean = 0.9, range = 0-5, S.D. = 1.6) with the Phone trial resulting in the fewest trials scored as IJA (total = 11, mean = 0.7, range = 0-6, S.D. = 1.7).

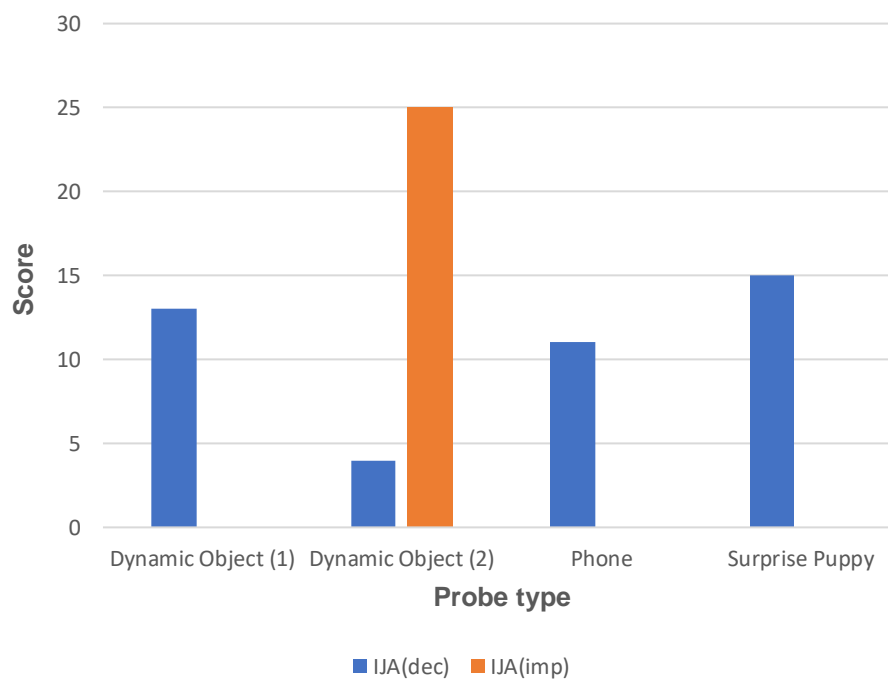


Figure 8.22. IJA(dec) and IJA(imp) scores for each probe.

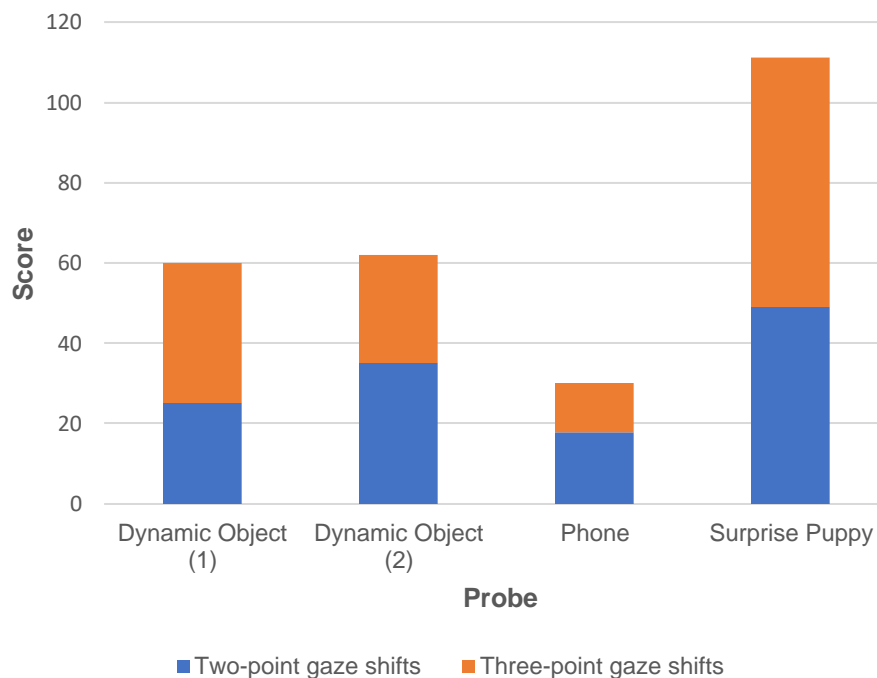
Judgements of IJA(imp) were only made for trials of the Dynamic Object (2) probe which is as expected since this was the only probe providing a context designed to elicit this behaviour.



In summary, all four types of probe elicited some instances of IJA, with the Dynamic Object (2), which successfully elicited the requesting it was designed for; eliciting in the highest number.

#### 8.5.3.4. The impact of probe type on the use of two and three-point gaze shifts.

Since two-point gaze shifts can signal the occurrence of JA in bottom-up but not top-down context, it was hypothesized that they would comprise a greater proportion of the gaze shifts demonstrated during the probes involving a bottom-up context (Dynamic Object (1), Dynamic Object (2) and Phone probes). However, descriptive analysis of the data did not provide evidence to support this hypothesis (See Figure 8.23).



*Figure 8.23. Frequency of two-point and three-point gaze shifts demonstrated in response to each probe.*

Two-point shifts did comprise a slightly higher proportion of gaze shifts in Phone trials (where 60% of gaze shifts were two-point) and Dynamic Object (2) trials (where 56% of gaze shifts were two-point) but not during Dynamic Object (1) trials (where 44% of gaze shifts were two-point).

## 8.6. Discussion

This thesis was motivated by issues identified in SLT's approaches to assessment of pre-linguistic communication in people with PIMD. Appraisal of current literature relating to pre-linguistic communication development and methods used to investigate this suggested that assessment might be enhanced by focusing on JA and making use of structured sampling techniques for gathering information. The research described in the thesis was designed to explore these areas by answering the following research questions:

- 1) What patterns of JA behaviour are demonstrated by young people with PIMD?
- 2) Are the JA behaviours demonstrated by young people with PIMD affected by their functional ability to fix and shift gaze?
- 3) Are structured probes an effective means of eliciting information about JA behaviours?

I will begin this final discussion by addressing each of these research questions, discussing the significance of results from this study and relating this to previous studies of JA in people with PIMD. Finally, in Chapter 10, I will evaluate the contribution which this makes to the field of pre-linguistic communication assessment in speech and language therapy.

### **8.6.1. Research question 1: What patterns of JA behaviour are demonstrated by young people with PIMD?**

The research carried out in this thesis indicated that children with PIMD show varying patterns of performance, not only in the frequency with which they engage in a state of JA but in their demonstration of a range of behaviours relating to JA. Profiles of performance on a continuum of JA behaviour derived from developmental literature may indicate an individual's capacity for ultimately developing the ability to engage in JA (or doing so with appropriate scaffolding) if they do not do so already. Such profiles of performance also have implications for the type of support and intervention they will require. Monitoring changes in performance along this continuum could provide a measure of progress over time.

The profiles of JA behaviour which were demonstrated by the participants in this study will now be discussed in terms of their relationship to theory around pre-linguistic development and findings of existing studies of JA in people with PIMD which were outlined in Table 4.2.

#### **8.6.1.1. Responding to JA**

Developing gradually from around the age of around 2-4 months in typical children, the ability to follow a head turn and, ultimately the gaze direction of another person facilitates JA, with demonstration of these early skills being related to a number of later developing language and social skills (e.g Mundy & Newell, 2007; see section 4.2.2.4.).

Participants in this study varied in their ability to respond to RJA as evidenced by following the direction of a head turn preceded by a cue (saying the participant's name). Although the majority (fourteen) were able to follow a head turn on some occasions, none did so consistently, with only two doing so in more than 50% of the trials administered to them. This contrasted with the reference group of typically developing children on whom the JABM was tested. Although this group of typically developing children was small, it was notable that five out of the six of them were able to follow gaze on 50% of trials or more with the two aged over twelve months performing at ceiling. There is very limited existing information about demonstration of this behaviour in people with PIMD although it was measured, amongst other JA

behaviours by Neerinckx & Maes (2016) using subtests of the ESCS. These authors reported a range of performance which was consistent with findings of the current study, with some participants failing to demonstrate RJA at all and others doing so in up to 29% of the time intervals they observed but none doing so consistently.

Given the lack of existing evidence around RJA in people with PIMD, its significance for this group is, as yet, unclear. However, there was a significant correlation between scores for RJA and scores for engaging in gaze shifts accompanied by communicative signals (AOP+) on the JABM suggesting that individuals who are most able to follow the direction of a head turn are also most likely to engage in communicative JA, perhaps indicting a general JA capacity. However, it was notable that the relationship between these skills was not absolute and there were individual participants who performed relatively well on the RJA trials but demonstrated few AOP+ gaze shifts and vice versa. This provides some weight to a suggestion by Mundy and Newell (2007) that RJA and IJA may constitute different neural systems which may be differentially impaired. Further research in this area would help clarify the significance of RJA abilities in this population and whether, for example, those that demonstrate relatively good RJA skills at an early age, go on to make the most progress in their communication skills. The results of this study relate to following a head turn, a behaviour which may result in lean JA where the result is sharing attention to the same referent. However, the extent to which participants who demonstrate this skill understood their partners state of mind (i.e. that they found the referent interesting) is unclear. Further work differentiating between the following of a head turn of a communication partner who has open or closed eyes as described by Brooks and Meltzoff (e.g. Brooks & Meltzoff, 2005)) might provide further knowledge in this area.

In functional terms RJA skills are significant since failing to follow the direction of another's attention will lead to fewer opportunities for engaging in episodes of joint attention about a shared referent. From the perspective of support, where individuals rarely or never follow the attentional focus of another person it will be particularly important for communication partners to focus on following that individual's focus of attention in order to facilitate JA

while intervention might target the development of RJA. This highlights the importance of including measurements of RJA as part of a comprehensive communication assessment.

#### **8.6.1.2. Initiating JA**

Participants in this study engaged in communicative JA very infrequently with raters judging only 5% of all JABM trials to demonstrate declarative JA and 3% to represent imperative JA. This is consistent with findings of other studies investigating JA in people with PIMD or developmental disabilities where JA was also observed to occur rarely (e.g. Arens, Cress, & Marvin, 2005; Hostyn, Neerinckx, & Maes, 2011; Neerinckx & Maes, 2016). Once again, some contrast is offered by the reference group of typically developing children used to test the JABM where 29% of trials were judged to represent either imperative or declarative JA.

The relatively rare occurrence of communicative JA in young people with PIMD means that, as a target measure, it can provide only a limited degree of differentiation between individuals given their generally low scores in this area and the fact that rater judgements around it lack consistency (inter-rater agreement being only fair for the study reported here). In the light of this situation, it is clearly important for assessment to consider the whole continuum of behaviours which relate to JA rather than focusing solely on the occurrence of JA itself. The behaviours on this continuum which were investigated by this study included whether or not individuals were able to attend to referents and, where they did so, whether they attended solely to single referents (people or objects) or whether they were able to integrate their attention by shifting gaze between them. Those that did shift gaze could be further differentiated by the extent to which they combined such gaze shifts with potentially communicative signals and whether these behaviours were perceived by raters to be communicative. Performance in each of these areas will now be discussed.

### **Failure to attend to referents**

Participants failed to fix their gaze on referents in 20% of the JABM trials. Whilst such failure to fixate was rarely observed in the reference group of typically developing children used in this study, a similar pattern of behaviour was reported by Arens, Cress and Marvin (2005) in their study of young children with developmental and physical disabilities. Indeed, participants in their study spent 54.8% of their observed time in 'unengaged behaviour', defined as being, '... not engaged in anything, looking off into space' (Arens, Cress & Marvin, 2005, p163). As with all the JA behaviours measured by the current study, there was variation between participants with three fixating on at least one referent in every trial and three failing to fixate in almost 50% or more of their trials. Failure to fixate also had an impact on scores for RJA since 31% of Look at the Ball trials could not be effectively administered due to participants failing to fix gaze on the tester's face.

From a functional perspective, identifying individuals who have particularly low rates of fixation is important since communication partners will find it challenging to identify and follow a focus of interest in these individuals, making it harder to provide scaffolding for JA and communication and it can be hypothesized that individuals showing low frequencies of fixation have the least capacity for engaging in JA, or certainly that which is manifested through gaze behaviour.

### **Attention to a single referent**

The most commonly demonstrated gaze behaviour on the JABM was attention to a single referent. Again, this is consistent with the findings of Arens, Cress and Marvin (2005) whose participants, when their attention was engaged, were most likely to be focused on single objects or people rather than integrating attention through gaze shifting. Results of the current study suggested that the focus of this attention may be significant. At a group level attention to an object was more commonly demonstrated than attention to a person, a finding which may well be explained by the context since the JABM probes involved objects which were novel and designed to be engaging. However, there was a group of six participants who were more likely to fix their gaze on a person rather than an object during trials in which they did

not shift gaze. This group of participants were also the most likely to engage in both types of gaze shifting (AOP and AOP+) during other trials as well as to be judged to engage in communicative JA. It seems likely, then, that young people with PIMD who show greater attention to people in general are also those who more likely to develop the ability to engage in JA than those whose predominant focus is on objects. Again, it is essential for the assessment process to consider this area since it has implications for support and intervention. Where individuals show an over-riding interest in objects, the presence of engaging objects will reduce opportunities for communication. Therefore, when the focus is on interaction it will be important that objects are removed and intervention should encourage increased interest in people through the use of rewarding person-person interaction, possibly using intensive interaction techniques.

### **Integrated attention to people and objects**

The integration of attention through gaze shifting between a person and object occurred in 32% of trials on the JABM and varied between participants with only two participants failing to demonstrate any gaze shifts at all. Detailed analysis of these gaze shifts and the behaviours accompanying them allowed for further differentiation between participants to be made.

### **Gaze shifts unaccompanied by communicative signals**

The most frequently observed gaze shifts were those which involved the shifting of attention between object and person in the absence of any additional behaviours which might indicate communication (AOP shifts). Fifteen of the seventeen participants in this study demonstrated at least some use of such gaze shifts. It is difficult to judge the underlying motivation for these shifts although the structured context of the JABM allows us to conclude that they were unlikely to have been a response to extrinsic cues such as the object suddenly becoming salient or the person saying something since these aspects were controlled. Indeed, such shifts often involved shifting gaze away from an engaging referent. For example, during the Dynamic Object (1) probe participants would sometimes disengage gaze from a visually engaging test item to look at the tester who was silent and visually neutral, possibly then shifting their gaze back and forth between the



item and the person. Despite the absence of communicative signals accompanying these shifts, it seems likely that they are driven by an intrinsic, social motivation – perhaps a desire to see if the tester was also looking at the object or in expectation of a response from them. In this respect they lend weight to the concept of ‘checking looks’ described by Hobson and Hobson (2007). Whilst reflecting a degree of social awareness, they do not meet the criteria for rich JA since they do not include an indicator of understanding that attention is being shared but might be classified as lean JA where an intentional behaviour results in the sharing of attention to a referent.

Regardless of the motivation underlying gaze shifts unaccompanied by communicative signals or the definitions which are applied to them, in functional terms they create an opportunity for an individual to witness their communication partner’s reaction to an object and offer the communication partner an opportunity to facilitate the sharing of such reactions. For this reason, it seems likely that individuals who engage in gaze shifting between objects and people, are more likely to progress to engagement in rich JA than those who do not, even where such gaze shifting is not accompanied by communicative signals. In this respect, AOP gaze shifts might be considered a ‘stepping stone’ toward JA, a suggestion reinforced by the finding that participants who most frequently demonstrated them were also those who showed the highest proportion of AOP+ shifts. This finding supports the notion that lean JA may be considered an earlier developing phenomenon which is related to the subsequent development of rich JA (Racine, 2011).

### **Gaze shifts accompanied by communicative signals and engagement in JA.**

The use of gaze shifts accompanied by communicative signals such as PCBs or sharing looks (AOP+) which might signify attempts to engage in JA, occurred infrequently across this sample of participants with PIMD, being demonstrated in only 14% of the trials overall although thirteen of the seventeen participants did demonstrate them on occasion. It seems significant that raters did not consider all these AOP+ shifts to be

communicative, with only 8% of trials being judged to represent declarative or imperative IJA. Again, this contrasted with the reference group of typically developing children who demonstrated AOP+ shifts in 35% of trials with most of these shifts being judged to represent communicative IJA.

The signals which participants used most commonly in conjunction with gaze shifts were potentially communicative behaviours (PCBs) such as changes in facial expression, vocalisations or body movements (including banging the table or rocking back and forth). Sharing looks were also observed but less frequently, and generally in combination with a PCB. Sharing looks tended to be used by participants who demonstrated the highest proportion of AOP+ gaze shifts suggesting that they may, indeed, be a frequent component of JA as suggested by Hobson and Hobson (2007) but are mainly used by those who show greater competence in this area. On the basis of observations made during the scoring the JABM it seemed that, although participants used communicative signals, they were rarely used with persistence and this may have accounted for the fact that they were not always considered to represent intentional communication. Potentially communicative behaviours were often idiosyncratic and not behaviours commonly associated with communication (e.g. rocking or banging the table). Informally it seemed that the behaviours used by the reference group of typically developing children were more conventional and more persistent which may account for their being more frequently judged to signify intentional communication.

In summary, the relatively low incidence of JA in this sample is consistent with findings from existing studies of JA in PIMD populations and has implications for support and intervention. The extent to which individuals use AOP+ gaze shifts and whether or not they engage in sharing looks provides a further degree of differentiation and may help identify individuals who have a greater capacity for learning to engage in JA or engaging in JA when appropriate support is provided.

### **The use of two and three-point gaze shifts**

Previous research by Arens, Cress and Marvin (2005) had suggested that the use of two-point gaze shifts may constitute a precursor to the use of more complex three-point gaze shifts as an indicator of JA in children with developmental disabilities. Findings from the current study indicated that two-point gaze shifts were, indeed, frequently demonstrated. They constituted 48% of all gaze shifts, being used to some extent by all participants with nine out of the fifteen participants who demonstrated gaze shifting using them more frequently than three-point shifts. Since two-point gaze shifts may signify JA in bottom-up contexts, it was hypothesized that they would be used most frequently in probes which created such a context. However, there was no evidence to support this.

It would seem that two-point shifts are less likely to be a behavioural component of JA than three-point shifts since the use of three-point gaze shifts was found to correlate significantly with the use of AOP+ shifts and with IJA scores while the use of two-point shifts was not. Therefore, it might be hypothesized that children who use two-point shifts more frequently than three-point shifts, may not be as far advanced along a continuum of JA development. However, to the author's knowledge, there is no current evidence relating to the use of two-point gaze shifts by typically developing children and such investigation was beyond the remit of this study. Further research in this area is required before the role of two-point gaze shifts in the development of JA can be established but, from a clinical perspective it seems that it might be beneficial to differentiate between children who use a high proportion of two-point rather than three-point gaze shifts. Engagement in two-point gaze shifts between a person and object indicates some social awareness (as opposed to not shifting gaze at all) and may lead to JA in bottom-up contexts if the sharing of affect is promoted by communication partner behaviour, for example, if a child looks from an engaging object to a communication partner who makes eye contact with them, smiles and says something in an excited tone to indicate their enjoyment. However, there should be awareness that further development may be needed before the child is ready to engage in JA in a whole range of contexts.

#### **8.6.1.3. Profiles of JA behaviour**

Overall, the results of this study suggested that children with PIMD can be differentiated by the patterns of JA behaviour they demonstrate and that these patterns may be viewed in terms of points on a continuum. Those at the most advanced end of this continuum tend to follow the head turn of another (although not necessarily consistently) and use gaze shifts accompanied by both PCBs and sharing looks which are interpreted as signifying communicative JA by observers. Those at the least advanced end of this continuum frequently fail to fix their attention on objects or people, being unlikely to follow gaze and when they do fix their gaze, are more likely to do so on objects, rarely shifting their gaze from object to person. Each individual's performance will vary, meaning that they will require different types of support and intervention. Therefore, effective communication assessment should seek to establish an individual's capacity for engaging in JA behaviours such as gaze fixation on people and objects and different types of gaze shifting as well as the extent to which they engage in JA as part of intentional communication.

### **8.6.2. Research question 2: Are the JA behaviours demonstrated by people with PIMD affected by their functional ability to fix and shift gaze?**

The finding that the children with PIMD demonstrate different patterns of behaviour relating to JA raises the question of what factors may underly and predict such variation. Given the involvement of gaze in typical developing JA behaviours, this study aimed to investigate the impact of functional vision on the demonstration of JA behaviours. Findings indicated that there was, indeed, a strong and significant link between functional vision and patterns of JA behaviour. The ability to fix and shift gaze on the FGBM, a measure of functional vision devised especially for this study to assess children with PIMD, was positively correlated with demonstration of AOP and AOP+ gaze shifts as well as with shifts judged to signify episodes of communicative JA. Participants who achieved low scores on the FGBM were also those who were least likely to fix on referents during the JABM indicating that failure to fixate may well be due to functional vision impairments rather than, for example, a general lack of interest in referents.

The identified association between functional vision abilities and gaze shifting is unsurprising – if an individual is unable to disengage and shift their gaze between stimuli it follows that they will be unable to use referential gaze shifting between objects and people as a means for engaging in JA. However, it was notable that those participants who were most competent at gaze shifting on the FGBM not only demonstrated more gaze shifts overall but also produced a higher proportion of gaze shifts which were judged to signify IJA. A potential explanation for this is that children who are functionally competent at gaze shifting, are most able to respond to caregiver prompts which encourage it, such as waving or tapping objects. Although the shifts resulting from these prompts may not initially be communicative, continued practice, accompanied by ongoing caregiver shaping provide opportunities for learning which eventually lead to their becoming intentionally communicative. If this is the case it implies that children with the best developed functional vision skills have the best capacity for learning to engage in JA as part of intentional communication. Such competence in functional vision is not only reflected in their ability to fix and shift their gaze

but in the extent to which they spontaneously use these skills to perform 'back and forth' shifts between stimuli since the use of such multiple shifts was associated with the greater use of three-point gaze shifts during the JABM, these being most likely to be judged communicative.

The association between functional vision and demonstration of IJA behaviours did not extend to RJA behaviours, there being no significant correlation between FGBM scores and RJA scores on the JABM. The reason for this is unclear since following the gaze of another person does involve the functional ability to disengage and shift gaze. However, the relatively small amount of data collected in relation to RJA may be a factor. As discussed, 31% of RJA trials could not be scored since the participant did not engage in eye contact with the tester for long enough to follow gaze and results showed that this was significantly more likely to be the case for participants who had shown poor performance on the FGBM. In this respect, functional vision skills were shown to have some impact on the ability to follow the focus of another's attention since failure to fix gaze on another person makes it impossible to follow changes in their gaze direction.

In summary, the findings of this study established a strong relationship between functional vision and the ability to engage in behaviours related to JA, highlighting a need to consider functional vision, particularly the ability to fix and shift gaze, when completing a comprehensive assessment of communication in children with PIMD.

### **The relationship between motor skills, cognition and communication and demonstration of JA behaviours**

Further results of this study indicated that functional vision skills were the only factor consistently associated with performance on the measure of JA used in this study. Although it might be expected that greater impairments in functional vision would be linked to greater degrees of physical disability, gross and fine motor skills as measured by the GMFCS and the MACS were not associated with performance on either the FGBM or the JABM. This suggests that functional vision and JA abilities cannot be predicted from motor skills, further emphasizing the need to assess these areas independently.

Cognitive and communication skills were found to be equally unrelated to performance on the JABM. This is a surprising finding given that both the measures used, the P-scales and the Communication Matrix, involve judgements about intentional communication which, in turn, is inextricably linked to JA. It is likely that the challenges raised by assessment for this population, as discussed in Chapter 3, have a role to play here.

Firstly, both the P-scales and Communication Matrix were completed on the basis of teacher judgements of a child's behaviour. Although the teacher knows the child well, their interpretation of criteria provided by assessments may differ. It was certainly the experience of the author, who was present while teachers scored the online Communication Matrix, that they varied in the flexibility with which they interpreted the statements provided by that assessment with some scoring more 'generously' than others. Based on findings from the exploratory phase of this study it might also be hypothesized that teachers accurately base their judgements on what they have observed in everyday contexts even though these contexts do not offer sufficient opportunity for participants to demonstrate their full capacity for JA.

Secondly, the P-scale and Communication Matrix scores for this group of children fell within the lowest levels of attainment measured by these assessments. P-scales range from levels 1-8 with participants in this study functioning mainly within the first two levels. The highest potential score on the Communication Matrix is 160 but 16 of the 17 participants in this study achieved a score of 35 or less and nine achieved a score of less than 10. The difficulties in using standardised assessments to differentiate between performance at these lowest levels of attainment is well documented (e.g. Crais, 2011; Tassé et al., 2013) and provided a partial motivation for the current piece of work. It may be, therefore, that scores on the P-Scales and Communication Matrix did not provide a sufficiently accurate measure of cognition or communication to use for comparison with JABM scores.

### **8.6.3. Research question 3: Are structured probes an effective means of eliciting information about JA behaviours?**

In judging whether the structured probes were effective it is necessary to consider whether they did, indeed, elicit JA behaviours and whether such information could have been gathered equally or more effectively using other methods such as observation in unstructured play.

All four probes used in the JABM were found to elicit the full range of target JA behaviours although they differed in the frequency with which they did so.

The Surprise Puppy was the most effective probe in terms of engaging participants' visual attention, resulting the smallest number of 'no fixation' trials and the highest number of gaze shifts. However, a relatively high proportion of these gaze shifts were scored as AOP, being unaccompanied by communicative signals. It is possible, then that this probe is particularly effective at eliciting checking looks, with participants looking to see if the tester had also noticed the puppy. However, it is also possible that the AOP gaze shifts used during this probe represented some dividing of visual attention between tester and puppy since this was the only probe in which both referents were actively engaging at the same time, the tester continuing to talk or sing while the puppy moved around above her shoulder. Adapting the probe so that the tester ceases to talk when the puppy appears would help to address this.

It was notable that, despite being the only truly 'top-down' context, participants rarely made active and persistent attempts to direct the tester's attention to the puppy during this probe. From observation, this contrasted with the behaviour of the typically developing reference group who tested the measure, the eldest of whom were very persistent in trying to share attention about the puppy, using pointing and vocalising and sometimes continuing to point in the direction of the puppy even after it had disappeared. This may be an indicator that children with PIMD have not yet reached a developmental phase in which they are motivated to direct a communication partner's behaviour for JA but it may also reflect the physical limitations on their ability to use conventional gestures such as pointing.



The Dynamic Object (2) probe elicited the highest proportion of AOP+ gaze shifts and trials judged to signify communicative JA. These were mainly judged to be requests which was unsurprising given the context, since the probe involved an engaging item which had become inactive making it likely that any attempts at communication would be interpreted as being requests.

The Phone probe was the least effective at eliciting JA behaviours, leading to a relatively high proportion of 'no fixation' trials and relatively few referential gaze shifts between person and object. Although it was designed to provide a context which might be familiar to participants, it is possible that ringing phones are such a routine occurrence that it did not engage their interest to a sufficient degree. The Phone probe was also subject to technical failure if there was no phone signal and this increased the occurrence of null trials.

The Look at the Ball probe elicited some instances of RJA in most participants, being easy to administer and, arguably more feasible than the 'poster' probes used for the ESCS. It also had the advantage of using real, 3D objects which were more suitable for young people with PIMD than picture book stimuli also used by the ESCS.

Although the probes succeeded in eliciting JA behaviours and providing some differentiation between participants in this regard, it was notable that they were far from eliciting referential gaze shifting or communicative JA in all trials. This was the case for both the reference group of typically developing infants on whom they were tested and the participants with PIMD. This may partially reflect limits in participants' capacity for engaging in such behaviours but is also a function of using structured sampling techniques where opportunities rather than demands for target behaviours are created. Therefore, in order to collect useful amounts of information, it is necessary for structured sampling procedures to include as many opportunities as possible for target behaviours to occur. Administering the JABM on three occasions facilitated this as well as meaning that variability in participant's responses caused by fluctuating engagement states did not limit the data obtained.

The scoring system for the JABM had been devised to focus on observable behaviour although some subjective judgements around whether communicative JA had occurred were also included. Analysis of inter-rater agreement indicated that, as might be expected, judgements about observable behaviours achieved a higher degree of agreement than judgements about whether communicative JA had occurred. Inter-rater agreement for JABM scores was rated as 'good' for focus of attention (AO and AP), the presence of gaze shifts (AOP and AOP+) and whether the gaze shift was accompanied by a PCB. However, judgements about whether JA had been part of intentional communication (IJA) and the type of communication used (IJA(dec) or IJA(imp) only achieved a 'fair' level of agreement, as did judgements about the presence of a sharing look. This finding highlights the need for reliable assessments to have a scoring system based on what can be observed. Those which rely solely on subjective interpretations of these observed behaviours will be less effective at reflecting progress over time since changes in scoring may be caused by changes in rater. That said, it could be argued that subjective interpretations are also significant since, if behaviours such as AOP+ gaze shifts are not perceived to be communicative, this will influence the responses of communication partners in everyday contexts. A comprehensive assessment might, then, include both types of score but should differentiate clearly between those which are based on behaviour and those which are based on interpretation.

The structured probes used in the JABM were successful in eliciting JA behaviours but within a context which was highly structured and controlled and which might, therefore, be deemed somewhat unnatural. This raises a question around the extent to which the performance of participants is representative of their 'everyday' behaviour which will form the focus for support and intervention. Administering such probes also requires a degree of preparation, time and knowledge on the part of the tester which is only worthwhile if it yields information which cannot be gained through less structured approaches. Comparing information gained from the unstructured play measure with that gathered by the JABM allowed these issues to be addressed.

The high degree of correlation between scores for JA behaviour on the unstructured play measure and JABM scores, suggested that participant performance on the probes were able to reflect performance in less structured settings. However, the structured probes had advantages over the unstructured play measure meaning that they could provide more accurate and detailed information about a wider range of behaviours. Firstly, by controlling the behaviour of referents (both object and person), the probes made it easier to differentiate between gaze shifts which might be intrinsically motivated from those which were caused by external factors such as referents becoming more engaging. Such differentiation was difficult to achieve during the unstructured play measure since partners talked and reacted freely, often doing so at the same time as an item was activating. As a result, if a participant looked from a light spinner to a communication partner who suddenly smiled and said “ooh, I like that!”, it was not possible to infer whether the shift had occurred because the communication partner had become more engaging than the light spinner when they started talking or because the participant was motivated to communicate their enjoyment of the spinner.

Secondly, the probes provided opportunities for an unexpected event to occur (the Surprise Puppy) and for gaze following, both of which are less likely to occur in unstructured settings, a suggestion supported by evidence from the exploratory phase of this study in which no opportunities for responding to JA were seen to arise during observation of everyday contexts. Relying solely on observations of unstructured play would mean that participants would be unable to demonstrate their capacity for JA behaviour in these contexts.

Finally, it should be noted that, whilst the unstructured play measure used in this study offered less structure than the probes, it was not a wholly unstructured or natural context. Indeed, it shared some features of the probes, with an adult actively introducing a number of engaging items and sessions being led by a tester who was both accustomed to administering the JABM and focusing on eliciting JA and who may, therefore, have been unconsciously biased towards encouraging JA behaviours. It is possible that

this led to participants showing a higher number of JA behaviours during this session than they would generally do.

The unstructured play measure differed from the JABM in involving a familiar adult rather than just the unfamiliar tester. As a group, participants remained more likely to engage the tester in their gaze shifts than the familiar adult, possibly because she was directing the session, however, a small number of participants were more likely to involve the familiar adult in their gaze shifts. This provides an argument for increasing the involvement of a familiar adult in administering structured probes, something that would be possible if time was allowed for giving them suitable training.

Overall, in addressing the research questions, the findings of this study indicated that children with PIMD demonstrate a variety of JA behaviours and that these might constitute a means of differentiating between behaviours for assessment purposes. Findings offered tentative support for the suggestion that RJA and IJA may be independent neural systems (Mundy & Newell, 2007). They also supported the concept that there are two types of qualitatively different gaze shifts - 'checking looks' and 'sharing looks' (Hobson & Hobson, 2007) which may reflect the difference between JA which is considered lean or rich. Functional vision abilities were found to have a strong and significant impact on JA behaviours which could not be predicted on the basis of motor or cognitive skills as assessed by the background measures used in this study. Finally, structured probes which provide a range of different contexts for JA behaviours to occur (including both top-down and bottom-up situations) were found to be an effective means of gathering information about JA behaviours and, thus, should be considered a useful addition to current means of assessing communication in children with PIMD. In the next section, the clinical issues which motivated this thesis will be re-visited in the light of the findings and conclusions discussed above.

## **9. The contribution made by this thesis to the field of SLT assessment for people with PIMD**

As outlined in Chapter 2, current approaches to SLT assessment for people with PIMD are characterised by inconsistency, with SLTs showing varied judgements about what areas should be assessed and relying heavily on indirect forms of evidence gathering. Where shared frameworks are used, these rely heavily on caregiver report and do not incorporate theoretical advances in the field of prelinguistic communication.

The findings of this thesis have addressed these issues by highlighting the growing understanding of joint attention as a significant area in pre-linguistic communication and the benefits of using structured sampling techniques to gather information for assessment. The potential implications of these findings for clinical practice will now be considered.

### **9.1. The significance of JA behaviours in communication assessment for people with PIMD**

The findings of this thesis combined with an appraisal of current thinking around the development of pre-linguistic communication indicate that communication assessment should include a focus on JA. Whilst JA has long been understood to be a feature of intentional communication, it does not appear to be a widely-used focus of assessment in its own right. Only 40% of SLTs surveyed in the current study reported assessing attention and some of these referred broadly to assessing ‘attention and listening’ and ‘attention span’ rather than aspects of joint attention. Whilst actual engagement in JA, whether lean or rich, appears to occur infrequently in this population, this thesis has demonstrated that there is a continuum of behaviour underlying this skill and that by assessing the behaviours on this continuum in detail it is possible to differentiate performance in children with PIMD. Differentiating in such a way provides a measure of an individual’s current capacity for JA and could be used to inform appropriate goal setting support and intervention including the relevance of various forms of Augmentative and Alternative Communication.

The importance of the functional vision skills of gaze fixation and gaze shifting in relation to demonstration of JA behaviours has also been highlighted by the findings of this thesis. Some SLTs surveyed as part of this thesis were aware of the need to understand the visual skills of individuals with PIMD, with 16% reporting that they considered them as part of their assessment. However, the current lack of an efficient tool for assessing gaze fixation and gaze shifting in this population suggests that these particular skills rarely provide a focus for visual assessment. The techniques adopted to measure functional gaze for this study, using real, 3D stimuli might provide a basis for an effective test of functional gaze although further work would be required to develop them into a clinical tool.

It should be noted, however, that JA is not necessarily dependent on gaze behaviour in all populations. While gaze shifting between people and objects is a key component of JA in typically developing infants, infants with visual impairments develop alternative means of sharing attention to referents. This might include changes in body orientation and touching the communication partner. For example Nunez (2016) describes a child who is profoundly deaf and blind bringing her mother's hand to a toy, placing her own right hand on her mother's chest and vocalising with pleasure. Given the high incidence of visual impairment in people with PIMD it is possible that they, too might use behaviours which are not gaze related to engage in JA. However, in this respect they may be limited by their physical abilities and it was notable that none of the participants in this study appeared to be using alternatives to gaze behaviour as a means of engaging in JA. Nevertheless, any clinical assessment of JA in people with PIMD should consider both gaze related and alternative behaviours when considering whether an individual is attending to people and objects or making attempts to integrate attention between the two.

The importance of a focus on JA as part of pre-linguistic communication assessment which is highlighted by the work in this thesis, provides support for the Communication Complexity Scale (CCS) , an assessment tool which, as noted in Chapter 3, has been under development during the period of time in which this thesis has been constructed. The CCS aims to assess early communication skills in pre-symbolic communicators with intellectual disabilities (Brady et al 2012). In doing so, it focuses on a similar sequence of JA behaviour to the current study since it is also based on a typical developmental continuum. The observable behaviours identified by the CCS are related to communication levels ranging from pre-intentional to intentional symbolic. The most recent version of the CCS, as reported by its authors, (Brady, 2018) may be seen in Figure 9.1.

| No. | Definition  | Communication level     |
|-----|---|-------------------------|
| 0   | No response   |                         |
| 1   | Alerting – a change in behavior, or stops doing a behavior  | Preintentional          |
| 2   | Single orientation only – on an object, event, or person; can be communicated through vision, body orientation, or other means                          | Preintentional          |
| 3   | Single orientation only and a PCB <sup>1</sup>  | Preintentional          |
| 4   | Single orientation only + more than 1 PCB   | Preintentional          |
| 5   | Dual orientation – shift in focus between a person and an object, between a person and an event using vision, body orientation, and so on (without PCB) | Preintentional          |
| 6   | Triadic orientation (e.g., eye gaze or touch from object to person and back)  | Intentional nonsymbolic |
| 7   | Dual orientation + 1 PCB (e.g., dual focus + gesture)   | Intentional nonsymbolic |
| 8   | Dual orientation + 2 or more PCB (e.g., dual focus + gesture + vocalization, and switch closure)  | Intentional nonsymbolic |
| 9   | Triadic orientation + 1 PCB (e.g., triadic + vocalization)  | Intentional nonsymbolic |
| 10  | Triadic orientation plus more than 1 PCB (e.g., triadic plus vocalization and differential switch closure)  | Intentional nonsymbolic |
| 11  | One-word verbalization, sign, or AAC symbol selection   | Intentional symbolic    |
| 12  | Multi-word verbalization, sign, or AAC symbol selection   | Intentional symbolic    |

*Note.* Copyright © 2017 by The University of Kansas. Printed with permission. CCS = Communication Complexity Scale; PCB = potentially communicative behavior; AAC = augmentative and alternative communication.

*Figure 9.1. The Communication Complexity Scale (Brady et al, 2018)*

The CCS, assigns individuals a single score based on the most complex behaviours they are seen to demonstrate. Scoring may be based on information from observation in any context but, to date, the authors of the CCS have described scoring it on the basis of ‘scripted protocols’. The exact nature of these protocols has not been reported in detail but, as with the structured probes in this study, it is reported that some are based on activities used by the ESCS and are designed to offer opportunities to make requests and engage in JA through the use of interrupting an engaging activity or introducing a novel event. It is clear, therefore, that the development of the CCS mirrors the evolution of the work carried out for this thesis in several respects including its focus on JA and an acknowledgement of the benefits of structured sampling. This fact supports the relevance of these areas to assessment of prelinguistic communicators with intellectual disabilities. However, the work contained in this thesis raises questions around certain aspects of the CCS.

Firstly, at the present time the CCS does not offer sufficient justification for the sequence of communicative complexity it proposes. It differentiates between ‘dual orientation’ (in which there is a shift of focus between a person and object or event) and ‘triadic orientation’ (in which focus shifts from an



object to a person and back) but the papers published to date (Brady et al., 2018; Brady et al., 2012) provide no theoretical explanation for this distinction. Indeed, there would appear to be some inconsistency in the suggested sequence of these behaviours which is described by the CCS since engaging in unaccompanied triadic orientation is scored more highly than engaging in unaccompanied dual orientation but engaging in dual orientation accompanied by two or more PCBs achieves a higher score than engaging in triadic orientation accompanied by only one PCB. This constitutes a somewhat confusing sequence based on the combination of orientation type and number of PCBs. The CCS also categorises unaccompanied dual orientation as 'preintentional communication' while unaccompanied triadic orientation is categorised as 'intentional communication' despite the absence of any PCBs, the reasons underpinning this distinction being unexplained. Whilst distinguishing between dual and triadic orientation is consistent with the findings of Arens and colleagues (2005) and those of the current study in suggesting that the predominant use of either two or three point gaze shifts may be a differentiating factor in this population, the significance of two-point gaze shifts or dual orientation has not been fully explored and the sequence of progression between dual and triadic focus, particularly when accompanied by communicative signals is not yet established. Indeed, the findings of the current study suggest that functional vision skills, rather than developments in communicative complexity may play a role in this sequence. Further clarification would be beneficial in order to ensure that the CCS becomes a useful clinical tool.

Secondly, the CCS focuses on observable behaviours which are considered to indicate JA and intentional communication. These behaviours, which involve integrated attention between person and object accompanied by PCBs, equate to the category of AOP+ which was used in work reported in this thesis. The current study found that raters did not judge all AOP+ behaviours to indicate intentional communication, suggesting that focusing solely on such behaviour patterns is not sufficient to reflect whether an individual is perceived as an intentional communicator by those interacting with them. Further investigation of the difference between AOP+ behaviours

which are or are not judged to indicate intentional communication would help to clarify this area.

In summary, the work within this thesis highlights the significance of joint attention behaviours and the need for communication assessment to include a measurement of capacity for these behaviours alongside consideration of an individual's functional ability to fix and shift gaze. This supports parallel work in the field carried out by Brady and colleagues.

## **9.2. The use of structured sampling for communication assessment with people who have PIMD**

The methodology used in this thesis has also demonstrated how structured sampling may be used to elicit and measure key communication behaviours in children with PIMD. The structured probes developed to measure JA behaviours as part of the reported studies effectively provided a context in which individuals could demonstrate their capacity to engage in such behaviours when optimal conditions were provided. Controlling communication partner behaviour and the physical environment increased the likelihood that variations in performance were due to individual capacity for engaging in the behaviours rather than external factors. While it remains important to assess an individual's performance in everyday contexts, evaluating the difference between an individual's capacity and their performance provides an essential foundation for planning intervention and support, ensuring that individuals are able to make use of their potential and enabling realistic, achievable goals to be set.

Structured sampling is currently underused in the clinical context of communication assessment for PIMD. Out of the SLTs surveyed in Chapter 3, only 20% reported using direct engagement with objects as a means of gathering information for assessment and there are no published or widely shared protocols incorporating such techniques which are specifically aimed at people with PIMD. This situation may reflect concerns about adopting a 'one-size-fits-all' approach for such a heterogeneous group who are characterised by fluctuating engagement. However, the findings reported here indicated that developing activities with the needs of children of PIMD in mind and allowing for some flexibility enables a suitable protocol to be developed although repeated administration is essential.

Underuse of structured sampling may also reflect appropriate awareness of a need for ecological validity when carrying out assessment and concerns that an individual's behaviour in an artificially structured situation does not

reflect their 'real life' performance. However, findings of this research suggested that children's behaviour demonstrated during the structured probes was broadly consistent with the skills they showed in less structured contexts such as an unstructured play session, with the structured probes enabling them to demonstrate behaviours more frequently and, in some cases, providing opportunities which occurred rarely in everyday contexts. As discussed, using structured probes provided a measure of capacity which could be compared to performance in naturalistic contexts. Structured sampling is, therefore, proposed as an addition to current methods of information gathering such as observation in everyday settings and caregiver interview rather than a replacement for them.

Finally, structured sampling would address the lack of consistency in SLT approaches to assessment. Adopting a standard protocol and scoring system alongside the use of video recordings could provide a form of measurement which can be repeated reliably over time regardless of changes in practitioner.

### 9.3. Review of the methodology

The structured probes designed for this study offered multiple opportunities for participants to initiate JA but included only one context in which RJA was measured, meaning that the amount of data relating to this skill was limited. Including a greater number of structured probes for measuring RJA as part of different activities would have enabled more detailed conclusions to be reached about this area as would work investigating whether individuals with PIMD are more likely to follow the turn of a head with open eyes than with closed eyes, a potential indicator of capacity for rich JA (Brooks & Meltzoff, 2015),

The probes which measured IJA varied in their effectiveness, with the Phone probe being of limited value since it did not elicit a great deal of communicative JA and was subject to technical issues. The Look at the Puppy probe proved engaging for participants and elicited high degrees of gaze shifting but better control of the socio-interactive context would have ensured that these shifts were not caused by extrinsic cues since the tester was talking at the same time as the puppy was being presented.

Whilst the unstructured play session offered an insight into the JA behaviours demonstrated by participants in less structured contexts, it was not wholly naturalistic, occurring solely for the purposes of the research, and it shared some characteristics with the JABM. Carrying out a more detailed analysis of the JA behaviours through the use of video recording in everyday contexts would enable a more detailed comparison between performance on the JABM and performance in naturalistic contexts to be carried out.

The presence of a familiar adult in the unstructured play context highlighted the possible effect that this might have on some participants suggesting that the inclusion of a familiar adult in the administration of all structured probes could be valuable.

The work described in this thesis has focused on visual behaviours and the effect which impairment in this area may have on joint attention. Hearing impairment is also known to be prevalent in children with PIMD but the

possible impact of hearing impairment on joint attention was not a focus of this study. Although none of the structured probes were reliant on hearing, some (such as marbles descending the marble tree) included auditory aspects. The hearing abilities of participants were not tested as part of the research protocol and, therefore, the extent to which any deficits in hearing or auditory processing affected outcomes of the study are unknown.

Finally, the results obtained from this study relate to young people with PIMD. It is likely that they might generalise to adults with PIMD, given the persistence of profound impairments in this population but further administration of measures to an older sample would be needed to ascertain this.

#### 9.4. Future Directions

The work carried out for this thesis has contributed to understanding of the JA behaviours demonstrated by children with PIMD. However, it has highlighted the need to investigate certain areas further. These areas include, firstly, the relevance of two-point gaze shifts (or 'dual orientation') to the development of JA in both typically developing children and people with PIMD. Secondly, the factors which lead raters to judge whether gaze shifts accompanied by potentially communicative signals do, indeed, serve a communicative function, should be explored and, finally, the significance of RJA to communication development of PIMD is currently underdeveloped and would merit further consideration.

From a clinical perspective, this thesis suggests that methods for assessing functional vision (particularly the ability to fix and shift gaze) in people with PIMD require further development with some of the procedures adopted in the FGBM providing a possible starting point, although adaptation would be needed for these feasible in a clinical context. A structured sampling protocol for assessing early communication skills including JA behaviours should also be developed and validated for clinical use. Again, the measures developed for the thesis, specifically the JABM, provide a useful foundation for ongoing work.

#### 9.5. Conclusion

This thesis has highlighted the significance of JA in pre-linguistic communication and contributed to knowledge about JA in people with PIMD. Its findings suggest that current approaches to SLT assessment for people with PIMD could be improved by focusing more consistently on behaviours relating to JA and that structured sampling should be included alongside other means of information gathering to provide a measure of capacity for communication behaviours under optimal conditions and increase consistency in approach between practitioners.

## 10. References

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## 11. Appendices

### Appendix A: Paper version of electronic questionnaire for Speech and Language Therapists working with clients with PIMD

Thank you for agreeing to take part in this survey. It asks questions about your clients who are described here as having 'Profound and Multiple Learning Disabilities' although they may also be described in other ways e.g. as having 'complex needs'. A definition of the group in question is included below to help you with your responses:

People with profound intellectual and multiple disabilities [or 'profound and multiple learning disabilities' - PMLD] are among the most disabled individuals in our community. They have a profound intellectual disability which means that their intelligence quotient is estimated to be under 20 and therefore that they have severely limited understanding. In addition, they have multiple disabilities which may include impairments of vision, hearing and movement as well as other problems like epilepsy and autism. Most people in this group are unable to walk unaided and many people have complex health needs requiring extensive help. People with profound intellectual and multiple disabilities have great difficulty communicating; they typically have very limited understanding and express themselves through non-verbal means or at most through using a few words or symbols.

Definition taken from the 'Raising our Sights: Services for adults with profound intellectual and multiple disabilities'

by Professor Jim Mansell (2009)

1. Into which age group do your clients with PMLD fall?

0-5 years

5-11 years

11-19 years

19+

2. For how many years have you been working as a Speech and Language Therapist?

0-2 years



2-5 years

5-10 years

10+ years

3. Please detail which key areas of communication and behaviour you consider when assessing clients with Profound and Multiple Learning Disabilities?
4. What methods do you use to assess the communication skills of clients who have Profound and Multiple Learning Disabilities?
5. Are there any other comments you would like to make about assessing clients with Profound and Multiple Learning Disabilities?

**Appendix B: Observation Checklist for use of target JA behaviours in everyday contexts**

Child's Initials: \_\_\_\_\_ Date: \_\_\_\_\_

Time: \_\_\_\_\_


Description of session/activity observed:

| Gaze behaviour   |   |                 |
|--|---|-----------------|
|  | <b>Scoring</b><br>0 = does not engage in target behaviour<br>1 = engages in target behaviour on at least one occasion | <b>Examples</b> |
| Attends to objects   |   |                 |
| Attends to people  |   |                 |
| Looks from an object to a person or vice versa, possibly looking back and forth. |   |                 |

| Initiating Joint Attention  |   |                 |
|---|---|-----------------|
|   | <b>Scoring</b><br>0 = does not engage in target behaviour<br>1 = engages in target behaviour on at least one occasion | <b>Examples</b> |
| Engages in joint attention to request or reject an object               |   |                 |
| Engages in joint attention to 'comment' or share affect about an object |   |                 |

| <b>Responding to Joint Attention</b><br><b>(Following another person's head turn, gaze or point)</b> |  |                        |
|--|--|------------------------|
|  | <b><u>Scoring</u></b><br><b>0 = does not engage in target behaviour</b><br><b>1 = engages in target behaviour on at least one occasion</b> | <b><u>Examples</u></b> |
| Looks from a person to an object that they turn to look and/or point at.                             |  |                        |

## Appendix C: The FunVis assessment: target behaviours and procedure.



### SCREENING PROCEDURE FOR SCHOOL STAFF

**Please return this to us in the envelope provided**

**OR Complete the questionnaire online: [www.ucl.ac.uk/gaze](http://www.ucl.ac.uk/gaze)**

To be completed by school staff (e.g. speech and language therapist). Please use the smiley faces provided to complete the following activities with the child. Please hold each smiley face by the handle (stick). Please hold the faces still when presenting them. Do not shake or jiggle the faces to attract the child's attention. Verbal prompts such as "Oh look! It's a face!" are acceptable.

Please **tick the box** corresponding to your answer from each question:

1. First gain the child's attention. Then present the smiley face (provided) to the child at their eye level and at arm's length away. Does the child look directly at the smiley face?  

|                              |                             |
|------------------------------|-----------------------------|
| Yes <input type="checkbox"/> | No <input type="checkbox"/> |
|------------------------------|-----------------------------|
2. Place the smiley face on a table or the child's wheelchair tray, about arm's length from the child. Does the child look directly at the smiley face?  

|                              |                             |
|------------------------------|-----------------------------|
| Yes <input type="checkbox"/> | No <input type="checkbox"/> |
|------------------------------|-----------------------------|
3. Hold up the two contrasting smiley faces at the child's eye level, approximately 30cm apart (12 inches), at arm's length from the child. Does the child look from one smiley face to the other in turn?  

|                              |                             |
|------------------------------|-----------------------------|
| Yes <input type="checkbox"/> | No <input type="checkbox"/> |
|------------------------------|-----------------------------|
4. Place two contrasting smiley faces on a table or wheelchair tray approximately 30cm apart (12 inches) and at arm's length from the child. Does the child look from one smiley face to the other in turn?  

|                              |                             |
|------------------------------|-----------------------------|
| Yes <input type="checkbox"/> | No <input type="checkbox"/> |
|------------------------------|-----------------------------|
5. Hold up the smiley face and the blank circle at the child's eye level, approximately 30cm (12 inches) apart, at arm's length from the child. The child may look at both but do they show a preference for the face by fixing their gaze on it?  

|                              |                             |
|------------------------------|-----------------------------|
| Yes <input type="checkbox"/> | No <input type="checkbox"/> |
|------------------------------|-----------------------------|
6. Hold up the smiley face at the child's eye level. Now, trying not to shake it, move it slowly and smoothly horizontally in front of the child, first to their left and then to the right. Does the child follow the smiley face with their eyes (head movement may also be involved)?  

|                              |                             |
|------------------------------|-----------------------------|
| Yes <input type="checkbox"/> | No <input type="checkbox"/> |
|------------------------------|-----------------------------|
7. When you smile at the child (without speaking), does the child return a smile?  

|                              |                             |
|------------------------------|-----------------------------|
| Yes <input type="checkbox"/> | No <input type="checkbox"/> |
|------------------------------|-----------------------------|
8. During these activities, did the child make direct eye contact with you?  

|                              |                             |
|------------------------------|-----------------------------|
| Yes <input type="checkbox"/> | No <input type="checkbox"/> |
|------------------------------|-----------------------------|
9. Does the child appear to have a squint? By a squint we mean that both eyes **do not** appear to be straight and looking together at the same time.  

|                              |                             |
|------------------------------|-----------------------------|
| Yes <input type="checkbox"/> | No <input type="checkbox"/> |
|------------------------------|-----------------------------|
10. Do you have any other observations or concerns you would like to add?

## Appendix D : Performance Descriptors for P-Scales 1 – 4 in English (Department for Education, 2014)

N.B. P Scales 1-3 in English have the same descriptor across subject. P-Scale 4 is differentiated for Speaking, Listening and Reading

### English

Performance descriptors across subjects:

#### **P1 (i) Pupils encounter activities and experiences**

- They may be passive or resistant
- They may show simple reflex responses, [for example, startling at sudden noises or movements]
- Any participation is fully prompted.

#### **P1 (ii) Pupils show emerging awareness of activities and experiences**

- They may have periods when they appear alert and ready to focus their attention on certain people, events, objects or parts of objects [for example, attending briefly to interactions with a familiar person]
- They may give intermittent reactions [for example, sometimes becoming excited in the midst of social activity].

#### **P2 (i) Pupils begin to respond consistently to familiar people, events and objects**

- They react to new activities and experiences [for example, withholding their attention]
- They begin to show interest in people, events and objects [for example, smiling at familiar people]
- They accept and engage in coactive exploration [for example, focusing their attention on sensory aspects of stories or rhymes when prompted].

#### **P2 (ii) Pupils begin to be proactive in their interactions**

- They communicate consistent preferences and affective responses [for example, reaching out to a favourite person]
- They recognise familiar people, events and objects [for example, vocalising or gesturing in a particular way in response to a favourite visitor]
- They perform actions, often by trial and improvement, and they remember learned responses over short periods of time [for example, showing pleasure each time a particular puppet character appears in a poem dramatised with sensory cues]
- They cooperate with shared exploration and supported participation [for example, taking turns in interactions with a familiar person, imitating actions and facial expressions].

## Appendix D: Performance descriptors for P-Scales 1-4 in English (cont.)

### **P3 (i) Pupils begin to communicate intentionally**

- They seek attention through eye contact, gesture or action
- They request events or activities [for example, pointing to key objects or people]
- They participate in shared activities with less support. They sustain concentration for short periods.
- They explore materials in increasingly complex ways [for example, reaching out and feeling for objects as tactile cues to events]
- They observe the results of their own actions with interest [for example, listening to their own vocalisations]
- They remember learned responses over more extended periods [for example, following the sequence of a familiar daily routine and responding appropriately].

### **P3 (ii) Pupils use emerging conventional communication**

- They greet known people and may initiate interactions and activities [for example, prompting another person to join in with an interactive sequence].
- They can remember learned responses over increasing periods of time and may anticipate known events [for example, pre-empting sounds or actions in familiar poems]
- They may respond to options and choices with actions or gestures [for example, by nodding or shaking their heads]
- They actively explore objects and events for more extended periods [for example, turning the pages in a book shared with another person]
- They apply potential solutions systematically to problems [for example, bringing an object to an adult in order to request a new activity].

#### **Speaking**

### **P4 (Speaking) Pupils repeat, copy and imitate between 10 and 50 single words, signs or phrases or use a repertoire of objects of reference or symbols**

- They use single words, signs and symbols for familiar objects [for example, cup, biscuit], and to communicate about events and feelings [for example, likes and dislikes].

### **P4 (Listening) Pupils demonstrate an understanding of at least 50 words, including the names of familiar objects**

- Pupils respond appropriately to simple requests which contain one key word, sign or symbol in familiar situations [for example, 'Get your coat', 'Stand up' or 'Clap your hands'].

### **P4 (Reading) Pupils listen and respond to familiar rhymes and stories**

- They show some understanding of how books work [for example, turning pages and holding the book the right way up]

**Appendix E: Protocol for the structured probes used in the exploratory phase of study**

| <b>Trial</b> | <b>Probe</b>       |
|--------------|--------------------|
| <b>1</b>     | Dynamic object     |
| <b>2</b>     | Look at the puppet |
| <b>3</b>     | Pencil tin         |
| <b>4</b>     | Look at the puppet |
| <b>5</b>     | Dynamic object     |
| <b>6</b>     | Pencil tin         |
| <b>7</b>     | Dynamic object     |
| <b>8</b>     | Look at the puppet |
| <b>9</b>     | Pencil tin         |
| <b>10</b>    | Look at the puppet |
| <b>11</b>    | Dynamic object     |
| <b>12</b>    | Pencil tin         |
| <b>12</b>    | Dynamic object     |
| <b>14</b>    | Look at the puppet |
| <b>15</b>    | Pencil tin         |

## Appendix F: Protocol and score sheet for the JABM

| <b>Trial</b> | <b>Probe</b>                           | <b>Gaze fixation</b><br><b>O, P O+P</b> | <b>Gaze Shifting</b><br><b>OP, PO, OPO,POP</b> | <b>Additional Signals</b><br><b>e.g. vocallisation</b><br><b>body movement</b> | <b>Looks to same object as assessor?</b> | <b>Seems to communicate to share attention or request?</b> | <b>Sharing Look?</b> |
|--------------|--|---|--|--|--|--|----------------------|
| 1            | Dynamic Obj 1<br>(on right):           |   |  |  |  |  |                      |
| 2            | Dynamic Obj. 2<br>(on right)           |   |  |  |  |  |                      |
| 3            | Look at the ball:<br>(Object on Left)  |   |  |  |  |  |                      |
| 4            | Surprise Puppy<br>(on right)           |   |  |  |  |  |                      |
| 5            | Phone<br>(on left)                     |   |  |  |  |  |                      |
| 6            | Look at the ball:<br>(Object on Right) |   |  |  |  |  |                      |
| 7            | Surprise Puppy<br>(on left)            |   |  |  |  |  |                      |



## Appendix F: Protocol and score sheet for the JABM (cont.)

| Trial | Probe                                  | Gaze fixation<br>O, P O+P | Gaze Shifting<br>OP, PO,<br>OPO,POP | Additional Signals<br>e.g.<br>vocalisation<br>body movement | Looks to same<br>object as<br>assessor? | Seems to<br>communicate to<br>share attention<br>or request? | Sharing<br>Look? |
|-------|--|---------------------------|-------------------------------------|---|---|--|------------------|
| 8     | Phone<br>(on right)                    |                           |                                     |   |   |  |                  |
| 9     | Dynamic Obj 1<br>(on left)             |                           |                                     |   |   |  |                  |
| 10    | Dynamic Obj 2<br>(on left)             |                           |                                     |   |   |  |                  |
| 11    | Dynamic Obj 1<br>(on right)            |                           |                                     |   |   |  |                  |
| 12    | Dynamic Obj 2<br>(on right)            |                           |                                     |   |   |  |                  |
| 13    | Look at the ball:<br>(Object on Left)  |                           |                                     |   |   |  |                  |
| 14    | Surprise Puppy<br>(on right)           |                           |                                     |   |   |  |                  |
| 16    | Phone                                  |                           |                                     |   |   |  |                  |
| 16    | Look at the ball:<br>(Object on Right) |                           |                                     |   |   |  |                  |

**Appendix F: Protocol and score sheet for the JABM (cont.)**

| <b>Trial</b> | <b>Probe</b>                    | <b>Gaze fixation</b><br><br><b>O, P O+P</b> | <b>Gaze Shifting</b><br><br><b>OP, PO, OPO,POP</b> | <b>Additional Signals</b><br><b>e.g. vocallisation</b><br><b>body movement</b> | <b>Looks to same object as assessor?</b> | <b>Seems to communicate to share attention or request?</b> | <b>Sharing Look?</b> |
|--------------|---------------------------------|---|--|--|--|--|----------------------|
| 17           | Surprise Puppy (on left)        |   |  |  |  |  |                      |
| 18           | Phone                           |   |  |  |  |  |                      |
| 19           | Dynamic Object 1 (on left)      |   |  |  |  |  |                      |
| 20           | Dynamic Obj 2 Phase 2 (on left) |   |  |  |  |  |                      |

## Appendix G: Score sheet for the unstructured play measure

Name:

Date:

Visit:

School:

Assessor:

Assistant:

Sheet scored by:

| Gaze shift number | Point on video at which gaze shift is observed | Direction of shift<br><br>O, P, OP, PO, OPO, POP | Additional Signals<br><br>Facial Expr (F)<br>Vocalisation (V)<br>Gesture (G)<br>Other (O) | Person gaze shift includes (assessor (A) or familiar adult (FA)) | Seems to communicate to share attention or request? | Sharing Look? | Comments |
|-------------------|--|--|---|--|---|---------------|----------|
|                   |  |  |   |  |   |               |          |
|                   |  |  |   |  |   |               |          |
|                   |  |  |   |  |   |               |          |
|                   |  |  |   |  |   |               |          |

## Appendix H: Ethics Documentation

This appendix contains the following sections:

Section 1: Details of assent process

Section 2: Notification of original ethical approval and amendments

Section 3: Sample information letters and consent form

### **Section 1: Details of assent process**

The research studies described involved children with PIMD or typically developing infants aged 18 months or less. Since these groups were not deemed to have the necessary cognitive or linguistic skills to give informed consent, consent was obtained from their parents. Parents of participants with PIMD were approached by their schools and parents of typically developing infants were approached by their child's nursery or a member of the research team.

Parents were made aware of details of the research study through information letters and were provided with contact details should they wish to ask further questions. If they were happy to proceed they were asked to sign a consent form. They were informed of their right to withdraw consent at any time.

Participants' behaviour was monitored throughout the research protocol to ensure that they appeared happy to engage and were not demonstrating any signs that they wished to withdraw (such as making unhappy noises, grimacing, pulling away, pushing items away or closing their eyes). Since such engagement/withdrawal behaviours were unique to each individual, a familiar adult accompanied them at all times and was advised to tell the research team if they observed signals that the participant seemed unhappy. If either the research or familiar adult observed signs of disengagement or distress, the session would be paused. Efforts would be made to re-engage the participant (for example, by changing to a different activity allowed by the protocol) or make them comfortable (for example, by addressing physical needs, talking or singing to them or suggesting some interaction with the familiar adult). If signs of distress persisted, the

session was terminated. Attempts would be made to repeat the session on a future occasion but if signs of discomfort or disengagement were repeated, the participant would be withdrawn from the study.

## Section 2: Notification of original ethical approval and amendments

UCL RESEARCH ETHICS COMMITTEE  
ACADEMIC SERVICES



11 November 2015

Dr Michael Clarke  
Division of Psychology and Language Sciences  
UCL

Dear Dr Clarke

**Notification of Ethical Approval**

**Project ID: 7565/001: Communicative and non-communicative intention in children with profound and multiple learning difficulties**

I am pleased to confirm in my capacity as Chair of the UCL Research Ethics Committee (REC) that your study has been approved by the UCL REC for the duration of the project i.e. until November 2016.

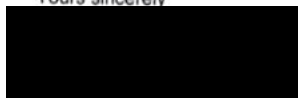
Approval is subject to the following conditions:

1. You must seek Chair's approval for proposed amendments to the research for which this approval has been given. Ethical approval is specific to this project and must not be treated as applicable to research of a similar nature. Each research project is reviewed separately and if there are significant changes to the research protocol you should seek confirmation of continued ethical approval by completing the 'Amendment Approval Request Form': <http://ethics.grad.ucl.ac.uk/responsibilities.php>
2. It is your responsibility to report to the Committee any unanticipated problems or adverse events involving risks to participants or others. The Ethics Committee should be notified of all serious adverse events via the Ethics Committee Administrator ([ethics@ucl.ac.uk](mailto:ethics@ucl.ac.uk)) immediately the incident occurs. Where the adverse incident is unexpected and serious, the Chair or Vice-Chair will decide whether the study should be terminated pending the opinion of an independent expert. The adverse event will be considered at the next Committee meeting and a decision will be made on the need to change the information leaflet and/or study protocol.

For non-serious adverse events the Chair or Vice-Chair of the Ethics Committee should again be notified via the Ethics Committee Administrator ([ethics@ucl.ac.uk](mailto:ethics@ucl.ac.uk)) within ten days of an adverse incident occurring and provide a full written report that should include any amendments to the participant information sheet and study protocol. The Chair or Vice-Chair will confirm that the incident is non-serious and report to the Committee at the next meeting. The final view of the Committee will be communicated to you.

On completion of the research you must submit a very brief report of your findings/concluding comments to the Committee, which includes in particular issues relating to the ethical implications of the research.

Yours sincerely



Professor John Foreman  
Chair of the UCL Research Ethics Committee

Academic Services, 1-19 Torrington Place (9<sup>th</sup> Floor),  
University College London  
Tel: +44 (0)20 3108 8216  
Email: [ethics@ucl.ac.uk](mailto:ethics@ucl.ac.uk)  
<http://ethics.grad.ucl.ac.uk/>



## Amendment Approval Request Form

|   |   |
|---|---|
| 1 | <b>Project ID Number:</b> 7565/001<br><br><b>Name and Address of Principal Investigator:</b><br>Dr Michael Clarke   |
| 2 | <b>Project Title:</b> Communicative and Non-Communicative Intention in Children with Profound and Multiple Learning Difficulties  |
| 3 | <b>Type of Amendment/s (tick as appropriate)</b><br>Research procedure/protocol (including research instruments) <input checked="" type="checkbox"/><br>Participant group <input type="checkbox"/><br>Sponsorship/collaborators <input type="checkbox"/><br>Extension to approval needed (extensions are given for one year) <input checked="" type="checkbox"/><br>Information Sheet/s <input checked="" type="checkbox"/><br>Consent form/s <input type="checkbox"/><br>Other recruitment documents <input type="checkbox"/><br>Principal researcher/medical supervisor* <input type="checkbox"/><br>Other <input type="checkbox"/><br><br><small>*Additions to the research team other than the principal researcher, student supervisor and medical supervisor do not need to be submitted as amendments but a complete list should be available upon request.</small>  |
| 4 | <b>Justification (give the reasons why the amendment/s are needed)</b><br><br>The initial project involved behavioural testing of functional visual skills and intentional communication in children with Profound and Multiple Learning Disabilities. Following this work we wish to follow-up children and recruit new children to a gain more detailed information about the children's visual attention skills, in addition to the use of more formal measures to assess their level of cognitive and communicative development.<br><br>Two masters students working on the original project have now graduated and have been replaced by two new Masters students.<br><br>An extension of time is required to allow further investigation of skills identified as being of interest during the initial part of this project.   |
| 5 | <b>Details of Amendments (provide full details of each amendment requested, state where the changes have been made and attach all amended and new documentation)</b><br><b><u>Amendment 1</u></b><br><b>Applicants</b><br>Applicants (in addition to the principal researcher) for the initial ethics application were Lucy Van Walwyk, Gemma Day and Bethan Jones.<br>Amended applicants are Lucy Van Walwyk (DBS: 001482356766), Alison Nobes (DBS: 001486441484) and Sophie Payne (DBS: to be ascertained).<br><b><u>Amendment 2</u></b><br><b>Research procedure and instruments</b><br>Initial ethical permission was granted for children to attend a direct assessment session which used real objects and play-based activities to assess functional visual skills and the presence of intentional behaviour and intentional communication. In addition, a member of their teaching staff was questioned about their communication skills using a questionnaire designed for the project. |



|   |  |
|---|--|
|   | <p>For the amended procedure children will be visited on four separate occasions. On the first occasion they will be assessed using the Mullens Scales of Early Learning. Their vision will also assessed by a specialist orthoptist and a qualified optometrist. Vision assessment will involve:</p> <ul style="list-style-type: none"> <li>• <u>Visual acuity</u> The quantitative measure of vision will be assessed using preferential looking techniques where their gaze control ability allows. Either Keeler Acuity Cards or Cardiff Acuity Cards and/or iSight application depending upon developmental age.</li> <li>• <u>Refractive status</u> Children's refractive status (their need for glasses) will be objectively assessed using retinoscopy which may require the instillation of eye drops.</li> <li>• <u>Accommodation</u> Accommodation refers to the ability of the lens to change its shape to focus near and distant objects. This will be examined using dynamic retinoscopy.</li> <li>• <u>Eye movements</u> We will assess children's ability to move their eyes into different positions of gaze.</li> <li>• <u>Functional gaze control</u> We will assess children's ability to fix their gaze, transfer gaze between objects and follow moving objects.</li> <li>• <u>Other factors</u> We will note the presence of factors such as squint, nystagmus or gross visual field defects that may influence children's performance.</li> </ul> <p>On each of the subsequent visits the children will attend two sessions:</p> <p><u>Session 1:</u> Assessment of their visual attention skills in a controlled environment (Duration: 20 minutes or less).</p> <p>The child will be positioned in front of a black screen which will be placed on the table in front of them. Either one or two objects (e.g. balloon/squishy ball) or faces will emerge above the screen at intervals. Information will be gathered about whether the child looks at or between them.</p> <p><u>Session 2:</u> Assessment of visual attention, joint attention and intentionality/intentional communication in a naturalistic setting (Duration: 35 minutes or less)</p> <p>This will be a play based session involving activities to engage the child's interest, to provide opportunities for them to look at and between objects and people, to share their interest by looking at the adults in the room and to communicate intentionally to request more of preferred activities.</p> <p>On each visit, participation in the two sessions will be separated by a period of time during which the child may access their usual school day i.e. they will not be required to attend the two sessions consecutively.</p> <p>In addition, the children's school staff will be asked to evaluate the child's communication skills by completing an existing formal assessment – the Communication Matrix (Rowland, accessible online). This has an online format.</p> <p><b><u>Amendment 3</u></b></p> <p><b>Extension</b><br/>Initial approval was granted until July 2016.</p> <p>The amended request is for approval to extend up to December 2017.</p> |
| 6 | <p><b>Ethical Considerations</b> (insert details of any ethical issues raised by the proposed amendment/s)</p> <p>Additional ethical considerations to those detailed in the original application are that the children are now required to participate in two assessment sessions during a day. One with a duration of 20 minutes or less and a second with a duration of 35 minutes or less. To avoid the child becoming tired the sessions will not be administered consecutively and the child will be allowed a period of time (at least 30 minutes) to rest and/or return to their usual classroom environment. A familiar adult will stay with the child. We will closely monitor the children for any signs of distress or fatigue and withdraw assessment for a suitable period if this is identified.</p>  |
| 7 | <p><b>Other Information</b> (provide any other information which you believe should be taken into account during ethical review of the proposed changes)</p>   |



**Declaration** (to be signed by the Principal Researcher)

- I confirm that the information in this form is accurate to the best of my knowledge and I take full responsibility for it.
- I consider that it would be reasonable for the proposed amendments to be implemented.
- For student projects I confirm that my supervisor has approved my proposed modifications.

Signature:

Date: 12.01.17

**FOR OFFICE USE ONLY:**

Amendments to the proposed protocol have been *agreed* by the Research Ethics Committee.

Signature of the REC Chair, Professor John Foreman:

Date: *18/1/2017*

## Amendment Approval Request Form

|          |  |   |
|----------|--|---|
| <b>1</b> | <b>Project ID Number:</b> 7565/001   | <b>Name and Address of Principal Investigator:</b><br>Dr Michael Clarke |
| <b>2</b> | <b>Project Title:</b> Communicative and Non-Communicative Intention in Children with Profound and Multiple Learning Difficulties   |   |
| <b>3</b> | <b>Type of Amendment/s (tick as appropriate)</b><br>Research procedure/protocol (including research instruments) <input type="checkbox"/><br>Participant group <input checked="" type="checkbox"/><br>Sponsorship/collaborators <input type="checkbox"/><br>Extension to approval needed (extensions are given for one year) <input checked="" type="checkbox"/> * To 11/12/2018<br>Information Sheet/s <input checked="" type="checkbox"/><br>Consent form/s <input checked="" type="checkbox"/><br>Other recruitment documents <input type="checkbox"/><br>Principal researcher/medical supervisor* <input type="checkbox"/><br>Other <input checked="" type="checkbox"/><br><br><i>*Additions to the research team other than the principal researcher, student supervisor and medical supervisor do not need to be submitted as amendments but a complete list should be available upon request *</i>  |   |
| <b>4</b> | <b>Justification</b> (give the reasons why the amendment/s are needed)<br><br>The original proposal concerned the assessment of looking and communication skills in children with profound and multiple learning difficulties (PMLD). We wish to replicate the research protocol with a small comparison group of typically developing children of a similar developmental age to the original participants with PMLD. This will allow us to establish the validity of the original protocol.  |   |
| <b>5</b> | <b>Details of Amendments</b> (provide full details of each amendment requested, state where the changes have been made and attach all amended and new documentation)<br><br><b>Extension to approval needed</b><br>A previous amendment provided ethical approval for the project up to December 2017. An extension to this period is requested to allow for testing of the protocol with a group of typically developing children.<br><br><b>Participant Group</b><br>The current amendment requests <u>that we be allowed to work with infants who are at a comparable level of cognitive functioning with the original participant group i.e. up to 30 infants aged between 6 and 18 months old.</u> Participants will be sought by approaching day nurseries offering services to infants of this age.<br><br><b>Information Sheets and consent forms</b><br><u>A new information sheet and consent form have been drafted to be appropriate for this participant group (attached).</u><br><br><br><br><b>Other</b><br>We would like to change the title of the project from 'Communicative and Non-Communicative Intention in Children with Profound and Multiple Learning Difficulties' to 'Assessing Communication in Children with |   |



|   |  |
|---|--|
|   | Profound and Multiple Learning Disabilities'. Whilst the nature and purpose of the project have not changed we feel that this broader and simpler title will be more accessible to parents and staff reading information about the project.  |
| 6 | <b>Ethical Considerations</b> (insert details of any ethical issues raised by the proposed amendment/s)<br>The proposed amendment to recruitment does not raise any major ethical concerns. All families will receive full information about the project procedures (see information sheets etc. attached) and will have opportunities to discuss the project with research staff and to withdraw their consent. It is expected that infants will find activities enjoyable but, in the event that they show signs of distress, activities would be suspended. A familiar adult would accompany the infant at all times to ensure that all signs of distress are recognised. |
| 7 | <b>Other Information</b> (provide any other information which you believe should be taken into account during ethical review of the proposed changes)  |

**Declaration** (to be signed by the Principal Researcher)

- I confirm that the information in this form is accurate to the best of my knowledge and I take full responsibility for it.
- I consider that it would be reasonable for the proposed amendments to be implemented.
- For student projects, I confirm that my supervisor has approved my proposed modifications.

Signature:

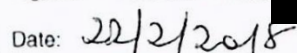


Date: 31<sup>st</sup> January 2018

**FOR OFFICE USE ONLY:**

Amendments to the proposed protocol have been *approved* by the Research Ethics Committee.

Signature of the REC Chair:



Date: 22/2/2018

*(Prof. M. Heinrich)*

## Section 3: Sample information letters and consent form

### 3.1. Sample information letter for parents of children with PIMD

UCL  
LANGUAGE AND COGNITION DEPARTMENT



#### INFORMATION SHEET for PARENTS/CARERS

## Assessing Communication in Children with Profound and Multiple Learning Difficulties

This project has been approved by the UCL Research Ethics Committee, Project ID  
Number: 7565/001

5<sup>th</sup> January 2017

Dear \_\_\_\_\_

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

This study is investigating looking skills in people with profound and multiple learning disabilities (PMLD). Skills such as looking at people and objects and being able to look between them (sometimes known as 'joint attention' are considered to be essential for the development of communication. For children with physical disabilities, the ability to use such looking skills is especially important as they may not be able to use other strategies such as reaching or pointing to communicate.

Existing research suggests that the ability to engage in joint attention by looking between people and objects may be particularly difficult for people with PMLD (Arens, Cress and Marvin, 2007; Neerincx and Maes, 2015). However, further work is needed to confirm this and to look at patterns of looking behaviour in more detail.

In this study we will be looking at whether children with PMLD have better looking skills, including the ability to look from one thing to another, in a distraction-free environment than in a natural setting. We will also be looking at the link between their looking skills and their overall communication skills.

We hope that gaining a better understanding of shared attention in children with PMLD and the effect this has on their wider communication skills will improve the type of support that can be offered to them.

### **WHY HAS MY CHILD BEEN INVITED TO TAKE PART?**

We are writing to you because your child attends XXXX School, which has agreed to participate in this research.

Please note: This letter has been passed to you by your child's school. No one outside your school has had access to your personal details in preparing this invitation.

### **WHAT WOULD PARTICIPATION INVOLVE?**

If you are willing for your child to take part in the study, a team of researchers will visit your child in school on four separate occasions.

The visits would involve the following:

#### **Visit 1**

- An assessment of your child's visual and communication skills using the Mullens Scales of Early Learning (Pearson, 1995). This will take around 15-20 minutes and involve showing your child some pictures, patterns or real items and noting how they respond.
- An assessment of your child's vision carried out by a qualified optometrist and ophthalmologist. This may include an examination of your child's eye-movements, visual acuity and a refraction test (testing for short or long sightedness and astigmatism). This assessment will take no longer than 10 minutes.

#### **Visits 2, 3 and 4**

On each of these visits your child will attend two sessions which will take place in a quiet room in school. The sessions will be separated by at least half an hour during which your child can return to their normal day.

During all sessions your child will be accompanied by a member of school staff who is familiar to them to ensure that they seem happy and comfortable. If your child seems unhappy or tired the session will be stopped and may be resumed later. Sessions will be carried out by members of the research team who are either speech and language therapists or speech and language therapy students (under supervision). All members of the team have experience in working with children with PMLD/complex needs. If you would like to observe the sessions in school please do let us know.

#### **Session 1 (lasting 20 minutes or less)**

In this session your child will see real objects (e.g. balloons) and/or real faces appear in front of a black background. We are interested to see if your child looks at these items and whether they can shift their gaze to look between two items.

**Session 2 (lasting 35 minutes or less)**

This will be a play-based session carried out by one of the research team. Your child will be shown some interesting items and toys with the option to watch and touch them if they are able to. We are interested in how your children looks at items and people and whether they seem to request more of something if it stops. A member of school staff will also be asked to show the children some toys and play with them.

**Communication Assessment**

As well as seeing your child during our visits, we would also like to ask a member of your child's school staff to complete an online assessment of communication skills called the 'Communication Matrix' (see [www.communicationmatrix.org](http://www.communicationmatrix.org) ).

**WILL YOU VIDEO RECORD MY CHILD?**

Yes, with your permission we would like to video record some of our 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:

**A. research** participation

and

**B. wider** participation

**A. Research participation** level of consent means that the video recordings will be used for the research study only. All video recording will be stored securely using a unique code to label the video file (e.g. P123) and will be destroyed when the project has been completed.

**B. Wider participation** level of consent means that video recordings might be used for teaching within UCL (e.g. undergraduate and postgraduate students, and health and education professionals), and at presentations outside University College London, such as international meetings.

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 Language and Cognition Department, University College London (020 7679 4253).

**DOES MY CHILD HAVE TO TAKE PART?**

It is up to you to decide. If, after reading this information sheet, you decide that your child can take part in the study, we ask that you sign and return the enclosed consent form in the envelope provided. Please do not hesitate to contact us to ask any questions you may have.

**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?**

Following the study we will be give you feedback about how your child got on. The results will contribute towards PhD research being carried out by Speech and Language Therapist Lucy Van Walwyk and to the dissertations of the two student Speech and Language Therapists. This research may also be publicised through journal articles and/or 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 Language and Cognition Department, University College London.

**WHO HAS REVIEWED THE STUDY?**

This research study has been looked at and given a favourable opinion by an independent group of people in the UCL Research Ethics Committee to protect you and your child's 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](mailto:m.clarke@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 or your child have been approached or treated by members of staff or about any side effects (adverse events) you or your child may have experienced due to your / their participation in the research, the normal University College London complaints mechanisms are available to you. Please ask members of the research team if you would like more information on this.

**WHAT NEXT?**

If you are interested in taking part with your child after reading about the study, please return the signed consent form in the envelope provided. We will then arrange a convenient time to visit your child in school.



### 3.2. Sample Information Letter for Parents of Typically Developing Infants

UCL  
LANGUAGE AND COGNITION DEPARTMENT



#### INFORMATION SHEET for PARENTS/CARERS

**Project Title: Assessing Communication in Children with  
Profound and Multiple Learning Disabilities: How typically  
developing children react to assessment activities.**

**31<sup>st</sup> January 2018**

**Researchers:** Lucy Van Walwyk, Georgia Cousins

**Principal Supervisor:** Dr Michael Clarke

Language and Cognition Department,  
Chandler House  
2 Wakefield Street,  
London WC1N 1PF  
Tel: +44 (0) 20 7679 4253  
Email: [m.clarke@ucl.ac.uk](mailto:m.clarke@ucl.ac.uk)

Dear Parents and Carers,

Your child is being invited to take part in a research study. Before you decide whether you agree to them taking part it is important for you to understand why the research is being done and what participation will involve. Please ask us if there is anything that is not clear or if you would like more information.

This project has been approved by the UCL Research Ethics Committee, Project ID Number: 7565/001. It has been organised by the Department of Language and Cognition at UCL.

#### **What is our study about?**

Our study is looking at ways in which children with profound learning disabilities communicate and how we might assess these children's communication skills. One of the areas we have been investigating is how children with profound learning disabilities use

their eyes to look at objects and people and to communicate. This is an especially important skill for this group of children as they have limited use of their limbs and do not use speech. Understanding how their looking skills are affected can help us make sure that we are supporting and teaching them in the right way.

We have developed a simple set of play-based activities to investigate the looking skills of the disabled children taking part in our project. However, to make sure that these activities are effective at measuring looking and communication skills we would also like to see how typically developing children (children without disabilities) react to them.

#### **Why has my child been invited to take part?**

We will be working with typically developing children, aged between 6 and 18 months. Your child has been asked to take part because they are the right age and their nursery has agreed to support us in this research. Since the study focuses on looking skills we will not be including any children known to have vision problems.

#### **What would participation involve?**

If your child takes part in the study, he or she will attend a single research session in a quiet space at their nursery. A familiar adult will remain with them throughout the session. Activities are designed to be interesting and fun and the whole session will last no longer than 25 minutes.

Firstly, your child will be seated in a high chair or on the knee of the familiar adult depending what is most comfortable for them. There will be a black screen on a table in front of them. Faces or balloons will then pop up in turn above the screen and we will be observing whether your child looks at them.

Secondly, a member of the research team will carry out some play-based activities which include showing your child some interesting toys and talking and singing to them. During these activities we will be observing where your child is looking.

We anticipate that the session will be enjoyable for your child. However, if he or she shows any signs of distress or concern the activities will be stopped.

#### **Who will run the research session?**

The research team consists of Lucy Van Walwyk, a PhD researcher and experienced speech and language therapist and Georgia Cousins, a student speech and language therapist. Both have satisfied Disclosure and Barring Service (DBS) checks for working with children.

#### **Will you video record my child?**

Yes, with your permission we would like to video record your child while he or she is taking part in the research session. This is so that we can analyse information about where your child was looking during activities - information which is hard to capture without video. Recordings will be used for the purposes of the research study only and will not be shown to anybody outside the research team.

All video recordings will be stored securely using a unique code to identify them and will be destroyed once the project has been completed.

#### **Does my child have to take part?**

It is up to you whether or not your child takes part. If you are happy for them to do so you will be asked to sign a consent form and you may withdraw them from the study at any time without giving a reason.

**What are the benefits of my child taking part in the study?**

Taking part in the study will not have direct benefits for your child although we would anticipate that they will find the session interesting and engaging. We hope that the work done as part of the study will have a positive impact on teaching and therapy support for children with profound and multiple learning disabilities.

**Confidentiality**

All information from the study will be kept strictly confidential. All children are free to withdraw from the study at any time. All data will be collected and stored in accordance with the Data Protection Act, 1998.

**What will happen to the results of this study?**

The results of this study will be used to inform a Masters research project for Georgia Cousins and will contribute towards PhD research being carried out by Lucy Van Walwyk. This research may also be publicised through written academic articles and conference presentations in the UK and abroad. Your child will not be identified in any publication or presentation.

**What if I have questions about the study?**

Please do not hesitate to contact Dr Michael Clarke if there is anything that is not clear, or if you would like more information (see contact details at the top of this sheet).

**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 or your child have been approached or treated by members of the research team please contact Dr Michael Clarke or, if you feel that a complaint has not been adequately handled, the Chair of the UCL Research Ethics Committee – [ethics@ucl.ac.uk](mailto:ethics@ucl.ac.uk).

**Data Protection Privacy Notice**

The data controller for this project will be University College London (UCL). The UCL Data Protection Office provides oversight of UCL activities involving the processing of personal data and can be contacted at [data-protection@ucl.ac.uk](mailto:data-protection@ucl.ac.uk). UCL's Data Protection Officer is Lee Shailer and he can also be contacted at [data-protection@ucl.ac.uk](mailto:data-protection@ucl.ac.uk).

Your personal data will be processed for the purposes outlined in this notice. The legal basis that would be used to process your personal data will be the provision of your consent. You can provide your consent for the use of your personal data in this project by completing the consent form that has been provided to you.

Your personal data will be processed so long as it is required for the research project and no later than March 2019. If we are able to anonymise or pseudonymise the personal data you provide we will undertake this and will endeavour to minimise the processing of personal data wherever possible.

If you are concerned about how your personal data is being processed, please contact UCL in the first instance at [data-protection@ucl.ac.uk](mailto:data-protection@ucl.ac.uk). If you remain unsatisfied, you may wish to contact the Information Commissioner's Office (ICO). Contact details, and details of data subject rights, are available on the ICO website at: <https://ico.org.uk/for-organisations/data-protection-reform/overview-of-the-gdpr/individuals-rights/>

### 3.3. Sample Consent Form

UCL  
LANGUAGE AND COGNITION DEPARTMENT



#### Informed Consent Form for Parents/Carers

Project Title: Assessing Communication in Children with Profound and Multiple Learning Disabilities

|   |  | Tick Box |
|---|--|----------|
| 1 | I confirm that I have read and understood the Information Sheet for the above study. I have had an opportunity to consider the information and to ask questions which have been answered to my satisfaction.   |          |
| 2 | I voluntarily agree that my child can take part in the study described on the Information Sheet.   |          |
| 3 | I understand that my child's participation is voluntary and that I am free to withdraw them at any time without giving a reason. I understand that if I decide to withdraw, any personal data I have provided up to that point will be deleted unless I agree otherwise.   |          |
| 4 | I consent to the processing of my personal information for the purposes explained to me (i.e the research team will see my name and my child's name on this consent form, also video recording will be used to collect information for the project). I understand that such information will be handled in accordance with all applicable data protection legislation. |          |

|   |   |  |
|---|---|--|
| 5 | I understand that all personal information will remain confidential and that all efforts will be made to ensure that my child cannot be identified.                 |  |
| 6 | I understand that my child's data gathered in this study will be stored anonymously and securely. It will not be possible to identify my child in any publications. |  |
| 7 | I consent to my child being video recorded and understand that the recordings will be destroyed no later than twelve months after the data has been collected.      |  |
| 8 | I am aware of who I should contact if I wish to lodge a complaint.  |  |

**Principal Researcher:** Dr Michael Clarke

**Contact details:** Telephone: 020 7679 4253/ Email: m.clarke@ucl.c.uk

This study has been approved by the UCL Research Ethics Committee (Project ID Number 7565/001):

Thank you for your interest in allowing your child to take part in this research. It is important that you have read the Information Sheet and understand the project before you agree to them taking part. If you have any questions, please ask the researcher before you decide whether to let your child join in. You will be given a copy of this Consent Form to keep and refer to at any time.

I understand that by ticking each box below I am consenting to this part of the study. I understand that it will be assumed that unticked boxes means that I DO NOT consent to that part of the study. I understand that by not giving consent for any one part might mean that my child might not be eligible for the study.

**Child's name:** \_\_\_\_\_

**Your name:** \_\_\_\_\_  
**child:** \_\_\_\_\_

**Your relationship to the**

**Your signature:** \_\_\_\_\_

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