

Early Intervention for Infants with Down Syndrome

Thesis submitted for the degree of Doctor of Philosophy

School of Psychology and Clinical Language Sciences

Emily Seager

December 2017

Declaration

I confirm that this is my own work and the use of all material from other sources had been properly and fully acknowledged.

Emily Seager

Contents

Acknowledgements.....	14
Abstract.....	15
Chapter 1: Introduction and Literature Review	16
1.1. Overview	16
1.2. Hearing Deficits	17
1.3. Fine and Gross Motor Skills.....	18
1.4. Co-occurrence of Additional Developmental Disorders	19
1.5. Cognitive Ability and Language Development.....	20
1.6. Language Profile	21
1.6.1. Early Language Development – First Word and Multi-word Utterances.....	21
1.6.2. Expressive and Receptive Language	23
1.6.3. Language at School Age and Beyond.....	26
1.6.4. Precursors to Language.....	28
1.7. Summary	43
Chapter 2 – Early Precursors to Language	45
2.1. Rationale for Study.....	45
2.2. Research Questions	46
2.3. Hypotheses	47
2.4. Method	47
2.4.1. Ethics, Recruitment and Consent.....	47
2.4.2. Participants.....	47
2.5. Measures.....	50
2.5.1. Maternal Measures.....	50
2.5.2. Child Measures	52
2.6. Procedure.....	54
2.7. Results	55
2.7.1. Results Strategy	55
2.7.2. Between Group Comparisons	55
2.7.3. Correlation Analysis	57
2.7.4. Regression Analysis - Predictors of Total Language Scores	59
2.8. Discussion	62
2.8.1. Summary of Results.....	62

2.8.2.	Differences Between Groups	63
2.8.3.	Joint Attention and Concurrent Language Scores	65
2.8.4.	Maternal Measures and Concurrent Language Status	66
Part 2-	Literature Review and Intervention Study	68
Chapter 3 -	Intervention Studies	68
3.1.	Case Studies/Case Series: Multiple Baseline Design.....	69
3.2.	Summary	72
3.3.	Intervention Studies Using a Control Comparison Group	72
3.3.1.	Early Stimulation Programme.....	72
3.3.2.	Published Scheme/Clinical Methods	74
3.3.3.	Responsive Teaching/Communication Training.....	76
3.4.	Summary	79
3.5.	Training Joint Attention	80
3.5.1.	Improving Joint Attention – Multiple Baseline Design.....	80
3.5.2.	Improving Joint Attention – Control Group Design	83
3.5.3.	Parent Administered Intervention	85
3.6.	Summary	85
3.7.	The Current Study (Intervention Study) – Rationale	86
3.7.1.	Aims.....	87
3.7.2.	Research Questions	87
3.7.3.	Outline.....	88
Chapter 4 -	Methodology	88
4.1.	Ethics, Recruitment and Consent	88
4.2.	Participants	89
4.2.1.	Demographics	90
4.3.	Pilot Test	92
4.4.	Pre-Test	93
4.5.	Measures.....	93
4.5.1.	Language and Vocabulary	94
4.5.2.	Non-Verbal Mental Age	95
4.5.3.	Joint Attention: Early Social Communication Scales	95
4.5.4.	Parental Questionnaires	101
4.5.5.	Parent-Child Interaction.....	102

4.6.	Intervention	103
4.6.1.	Intervention Levels	104
4.6.2.	Parent Sessions.....	107
4.6.3.	Treatment Fidelity.....	107
4.7.	Post-Test.....	109
4.8.	Six Month Follow Up.....	109
4.9.	One Year Follow Up	109
Chapter 5 – Results Time Point One/Pre-test Assessment Comparison of Intervention Group and Control Group		110
5.1.	Pre-Test Assessment Comparison of the Intervention Group and the Control Group 110	
5.2.	The Hospital and Anxiety Depression Scale.....	115
5.3.	M-CHAT	116
Chapter 6 - Pre-Post-Test Results		117
6.1.	Responding to Joint Attention.....	117
6.2.	Receptive and Expressive Language (PLS-4).....	120
6.3.	Vocabulary and Use of Signs	122
6.4.	Factors Associated with Post-Test Responding to Joint Attention Scores.....	124
6.4.1.	Gaze Following Post-Test.....	125
6.4.2.	Proximal Point Post-Test	126
6.4.3.	Total Responding to Joint Attention Post-Test.....	127
6.5.	Parental Questionnaires.....	128
6.6.	Parent diary	129
6.7.	Summary	129
Chapter 7 - Results – Follow Up One.....		130
7.1.	Responding to Joint Attention (Total).....	131
7.1.1.	Assumptions.....	131
7.2.	Receptive Language	133
7.3.	Expressive Language.....	134
7.4.	Total Language Scores	135
7.5.	Receptive Vocabulary	136
7.6.	Expressive Vocabulary.....	137
7.7.	Use of Sign	138
7.8.	Intervention Effects	139

7.9. Summary	140
7.9.1. Intervention Results	140
Chapter 8 - Results – Final Follow Up	141
8.1. Receptive Language	143
8.2. Expressive Language.....	144
8.3. Total Language.....	145
8.4. Receptive Vocabulary	146
8.5. Expressive Vocabulary.....	147
8.6. Use of Signs	148
8.7. Summary	149
Chapter 9- Discussion.....	150
9.1. Gains in Responding to Joint Attention	150
9.2. Language and Vocabulary Outcomes 6 months Post Intervention Onset.....	153
9.3. Language and Vocabulary Outcomes 12 months Post Intervention Onset.....	154
Part 3 – Early Predictors of Language Development.....	155
Chapter 10 – Concurrent and Longitudinal Predictors of Language	155
10.1. Baseline Predictors of Time Point 2 Language	156
10.2. Regression Analyses.....	158
10.2.1. Predictor Variables of Receptive, Expressive and Total Language	159
10.2.2. Predictor Variables of Receptive Vocabulary and Use of Signs.....	162
10.3. Summary – Time Point 1 Predictors of Language Scores at Time Point 2	163
10.4. Time Point 2: Concurrent Predictors of Language/Vocabulary	164
10.4.1. Regression Analysis: Predictors of Language Scores	165
10.4.2. Regression Analysis: Predictors of Receptive and Expressive Vocabulary and Use of Signs	168
10.5. Summary - Concurrent Predictors of Language at Time Point 2	171
10.6. Baseline Predictors of Time Point 3 Language Scores.....	172
10.6.1. Regression Analysis – Predictors of Expressive and Receptive Language .	174
10.6.2. Regression Analysis – Predictors of Receptive and Expressive Vocabulary and Use of Signs	177
10.7. Summary – Time Point 1 Predictors of Language Outcomes at Time Point 3 ...	179
10.8. Time Point 2 Predictors of Time Point 3 Outcomes.....	180
10.8.1. Regression – Predictors of Receptive and Expressive Language Scores.....	182
10.8.2. Regression – Predictors of Receptive Vocabulary and Use of Signs.....	184

10.9.	Summary – Time Point 2 Predictors of Language/Vocabulary Outcomes	186
10.10.	Discussion - Concurrent and longitudinal predictors of language and vocabulary	187
10.10.1.	Factors Associated with Language/Vocabulary Development at 23-30 months	187
10.10.2.	Factors Associated with Vocabulary/Language Development at 30-35 Months	193
Chapter 11 – Discussion		195
11.1.	Theoretical implications	195
11.2.	Practical/clinical implications.....	198
11.3.	Limitations and ideas for future research	199
11.4.	Conclusion	202
References.....		204
Appendix A: Information letters and consent forms.....		235
A.2 Information sheet for intervention study.....		236
A.3 Consent form.....		239
Appendix B: Maternal interaction style coding scheme		240
Appendix C: Demographic questionnaire.....		248
Appendix E: Parent materials for intervention		254
E.1 Intervention manual and weekly instructions for parents.....		254
E.2 Intervention diary for parents		263
Appendix F: Parent questionnaire – satisfaction of intervention.....		264
Appendix G: Normality tests		266
Appendix H: Full Correlation Matrices		270

List of Figures

Figure 2.1: Scatterplot of positive expressed emotion and total language scores showing the regression line for group.....	61
Figure 2.2: Scatterplot of responding to joint attention and total language scores showing the regression line for group.....	62
Figure 4.1: Diagram to show which measures are used at which time point.....	94
Figure 4.2: Diagram of room layout for ESCS.....	96
Figure 4.3: Stages of study for the intervention group and the control group.....	110
Figure 5.1: Maternal Scores for Anxiety and Depression from the HADS.....	115
Figure 5.2: Participant Scores for ASC Risk (items removed).....	116
Figure 6.1: Box plot showing mean % scores for the proximal point task pre and post intervention.....	118
Figure 6.2: Box plot showing mean scores for the gaze following task pre and post intervention.....	119
Figure 6.3: Box plot showing mean scores for total responding to joint attention pre and post intervention.....	120
Figure 6.4: Box plot showing mean scores for receptive, expressive and total language scores pre and post intervention.....	122
Figure 6.5: Boxplot showing mean numbers of words and signs understood and produced pre and post intervention.....	124
Figure 6.6: Parental satisfaction with intervention.....	129
Figure 7.1: Responding to joint attention scores at pre-test and at first follow up for the intervention and control group.....	132

Figure 7.2: Receptive language scores pre-test and at first follow up for the intervention and control group.....	133
Figure 7.3: Expressive language scores pre-test and at first follow up for the intervention and the control group.....	134
Figure 7.4: Total language scores at pre-test and first follow up for the intervention and control group.....	135
Figure 7.5: Receptive vocabulary scores pre-test and at first follow up for the intervention and control group.....	136
Figure 7.6: Expressive vocabulary scores at pre-test and at first follow up for the intervention and control group.....	138
Figure 7.7: Use of signs pre-test to first follow up comparing the intervention and control group.....	139
Figure 7.8: Total responding to joint attention scores at pre-test, post-test and first follow up for the intervention group.....	140
Figure 8.1: Receptive language scores on PLS-4 at baseline assessment and final follow up comparing the intervention and control group.....	144
Figure 8.2: Expressive language scores on PLS-4 at baseline assessment and at final follow up comparing the intervention and control group.....	145
Figure 8.3: Total language scores on PLS-4 at baseline assessment and at final follow up comparing the intervention and control group.....	146
Figure 8.4: Receptive vocabulary scores on CDI baseline to final follow up comparing the intervention and control group.....	147
Figure 8.5: Bar chart showing expressive vocabulary from baseline to final follow up comparing the intervention and control group.....	148
Figure 8.6: Number of signs at baseline and at final follow up comparing the intervention and the control group.....	149

List of Tables

Table 2.1: Demographic variables of parents of children of DS and parents of TD children.....	49
Table 2.2: Descriptive statistics for language, joint attention and maternal interactive style in DS and TD (raw scores).....	56
Table 2.3: Correlations between language, nonverbal mental age, joint attention and maternal interactive style for the DS and TD group (Spearman's rho).....	58
Table 2.4: Results from hierarchical regression analysis assessing association between joint attention, positive expressed emotion and total language scores.....	60
Table 4.1: Demographic variables for intervention and control group.....	92
Table 5.1: Means and standard deviations for the intervention group and control group for age and non-verbal measures.....	111
Table 5.2: Descriptive statistics for PLS-4 for the intervention and the control group.....	112
Table 5.3: Means and standard deviations for receptive/expressive language and vocabulary scores for the intervention and control group.....	112
Table 5.4: Means and standard deviations for Early Social Communication Scale scores for the intervention group and the control group.....	114
Table 5.5: Means and standard deviations for maternal interactive style for the intervention group and the control group.....	115
Table 6.1: Regression analysis for post-test gaze following scores.....	127
Table 6.2: Regression analysis for proximal point post-test scores.....	128
Table 6.3: Regression analysis showing significant predictors of post-test total responding to joint attention scores.....	129
Table 8.1: Descriptive statistics for age, non-verbal mental age, language and vocabulary at the 2 nd follow up.....	143

Table 10.1: Spearman’s rho correlations between baseline language scores and time point 2 language scores.....	158
Table 10.2: Significant correlations between language/vocabulary scores at time point 2 and baseline predictors.....	159
Table 10.3: Regression analysis showing time point 1 predictors of receptive language.....	160
Table 10.4: Regression analysis showing significant predictors for expressive language.....	161
Table 10.5: Regression analysis showing significant predictors for total language scores.....	162
Table 10.6: Regression analysis showing significant predictors of receptive vocabulary.....	163
Table 10.7: Regression model showing significant predictors of number of signs used and understood.....	164
Table 10.8: Significant correlations between language/vocabulary scores and concurrent predictor variables.....	166
Table 10.9: Regression model showing significant predictors of receptive language.....	167
Table 10.10: Regression model showing concurrent predictors of expressive language.....	168
Table 10.11: Regression model showing significant predictors of total language scores.....	169
Table 10.12: Regression model showing significant predictors of receptive vocabulary.....	170
Table 10.13: Regression model showing significant predictors of expressive vocabulary.....	171
Table 10.14: Regression model showing predictors of use of sign.....	172

Table 10.15: correlation analysis between baseline language scores and language scores at time point 3.....	173
Table 10.16: Significant correlations between language/vocabulary scores and baseline predictor variables.....	174
Table 10.17: Regression analysis showing significant predictors of receptive language.....	175
Table 10.18: Regression analysis showing significant predictors of expressive language.....	176
Table 10.19: Regression analysis showing significant predictors of total language.....	177
Table 10.20: Regression analysis showing significant predictors of receptive vocabulary.....	178
Table 10.21: Regression analysis showing significant predictors of expressive vocabulary.....	179
Table 10.22: Regression analysis showing significant predictors of number of signs used and understood.....	180
Table 10.23: Spearman's rho correlations between language/vocabulary scores at time points 2 and 3.....	181
Table 10.24: Significant correlations between language/vocabulary scores and time point 2 predictor variables.....	182
Table 10.25: Regression model showing significant time point 2 predictors of receptive language outcomes.....	183
Table 10.26: Regression model showing significant time point 2 predictors of expressive language outcomes.....	184
Table 10.27: Regression model showing significant time point 2 predictors of total language outcomes.....	185

Table 10.28: Regression model showing significant time point 2 predictors of receptive vocabulary outcomes.....	186
Table 10.29: Regression model showing significant time point 2 predictors of use of sign.....	187

Acknowledgements

Thank you to my supervisor Vesna Stojanovik for your constant support throughout the past 3 years. I feel very fortunate to have had a supervisor who is so supportive and it has made the process very enjoyable. Thank you also to Courtenay Norbury for all your valuable knowledge and feedback throughout the many drafts! Finally, thank you to all the children and parents who took part and were extremely dedicated to completing the intervention – without you this wouldn't have been possible.

Abstract

Most individuals with Down syndrome (DS) have some sort of speech and language deficit and this is prominent from an early age. Previous research has identified that various early precursors may be important for language development for typically developing (TD) children including: joint attention and maternal interactive style. However, there is limited research in this area for children with DS. Part 1 of the thesis includes a study looking at whether joint attention and maternal interactive style are important for concurrent language outcomes for a group of children with DS aged 17-23 months and a TD group with comparable non-verbal mental age. The results found that responding to joint attention was a significant predictor of concurrent language scores for infants with DS and maternal positive expressed emotion was a significant predictor for TD children. Part 2 of the thesis was a longitudinal intervention study focusing on improving responding to joint attention for infants with DS at 17-23 months with the view to improving speech and language outcomes at 30-35 months. The results of the intervention found responding to joint attention could be improved through an early intervention and at 30-35 months there was a significant difference for receptive vocabulary with the intervention group being reported to understand more words than the control group. Finally, part 3 investigated which early precursors were associated with concurrent and longitudinal language and vocabulary outcomes at 24-30 months and 30-35 months for children with DS. Non-verbal mental age emerged as an important predictor. The results are discussed in line with previous research. The theoretical and practical implications are discussed as well as ideas for future research.

Chapter 1: Introduction and Literature Review

1.1. Overview

The PhD is split into three parts. The first part focuses on the language profile of individuals with Down syndrome and investigates which early precursor skills are associated with concurrent expressive and receptive language scores as measured by standardised tests for children with Down syndrome compared to typically developing children. The second part is a longitudinal early intervention study which aims to improve longitudinal language and vocabulary outcomes. Finally, part three focuses on concurrent and longitudinal predictors of language development for children with Down syndrome.

Down syndrome (DS) is a genetic disorder that is caused by an additional copy of chromosome 21 and is a genetic abnormality which results in a distinct facial expression, heart and respiratory problems and a learning disability (Chapman & Hesketh, 2000). DS is estimated to affect approximately 1 in 700 births (Martin, Klusek, Estigarribia & Roberts, 2009). It can be diagnosed by prenatal screening using amniocentesis and karyotyping of the foetal cell, or at birth using diagnostic tests (Fiddler, 2005). It is caused by cell errors of which three types have been identified: trisomy 21 (the most common), translocation and mosaics (Mutton, Alberman & Hook, 1996). DS makes up the largest identified population of those with a learning disability (O'Toole & Chiat, 2006) and is the most common non-inherited cause (Rice, Warren & Betz, 2005). The average IQ of an individual with DS is 50 and the range is approximately 30 to 70 (Chapman & Hesketh, 2000). Deficits in communication, language and memory have been identified (Laws, Byrne & Buckley, 2000). However, an individual with DS may only experience a few of the deficits outlined as there is a high level of variability within the condition (Davis, 2008).

Individuals with DS often suffer from various health issues, such as: congenital heart disease, middle ear disease and endocrine system abnormalities (Pueschel & Pueschel, 1992). It is also associated with impairments in physical features, including: dysmorphic facial features, limited growth, hypotonia, epicanthal folds and broad hands (Robert, Price & Malkin, 2007). The life expectancy of individuals with DS has dramatically increased from just 10 years in the 1960s to around 47 years in 2007 (Presson et al., 2013). It is believed that this change in life expectancy is due to better medical assistance and that it is more common for individuals with DS to be integrated in the community instead of living in institutions (Bittles & Glasson, 2004).

1.2. Hearing Deficits

It is estimated that between 40-80% of individuals with DS have some form of hearing impairment. Young children with DS commonly suffer with otitis media with effusion (Laws & Hall, 2014). Research investigating early hearing loss for infants with DS found 26% of 332 children failed their new-born screening tests (Park, Wilson, Stevens, Harward & Hohler, 2012). Over 43% of those who passed the test went on to develop conductive hearing loss. Chapman, Seung, Schwartz and Bird (2000) reported that hearing loss in DS can be a long existing problem as 60% of adolescents and young adults with DS have been found to have mild to moderate hearing loss in one or both ears.

Various studies have linked the hearing and motor deficits in DS to their delayed language development. For example, Rice et al. (2005) suggested that speech difficulties may occur because of periodic hearing loss due to otitis media which commonly occurs for individuals with DS. Laws and Hall (2014) investigated retrospective data to consider a link between history of hearing loss between 2-4 years and current speech and language abilities. The speech and language of 51 children with DS aged 3-11 years old were assessed and prior hearing loss was recorded through parental questionnaire and audiology reports. The children

were then divided into two groups based on whether they had experienced severe hearing loss and received treatment versus those who had not received treatment and had milder issues. A statistically significant difference was found between the two groups for language comprehension, expressive language, receptive vocabulary and speech accuracy control with those who had experienced mild hearing issues having better outcomes controlling for chronological age and non-verbal mental age.

However, hearing loss alone cannot be the sole reason for language difficulties as some individuals with DS without hearing problems also experience language impairments (Laws & Bishop, 2004). Furthermore, research has found that the severity of hearing loss does not correlate reliably with the level of language difficulties (Chapman, Seung, Schwartz & Bird, 1998).

1.3. *Fine and Gross Motor Skills*

Difficulties in fine motor skills have been identified for children with DS, including deficits in hand-eye coordination (Frith & Frith, 1974). Fidler, Hepburn, Mankin and Rogers (2005) assessed 16 children with DS ($M= 33$ months) and compared them to a group of developmentally delayed children (matched for chronological age and mental age) and a TD group (matched for mental age). Results showed that the DS group obtained significantly worse overall scores on the motor subsection from the Vineland Adaptive Behaviour Scales (Sparrow, Balla & Cicchetti, 2005) than the developmentally delayed group but not the TD group. Children with DS obtained lower age equivalent scores for both fine and gross motor in comparison to the developmentally delayed group. The authors reported that the DS group demonstrated particular difficulties on tasks that involved fine motor skills such as reaching or grasping items and stringing beads.

Gross motor skills can also be delayed. Palisano et al. (2001) created gross motor curves to identify when motor functions may be achieved for children with DS. One-hundred and twenty-one children with DS aged 1 month to 6 years were assessed. The results suggested that there is a 51% chance that children with DS will roll over by 6 months, a 78% chance they will sit by 12 months and a 34% chance they will be crawling by 18 months. By 24 months, there is a 40% chance that they will be walking, and a 45-52% chance that they will be able to walk upstairs and jump by 5 years. This demonstrates that not all children achieved all of the milestones by 6 years of age. Furthermore, the authors report that development tends to be faster at a younger age and suggest that this may be to do with the balance needed to be able to stand alone and walk. Muscle hypotonicity and hypermobility of joints is not uncommon in individuals with DS, which may affect their gross motor development (Hayes & Batshaw, 1993). In summary, the research suggests that children with DS may have difficulties with both fine and gross motor skills, particularly in the early years.

1.4. Co-occurrence of Additional Developmental Disorders

Individuals with DS may show similar profiles to, or have a co-occurring disorder, for example Developmental Language Disorder (DLD) or Autism Spectrum Conditions (ASC). Some individuals with DS share a similar language profile to those with DLD, such as: problems using morphemes and marked syntactic weaknesses in comparison to other linguistic domains, such as semantics and pragmatics (Chapman et al., 1998). Laws and Bishop (2004) suggest that the language profiles of individuals with DS are similar to those of children with DLD; with the main deficits being in phonology and grammar (the language profile of individuals with DS will be discussed in further detail in the next section 1.6). Some have suggested that a small group of individuals with DS exhibit behavioural profiles which are consistent with those displayed by individuals with ASC (Pary & Hurley, 2002). It has been estimated that the prevalence of those with DS who show social interaction deficits

that would meet the criteria for ASC ranges from 5 to 9% (Ghaziuddin, Tsai & Ghaziuddin, 1992). However, it has been suggested that such co-morbidity is considered uncommon and that health care professionals are sometimes reluctant to recognise/diagnose ASC in those with DS (Carpone, Grados, Kaufmann, Bernad-Ripoll & Jewell, 2005).

1.5. Cognitive Ability and Language Development

There has been a long standing theoretical debate regarding the acquisition of language, the influences of genetics and the environment and whether language develops alongside general cognitive ability or whether it is not related to general cognitive abilities. The nativist theory originated from Chomsky and argues that the child is born with an innate ability to develop language (Braine, 1994; Chomsky, 1976). Only minimal input from the environment is therefore required. Further, the nativist theory suggests that language is a domain specific module and therefore the language system is independent from other cognitive processes (Chomsky, 1976; Fodor, 1983). Therefore, if an individual has a language delay/disorder, the nativist theory would argue that this is due to an impaired language module (Pinker, 1994). Evidence for the nativist theory comes from individuals with acquired brain injuries who show a dissociation of impairments (Karmiloff-Smith, 2009), or from genetic disorders such as DS and Williams syndrome where patterns of strengths and weaknesses are evident (e.g. Jarrold, Baddeley & Hewes, 1999). However, this approach would suggest that if a module such as language was 'impaired', then this is genetically pre-determined and could not be rectified by the environment (Karmiloff-Smith, 2009).

Other theories, such as the neuro-constructivist approach, is a developmental approach which suggests that the brain develops over time due to the influences of genes, the brain, cognition and the environment and that all these components interact with each other during development (Karmiloff-Smith, 2009). Therefore, outcomes are not pre-determined

due to modularisation or genetics. If modules are evident, this is thought to be a result of development over time (Karmiloff-Smith, 1992; 1998). This approach would argue that language delays seen in genetic disorders, such as DS, are due to an atypical development trajectory (e.g. Laing et al., 2002; Steele, Scerif, Cornish & Karmiloff-Smith, 2013).

From an intervention perspective – if an individual scored within the typical range in a specific domain, the nativist approach would not target that area in intervention as it would be perceived as being ‘intact’. In contrast, the neuro-constructivist approach considers a more dynamic approach to intervention. Therefore, this theory would consider intervention in an ‘intact’ domain with a view to improving another skill. For example, to target language, low level processes that influence language development may be focused on in intervention, such as early communication skills (Karmiloff-Smith, 2009).

1.6. *Language Profile*

This section will discuss the language profile and language development of individuals with DS, including the relative strengths and weaknesses, as well as pre-linguistic precursors to language acquisition. Research suggests that those with DS show a different language profile in comparison to TD children, and a deficit in language is evident from an early age.

1.6.1. *Early Language Development – First Word and Multi-word Utterances*

Children with DS pass through similar stages of language development as TD children but those with DS progress at a slower pace and spend longer in transitional periods (O’Toole & Chiat, 2006). TD children start to intentionally communicate between 9-12 months and then communicate symbolically between 12-18 months of age. For children with DS however, the transition to intentional communication can occur between 24-36 months

(Rondal, 2003) and by three years some children are still not talking (Abbeduto, Warren & Conners, 2007).

TD children commonly produce their first word around their first birthday (Tomasello, 2003) and begin to combine words around 24 months (Capone, 2010). Between 18-30 months TD children start to use phrases and sentences (Atkinson, Atkinson, Smith, Bem & Hoeksema, 1996). In comparison children with DS produce their first word at around 21 months (Stoel-Gammon, 2001), demonstrating a significant early delay. The initial delay in producing their first word is followed by slow progress in expressive language (Fabbretti, Pizzuto, Vicari & Volterra, 1997). For example, Berglund, Eriksson and Johansson (2001) assessed 330 Swedish children with DS between 1;0-5;6 years and compared them to a TD group aged 1;4-2;4. Participants were assessed on 1-3 occasions over a period of 6 months. The Swedish Early Communicative Development Inventory (SECDI) was used to assess vocabulary as well as grammar and pragmatics. Approximately 10% of the children with DS produced their first word at 12 months and after 2 years 80% had begun to talk. By 5 years between 10-20% of children produced fewer than 10 words and some had not started to speak. By 3 years of age, 25% of the DS sample were able to produce 50 words, by 4 years 50% produced 50 words and finally by 5 years 75% produced 50 words. This still leaves 25% of the sample who at 5 years were not able to produce 50 words. The average vocabulary of the sample at 3 years was 36 words and the majority of the sample were using speech and sign concurrently. The authors concluded that at 36 months those with DS showed the same performance as the TD sample showed at 16 months. This study demonstrates a significant early delay in expressive language.

Further delays are evident as children begin to combine words. Rondal (1998) reported that children with DS do not tend to use multi-word utterances until they are between 4;0-5;0 years old, whereas the majority of TD children reach this stage by 2 years (Lieven, Salomo &

Tomasello, 2009). Oliver and Buckley (1994) tracked the progress of 17 individuals with DS aged 1;0 to 4;0 years ($M= 25.6$ months) from the time they produced their first word to the time they produced a two-word utterance. The results relied on parent kept diary records. All children produced their first word between 19-38 months, and on average at 27.3 months. Five of the twelve children demonstrated a vocabulary spurt of 16-36 words when they reached 28-32 months ($M= 30$ months). Children started to combine two words at an average age of 36.9 months (range 25-52 months) and at this stage their expressive vocabulary ranged from 21-109 words. These results show an 18 month delay relative to the children's chronological age.

1.6.2. *Expressive and Receptive Language*

The majority of individuals with DS will have some form of language deficit which is prominent from an early age and a pattern of strengths and weaknesses is evident. For children with DS language production fails to keep up with the child's mental age but language comprehension keeps up with and sometimes exceeds the levels expected from the child's mental age (Abbeduto, Murphy, Cawthon et al., 2003; Caselli et al., 1998; Fidler & Nadel, 2007; Miller, 1999). The gap between production and comprehension skills has been found to continue to widen throughout childhood and into adolescence (Chapman et al., 1998; 1999). Expressive language is exceptionally delayed in comparison to overall functioning (O'Toole & Chiat, 2006). This may lead to frustration as children may constantly have to repeat themselves due to poor speech intelligibility and may be treated based on their expressive abilities, which may underestimate their academic potential and general understanding of language (Fidler & Nadel, 2007).

With regard to receptive language skills, there is a pattern of relative strengths and weaknesses. Fowler (1990) reported a divide between lexical knowledge (vocabulary comprehension) and grammatical knowledge (morphology and syntax comprehension), with

lexical knowledge being superior to grammatical. By adolescence, comprehension of syntax lags behind non-verbal mental age but comprehension of vocabulary keeps up with or even exceeds non-verbal mental age (Chapman, Schwartz & Kay-Raining Bird, 1991). Other areas of weakness that have been identified are expressive grammar (Fowler, Gelman & Gleitman, 1994), verbal short term memory (Jarrold & Baddley, 1997) and receptive syntax (Abbeduto et al., 2003).

Further evidence has shown that individuals with DS experience worse deficits in expressive language than those with other genetic disorders who have similar levels of non-verbal functioning, such as individuals with Williams syndrome. Levy and Eilam (2013) conducted a naturalistic study comparing the language development of 9 children with DS ($M= 46.8$ months), 9 children with Williams syndrome ($M= 54.7$ months) and a TD group with similar mean length utterance. A mixed longitudinal and cross-sectional design was used and Hebrew was the first language of all the children included. TD children started to combine words at 22.8 months, children with Williams syndrome at 46.8 months and children with DS at 54.7 months. The children with Williams syndrome exhibited an expressive language delay of 24 months whereas the children with DS were delayed by 32 months. The children with DS in this study continued to have the slowest acquisition pace.

In another study, Mervis and Robinson (2000) compared the expressive vocabulary of 28 children with DS and 24 with Williams syndrome ($M= 32$ months) using the Communicative Development Inventory (CDI; Fenson et al., 1993). A significant difference was found between the two groups – individuals with DS had a mean expressive vocabulary of 66.35 words (range 0-324) and those with Williams syndrome had a mean vocabulary of 132.5 words (range 3-391). The same result was found when the authors focused on those aged 24-27 months. Children with Williams syndrome had a significantly higher language age (median 18.5 months, range 10-25 months) in comparison to children with DS (median

15.5 months, range < 8-23 months). It was found that 57% of the children with Williams syndrome had expressive vocabulary sizes below the 5th percentile for their chronological age whereas 92% of children with DS fell below the 5th percentile.

1.6.2.1. Speech sound disorders and expressive language

The majority of individuals have inconsistent production of specific speech sounds and this is prevalent by around 3 years (Kent & Vorperian, 2013). Children with DS commonly have physical features which may have an impact on their expressive language (Kumin, Councill & Goodman, 1994). Examples include: midface hypoplasia (where facial features including the bridge of the nose and upper jaw are not fully developed) as well as lower muscle tone in areas such as the lips and tongue (Kumin, 2006). Physical features such as the ones mentioned above can affect the pronunciation of various speech sounds; in addition, previous research has shown that individuals with DS have poor motor control in speech production (Kumin, 1994). Such anatomical differences and poorer motor control may have a negative effect on the production of speech sounds. For example, an oversized and muscular tongue may have a negative effect on the production of lingual consonants (Stoel Gammon, 2001).

A study by Miller, Miolo, Sedey, Pierce and Rosin (1989) assessed 43 children with DS aged 18-60 months. A positive correlation was found between speech motor function and expressive vocabulary (measured in a spontaneous speech sample). This suggests that children with more speech motor difficulties produced fewer words. Further research, from Kumin et al. (1994) evaluated the clinical records of 60 children with DS aged 9 months to 9 years to look at how children with DS acquired speech sounds. Great variation was found for the development of speech sounds, e.g. some children developed the /th/ sound at 8 months but others did not develop the sound until 11 years. Furthermore, the order in which speech

sounds emerged was different to the pattern seen in TD children suggesting that the trajectory is disordered.

A further study by Cleland, Wood, Hardcastle, Wishart and Timmins, (2010) assessed the relationship between speech, oromotor skills, language and cognitive abilities of 15 children with DS aged 9-18 years. A battery of standardised tests for language and cognition was administered as well as measures of phonology. The results found that two thirds of the group did not meet the basal equivalent of 3 years on the Diagnostic Evaluation of Articulation and Phonology (Dodd, Hua, Crosbie & Holm, 2002); therefore suggesting that two thirds of participants had severe speech sounds deficits. A correlation analysis found a significant association between oromotor skills and speech errors/intelligibility with those with poorer oromotor skills producing a higher rate of speech sound errors and having poorer speech intelligibility. This supports research which has suggested that anatomical differences/difficulties with motor skills may have a negative influence on the production of speech sounds and speech intelligibility. The average intelligibility of the group was found to be 52% and all children in the study made at least one atypical error. Cluster reduction was found to be the most common error, followed by final consonant deletion and initial consonant deletion. A similar pattern of errors has also been reported by other researchers (e.g. Roberts et al., 2005; Stoel-Gammon, 2001).

1.6.3. *Language at School Age and Beyond*

Many children with DS enter school with limited spoken language. Buckley and Bird (2001) report that many children aged 5 years with DS enter school with a spoken vocabulary of approximately 300 words and most will communicate in 2-4-word key word sentences. These children are not able to use correct word endings or efficiently use joining words in sentences. By 8-9 years the average spoken vocabulary reportedly increased to 450-500 words. However, this can range from 150-600, so by 9 years of age some children have less

than 200 words. Intelligibility is also an issue as their speech is often not clear and they may have problems with speech sound production and speech-motor planning (saying longer sentences). The authors also report that it is common for these children to speak much less than TD children of the same age, which means they get less practice speaking.

Further difficulties with speech and language are evident for adolescents with DS. A study by Buckley and Sachs (1987) recorded developmental information of 90 adolescents with DS, three of which had no speech. Seventy percent of the girls and older boys (over 14 years) were reported to frequently use utterances of at least 5 words. Only 50% of the younger boys were found to also do this. Of the younger children: 18% of girls and 33% of boys only communicated using utterances of 3 words or less. This was also recorded for 10% of the older children. Although intelligibility was generally reported as 'good' by parents, the adolescents had trouble being understood by strangers, for example in shops. This may suggest that the parents were able to understand their own children due to repeated exposure and therefore were not able to reliably report on their intelligibility.

There are few studies documenting the language and communication skills of adults with DS (Martin et al., 2009). However, the available studies suggest that communication difficulties are still prominent and present issues such as accessing the community, employment and communicating with health care staff. Furthermore, many adults with DS still access speech and language therapy due to issues with intelligibility (Smith, 2001).

Typically, adults with DS communicate using short utterances and their mean length utterance is around 5 words. Their use of grammatical morphology is limited and inconsistent (Rondal & Comblain, 1996). Adults with DS are at an increased risk of early onset Alzheimer's from around 30 years of age and it is thought to affect approximately 70% of adults with DS over 60 years of age (Lai & Williams, 1989). This can lead to further

language problems including slow production and comprehension of language, reduced fluency and an increased difficulty with word discrimination (Rondal & Comblain, 1996).

The available research demonstrates that speech and language difficulties for individuals with DS persist throughout childhood and into adolescence/adulthood, therefore emphasising the importance of developing and administering early interventions for children with DS so that they can develop speech and language as early as possible. If a child enters school at 5 years with no or few words they will struggle to access the curriculum. Basic literacy skills can promote independence and the ability to be employed which will in turn improve the quality of life of individuals with DS (Miller, Leddy & Leavitt, 1999). Research that has considered the language ability of children with DS at school age or later highlights the importance of improving their speech and language from a young age.

1.6.4. *Precursors to Language*

1.6.4.1. *Babbling*

Prior to using spoken words, TD children use other forms of communication; one example of this is canonical babbling. Canonical babbling is a type of multisyllabic babbling which includes varied use of both consonants and vowels (Fidler, Philofsky & Hepburn, 2007). In TD children this occurs by the time the child is 7-8 months old (Ejiri, 1998). Babbling is predictive of later language for TD children (Whitehurst, Smith, Fischel, Arnold & Lonigan, 1991) and correlates with early social communication behaviours for children with DS (Lynch, Oller, Steffens, Levine, Basinger & Umbel, 1995). Current research on babbling in DS has been mixed in that some studies have suggested that the level of babbling in infants with DS is normal (Buckley, 1993), whereas others have reported a delay in canonical babbling and suggest that it does not occur until around 9 months of age (Fidler, Philofsky, Hepburn & Rogers, 2005; Lynch et al., 1995). There are also reports that canonical

babbling is less stable in infants with DS and that the babbling period is longer than for TD children and often occurs through their second year (Lynch et al., 1995; Stoel-Gammon, 2001). Delays in babbling have been linked to general motor delays and oral structure problems (Stoel-Gammon, 2001).

To summarise, early babbling appears to be associated with later communication and language skills for TD children and children with DS. Some research suggests that the onset of babbling may be delayed for infants with DS and that the babbling period is longer.

1.6.4.2. Gestures

Another form of communication used prior to and in combination with spoken words is the use of gestures. TD infants use gestures to communicate information until they can do this verbally (Iverson & Goldin-Meadow, 2005). The use of gestures has been reported as a relative strength for children with DS, as it enables them to use gestures as an alternative form of communication and this has been encouraged by professionals (Abbeduto et al., 2007).

During early stages of development it has been suggested that children with DS show a preference for using gestures as an alternative to spoken words and that they may use gestures for a longer period than TD children to compensate for their lack of speech (Zampini & D'Ordico, 2008). A study of twenty Italian children with DS aged thirty-six months recorded gesture use and vocabulary production in a twenty minute free play session between the mother and child. For both gesture and verbal production high individual variability was found. Some of the gestural processes that prevailed were similar to those observed in TD children. The researchers observed that gestures appeared to be related to verbal comprehension, not verbal production (Zampini & D'Odorico, 2008).

Further research supporting a gesture advantage shows that children with DS used more gestures than children with other genetic disorders such as Williams syndrome. Singer-Harris, Bellugi, Bates, Jones and Rossen (1997) compared 54 children with Williams syndrome and 39 children with DS aged 12-76 months and found that children with DS used significantly more gestures than children with Williams syndrome.

In summary, the use of gestures is reported to be a relative strength for infants with DS in comparison to individuals with Williams syndrome. Infants with DS frequently use gestures to support communication attempts and research has found early gesture use to be related to verbal comprehension.

1.6.4.3. Play

The pattern of play that is seen in TD children was first identified by Piaget (1962) who suggested that the stage of play the child is in reflects their level of cognitive development. Initially children participate in object play where they explore and manipulate objects focusing on their properties and functions. Around 2-4 years of age symbolic play can be observed. In this stage the child is able to engage in pretend play where they can engage in pretence and use toys symbolically (Venuti, Falco, Esposito & Bornstein, 2009). Symbolic play has been shown to be an indicator of the child's cognitive development and achievements (Bornstein, 2006; Piaget, 1962; Werner & Kaplan, 1963). Research has found symbolic play to be an influential predictor of early language development and to be linked to the ability to produce multiword utterances (Casby & McCormack, 1985; McCathren, Warren & Yoder, 1996). A study by Casby (1980) looked at the development of symbolic play in relation to early language by assessing a TD and a language delayed group. It was found that regardless of age or developmental status, all children's use of multiword utterances in speech was linked to their symbolic play abilities.

Children with DS may differ from TD children in terms of play. For example, children with DS have been reported to have a narrow range of play skills, often involving themselves in stereotypical and repetitive acts during play and to be less likely to initiate play (Hines & Bennett, 1996). Others have found that children with DS follow the same developmental trajectory for play as TD children when matched for mental age (Cielinski, Vaughn, Seifer & Contreras, 1995). This supports the view that children with DS follow the same pattern of play as TD children but at a slower rate (O'Toole & Chiat, 2006; Venuti et al., 2009).

Play skills have also been found to be associated with language development in infants with DS. When the play skills and language scores of 19 children with DS aged 15-54 months were assessed, play skills significantly correlated with expressive language, receptive language and mean length utterance controlling for the child's chronological age (Fewell, Ogura, Notari-Syverson & Wheedon, 1997).

In summary, it appears that infants with DS may go through the same stages of play as TD children but at a slower rate. There is also evidence that play is associated with language development for both children with DS and TD children.

1.6.4.4. Early social communication skills – requesting and joint attention

1.6.4.4.1. Behavioural requests

Behavioural requests are often used by children to obtain something e.g. a toy, or to obtain help with a task. Two main types have been identified – initiating behavioural requests and responding to behavioural requests. Initiating behavioural requests are defined as using gestures (such as pointing) and gaze, to elicit help from a social partner to obtain an object/event. Instrumental tasks include obtaining help with a task whereas social requests are

turn taking or indicating that you would like something to happen again. Responding to behavioural requests refers to responding correctly to such a bid. For example, if a parent requested a particular toy and the child gave it to them (Mundy, Block, Delgado, Pomares, Van Hecke & Parlade, 2007).

Initiating behavioural requests develop between 9-13 months in TD children (Fidler, 2006). However, a delay in behavioural requests has been identified for children with DS. Mundy, Kasari, Sigman and Ruskin (1995) compared 37 children with DS aged 22 months and 25 TD children aged 14 months (matched for mental age). The participants were separated into two sub-groups for low mental age (< 18 months) and high mental age (> 18 months) and were assessed using the Early Social Communication Scale (ESCS; Mundy, Hogan & Doehring, 1996). There were no differences between groups for social interaction behaviours but a significant difference for non-verbal requests with the DS group using fewer than the TD group. Furthermore, Mundy, Sigman, Kasari and Yirmiya (1988) found that children with DS exhibited more social interaction behaviours than the TD and developmentally delayed groups but less instrumental requests.

Further research from Fidler, Philofsky, Hepburn and Rogers (2005) found a significant difference between children with DS and a TD group with the DS group showing significantly fewer instrumental requests. In summary, the research suggests that children with DS have a delay in initiating behavioural requests in comparison to TD children matched for mental age and children with developmental delays. However, further analysis seems to suggest that this deficit is applicable to instrumental requests only and not social requests.

1.6.4.4.2. *Joint Attention (Responding to and Initiating)*

Joint attention is defined as a triadic interaction between an individual, another person and an object or event (Tomasello, 1995). For example, both a mother and child may be focused on the same toy. Two main types of joint attention have been identified – responding to joint attention refers to the infant following another person’s bid. For example, if an infant turns their head to follow the eye gaze or point of another person. The second type is initiating joint attention where the infant directs another person’s attention to an object/event by pointing or turning their head. Initiating joint attention is characterised as using eye contact, pointing and showing to share attention with another individual (Mundy et al., 2003).

In TD children, joint attention begins to develop when they are aged between 2-6 months old (Scaife & Bruner, 1975) and continues to develop until approximately 36 months (Carpenter, Nagello, Tomasello, Butterworth & Moore, 1998). Initially, infants will follow the gaze or head turn of another person. TD infants aged 6 months are able to follow the gaze of another person but cannot follow a gaze to a target that is behind them (Morales, Mundy & Rojas, 1998). This skill continues to emerge and develop when the child is aged between 6-12 months (Butterworth & Jarrett, 1991). By 12 months infants are initiating joint attention and directing an adult’s attention to objects/events in their environment (Carpenter et al., 1998). Infants may point to objects to ‘show’ them to their care-giver.

Some research suggests that children with DS may have a deficit in joint attention, for example infants with DS sometimes struggle with making reciprocal eye contact (Berger & Cunningham, 1983). Other studies have reported that infants with DS are as likely to initiate and respond to joint attention when compared to TD infants of comparable mental age (Adamson, Bakeman, Deckner & Ronski, 2009).

Mundy et al. (1995) assessed 37 children with DS (aged 12-36 months) and 25 TD children (aged 8-28 months). The two groups were matched for mental age and language scores. Participants were divided into a low mental age group (mental age < 18 months) and a high mental age group (mental age > 18 months). No differences were found between the two groups for initiating joint attention but children with DS scored significantly lower for responding to joint attention than the TD group.

Conflicting results were found by Sigman and Ruskin (1999) who used the same measure of responding to joint attention (the gaze following task from the Early Social Communication Scale; ESCS, Mundy et al., 1996) to compare children with DS aged 24-48 months and a TD group of equivalent mental age. They found no differences between the two groups for responding to joint attention. Furthermore, Sigman and Ruskin (1999) found that children with DS had significantly better initiating and responding to joint attention skills than children with Autism Spectrum Conditions (ASC) of the same chronological age (24-48 months). Further studies have identified that children with ASC have significantly worse joint attention skills compared to individuals with DS (Adamson, Bakeman, Deckner & Ronski, 2009).

The differences in results found by the two studies are likely to be due to participant characteristics since both studies used the ESCS to measure joint attention. A wider age range was used in the Mundy et al. study (12-36 months) in comparison to the Sigman and Ruskin study (24-48 months). Furthermore, some of the infants in the latter study were twice the age of participants in the Mundy et al. study. It is therefore likely that children in the Sigman and Ruskin study had more developed joint attention due to their older age and greater exposure to interaction. This may explain why a deficit was found for the younger group in the Mundy et al. study only.

In summary, mixed results have been found regarding whether infants with DS have a deficit in joint attention skills relative to their mental age. However, it has been identified that infants with DS have significantly better joint attention skills than infants with ASC.

1.6.4.4.3. Joint Attention and Language Development

The relationship between initiating and responding to joint attention and concurrent and longitudinal language skills has been investigated extensively in TD children. Multiple studies have found that early joint attention skills are associated and sometimes predictive of concurrent and longitudinal language outcomes for TD children (Crowson, Markus, Yale & Schwartz, 2002; Delgado, Mundy, Colonesi, Stams, Koster & Noom, 2010; Markus, Mundy, Morales, Delgado & Yale, 2000; Tomsello & Farrar, 1986). For example, Morales, Mundy and Rojas (1998) assessed responding to joint attention in 21 TD infants when they were 6 months old. They found that responding to joint attention significantly positively correlated with receptive vocabulary at 12 months and with expressive vocabulary at 18 and 24 months, suggesting that those with better early responding to joint attention skills had better language outcomes.

Furthermore, Mundy and Gomes (1998) assessed 24 TD infants aged 14-17 months. Joint attention was assessed using the ESCS and language scores were computed using the Reynell Developmental Language Scales (Reynell & Graber, 1990). Both initiating joint attention and responding to joint attention significantly positively correlated with receptive and expressive language scores measured 16 weeks later. A regression analysis showed that responding to joint attention was a significant predictor of receptive language when controlling for chronological age, mental age, baseline scores and initiating joint attention, whereas, initiating joint attention was a significant predictor of expressive language when controlling for chronological age, mental age, baseline scores and responding to joint attention.

The research suggests that for TD infants both initiating joint attention and responding to joint attention are related to expressive and receptive language scores both concurrently and longitudinally. However, there has been hardly any research in this area which has considered the role of joint attention and language development for infants with DS.

Mundy et al. (1995) found that initiating joint attention significantly correlated with receptive and expressive language concurrently and longitudinally for the DS group. However, a significant relationship was not found between responding to joint attention and language. For the TD group, initiating joint attention significantly correlated with concurrent expressive language and responding to joint attention was found to correlate with both receptive and expressive language, concurrently and longitudinally.

On the other hand, Sigman and Ruskin (1999) found that for the infants with DS, responding to joint attention significantly correlated with concurrent language skills (combining receptive and expressive). However, responding to joint attention was not significantly associated with expressive or receptive language gains 12 months later.

Further support that joint attention is related to language gains for children with DS comes from Harris, Kasari and Sigman (1996). The joint attention and language development of 28 children with DS and a TD group matched for mental age were assessed. Children with DS were tested at 23 and 36 months. Joint attention was measured by coding a parent child interaction at the first time-point. Children with DS spent 20% more time in joint attention episodes than TD children. However, there were no significant differences between the two groups with regard to frequency of episode or average length. The parents of children with DS maintained attention more to care-giver selected toys than parents of TD children. Over the 13 months between the first and second assessment the infants with DS averaged a 3.68 month growth in expressive language compared to 15.06 months for the TD group. For

receptive language the DS group exhibited an average 5.21 month growth and the TD group 18.82 months.

Further analysis revealed a positive significant correlation between the total number of seconds spent in joint attention episodes and receptive language gains for TD children. The average length of a joint attention episode positively correlated with receptive language gains for children with DS. Joint attention duration, frequency of episodes or average length of episodes were not significantly associated with gains in expressive language for either group. Receptive language development positively correlated with the frequency with which the caregiver maintained attention to child selected toys and to toys in general. A negative correlation was found between receptive language development and the frequency which the caregiver re-directed the child's attention from child selected toys and toys in general.

Other research has considered the link between time spent in joint attention episodes and vocabulary development for children with DS. Zampini, Salvi and D'Odorico (2015) assessed the joint attention and concurrent and longitudinal vocabulary of 18 infants with DS aged 24 months. When developmental age was controlled for, time spent in joint attention episodes was found to significantly correlate with receptive vocabulary at 24 months. Further analysis revealed that joint attention at 24 months was a significant predictor of receptive vocabulary at 30 months.

Furthermore, a longitudinal study, from Mason-Apps (2013) assessed precursors of language development in a group of TD and DS children who were matched for non-verbal mental age. The precursors assessed included: speech segmentation, responding to joint attention, initiating joint attention, initiating behavioural requests, parental responsivity and non-verbal mental ability. Assessments of expressive and receptive language were also included. Participants were assessed at three time points, initially when the DS group was 18-

21 months, then at a 6 month follow up (M age= 26 months) and finally a year after the initial assessments (M age= 32 months). The results for the DS group showed that there was a concurrent relationship between non-verbal mental age and receptive language and vocabulary, and between responding to joint attention and expressive language and receptive vocabulary. Longitudinally, non-verbal mental age at time 1 predicted receptive language at times 2 and 3, whereas responding to joint attention at time 1 predicted both expressive and receptive language outcomes at time 3. Non-verbal mental age at time 1 was found to be a unique predictor of receptive language at time 2 and responding to joint attention at time 1 was a unique predictor of expressive communication and vocabulary scores at time 3. In comparison, for the TD group – initial responding to joint attention and non-verbal mental age were significant predictors of concurrent receptive vocabulary scores. Time point 1 initiating joint attention and speech segmentation were significant predictors of expressive language and vocabulary at time point 2. Finally, speech segmentation at time point 1 was a significant predictor of receptive language, expressive language and receptive vocabulary at time point 3.

In summary, it appears that multiple studies have established relationships between joint attention and language/vocabulary outcomes for infants with DS.

1.6.4.5. Maternal Interactive Style and Links to Language Development

Early research tends to suggest that mothers whose children have developmental delays often interact differently than mothers of TD children and that they tend to act in a more directive, intrusive and controlling way during interactions with their child (Glenn, Dayus, Cunningham & Horgan, 2001; Tannock, 1988). Various explanations are offered for this potential difference – including mothers compensating for their child's lower cognitive level (Krakow & Kopp, 1982).

Nevertheless, a difference between how mothers of TD and mothers of DS children interact with their offspring has not always been found. Gilmore, Cuskelly, Jobling and Hayes (2009) found no differences between how supportive or directive the mothers of children with DS were compared to mothers of mental age matched TD children. Similarly, Sterling and Warren (2014) reported that, although mothers of children with DS used more directive type behaviours (e.g. requests for behavioural compliance) compared to mothers of TD children, they did not increase the directive behaviours thought to hinder language development (e.g. redirecting the child's attention). However, in the same study, Sterling and Warren found that mothers of children with DS differed from mothers of TD children in that they used facilitative behaviours more frequently with older than with younger children. Examples of facilitating behaviours included giving verbal praise in response to a child's action, or giving a verbal interpretation of something the child said. The reverse was seen in the TD group whereby mothers used more facilitative behaviours with younger children. The authors suggest that the mothers of DS children adapted their style to meet the linguistic needs of their child. This has also been found in early research (e.g. Marfo, 1990). Within the literature on maternal interactive style, different concepts have been discussed, including directiveness, sensitivity and warmth (e.g. Clarke-Stewart & Apfel, 1979; Leigh, Neivar & Nathans, 2011; Sterling & Warren, 2014) and each of these and the effect on language will be discussed below.

1.6.4.5.1. Directiveness

Directiveness is defined by high levels of parental control over their child's behaviour and/or attention (Sterling & Warren, 2014). Higher levels in adult directiveness have been negatively associated with children's language development. For example, maternal directiveness was found to be negatively correlated to the number of object labels in a TD child's vocabulary (Tomasello & Farrar, 1986; Tomasello & Todd, 1983). Tomasello and

Todd (1983) suggested that high levels of adult directiveness make it more difficult for a child and adult to have a joint focus on an object. Tomasello (1988) also argued that by trying to redirect the child's attention, the adult alters the learning conditions for acquiring individual referential words as the child has to attempt to shift their focus and try to coordinate with the adult. This was demonstrated in a lexical training study where an adult tried to teach novel words to 17-month-old TD children. There were two conditions: 1) a 'follow in' condition, in which the experimenter referred to an object which the child was already focussed on; and 2) a 'direct condition' in which the experimenter used an object which was not the child's current focus and they were therefore required to shift their attention. The researchers reported that those in the 'follow in' condition were more successful than the 'direct condition' group in learning to understand the novel words (Tomasello & Farrar, 1986).

Further research has identified that since children with DS are found to be less motivated in a task and more passive than their TD peers, over time maternal directiveness may prevent a child's motivation to persist independently when they face a challenging task (Gilmore, Cuskelly, Jobling & Hayes, 2009). In their problem-solving study, Gilmore et al. (2009) reported that, in the DS group, those children with 'supportive' mothers (mothers who were supportive of their child's autonomy on the task) demonstrated greater persistence than those with 'directive' mothers (mothers who tried to control and direct their child's behaviour). They therefore suggested that early interventions which facilitate autonomy for the child and less directive behaviour from parents may have significant benefits for the child's motivation and ability (Gilmore et al., 2009).

However, not all 'directive' parental behaviours hinder language development. A longitudinal study by Akhtar, Dunham and Dunham (1991) reported a significant positive relationship between 'follow prescriptives' (the mother directing the child's behaviour to

something the child was already engaged in: for example if the child was holding a ball and the mother said: “roll the ball to mummy”) at 13 months and the child’s vocabulary at 22 months. In contrast, a significant negative correlation was found between the child’s vocabulary at 22 months and the number of the times the mother attempted to change the child’s attentional focus. This suggests that directives that follow the child’s interest may be beneficial for language development but those which aim to change the child’s focus may have a negative association with language development. These findings are corroborated by McCathren, Yoder and Warren (1995).

In terms of atypical development, research is quite scarce. An early study by Crawley and Spiker (1983) assessed two-year-old children with DS and their mothers using a semi structured mother-child play interaction and maternal, child and dyadic qualities with the Mental Development Index Scores on the Bayley Scales of Infant Development (Bayley, 1969). The interactions were coded using a rating scale for maternal qualities such as directiveness and sensitivity; child qualities, including play maturity, social initiative, social responsiveness; and one dyadic quality, which was mutuality, i.e. the degree to which the mother and child shared the same intentions. They found that maternal directiveness and sensitivity were partially orthogonal and that mothers who combined sensitivity and directiveness for the purposes of stimulating the child had children with higher mental development index scores. However, the relationship between maternal directiveness and sensitivity and language per se is not clear because only general mental age scores are provided.

Some of the research presented would suggest that parental directiveness can potentially impede language development, motivation and overall competence but this depends on the type of directives used. As argued by Gilmore et al. (2009), directiveness is often operationalised differently and there are differences in the methodology (e.g. how

behaviours are observed, free play session vs. task) which may account for the variations in findings. Furthermore, the distinction between directives which follow the child's interest and directives which attempt to change the child's focus seems to be important.

1.6.4.5.2. Sensitivity and Warmth (Positive Expressed Emotion and Affection)

Previous research examining the relationship between maternal interactive style and child outcomes shows that a healthy, warm, nurturing and stable relationship between the caregiver and the child, along with contingent, prompt and appropriate caregiver reactions to child behaviours, has a positive impact on different aspects of a child's development including language (Landry, Smith, Swank, Assel & Vellet, 2001). Sensitivity, defined as being attuned to infant signals, needs and direction of interest, and responding promptly and appropriately to them, is one dimension of maternal interactive style that has been associated with expressive and receptive language abilities (Leigh, Neivar & Nathans, 2011). For example, maternal sensitivity at 9 months has been found to predict child language comprehension at 13 months and the development of language milestones when controlling for child behaviours at 9 and 13 months, such as vocalisations and play (Baumwell, Tamis-LeMonda & Bornstein, 1997; Tamis-LeMonda, Bornstein & Baumwell, 2001). Additionally, maternal sensitivity between 6 and 18 months has been found to positively correlate with expressive language skills at 30-36 months (Leigh, Nievar & Nathans, 2011; Nozadi et al., 2013). Of particular relevance is the fact that, maternal sensitivity is reported to be an important factor for fostering child language development when children are at risk of language delay (Hoff-Ginsberg, 1987), or with children who have lower language comprehension levels (Baumwell et al., 1997).

A positive relationship has also been reported between maternal warmth (e.g. expressing positive emotions, praising the child) and language ability (Clarke-Stewart &

Apfel, 1979; Landry, Smith, Swank, Assel & Vellet, 2001; Steelman, Assel, Swank, Smith & Landry, 2002). Both maternal sensitivity and warmth contribute to creating a stimulating social environment for the child, whereby they feel supported, guided, encouraged to engage in joint attention and motivated to learn and use appropriate language (Bigelow et al., 2010). In contrast, a lack of sensitivity and warmth has been found to be negatively correlated with language abilities (Landry, Smith, Miller-Loncar, & Swank, 1997).

However, the relationship between maternal interactive style and language development has not been previously investigated for children with DS. We are therefore unsure as to whether maternal interactive style is related to language development for this group and if so what the 'optimum' interactive style is.

1.7. *Summary*

The available research shows that, commonly, a pattern of strengths and weaknesses is observed for individuals with DS in terms of their language development. Receptive language tends to be better than expressive and can exceed the level expected from the child's mental age (Abbeduto et al., 2003; Caselli et al., 1998; Fidler & Nadel, 2007; Miller, 1999). On the other hand, delays in early expressive language and the emergence of first words are evident, with some children not producing any words until 3 years of age (Abbeduto et al., 2007). Such delays continue through the early years and into childhood (Rondal, 1998) and are evident when children start school (Buckley & Bird, 2001). Further issues have been reported with speech intelligibility (Kumin, 2006), expressive grammar (Fowler et al., 1994) and receptive syntax (Abbeduto et al., 2003).

Various studies have investigated early precursor skills to language and the effect they have on concurrent/longitudinal language for infants with DS. Deficits have been identified for instrumental requests but not for social requests (Fidler et al., 2005; Mundy et al., 1988).

There are contradictory findings regarding an early deficit in responding to joint attention (Mundy et al., 1995; Sigman & Ruskin, 1999) but some evidence points to the fact that responding to joint attention may be related to concurrent and longitudinal language abilities for children with DS (Mason-Apps, 2013; Sigman & Ruskin, 1999).

In terms of maternal interactive style, research suggests that mothers of developmentally delayed children are less sensitive/more intrusive during play (Glenn, Dayus, Cunningham & Horgan, 2001; Tannock, 1988) but this pattern is not always observed in DS (e.g. Gilmore et al., 2009; Sterling & Warren, 2014). Being directive/intrusive can have a negative impact on language development (Tomasello & Farrar, 1986; Tomasello & Todd, 1983) but this depends on how directiveness is being measured, e.g. re-directing the child's attention vs directing them on current task. Maternal sensitivity and warmth have been found to be positively associated with language development (Landry, Smith, Swank, Assel & Vellet, 2001; Leigh, Nievar & Nathans, 2011; Nozadi et al., 2013; Steelman, Assel, Swank, Smith & Landry, 2002). However, there is little research in this area for children with DS so it is not clear if sensitivity/warmth has the same impact on language development as for TD infants.

Furthermore, parents can enhance and support language development by following their child's lead during interactions and using directive speech that follows the child's current interest (Akhtar, Dunham and Dunham, 1991; Tomasello, 1988). Research has found that parental sensitivity and warmth creates a stimulating environment which in turn facilitates language development (Bigelow et al., 2010). This supports the transaction model of development (Sameroff, 1975) which has found that language development is supported through reciprocal interactions of a caregiver and child (Warren & Brady, 2007).

Chapter 2 – Early Precursors to Language

This chapter will investigate which early precursors (including joint attention and maternal interaction style) are related to concurrent language scores for children with DS.

2.1. Rationale for Study

The current study focuses on the relationship between early social communication skills and concurrent language scores for children with Down Syndrome (DS) aged 17-22 months and a typically developing (TD) group aged 9-11 months with comparable non-verbal mental age and language scores. Specifically, the study focuses on joint attention (both initiating and responding to) and maternal interactive style. These variables were focused on as they have both been linked to early language development for TD children (Baumwell, Tamis-LeMonda & Bornstein, 1997; Crowson, Markus, Yale & Schwartz, 2002; Delgado, Mundy, Colonnesi, Stams, Koster & Noom, 2010; Markus, Mundy, Morales, Delgado & Yale, 2000; Tamis-LeMonda, Bornstein & Baumwell, 2001; Tomasello & Farrar, 1986). It was therefore, of interest, to investigate if the two groups differed on any of these variables and if these variables were associated with concurrent language abilities. Children with DS are likely to have significant language delays in their early life; hence it is of importance to understand which early social communication factors are related to early language development. This could have implications for early intervention.

The aim of the study is to compare the two groups on joint attention and maternal interactive style to investigate if there are any differences and if any aspect of joint attention or maternal interactive style is related to concurrent language scores for children with DS or TD children. The study will enable us to see which factors are related to for early language development for these two groups of children.

Based on previous research which has identified some differences in terms of interactive style between parents of children with DS and parents of TD children, we predict that the mothers of children with DS may be rated as less sensitive than the parents of TD children (e.g. Glenn, Dayus, Cunningham & Horgan, 2001; Tannock, 1988). Only maternal interactions were compared since research shows that fathers may interact with their children differently from mothers (e.g. Hallers-Haalboom et al., 2014). Since numerous studies have identified that both initiating and responding to joint attention are related to language outcomes for both children with DS and TD children (see 1.6.4.4.3), we predict that joint attention will be related to concurrent language scores in both groups (e.g. Mundy et al., 1998; Mundy & Gomes, 1998; Sigman & Ruskin, 1999). Finally, since previous research has identified that maternal sensitivity and warmth are associated with better language outcomes for TD children (Leigh et al., 2011; Tamis-LeMonda et al., 2001) we predict that there will be a relationship between a positive maternal interactive style and concurrent language scores for this group. However, as to our knowledge no previous research has investigated this for children with DS, we are not able to predict if a relationship will be found between maternal interactive style and language abilities as measured by standardised assessments for children with DS.

2.2. Research Questions

1. Will there be differences between the two groups of parents in terms of maternal interactive style?
2. Will joint attention or maternal interactive style be related to concurrent language scores for children with DS or TD children?

2.3. *Hypotheses*

1. Joint attention will be associated with concurrent language scores for both TD children and children with DS.
2. A positive maternal interactive style will be related to concurrent language scores for the TD children. Based on the previous research, we are unable to make a prediction for the relationship between maternal interactive style and language for children with DS.

2.4. *Method*

2.4.1. *Ethics, Recruitment and Consent*

Ethical approval was gained from the University of Reading's Research Ethics Committee and given favourable opinion to proceed. Parents were required to give consent on their child's behalf. However, testing sessions were stopped or postponed if a child became visibly upset.

2.4.2. *Participants*

Participants included 25 children with DS (11 girls and 14 boys) and 30 TD children (14 girls and 16 boys). The mean age of the children with DS was 19 months (range 17-22 months) and the mean age of the TD children was 10 months (range 9-11 months).

The TD children were recruited using the University of Reading Child Development Database (a database which contains contact details for children in the surrounding areas who have expressed an interest in participating in research studies). The children with DS were recruited through local charities and support groups; a flyer with the researcher's contact details was given to interested parents explaining the aims and purpose of the study. Two of the infants with DS were exposed to another language but English was their dominant language. All other participants across both groups were monolingual native English

speakers. All children were participating in either a longitudinal or intervention study and the data from this experiment was taken from their baseline assessment.

The two groups had equivalent non-verbal mental age (calculated by combining the Visual Reception and Fine Motor sub-scales on the Mullen's Scale of Early Learning; Mullen, 1995) and total language scores which were assessed using the Pre-school Language Scales 4 (Zimmerman, Steiner & Pond, 2002).

2.4.2.1. Parent demographics

Eight percent of the mothers of DS children reported no formal qualifications, 12% had GCSE's, 20% had A-levels or an NVQ and 56% had been educated to degree level or higher. Seventeen percent of the mothers of TD children had GCSE's, 17% had A-levels or an NVQ and 67% had been educated to degree level or higher. Eight percent of the fathers of DS children had GCSE's, 33% had A-levels or an NVQ and 50% were educated to degree level or higher. Finally, 6% of the fathers of TD children had GCSE's, 13% had A-levels or a NVQ and 80% had been educated to degree level or higher.

In terms of occupation, the majority of the mothers of children with DS were not currently working (62.5%), one mother was on maternity leave, and the remaining were employed (33%). The mothers of TD children were mostly split between not currently working (25%), employed (50%) and on maternity leave (29%). All the fathers in both groups were either employed full time or self-employed. Data was dichotomised so that Fischer's exact tests could be used to identify any significant differences between the two groups (see table 2.1). Education was split into compulsory education (up to 16 years old) and further education. Employment was split into currently employed (including full-time, part-time and self-employed) and not currently working (including parents on maternity leave or full-time parents).

Table 2.1: Demographic variables of parents of children of DS and parents of TD children

	Levels	Frequency		<i>p</i>
		DS (25)	TD (30)	
Gender	Male	14	16	
	Female	11	14	.530
Maternal ed	Compulsory	3	5	
	Further	21	25	.487
Maternal employ	Employed	8	19	
	Unemployed	16	8	.009*
Paternal ed	Compulsory	8	2	
	Further	14	28	.01*
Paternal employ	Employed	22	27	
	Unemployed	0	0	--

Note. Maternal ed= maternal education level, Maternal employ= maternal employment status, Paternal ed= paternal education level, Paternal employ= paternal employment status, * - highlights significant difference

The results show that there were no significant differences between the two groups for gender, maternal education or paternal employment. However, a significant difference was found for maternal employment with more mothers of TD children currently working in comparison to the DS group. Similarly, a difference was also found for paternal education with more fathers in the TD group having received further education.

2.5. Measures

2.5.1. Maternal Measures

To measure maternal interactive style each parent-child dyad was video recorded in a child friendly room for 5 minutes whilst engaging in a play interaction. This interaction was then coded using the 'Coding scheme for structured mother-infant play interaction' (Murray & Karpf, 2000), which is an adaptation of the Global Rating Scales (Murray, Fiori-Cowley, Hooper & Cooper, 1996a). A different version of the scheme has been previously used with children with DS (Slonims & McConachie, 2006). The author and a research assistant (who was blind to the study's aims and hypotheses) were provided with training materials by Murray and Karpf. Once the training was completed, they independently coded 20% of the sample and good inter-rater reliability was found: $\kappa = .737$, $p < .001$, $CI = -0.59-0.88$. Only certain subsections were chosen and these were ones which focused on the mother: verbal control, positive expressed emotion, negative expressed emotion, coercions/intrusions, verbal elaboration, emotional tone, sensitivity and the reciprocity of the interaction.

The verbal control category was coded by analysing the utterances by the mother and coding how many of these were 'strong verbal control' or 'mild verbal control' directed at the infant. An example of strong control was: 'come here!' and was generally a directive or demand. Examples of mild control were statements such as: 'would you like to...' or 'shall we...?'

For positive expressed emotion any instances of verbal praise or terms of endearment/affection displayed by the mother towards the child were scored. For example, 'good girl' (praise) or 'you silly billy (endearment)'. Furthermore, if the mother clapped in response to something the child had done or showed physical affection this was also scored. In contrast negative expressed emotion was scored if the mother was critical or showed

frustration towards the child. For example, 'don't do that' in a negative tone of voice when there is no obvious reason (i.e. not a safety issue).

Finally, coercions/intrusions were coded by counting how many times the mother physically interrupted or disrupted the child's play. For example, if the child was sitting playing with a toy and the mother moved the child or took away the toy.

The rest of the subsections were coded on a scale of 1-5. For example, for verbal elaboration 1= no verbal elaboration, 2= little verbal elaboration, 3= moderate verbal elaboration, 4= good verbal elaboration and 5= very good verbal elaboration. Rather than counting individual instances to code such categories it was necessary to watch the whole interaction and then make a judgement. For verbal elaboration, the coder had to pay attention to the quantity and quality of elaborations by the mother. For example, a mother at the lower end of the scale may display few elaborations and these would be simple descriptive statements, such as naming the toy and describing the colour. Mothers at the higher end would be frequently elaborating and using a combination of simple and higher level educational elaborations. An example of a higher level elaboration could be: 'the ball is blue, the same colour as your t-shirt' or 'daddy's on the phone, he's calling from work'.

Similarly, emotional tone was coded on a 5 point scale ranging from very unhappy to very happy. Mothers at the lower end of the scale were described as either showing a verbal protest or being neutral/distant for the majority of the interaction. Whereas those at the higher end of the scale were happy and animated throughout.

Sensitivity was coded using a similar five point scale ranging from highly insensitive to highly sensitive. To code this variable, many elements of the interaction were considered, for example: 1) whether the mother let the infant explore but offered help when needed, 2) whether the mother responded quickly and efficiently to child cues; 3) the physical

positioning of the infant and child, i.e. whether the mother was sitting with the child and able to make eye contact. Mothers who were scored as being insensitive frequently missed their child's cues or responded inappropriately and seemed to focus on their own preferences rather than the child's. On the other hand, mothers who were scored as sensitive responded to their child's cues consistently and appropriately and allowed their child to explore the room and toys, offering help and guidance when needed.

Finally, reciprocity was scored in the same way as sensitivity but was slightly different to the other variables in that the focus was on the interaction as a whole rather than solely the mother's behaviour. Interactions that were coded at the lower end of the scale involved solitary activity/play. For example, if the mother and child focused on different toys and never engaged in goal oriented activities, such as stacking blocks together. Those at the higher end of the scale were interactions where there was joint engagement throughout and the mother and child played together with a toy (see appendix B for the coding scheme).

2.5.2. *Child Measures*

To assess non-verbal mental age, the visual reception and fine motor sub-sections of the Mullen's Scale of Early Learning (MSEL; Mullen, 1995) were combined into a composite variable as used by Wetherby et al. (2004) for children with developmental delay. The MSEL is a standardised assessment and has been used previously with both TD children and children with DS (e.g. Klee et al., 1998; Sterling & Warren, 2014; Wright, Kaiser, Reikowsky & Roberts, 2013)

For language, a direct assessment was also used – the Pre-school Language Scales-4 (PLS; Zimmerman, Steiner & Pond, 2002) which is standardised for children aged 0 to 6;5 years. The PLS consists of a receptive language and expressive language subscales. These

can be combined to form a total language score. The measure has been used previously with TD children and children with DS (Bird et al., 2005; Chiat & Roy, 2008; Wright et al., 2013).

Joint attention was assessed using the Early Social Communication Scales (ESCS; Mundy et al. 1996) with tasks relating to initiating and responding to joint attention. This measure has been used with TD children and children with DS (Mundy et al. 1995; Mundy & Gomes, 1998). This assessment was video recorded and later coded using the specified coding scheme. Initially the researcher and a research assistant independently coded 20% of the data and good inter-rater reliability was found $r(12) = .973, p < .001$.

Materials used for the ESCS included three wind-up toy animals, three mechanical toys (a telephone, a spinning top and a light up hand-held wind-mill) a picture book and four posters of known children's characters. The posters were placed to the left, right, behind left and behind right of the child. To elicit initiating joint attention an object spectacle task was used. Each toy was activated in front of the child for approximately 6 seconds and then given to the child for around 10 seconds. This was repeated three times for each toy. During this task, any of the following behaviours were coded as the child attempting to initiate joint attention with either the researcher or the caregiver. Low level initiating joint attention behaviours included the child alternating their gaze between an active toy and the researcher or the child making eye contact with the researcher whilst they (the child) were manipulating the toy. High level initiating joint attention behaviours included pointing to an active object or picture and showing the toy by raising a toy up to the researcher or caregivers face. A tally was made of how many times the child exhibited each type of initiating joint attention behaviour.

Responding to joint attention was scored during a proximal point task and a gaze following task. For the proximal point task, a picture book was used. The researcher

presented the child with the book and asked 'what can you see'. The child was then able to look through the book for around 10 seconds. The researcher then retrieved the book and pointed to six pictures in the book on consecutive pages whilst saying the child's name. For a correct response, the child was required to turn their head and look at the selected picture. A score out of six was obtained for each child and this was converted into a percentage.

The gaze following task used the four posters which had been placed to the side and behind the child (a diagram of the room can be seen in section 4.5.3). The researcher obtained the child's attention, made eye contact and then turned to point to the poster to the left of the child. Whilst pointing at the poster the researcher called the child's name three times. This process was then repeated for the posters on the right, behind left and behind right. To obtain a correct response the child had to turn their head and look at the appropriate poster. This task was administered twice during the ESCS and so each child was given a score out of eight. This was then converted into a percentage. A total responding to joint attention score was made by combining the scores from both the proximal points and gaze following tasks.

2.6. Procedure

The majority of participants were assessed in a child friendly room within the Speech and Language Therapy Clinic at the University of Reading. Those who were unable to travel were assessed in their own home. The testing session took approximately 1 hour and breaks were given when needed. For the non-verbal mental age, joint attention and language assessments children were seated on their parents lap or on a small chair at a table opposite the researcher. At the end of the testing session parents were asked to engage in a 5-minute play interaction with their child. They were left with a variety of toys including a shape sorter, stacking blocks, balls and books. Parents were instructed to play with their child normally as they would at home and to use any of the available toys. All the play interactions

took place on the floor. The researcher watched and recorded the play interaction from an observation room and then coded the interaction at a later date.

2.7. Results

2.7.1. Results Strategy

Throughout the thesis regression models are used to look at concurrent and longitudinal predictors of outcome measures. Due to the study being exploratory in nature, we chose to build our regression models based on significant associations that we found between the predictor variables and outcome measures as opposed to a more theoretical approach. A similar approach has been used in previous studies (e.g. Zampini, Salvi & D'Odorico, 2015). This approach will be used throughout the thesis.

2.7.2. Between Group Comparisons

Table 2.2 shows the descriptive statistics and statistical analysis for language, joint attention and maternal interactive style for both the DS and TD groups. In order to investigate if there were any differences between groups in terms of their language profile, joint attention skills and maternal interactive style, mean group scores were compared. In addition to total responding to joint attention and initiating joint attention scores we also looked at specific tasks including low and high level initiating joint attention behaviours and both the proximal point and gaze following task. For the gaze following task, the child's ability to follow a target to the side and behind them were analysed together and separately. Normality tests revealed that only the total responding to joint attention variable was normally distributed (see appendix G). Therefore, a combination of independent t-test and Mann Whitney U tests were used to investigate if there were any differences between groups for the two variables.

Table 2.2: Descriptive statistics for language, joint attention and maternal interactive style in DS and TD (raw scores)

Variable	<i>M(SD)</i>		<i>t</i>	<i>d</i>	
	DS (n= 25)	TD (n= 30)			
Total RJA	42.15 (23.4)	45.55 (23.92)	.530	-.14	
Mild VC	21.33 (13.46)	17.48 (9.26)	-1.146	.033	
			<i>U</i>	<i>Z</i>	<i>r</i>
Gaze follow	32.29 (21.47)	24.58 (21.14)	287.5	-1.29	-.17
Left and right	56.25 (34.77)	45.83 (37.76)	300	-1.069	-.15
Behind	8.33 (15.93)	3.33 (8.64)	314	-1.186	-.16
Book point	53.31 (34.68)	70.27 (35.85)	258	-2.02	-.27
Low IJA	16.33 (10.37)	15.69 (10.92)	325.5	-.403	-.06
High IJA	2.00 (4.00)	2.38 (3.13)	294.5	-1.028	-.14
IJA total	19.73 (11.94)	17.76 (13.12)	268	-.971	-.13
Strong VC	17.57 (8.87)	8.96 (7.76)	114	-3.280**	-.48
PEEM	5.33 (4.71)	3.52 (2.57)	215.5	-1.05	-.14
NEEM	0.33 (0.58)	0.24 (.6)	233.5	-.889	-.13
Intrusions	1.71 (2.45)	1.20 (1.32)	259.5	-.070	-.01
VE	2.76 (1.14)	2.92 (1.12)	243	-.450	-.07
Emo T	3.24 (1.18)	3.24 (1.09)	260	-.057	-.01
Sensitivity	2.95 (1.2)	3.04 (1.24)	256.5	-.136	-.02
Recip	3.14 (1.2)	3.08 (1.04)	243	-.448	-.07
PLS AC	19.52 (2.6)	17.53 (1.01)	192.5	-3.14**	-.42
PLS EC	18.96 (2.78)	19.2 (2.01)	324	-.876	-.12
PLS TL	38.48 (4.74)	36.73 (2.53)	306	-1.18	-.16

Note. RJA – responding to joint attention, VC= verbal control, IJA – initiating joint attention, PEEM – positive expressed emotion, NEEM= negative expressed emotion, VE= verbal elaboration, Emo T= emotional tone, Recip= reciprocity, PLS AC – auditory component, PLS EC – expressive component, PLS TL – total language, t= t-test, d= Cohen's d, U = Mann Whitney U, r= effect size, 0.1= small size, 0.3= medium size, 0.5= large size (Field, 2009), ** - $p < .01$

There were no significant differences between groups for any aspects of joint attention ($p > .05$) including low level and high level behaviours of both initiating joint attention and responding to joint attention. Similarly, there were no differences between groups for mild verbal control, maternal expressed emotion, intrusions, verbal elaboration, emotional tone, sensitivity and reciprocity. However, a significant difference was found for strong verbal control with mothers of children with DS using significantly more strong verbal control phrases than parents of TD children. There was no significant difference for total language scores or for expressive language skills. However, a between group difference was found for receptive language thus reflecting the receptive language advantage exhibited by children with DS.

2.7.3. *Correlation Analysis*

Our next question focused on the relationship between joint attention and concurrent language scores as measured on the PLS-4 for children with DS and TD children, as well as, the relationship between maternal interactive style and concurrent language for the two groups. Initially we split the groups (children with DS and TD children) and conducted a correlation analysis to determine if there was a relationship between any of the joint attention and maternal interaction style variables with the concurrent language scores. As the majority of the variables, including the PLS scores, were not normally distributed, Spearman's rho was used (see table 2.3, see Appendix H for full correlation matrix).

Table 2.3: Correlations between language, nonverbal mental age, joint attention and maternal interactive style for the DS and TD group (Spearman's rho)

	DS			TD		
	AC	EC	TL	AC	EC	TL
Age in days	.257	.012	.201	.170	.376*	.319
NVMA	.554**	.303	.494*	.218	.033	.115
RJA (total)	.528**	.512**	.597**	-.022	.182	.131
Book point	.465*	.440*	.512**	.008	.068	.057
Gaze follow	.440*	.496*	.531**	-.070	.236	.159
Left and right	.492*	.565**	.599**	-.075	.216	.141
Behind	.112	.104	.122	-.013	.209	.160
IJA (total)	.298	.303	.375	.003	-.020	.018
IJA low	.172	.286	.261	-.081	.051	.006
IJA high	.202	.456*	.378	.126	.258	.255
Strong VC	-.141	.145	.019	.178	.063	.145
Mild VC	-.089	-.314	-.253	.107	.178	.193
PEEM	-.222	-.111	-.205	.460*	.607**	.643**
NEEM	.128	.017	.076	.047	.099	.10
Intrusions	.266	.242	.346	.058	.190	.214
VE	.110	.066	.099	-.473*	-.017	-.123
Emo T	-.122	.147	-.030	-.141	.187	.106
Sensitivity	.054	.244	.137	-.165	.101	.011
Recip	.069	.300	.200	-.238	.177	.057

Note. * - $p < .05$, ** - $p < .01$, *** - $p < .001$, NVMA – non-verbal mental age, RJA-responding to joint attention, IJA – initiating joint attention, VC= verbal control, PEEM – positive expressed emotion, NEEM= negative expressed emotion, VE= verbal elaboration, Emo T= emotional tone, Recip= reciprocity, AC – auditory comprehension, EC – expressive communication, TL – total language PLS.

For the DS group, a significant positive correlation was found between non-verbal mental age and receptive and total language scores, as well as high level initiating joint attention behaviours (e.g. pointing and showing) and expressive language. Further significant positive correlations were found between total responding to joint attention scores as well as the scores for the two separate tasks (proximal points and gaze following) with expressive, receptive and total language scores. Focussing on the gaze following task – the overall score correlated with language as did the child's score on left and right trials only. However, there was not a significant correlation between the behind trials and any of the language scores.

On the other hand, for the TD group, a significant positive correlation was found between the child's chronological age and expressive language score. Further significant positive correlations were found between maternal positive expressed emotion and receptive, expressive and total language scores. A significant negative correlation was found between verbal elaboration and receptive language. None of the joint attention variables correlated with language for this group.

2.7.4. *Regression Analysis - Predictors of Total Language Scores*

The next step was to investigate if any of the variables which significantly correlated with total language scores were significant predictors. A hierarchical regression was run with the total language score as the dependent variable. Age and non-verbal mental age were put into the first model, followed by group, initiating joint attention, responding to joint attention and positive expressed emotion in the second model. Two interaction variables were then computed involving group x responding to joint attention and group x positive expressed emotion (see table 2.4).

Table 2.4: Results from hierarchical regression analysis assessing the association between joint attention, positive expressed emotion and total language scores

Variable	Model 1			Model 2			Model 3		
	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β
Age	.005	.004	.199	-.010	.017	-.377	-.013	.016	-.481
NVMA	.406	.233	.257	.347	.218	.219	.265	.200	.168
Group				4.122	4.881	.524	3.273	5.009	.416
IJA				.075	.042	.243	.057	.038	.184
RJA				.073	.023	.422**	-.095	.066	-.548
PEEM				.035	.146	.033	1.370	.531	1.303*
Group*RJA							.109	.043	1.190*
Group*PEEM							-.756	.296	-1.427*
<i>R</i> ²		.132			.350			.502	
<i>F</i> for <i>R</i> ² change		3.268*			3.268*			5.639**	

Note. NVMA= non-verbal mental age, IJA= initiating joint attention, RJA= responding to joint attention, PEEM= positive expressed emotion, * $p < .05$, ** $p < .01$, *B*= unstandardized Beta, *SE*= standard error, β = standardised Beta

The first model was significant $F(2, 43) = 3.268$, $p = .048$, but neither age or non-verbal mental age were significant predictors. The second model was also significant $F(6, 39) = 3.498$, $p = .007$, and responding to joint attention was a significant predictor $\beta = .422$, $t =$

3.183, $p = .003$. The final model was significant $F(8, 37) = 4.657$, $p = .001$ and positive expressed emotion ($\beta = 1.303$, $t = 2.579$, $p = .014$), group x responding to joint attention ($\beta = 1.190$, $t = 2.563$, $p = .015$) and group x positive expressed emotion ($\beta = -1.427$, $t = -2.557$, $p = .015$) were all significant predictors.

The results from the regression analysis therefore confirm the findings from the correlation analysis and suggest that responding to joint attention is predictive of concurrent language scores for the DS group and positive expressed emotion is predictive of concurrent language skills for the TD group. This is further demonstrated in figures 2.1 and 2.2 below.

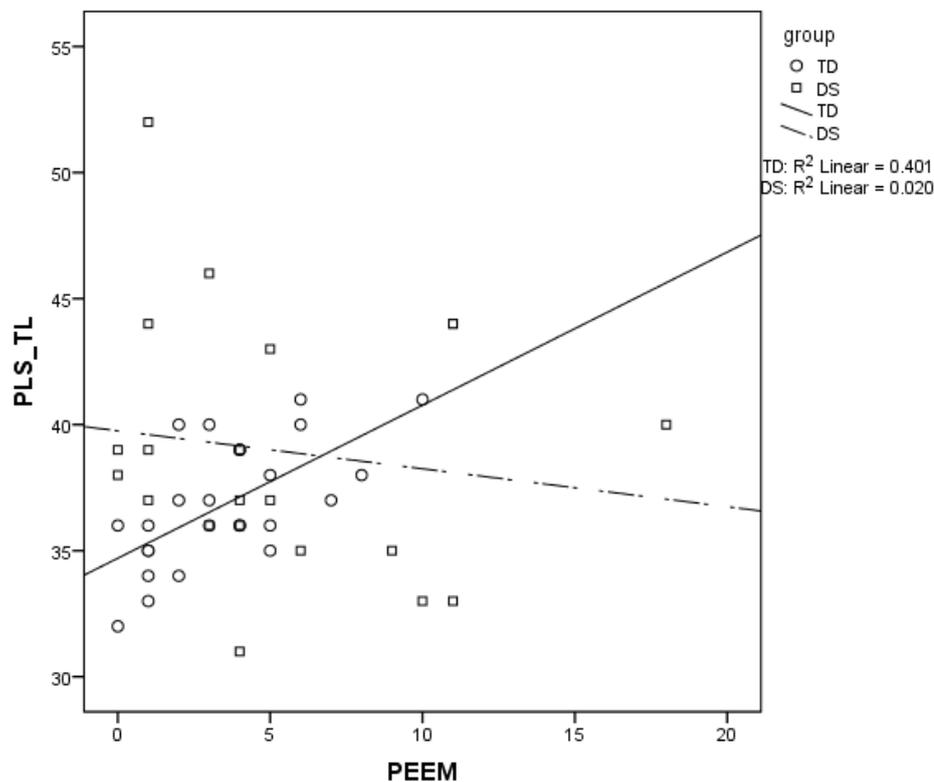


Figure 2.1: Scatterplot of positive expressed emotion and total language scores showing the regression line for group

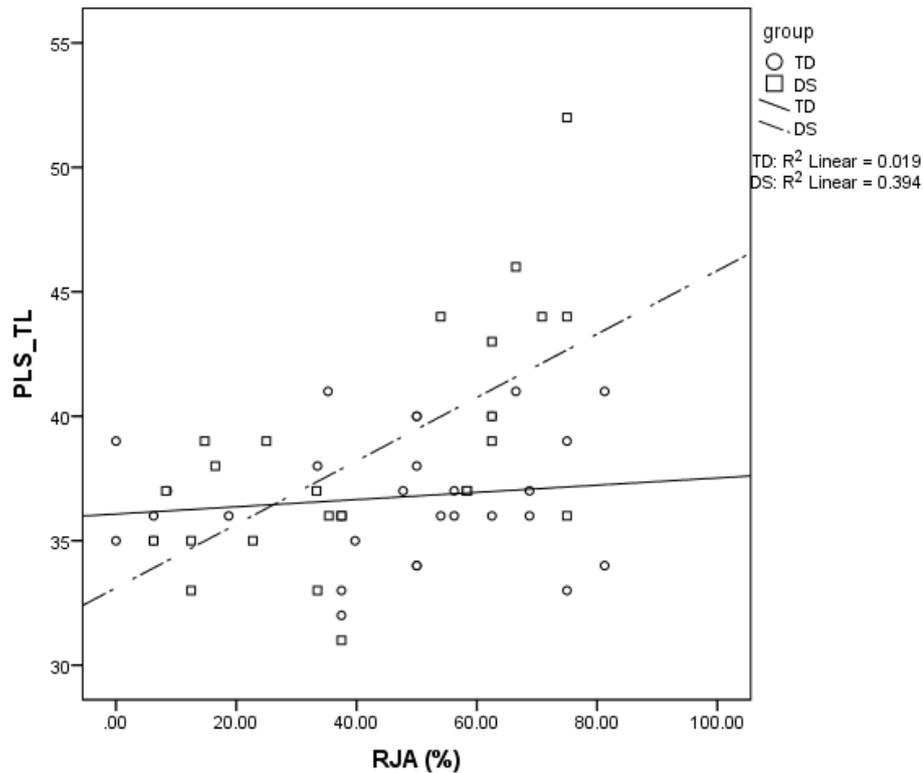


Figure 2.2: Scatterplot of responding to joint attention and total language scores showing the regression line for group

2.8. Discussion

2.8.1. Summary of Results

The results of the study suggest that, at this age (children with DS 17-22 months and TD children 9-11 months) different early social-communication factors seem to be related to language development in the two groups. We found that responding to joint attention was a significant predictor of concurrent language scores accounting for chronological age, non-verbal mental age, initiating joint attention and positive expressed emotion for children with DS aged 17-22 months. On the other hand, maternal positive expressed emotion was a significant predictor of language scores controlling for chronological age, non-verbal mental

age, initiating joint attention and responding to joint attention for TD children aged 9-11 months. These findings will be discussed in relation to previous research.

2.8.2. *Differences Between Groups*

2.8.2.1. *Maternal Measures*

There were no differences between the two groups of parents in terms of how much positive expressed emotion they used or how sensitive they were during interactions with their child. This is different from early research (e.g. Glenn, Dayus, Cunningham & Horgan, 2001; Tannock, 1988) which reports that mothers of children with developmental disabilities such as DS are less sensitive during interactions with their child. However, our results are in line with more recent research (Gilmore et al., 2009) which has also found no differences between mothers of children with DS and mothers of TD children in terms of sensitivity. Furthermore, no differences were found between groups for positive expressed emotion. This is in line with previous research which has identified a ‘Down syndrome advantage’ for parents of children with DS in comparison to other developmental disabilities. For example, Stoneman (2007) reported that parents of children with DS were rated as exhibiting more warmth during interactions with their child in comparison to parents of children with other developmental disabilities, such as fragile X syndrome. The differing results in sensitivity and positive expressed emotion likely reflect changes over time, with more information being available about children with DS, particularly focusing on parent-child strategies in the early years.

However, a significant difference was found for strong verbal control with mothers of children with DS using more strong verbal control utterances during interactions with their child in comparison to mothers of TD children. Previous research identified that using ‘follow prescriptives’ i.e. directive statements concerning the child’s current object of interest

aided later expressive language development for TD children (Akhtar et al., 1991). Therefore, such techniques may have been adopted by the parents of children with DS.

2.8.2.2. *Joint Attention*

No differences were found between the two groups for initiating joint attention or responding to joint attention, or for high and low level behaviours. This in line with previous research from Sigman and Ruskin (1999) who also found no differences between children with DS and TD children using the gaze following task of the ESCS. However, our findings are different from Mundy et al.'s (1995), who also used the gaze following task and found that children with DS performed significantly worse than the TD group. The reason for this difference could be attributed to a number of factors. In the current study both the DS and TD children were sometimes able to follow targets that were placed to their left and right, however only a very small number of children in both groups were able to follow a point to a target that was behind them. The percentages correct for the behind trials were actually higher for the DS group in comparison to the TD group – but this difference was not significant. The ability to follow a point to a target that is outside of a child's visual field usually develops when they are aged between 12 and 18 months. The TD children in our sample were 9-11 months and therefore it is highly likely that this skill had not fully developed yet, or was just emerging. In contrast, a much larger age range was used in the Mundy et al.'s study (12-36 months). This means that some of the children in the Mundy et al. study were considerably older than the children in our sample and would have had more exposure to social interactions. It is therefore also likely that some of the TD children would have fully developed this skill. Another consideration is that our study used two responding to joint attention tasks (proximal points and gaze following) whereas both Sigman and Ruskin (1999) and Mundy et al. (1995) only used the gaze following task.

2.8.3. *Joint Attention and Concurrent Language Scores*

Moderate to strong correlations were found between high level initiating joint attention behaviours and expressive language as well as responding to joint attention and receptive, expressive and total language scores for children with DS. When the results were put into a regression analysis, responding to joint attention emerged as a significant predictor of total language skills for this group. This is in line with previous research which has found that responding to joint attention is related to concurrent language skills for children with DS (Sigman & Ruskin, 1999).

Contrary to our prediction, there were no significant correlations between any aspect of joint attention and concurrent language scores for TD children, despite this being reported previously (Markus et al., 2000; Mundy et al., 2007). This is likely due to the narrow age range included in our study (9-11 months) which meant the children were very young and thus their joint attention skills were still developing. Further evaluation of the data revealed that the TD group were initiating less often than the DS group and performing worse on the gaze following task (although no significant differences were found between groups). Previous research has identified that the ability to initiate joint attention starts developing at approximately 9 months (Bakeman & Adamson, 1984). Therefore, the TD infants in our sample may have still been developing this skill which may explain why there was not a concurrent relationship between initiating joint attention and language for the TD children. On the other hand, as the two groups were matched for non-verbal mental age, the children with DS were considerably older and would have been exposed to more social interaction. Furthermore, the available research in this area demonstrating a link between joint attention and language for TD children has often focused on a longitudinal relationship and not a concurrent one (e.g. Brooks & Meltzoff, 2005; Morales et al., 2000; Mundy et al., 2007), or has assessed older children (e.g. Delgado, Mundy, Crowson, Markus, Yale & Schwartz,

2002, assessed infants aged 15 and 24 months) who would likely have more developed joint attention skills.

2.8.4. *Maternal Measures and Concurrent Language Status*

Moderate and strong correlations were found between positive expressed emotion and receptive, expressive and total language scores for TD children only. When entered into a regression analysis, positive expressed emotion was a significant predictor of total language scores for the TD children when controlling for chronological age, non-verbal mental age, initiating joint attention and responding to joint attention. This supports previous research which has found that maternal warmth is related to concurrent and longitudinal language development (e.g. Steelman et al., 2002).

A significant correlation was not found between sensitivity and concurrent language scores, which goes against previous research where an association between maternal sensitivity and language development for TD children has been found (e.g. Baumwell, Tamis-LeMonda & Bornstein, 1997; Leigh et al., 2011; Tamis-LeMonda, Bornstein & Baumwell, 2001). However, previous research has established a longitudinal relationship and not a concurrent one.

Furthermore, a significant negative correlation was found between verbal elaboration and receptive language for the TD group suggesting that the children with the lowest language skills had mothers who used more verbal elaboration. To gain a high rating of verbal elaboration, both the frequency and complexity of the elaboration had to be high. This links to previous research which has identified that early maternal speech usually consists of short, simple utterances to optimise the chances of the message being understood (Snow, 1995). The TD children in the sample were 9-11 months, therefore it is possible that children whose parents were using a more complex linguistic style was not suited to their current

language level. This may explain the negative correlation seen between maternal verbal elaboration and concurrent receptive language.

None of the maternal measures were associated with concurrent language scores for the DS group. Anecdotally, it was observed that some of the mothers of children with DS tended to intermittently praise their child and this was not always in response to an action or initiation from the child. On the other hand, mothers of TD children seemed to praise their child more often in response to an achievement e.g. rolling the ball. Further analysis of the results shows that, on average, mothers of children with DS used more positive expressed emotion during interactions with their child than mothers of TD children; however this difference was not significant. An explanation for such findings could be that, since the children with DS are not verbalising much, their mothers use more positive expressed emotion to try and motivate their language delayed child. This pattern has been observed previously in research with developmentally delayed children (e.g. Breiner & Forehand, 1982; Kogan & Tyler, 1973).

From a theoretical point of view, the results may support the idea of atypical development by suggesting that different early social communications skills predict language development for TD children and children with DS. Different developmental trajectories have been reported for children with other genetic disorders, e.g. Williams syndrome (Laing et al., 2002) and atypical language profiles have been identified for various disorders including DS, developmental language disorder and ASC (Caselli, Monaco, Trasciani & Vicari, 2008; Charman, Drew & Baird, 2003). On the other hand, a positive correlation was found between non-verbal mental age and language for children with DS suggesting that language develops in-line with general cognitive ability, which may suggest that language abilities are delayed as opposed to following an atypical trajectory (Thomas et al., 2009). Furthermore, although different predictors were found for the two groups, there is evidence from previous research

that responding to joint attention is a significant predictor of language for TD children also (Morales, Mundy and Rojas, 1998; Mundy & Gomes, 1998). We did not replicate this as the TD group was younger and hence had less social experience.

In conclusion, it seems that different early social communication factors are important for early language development for DS children aged 17-22 months and TD children of comparable non-verbal mental age. In particular, early responding to joint attention seems to be important for language development for children with DS which may suggest that future interventions for this group should focus on responding to joint attention. The results from this study suggest that responding to joint attention is not a weakness for children with DS relative to their non-verbal mental age, since no significant differences were found for responding to joint attention between the DS group and the TD group who had equivalent non-verbal mental age. However, one approach to intervention is to target a relative strength that is related to the goal which is the weakness (Capone, 2010). Since responding to joint attention is a precursor which underpins language development, targeting such a skill may have positive cascading effects on language abilities. This is in line with the neuro-constructivist approach which argues that basic level processes should be targeted in intervention to improve developmental progress and outcomes (D'Souza & Karmiloff-Smith, 2017).

Part 2- Literature Review and Intervention Study

Chapter 3 - Intervention Studies

This chapter is organised in to three sections. Sections 3.1 and 3.3 focus on early interventions for infants with DS that have looked to improve speech/language outcomes. Section 3.5 focuses on case studies/case series studies which have used a multiple baseline

design and section 3.2 focuses on studies which used a controlled comparison group. Section 3.3 focuses on interventions that aim to improve responding to joint attention.

Despite the majority of individuals with DS having a speech/language deficit, currently, there are few published treatment studies which focus on early speech/language interventions for children with DS. The available studies focus on primary or secondary school children (e.g. Burgoyne et al., 2012; Camarata, Yoder & Camarata, 2006) suggesting that interventions start primarily once a child has reached a specific age or communication milestone (Kumin, 2002). However, it has been emphasised that intervention should concentrate on targeting and preventing expressive language impairment before it becomes a prominent deficit (Miller, 1999).

3.1. Case Studies/Case Series: Multiple Baseline Design

A number of studies have used a multiple baseline design. Carbone, Sweeney-Kerwin, Attanasio and Kasper (2010) used manual sign training and prompt delay to try to increase the vocal responses of children with ASC and DS. In this study individual results were given for each participant. One participant was a 4 year old with DS and two other participants were aged 4 years and 6 years had ASC. During an initial pre-test assessment, six target objects were chosen for each individual. Intervention sessions occurred twice per day. During each intervention session there were 50 trials where the chosen target items were shown to the child in a random order. If a child gave no response then the next item was shown; if the child showed physical motivation (e.g. signed/reached for item) they were given a prompt to also vocalise. The 'correct response' was a sign and a vocalisation. During the baseline sessions, the participant with DS frequently signed but few vocalisations were observed. However, during the intervention sessions the number of vocalisations increased. The participant gained 7 new vocalisations during the treatment.

The authors reported that some participants did better than others and that those with better initial language made greater gains during treatment. However, it is unclear how many intervention sessions each child had and from the graphs provided it seems the number of intervention sessions varied for each child. To gain further information it would be beneficial to have an additional measure of language – it may also be that receptive language has improved. Furthermore, more information could be gathered to see if the new skills have generalised to a different setting, e.g. at home. Finally, it would be useful to have a further follow up of the participants to see if treatment effects continued or declined after intervention sessions had finished.

Wright et al. (2013) again focused on improving expressive language but this time through a naturalistic sign intervention. Enhanced Milieu Techniques (EMT) and JASPER techniques (joint attention, symbolic play and emotional regulation) were used to teach spoken words and signs to four infants with DS aged 23-29 months. EMT techniques used in the intervention included: following the child's lead during sessions and imitating the child's actions and mapping words. JASPER techniques involved teaching the child new play actions and sequences as well as focusing on joint attention skills such as pointing, giving and showing. A multiple baseline design was used in the study. Each child had 20 play based treatment sessions twice a week which each lasted for 20-30 minutes. The aim of the sessions was to teach participants 32 signs paired with a spoken word. The 32 words were taken from the CDI and were early occurring words. The intervention sessions were delivered by therapists in a clinic setting. The paired signs and words were modelled by the therapist using EMT and JASPER strategies. Intervention sessions were recorded and coded for signs/vocalisations used as well as joint engagement. A parent child interaction was also filmed in the child's home to see if skills had generalised.

All participants increased their use of spontaneous signing and the number of expressive words after the intervention ranged from 3-9 words. All participants generalised the skills and used more signs at home. However, only minor changes were found for expressive vocabulary outside of the clinical setting. Gains in joint attention was also seen for three of the participants. However, joint attention data was only collected for half of the intervention sessions and only one child showed a significant improvement. A drawback of the study is that the authors didn't distinguish between initiating and responding to joint attention when measuring outcomes.

A further study used a naturalistic approach to try to improve expressive vocabulary (Vilaseca & Del Rio, 2004). Three Spanish children with DS participated in the intervention (aged 4;6 – 5;11 years). The study used an ABA design. For the first four months of the study, participants were videotaped at home for 20 minutes once per month. Intervention sessions were then carried out for the next 4 months and observations took place once per month. For the final four months intervention sessions were withdrawn and participants had 4 observation sessions.

The intervention was researcher led and took place in the child's school. Sessions lasted for 35-45 minutes and took place 3 times a week over the 4 months. Intervention sessions were carried out within natural play routines e.g. drawing or looking at book. Intervention strategies focused on verbal expansions, imitating and informative-corrective feedback, e.g. each child's utterance was expanded by the researcher to include a verb. For some children the rate of utterances with a verb increased as did morpho-syntactic complexity. However, for some children the rate of these skills decreased and didn't generalise to home. The authors reported that those with higher cognitive skills saw better gains from the intervention.

3.2. *Summary*

In summary, various methods have been used in multiple baseline studies to improve speech/language outcomes including: sign training, ABA and more naturalistic approaches. The number of intervention sessions varied, and the exact number was not always reported (e.g. Carbone et al., 2010). In some studies intervention sessions occurred twice a day (Carbone et al., 2010), whereas in another study sessions took place twice a week (Wright et al., 2013).

Varying results were reported in the studies: Carbone et al. (2010) reported that the participant with DS increased their use of vocalisations with signs and learnt 7 new vocalisations during the course of the intervention. Wright et al. (2013) found that all participants used more spontaneous signs and their expressive vocabularies increased. However, these gains didn't generalise to other settings, e.g. home. Finally, Vilaseca & Del Rio (2004) reported that gains in expressive language were seen for participants with higher cognitive ability but that for those with lower cognitive ability a decrease in language was seen.

3.3. *Intervention Studies Using a Control Comparison Group*

3.3.1. *Early Stimulation Programme*

A group of studies from Spain all cite using an 'early stimulation' method which included the following materials: Intervention programme for DS children (Hanson, 1979), the Early stimulation programme (UNICEF, 1982) and the Bayley Child Development Scale (Bayley, 1959). However, very little information about the intervention strategies is provided and it is not clear how these materials were used or combined. In one of the selected studies (Sanz, Menendez & Rosique, 2010) slightly more information is given. The programme is described as being individualised and designed to 'remedy' developmental delay and provide

maximum stimulation at a critical time during development. It aims to target four main areas of development – language, gross motor, fine motor and social development skills. The stimulation is designed to begin shortly after birth and to continue until the child reaches 24 months. Parents are trained as part of the process, and treatment sessions are observed so that parents can use the techniques in their own home (Sanz et al., 2010). The group have used variations of the intervention – comparing how they praise children, how they provide instructions to parents and the child's age at the beginning of the intervention. In all studies, language is measured using the subscale from the Brunet-Lezine's First Childhood Scale (Brunet & Lezine, 1976).

Sanz (1996) and Aparicio (1989) both compared the effectiveness of the programme when parents were either provided with vicarious (parents learning intervention techniques through observing the researcher) or written instructions. Children were randomly assigned to either group. In both studies children received direct training from a clinician twice a week for 1 hour and their mothers practised on the remaining days for 1 hour. In the Sanz (1996) study children were aged 0-2 months at the start of the study. The results showed that children whose parents received vicarious training had better language outcomes. As well as comparing the two parental training techniques, Aparicio (1989) also compared the age at which children started the intervention (ranging from newly-born to 18 months). Similarly, those whose parents received vicarious training had better language outcomes and this was seen regardless of when children started the intervention.

Aparicio and Balana (2002) included children with DS from birth to seven months. Each child was given an individualised language stimulation programme, however the time, length and any further details about the intervention itself are not provided. The researchers concluded that those in the youngest age group (birth to one month) obtained the best results at 24 months; gains were measured by increases in developmental language quotients.

However, a control group was not included in the study. Also, those who started the intervention earlier would have received considerably more intervention sessions since children stayed in the intervention until 24 months of age regardless of when they started. It is therefore difficult to conclude whether it was the 'early' start of the intervention, or the sheer number of sessions which led to better language gains for the younger group.

Finally, Sanz et al. (2010) compared using verbal (e.g. saying 'well done') versus physical (e.g. clapping) feedback with the children participating in the intervention. Twenty children with DS aged 5-8 months participated in the intervention. Parents were given training and observed sessions so that they were able to provide the appropriate feedback also. The intervention lasted for 10 months. No significant differences were found for language outcomes between the two groups.

Despite numerous studies being published using these techniques, the actual intervention is not clearly described, and it is difficult to understand what specific techniques and targets were used. Furthermore, as the studies only compare variations of the same intervention and do not use a no-treatment control group the results have to be interpreted with caution.

3.3.2. *Published Scheme/Clinical Methods*

Some intervention studies have been based on published programmes and clinical methods. Andrade and Limongi (2007) used an intervention based on the dialectic-didactic method which aims to build a child's knowledge through problem situations. Eight participants with DS (aged 33-52 months) received 40 intervention sessions once per week by the researcher. Participants were assigned to either the treatment group or a control group. The aim of the intervention was to improve the child's expressive language. At the start of the session the researcher and child would partake in spontaneous play. The researcher would

then engineer a problem-situation. After the intervention, no significant differences were found for oral language or total number of words used for the intervention and control group. This was scored by coding a transcription from the testing session. However, there are methodological issues which may have compromised the results. Firstly, a very small sample size was used and there were only 4 participants in each group. Secondly, the specific intervention targets and procedure were not clear from the information provided. Thirdly, participants in the control group were receiving early speech-language stimulation outside of the intervention and it was stated that this intervention had a different theoretical/methodological approach. However, no further information was given with regards to the focus, duration or intensity. It is therefore difficult to determine if the alternative intervention could have also affected expressive vocabulary.

Another intervention (Weller & Mahoney, 1983) trained children using the Environmental Language Intervention programme (MacDonald & Nickols, 1978). Fifteen children with DS (aged 18-36 months) were assigned to either an oral language group or a total language group (incorporating both speech and sign). Specific words were chosen to reflect the specific interests of each individual. Mothers were taught to use techniques at home daily including imitation, conversation training and structured play. However, after the intervention no significant differences were found between the two treatment groups for expressive or receptive language. Again, a relatively small sample size was used (7 children in the oral language group and 8 in the total language group). The authors concluded that there were no differences in intervention effects between the two modalities. However, without the inclusion of a no treatment control group it is not possible to conclude which treatment modality yields better effects or if either treatment modality is preferable to no treatment.

Two studies used the Hanen programme (Manolson, 1992) as their intervention tool. Both used a randomised control wait-list design. In the first study (Girolametto, 1988), parents were trained to use the programme in 8 group sessions and also received 3 home visits. Eleven children with DS (aged 15-62 months) participated in the study. After the intervention there were no significant differences between the two groups for receptive or expressive language. However, analysis of a mother-child interaction revealed that, during interactions, children in the intervention group used significantly more examples of turn taking and a more diverse vocabulary. This suggests that the intervention did have some positive effects on participants' communication skills, however this did not generalise to standardised measures of receptive and expressive language.

In the second study carried out 10 years later (Girolametto, Weitzman & Clements-Baartman, 1998), which included 12 children with DS (aged 29-46 months), parents were trained to use the programme in 9 group sessions and 4 individual home visits. After the intervention it was found that the intervention group used significantly more words during a parent-child interaction and as measured by parental report. There were no differences between groups for the use of target words during a semi-structured task with a different conversational partner. Although during this interaction the intervention group used more words overall than the control group. The results from these two studies which used the Hanen programme may suggest that although a positive impact on communication skills was found this did not generalise to standardised language assessments in either study.

3.3.3. *Responsive Teaching/Communication Training*

In recent years, early intervention studies focusing on improving communication skills of children with additional needs have used responsive teaching/education and milieu communication teaching techniques. Responsive teaching interventions focus on parent-child relationships and look to encourage parents who have children with learning disabilities to

engage in highly responsive interactions with the child to facilitate the development and social emotional functioning of the child (Mahoney, Robinson & Powell, 1992). Milieu communication training targets the child's pre-linguistic and/or communication skills (Yoder, Woynaroski, Fey & Warren, 2014).

Karaaslan and Mahoney (2013) focused on responsive teaching. They undertook a randomized control trial in Turkey with a group of 15 children with DS aged between 2-6 years of age and their mothers. They replicated a previous study which used the same responsive teaching strategies but with a group of children with a range of developmental disabilities (Karaaslan, Diken & Mahoney, 2013). All participants received standard classroom special education services and those in the treatment group received individualised parent-child sessions once per week for 1.5-2 hours over 6 months. During these sessions a researcher trained in administering interventions worked with the mothers to help them use responsive teaching techniques to promote their child's use of their individual pivotal behaviour objectives.

Developmental assessments and observations took place at the beginning of the intervention and two months after it had finished. Assessments included the Turkish versions of: the Denver Developmental Test (Anlar & Yalaz, 1996), Ankara Developmental Screening Inventory (ADSI; Savasir, Sezgin & Erol, 2005), Maternal Behaviour Rating Scale (Mahoney, 1999) and the Child Behaviour Rating Scale (Mahoney & Wheeden, 1998).

There were significant differences between attention and initiation with children in the intervention group scoring higher than those in the control group. These changes positively correlated with changes in their mother's responsiveness. Furthermore, children in the intervention group scored significantly higher on subscales of language in both the Denver Developmental Test and the ADSI.

A further two studies used a combination of responsive education and pre-linguistic milieu teaching. In the first study 17 children with DS (median age= 22 months, a range is not provided) were included in the study as part of a mixed aetiology group (Yoder & Warren, 2002). Children in the intervention group received 3-4 sessions a week of pre-linguistic milieu teaching for 6 months and parents had 12 sessions of responsive education. After the intervention, it was found that children in the intervention group actually had decelerated growth in requesting and commenting compared to the control group, suggesting that participating in the intervention had actually made their performance worse. This highlights the importance of having interventions which are specifically aimed at children with DS.

The second study used a combination of responsive education and pre-linguistic milieu teaching and included 26 children with DS (aged 24-33 months) as part of a mixed group (Fey et al. 2006). Children in the intervention group had 4 weekly sessions of pre-linguistic milieu teaching and parents had 8 sessions of responsive education. Children remained in the intervention for 6 months. After the intervention, no significant differences were found between the control group and the intervention group for receptive or expressive language.

The final study focused only on milieu communication training and investigated if there would be a difference in outcomes for children receiving higher or lower doses of treatment (Yoder et al., 2014). Thirty-five children with DS (aged 18-27 months) were included in the study as part of a mixed group. The intervention lasted for 9 months. During this time, one group received 5 hours per week of milieu communication training and the other group received just 1 hour per week. After the intervention, it was found that children who had received the higher dose (5 hours per week) produced significantly more words (measured by the CDI) than those in the low dose group. However, as there was not a no

treatment control group, the study does not show whether milieu communication training is an effective intervention for the group who took part in the intervention.

3.4. *Summary*

The reported literature demonstrates that there are few existing early intervention studies that focus on improving speech and language skills for pre-school children with DS. A range of techniques have been used including published protocols such as the Hanen programme, the Environmental Language Intervention programme and the dialectic-didactic method. Further intervention strategies include responsive teaching, milieu communication training, improving gestures and signs, early stimulation and behaviour modification. Some of the interventions targeted both parents and children whereas others targeted parents and children.

The majority of reported studies primarily focus on improving pre-linguistic skills (e.g. gestures, imitation) or encouraging vocalisations. The methodology of intervention was not always clearly reported (e.g. Aparicio, 1989; Aparicio, 2002; Sanz, 1996; Sanz et al., 2010), which makes it difficult to interpret the results and also causes issues for replication. Primary outcome measures included speech/language as measured by standardised language assessments, parental questionnaires and/or observation and transcription of speech. Intervention duration ranged from 11 weeks to two years and the starting age of participants ranged from birth to 72 months.

A range of results were reported, with some studies reporting gains in early social communications and/or language, whereas others found no significant gains for the intervention group. One study (Yoder & Warren, 2002) found that those in the intervention group had significantly worse outcomes than children in the control group. In this study children with DS were part of a mixed aetiology group but this pattern was seen specifically

for children with DS. This finding highlights the importance of having interventions which are specifically tailored for individuals with DS.

In conclusion, the research suggests that early communication interventions can be beneficial for infants with DS. However, based on the available research in the area an 'optimum' intervention cannot be highlighted.

3.5. Training Joint Attention

Previous research has identified that responding to joint attention is related to language scores for children with DS (Mason-Apps, 2013; Sigman & Ruskin, 1999). Therefore, this section will review existing interventions which target joint attention. To our knowledge, there are no published studies which focus on improving joint attention for children with DS. Although a deficit in responding to joint attention relative to non-verbal mental age, is not evident for individuals with DS, targeting the lower level skills underpinning language may have a positive impact on language scores, in line with the neuro-constructivist approach (Karmiloff-Smith, 2009). The studies included in the review focus on improving joint attention for children with ASC. Joint attention is a known and major deficit for individuals with ASC (Charman, Swettenham, Baron-Cohen, Cox, Baird & Drew, 1997; Mundy, Sigman & Kasari, 1990) and so there have been many intervention studies which have attempted to address it.

3.5.1. Improving Joint Attention – Multiple Baseline Design

Jones, Carr and Feeley (2006) used discrete trial training and pivotal response training techniques to attempt to improve responding to joint attention and initiating joint attention in children with ASC. Participants included 5 children with ASC aged 2-3 years. A multiple baseline design was used. To be included in the study children had to be able to show that they were capable of basic responding to joint attention skills, e.g. making eye contact when

their name was called, the instruction 'look' or when a primary re-inforcer was held in front of the researcher's eyes. Teachers were trained to administer the intervention. To achieve a correct response the child had to independently, without prompts, alternate their gaze between the object and the adult within 2 seconds of the adult's bid. During each intervention session the child had 10 opportunities to respond. To 'master the skill' children had to achieve 80% correct independently across two consecutive sessions administered by two different teachers over two days of intervention.

For each responding to joint attention bid the researcher placed a toy less than 1.5 metres away from the child, activated the toy and then alternated their gaze between the child and toy whilst pointing and commenting. The child was given two seconds to engage with the toy otherwise a prompt was used. Prompts included calling the child's name or using a re-inforcer to trace the path from the toy to the teacher's eyes and back to the toy. Over time prompts were faded using a most to least prompting procedure and a time delay.

All participants were able to master responding to joint attention within 19-78 sessions ($M= 39$ sessions). After the participants had mastered both responding to and initiating joint attention, each participant showed some ability when exposed to novel stimuli (range 11-100%, $M= 89\%$) and maintenance with original toys (range 20-100%, $M= 91\%$). This study does not provide any further assessments so it cannot be concluded that effects were maintained over time. Furthermore, there is large variability in the results for both novel and original stimuli suggesting that the intervention was not successful for all children. Although a multiple baseline procedure was used future research could include larger sample sizes and a no treatment control group.

A further attempt to train both responding to and initiating joint attention was made by Whalen and Schreibman (2003). They used a behavioural intervention which used

components of discrete trial training and pivotal response training. The participants consisted of 5 children with ASC aged between 4 and 4;4 years. A multiple baseline design was used.

The response training was divided into six levels and participants had to master one level to proceed to the next, to 'master' a level participants had to get 80% correct of 4/5 consecutive sessions. For the different levels the child was required to redirect their attention from a toy they were already playing with. For level 1 the child's hand was placed on a toy, if the child engaged or looked at a toy for 5 seconds this was scored as a correct response. If the child did not respond then all toys were removed for 5 seconds. If the child had two incorrect responses in a row a physical prompt was applied to keep the child's hand on the object for 5 seconds and the response was scored as a prompted response. The same protocol was used for levels 2 and 3.

Level 4 focussed on establishing eye contact which was trained using standard pivotal response training procedures, e.g. the child had to make eye contact with the researcher to gain access to the re-inforcer. For level 5 the researcher established eye contact with the participant and then turned their head and pointed at another toy in the room, the child had to turn their head in the same direction as the experimenter. If they did this correctly they were allowed to play with the toy. Level 6 followed the same format except the experimenter shifted their gaze only and did not point.

The results of the post-test found that the intervention was effective in teaching responding to joint attention to all participants. From pre to post test positive changes were observed for responding to showing, following a point and gaze following. Response training took 18, 23, 16 and 22 days for participants. To assess the generalisation an unstructured assessment was used and found that, at post training, all children showed gains in responding to joint attention. At the follow up assessment 3 out of 4 participants maintained their ability

to respond correctly to responding to joint attention above baseline levels, although a decline from post-test was seen. One participant decreased to baseline levels between the post-test and follow up assessment.

A further extension of this study assessed ‘non-targeted’ behaviours (Whalen, Schreibman & Ingersoll, 2006) and found at the post-treatment assessment all 4 participants showed significant improvements in spontaneous speech, which declined slightly by the follow up assessment. This, therefore, suggests that improving responding to joint attention and initiating joint attention had effects on other areas of development. However, a limitation of this study is that the intervention effects declined once sessions had finished, and the small sample size.

3.5.2. *Improving Joint Attention – Control Group Design*

A further study which looked at improving joint attention and symbolic play in children with ASC used a randomised control study design (Kasari, Freeman & Paparella, 2006). Fifty-eight children (aged 3-4 years) with ASC were assigned to one of three groups - a joint attention intervention group, a symbolic play intervention group or a no treatment group control group. The intervention lasted for around 5-6 weeks and experimenters worked with each child individually daily for 30 minutes. The intervention approach used applied behaviour analysis. Initially each child had approximately 5-8 minutes of discrete trial training to ‘prime’ their treatment goal. Then a table activity took place for around 5 minutes, the experimenter sat opposite the child and modelled the targeted joint attention/symbolic play target skill whilst engaging the child in social interaction. This activity was primarily adult directed.

The same activity then took place on the floor for around 20 minutes; this was less structured than the table activity. The techniques used for this part of the intervention were

similar to milieu teaching: following the child's lead and interests in stimuli/activity and commenting on what the child was doing. Direct instruction was immediately followed by naturalistic milieu instruction to enhance generalisation. Two further strategies included were: to imitate the child's actions on toys and using the child's activity interests to develop play routines. Goals were considered 'mastered' once a child showed the goal at least 3 different times at both the table and floor interactions.

The results from the Early Social Communication Scale (ESCS) showed that the joint attention group and the symbolic play group made significant improvement in initiating shows in comparison to the control group. There were no significant differences for shows between the joint attention and symbolic play group. All three groups (joint attention, symbolic play and control group) made significant gains in coordinated joint looks and there were no significant differences between groups for initiating gives or points. The joint attention group did significantly better in terms of responding to joint attention than the symbolic play or control group.

To see if the intervention effects had generalised, the researchers also assessed joint attention during a parent child interaction. The joint attention group used significantly more gives and shows. The joint attention and symbolic play group used significantly more coordinated joint looks. In summary, the results suggest that the intervention was beneficial in improving joint attention skills. However, some gains were seen in both the joint attention and symbolic play groups. It is therefore difficult to tease apart if the relative gains were seen due to the targeted joint attention intervention or because children were receiving some form of daily input. On the other hand, the gains seen for responding to joint attention were specific to the joint attention group – suggesting that responding to joint attention was improved by the joint attention intervention.

3.5.3. *Parent Administered Intervention*

A number of studies have used parents as agents of intervention. One such study is by Rocha, Schreibman and Stahmer (2007). The study included three children with ASC aged between 26-42 months and two TD children acted as a control group matched for developmental age. Data from a previous study acted as a chronological age match.

To ensure motivating toys were picked for the intervention, researchers used an informal preference assessment adapted from DeLeon and Iwata (1996). The training levels were taken from Whalen and Schreibman (2003). During training parents were asked to provide a joint attention bid at least once every 2 minutes. To assess generalisation of the joint attention skills 10-minute parent child interactions were filmed in the participants' homes once per week.

All the parents increased their use of joint attention bids and initiated more joint attention bids during training than they had at baseline. Positive results in all the child participants were reported during training and at generalisation sessions. Participants responded to a greater proportion of bids during later phases of training and at post-test in comparison to baseline. Two out of three children still responded to a greater number of bids at the follow up session. This suggests that training parents may help to maintain intervention effects.

3.6. *Summary*

Numerous intervention studies have looked to improve both initiating joint attention and responding to joint attention for children with ASC since it is a known deficit for this clinical group. To our knowledge there have been no previous intervention studies which have attempted to improve responding to joint attention for children with DS. Results from various systematic reviews suggest that interventions can improve responding to joint

attention for children with ASC (Lang, Machalicek, Rispoli & Regester, 2009; Meindl & Cannella-Malone, 2011; White et al., 2011). The majority of studies seem to use behavioural techniques, e.g. applied behaviour analysis, discrete trial training and pivotal response training whilst prompting the child and providing positive reinforcement. Parents/teachers have been successfully trained to administer interventions. One key issue for the reported studies is that children started to decline once the programme had stopped. In the first study (Whalen & Schreibman, 2006) parents were not trained so it was unlikely they would carry on the techniques; in the second study, although the parents were trained, the intervention sessions always took place in a structured clinical setting. Future research should consider combining interventions sessions in the clinic by a researcher with sessions at home by the parent. This may encourage generalisation and may mean that intervention effects do not decline once sessions have stopped.

3.7. The Current Study (Intervention Study) – Rationale

There is evidence that responding to joint attention is an important precursor for concurrent and longitudinal language development for children with DS (Harris et al., 1999; Mason-Apps, 2013; Mundy et al., 1995). Our language precursor study found further support for this in that responding to joint attention was a significant predictor of concurrent receptive and expressive language skills in a model accounting for age, non-verbal mental age, group, initiating joint attention and positive expressed emotion in a group of children with DS aged 17-22 months old. Since to our knowledge no previous intervention study has focused on improving responding to joint attention for children with DS, the feasibility of such a study needs to be considered.

The literature review of previous interventions targeting responding to joint attention suggests that many attempts have been made to improve the responding to joint attention skills of young children with ASC (Jasper et al., 2006; Jones et al., 2006; Whalen &

Schreibman, 2006). Such interventions have used behavioural techniques such as providing prompts and rewards and modelling behaviour. Applying such an intervention for children with DS may benefit their later speech and language outcomes and would therefore have implications for theories and practice. By attempting to improve responding to joint attention with the view of having a positive effect on later speech and language is in line with the neuro-constructivist theory (e.g. Karmiloff-Smith, 2009). Improving a precursor to language may lead to a positive cascading effect on language development. In our proposed study we intend to have both the researcher and parent delivering sessions concurrently. The researcher will administer sessions in a clinic setting and parents will deliver sessions at home. Researchers and parents will both be using the same activities and the aim is to encourage generalisation and long-lasting effects.

3.7.1. *Aims*

The aim of this study is to trial an intervention targeting responding to joint attention for children with DS. The aims are: 1) to find out if this type of study is feasible to be carried out with young children with DS; 2) to find out whether responding to joint attention can be improved; and 3) to find out whether targeting responding to joint attention may have a cascading effect on later language outcomes?

3.7.2. *Research Questions*

- Can responding to joint attention be improved for children with DS using a 10 week intervention?
- Will children with DS who have received the intervention between 17 and 23 months have better language outcomes 6 and 12 months later compared to children with DS who did not receive the intervention?

3.7.3. *Outline*

The intervention study is based on the methodology used by Whalen and Schreibman (2003). Due to constraints on time and resources needed to run the study with two groups (intervention and control), pre-existing data from Mason-Apps (2013) were used as the comparison control group. This group consisted of 14 children with DS (aged 18-21 months) who were assessed at three time points (18-21 months, 24-29 months and 30-35 months). A similar size group of children with DS was recruited for the intervention group and these children were also assessed at these three time points. The intervention group received a 10 week intervention focusing on improving responding to joint attention as well as an immediate post-test assessment. The two groups will then be compared on measures of non-verbal mental age, joint attention, language and vocabulary.

Chapter 4 - Methodology

4.1. *Ethics, Recruitment and Consent*

The study was given ethical approval from the University of Reading Research Ethics Committee. Participants were recruited in a number of different ways. Initially, parents from a language support group held at the University were sent a recruitment poster and information sheet asking if they would be interested in taking part. They were given the opportunity to discuss the project with the researcher and were then required to sign a consent form if they wished to take part (See appendix A).

Next local charities were emailed and sent the recruitment poster and information sheet and were asked to circulate this information to members of their group in the form of a flyer which contained contact details of the researchers. Any parents who were interested were able to get in touch with the researchers directly to express their interest and to discuss any queries. If they agreed to take part, they were given consent forms to complete. An

advertisement was also placed on the Down Syndrome Association's website which gave details of the study, a link to the information sheet and the contact details of the researchers.

As participants were aged 17-23 months and had a learning disability, their parents were required to give consent on their behalf. It was made clear to all parents, verbally and in an information sheet, that they did not have to take part and that they were free to withdraw from the study at any time, and that withdrawing from the study would not affect their future involvement in projects or educational provision/speech and language therapy. However, if a child got distressed or visibly upset during a session, this was taken as the child refusing to consent and testing/intervention sessions were postponed or stopped.

All hard copies of data were stored in a locked cabinet in a locked room at the University. Electronic data was stored on a secure computer at the University and the file was password protected. The data were anonymised so that personal details could not be linked to test results.

4.2. Participants

Participants in the intervention group were 16 children with Down syndrome (DS) aged 17-23 months at the start of the intervention (males= 9, females= 7). One child was being brought up in an English/German household but the dominant language was English. Three children were born prematurely and one had an additional diagnosis of West syndrome which results in infantile spasms and is the most common cause of epilepsy in children with DS (Hamouda et al., 2014). Although 11 parents reported that they were concerned about their child's hearing, none of them reported their child to have a history of ear infections. Ten of the children were receiving input from Portage workers and all children had received some sort of support from speech and language services but none were receiving weekly targeted intervention, or an intervention that targeted responding to joint attention.

The control group came from Mason-Apps (2013) and consisted of 14 children with Down syndrome (age range 18-21 months, 5 girls and 9 boys). Three of the children were being exposed to languages other than English. These children were assessed at 3 time points (18-21 months, 24-29 months, 30-35 months) using the battery of tests described in the materials section. During this time, these children did not receive any specific intervention targeting responding to joint attention. No significant differences were found between the two groups at the start of the study in terms of chronological age: $t(28) = .367, p = .716$. Further differences between groups are described in the results section.

4.2.1. *Demographics*

A demographic questionnaire was administered to all parents to check for any socio-economic differences between the intervention group and the control group. The same demographic questionnaire was used for the control group apart from the questions regarding support services which were added in for this project. The questionnaire asked various questions regarding the child including information about additional diagnoses, hearing/vision concerns and what support services the child was accessing. Further questions asked if the child had siblings and the education level and employment status of the parents. A copy of the questionnaire can be found in Appendix C.

The demographic questionnaire revealed that 12.5% of mothers in the intervention group had been educated to GCSE level, a further 12.5% had an NVQ or equivalent, 18.75% had A-levels, 25% had a degree, 25% had a post-graduate diploma/degree and 6.25% reported no formal qualifications. In comparison, 8.3% of mothers in the control group had GCSE's, 25% had a degree, 58.3% had a post-graduate diploma/degree and 8.3% had a PhD.

For paternal education, 13.3% of fathers in the intervention group had GCSE's, 26.7% had an NVQ or equivalent, 20% had A-levels and 40% had a degree. In comparison, 8.3% of

fathers in the control group had A-levels, 25% had a degree, 33.3% had a post-graduate diploma/degree, 25% had a PhD and 8.3% reported no formal qualifications.

In terms of occupation status, 68.75% of mothers in the intervention group were not currently working, 12.5% were self-employed, 12.5% were employed part-time and the remaining 6.25% were employed full-time. In comparison, 46% of the control group mothers were not currently working, 7.7% were self-employed, 30.77% were working part-time and 15.39% were working full-time. All fathers in both the intervention group and the control group were employed. Thirty-three percent of fathers in the intervention group were self-employed and the remaining 66.6% worked full-time, whereas 8.3% of the control group fathers were self-employed and 91.6% were working full-time.

For the sake of analysis, the data was dichotomised. Fischer's exact tests showed that there were no significant differences between the intervention group and the control group for gender ($p = .722$), history of ear infections ($p = .448$), additional languages ($p = .315$), maternal education ($p = .606$), maternal occupation ($p = .274$) or paternal education ($p = .586$). This could not be computed for paternal occupation since for both groups all fathers were currently employed (see table 4.1).

Table 4.1: Demographic variables for intervention and control group

Demographic variables	Levels	Frequency	
		IG	CG
Gender	Male	9	9
	Female	7	5
Hearing	Yes	0	1
	No	16	12
	Missing	0	1
Language	Yes	1	3
	No	15	11
Maternal Ed	Compulsory	3	1
	Further	13	12
Maternal Occ	Employed	5	7
	Unemployed	11	6
Paternal Ed	Compulsory	2	1
	Further	13	11
Paternal Occ	Employed	15	12
	Unemployed	0	0

Note. IG = intervention group, CG = control group, Ed= education, Occ= occupation

4.3. Pilot Test

The procedure was initially piloted on a 21-month-old female infant with DS to check for any issues with the timing and methodology of the intervention. This resulted in the levels of the intervention being extended from five to seven to allow for different types of

‘following a point’ to be included and trained. In the original intervention, we only focused on pointing in a book and pointing to the side and behind the child. After the pilot, we also included a level where the researcher would point to one of two toys in front of the child (the levels will be explained in more detail in a later section 4.6.1). The number of opportunities within a trial was also reduced from five to three to reduce the time of the intervention sessions and avoid child fatigue.

4.4. *Pre-Test*

Prior to the intervention starting, pre-test assessments were administered. These included measures of language, early social communication skills including joint attention, non-verbal mental age, parental responsiveness and maternal well-being. Parents and their children were asked to visit the University of Reading for this purpose, or if they were unable to travel, the researcher completed the testing in the participant’s home, and this was the case for two children. If they had not already done so, parents were required to complete a consent form before any testing took place. The pre-test assessments took place in the Speech and Language Therapy clinic or in an infant friendly lab at the University of Reading. The room was set up by the researcher and contained a table with one chair for the researcher and the other for the parent and child. In all sessions the child sat on the parent’s lap opposite the researcher. We made sure that the child’s face could be seen by the cameras as some parts of the session were video recorded for data analysis. The session took approximately an hour and a half to two hours including breaks.

4.5. *Measures*

The below diagram (figure 4.1) demonstrates which measures were used at which time point. These will be described in more detail in the following sections.

	MSEL	ESCS	PLS-4	CDI
Pre test	✓	✓	✓	✓
Post test	✓	✓	✓	✓
Follow up one	✓	✓	✓	✓
Follow up two	✓		✓	✓

Note. MSEL= Mullen's Scale of Early Learning, ESCS= Early Social Communication Scale, PLS-4= Preschool Language Scales, CDI= Communicative Development Inventory

Figure 4.1: Diagram to show which measures are used at which time point

4.5.1. *Language and Vocabulary*

4.5.1.1. *Pre-School Language Scales-4 (PLS-4)*

The first language measures used were the receptive and expressive sections of the Preschool Language Scales 4 (PLS; Zimmerman, Steiner & Pond, 2002). The PLS is used to measure a child's understanding (receptive) of language and their speech (expressive language) and is used for both clinical and research purposes. The measure is suitable from birth to 6;11 years and the manual provides norms and percentile ranks. Both sections took approximately 20 minutes to administer. The measure was coded using the specified coding scheme provided by the manual. This derived raw scores for receptive and expressive language which can be combined to give a total language score. The test was standardised

using a sample of 1,564 children and sensitivity and specificity for both the subscales and combined score range from .77 to .92.

4.5.1.2. *Reading Communicative Development Inventory (CDI)*

The second language measure used was the Reading CDI (Hamilton, Plunkett, & Schafer, 2000) which is a parental questionnaire which focuses on the child's vocabulary. Similarly, the questionnaire is designed to test what the child understands and what the child says, and parents are able to indicate this using a checklist format. We slightly adapted it so that parents also informed us as to what signs their child understood and used also. The questionnaire is suitable for 16-30-month-old infants. To score the questionnaire the checklists are counted up so that a numerical score is given for a child's receptive vocabulary, expressive vocabulary and use of signs.

4.5.2. *Non-Verbal Mental Age*

The Mullen Scale of Early Learning (MSEL; Mullen, 1995) was used to measure cognitive ability and motor development. The measure is suitable for use with children from birth to 68 months. For the purpose of this study three of the five scales were administered: fine motor, gross motor and visual reception as these were the three scales used with the control group. The subscales were scored using the specified coding scheme to generate three raw scores. The visual reception and fine motor subscales were combined to obtain a score for non-verbal mental age. The concurrent validity of the measure has been tested using the Bayley Scales of Infant Development (Bayley, 1969) and the PLS.

4.5.3. *Joint Attention: Early Social Communication Scales*

To assess non-verbal communication skills, we used the abridged version of the Early Social Communication Scales (ESCS; Mundy, Hogan & Doehring, 1996). To match the control group, only tasks relating to initiating joint attention, responding to joint attention and

initiating behavioural requests were used. The assessment was video recorded as it was coded by the researcher after the session. For the ESCS, the researcher sat opposite the child at a table and the child sat on their parent's lap. Four posters were positioned around the room: one to the child's left, one to the right, and one behind the child to the left and one behind the child to the right (see figure 4.1).

Cameras were positioned so that the full face of the infant could be visible as well as the profile of the researcher. During administration the researcher would respond naturally to the child but minimised their verbal interaction. They also made sure to reinforce effort rather than success. The procedure took around 10-15 minutes to administer. As advised by the manual tasks took place in the following order: 1) mechanical object spectacle, 2) hand operated object spectacle, 3) pointing trials, 4) book task, 5) hand operated object spectacle task, 6) mechanical object spectacle, 7) mechanical object spectacle, 8) pointing trials, 9) plastic jar and 10) hand operated object spectacle.

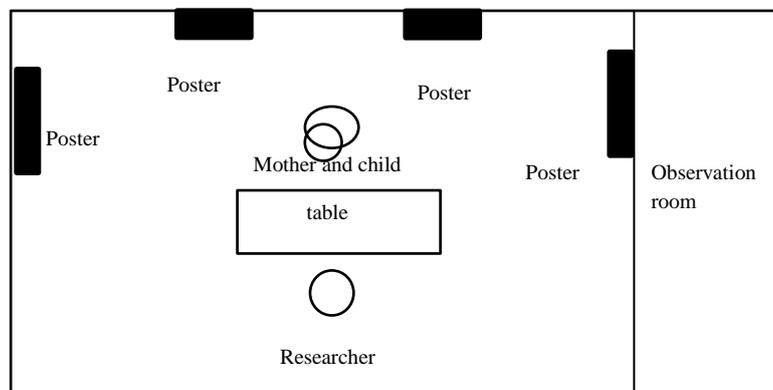


Figure 4.2: Diagram of room layout for ESCS

4.5.3.1. *Object Spectacle Tasks*

The target behaviours for this task were initiating joint attention and initiating behavioural requests. Six toys were used in total – three wind up mechanical toys and three hand operated mechanical toys which included a telephone, a spinning top and a light-up windmill toy. Each toy was presented to the child three times and each presentation followed the same format. A toy was activated in front of the child on the table but out of their reach for around 6 seconds. The researcher remained silent but was attentive to the child in case they initiated a bid for joint attention. If the child did initiate joint attention the researcher would give a natural, brief response: ‘yes I see’. If the child made a bid to get the toy the researcher would respond by placing the toy within their reach. Once the activation had finished the researcher would give the toy to the child regardless of whether they had made a bid or not. The child was then able to play with the toy for around 10 seconds or until they gave it back to the researcher. After 10 seconds, the researcher would verbally request the toy twice ‘give it to me’ and then they would use a palm up gesture whilst saying ‘give it to me’ a further two times. If the child still did not give the researcher the toy they would gently retrieve the toy from the child. The same procedure was then repeated for the same toy two more times.

4.5.3.2. *Look/Gaze Following Task*

The target behaviour for this task was responding to joint attention and this task used the four posters which were on the walls to the left and right of the child and behind the child to the left and right. Initially the researcher would get the child’s attention by calling their name and then touch their nose to establish eye contact. The researcher would start with the left and right trials and then follow with the behind left and behind right trials. The researcher always used a ‘short-arm point’ with the elbow of the pointing arm in contact with their side. If doing a left/right trial they would move their torso to be in line with the point and if it was

a behind trial the researcher would lean slightly forward and to the left/right. During the point the researcher would call the child's name three times with approximately a two second gap between each. They would not look back at the child until after they had said their name for a third time. It was ensured that the researcher's 'pointing' finger was at least two feet from the child. Two sets of pointing trials were completed in the assessment.

4.5.3.3. *Book Presentation Task*

For this task the target behaviours were responding to and initiating joint attention. The researcher opened a picture book and placed it on the table within reach of the child. The researcher said 'What do you see?' and the child was then left to examine the book for approximately 20 seconds. If the child pointed to anything in the book during this time the researcher responded naturally but briefly 'yes I see'. After the 20 seconds the researcher began pointing in the book. The researcher firstly pointed to the picture on the (child's) left hand side of the page and then on the right hand side. The researcher then turned the page and repeated this twice more and in total six pictures were pointed at. When pointing, the researcher would say the child's name and it was made sure that the point was at some distance from the picture. This task was coded live by the researcher administering the session since the video cameras were not always sensitive enough.

4.5.3.4. *Plastic Jar Task*

The target behaviour for this task was initiating behavioural requests. The researcher placed a transparent plastic jar on the table in front of the child but out of their reach. The jar was fastened with a screw lid and contained two novel wind-up mechanical toys inside. As the child was watching the researcher took the lid off the jar and tipped the toys onto the table. Next the toys were put back in the jar and the lid was fastened. The researcher then gave the sealed jar to the child for approximately 10 seconds, or until the child spontaneously gave them the jar. If the child did not return the jar, the researcher used verbal prompts using

the palm up gesture if needed. If the child still did not give the researcher the jar, they gently retrieved it. The researcher then un-screwed the lid and took out one of the toys and gave it to the child to play with for approximately 10 seconds. The same process was then repeated for the other toy in the jar. This task was presented once during the assessment.

4.5.3.5. *Scoring*

Coding was done in accordance with the ESCS manual and all coding apart from the book presentation task, was done by observing the video recordings. Initiating joint attention, responding to joint attention and initiating behavioural request behaviours were split into high level and low level behaviours. For initiating joint attention, low level behaviours consisted of eye contact and alternating. These were coded from the object spectacle task. Eye contact was coded if the child made eye contact with the researcher whilst touching or manipulating an inactive toy. An alternate was coded if the child alternated their look between an active object spectacle and the researcher's eyes. This could have occurred when the active object was on the table or in the researcher's hands.

Higher level behaviours included pointing and showing. A point was scored if the child pointed to an active object on the table. It was also coded if the child pointed to the pictures in the book or posters on the wall before the researcher had pointed at them. Pointing could occur with or without eye contact between the researcher and child. The index finger had to be clearly extended to be coded as a point. A show was scored when the child looked at the researcher and raised a toy up towards the researcher's face. To be scored as a show, the toy had to be initially held still for 1 to 2 seconds. This behaviour was sometimes difficult to score as the behaviour is very similar to a 'give'. However, if the researcher went to retrieve the toy and the child resisted, then it was seen that the intention was to 'show' not to 'give'. Usually a show was a brief behaviour where the child would quickly show the toy and then retract it. The initiating joint attention behaviours described could also be coded as a 'bid

to caregiver' if during the assessments any of these behaviours were directed towards the child's mother.

Responding to joint attention was scored from the book presentation task and gaze following task. The lower level behaviour for responding to joint attention was following proximal points and this was coded using the book presentation task. In this particular task the researcher pointed to six pictures in a book and the child's response was scored as 'correct' if they turned their head and eyes to the selected picture. The maximum score for this category was therefore six.

The higher level behaviour scored for responding to joint attention was if the child was able to follow the researcher's line of regard and this was coded during the gaze following task where the researcher pointed to 4 posters one to the left of the child, one to the right and two behind the child. To score a correct response, the child had to turn their eyes or head so that it was clear they were looking in the correct direction and past the end of the researcher's index finger. For the behind trials the child had to have turned their head so it was obvious they were looking at something behind them. This task was administered twice during each assessment and therefore a score out of eight was obtained for each child. For both responding to joint attention tasks the child's score was converted into a percentage and these percentages were combined to give a 'total responding to joint attention' score.

Initiating behavioural requests were coded when the child attempted to request something from the researcher/caregiver as opposed to showing/sharing something with them. For example, if the child focused their attention on or tried to obtain a toy that was out of their reach. Low level behaviours included: eye contact, reaching and appeals.

Eye contact was coded if the child made eye contact with the researcher after they had removed an object from the child or after an object spectacle had finished. A reach was coded

if the child extended their arm to a toy that was out of reach. If the child simply reached and obtained said toy then this was not scored. If the child attempted to get out of their seat to obtain a toy this was only scored if the researcher was holding the toy. If the child retracted their arms for over two seconds or laid their arm on the table and closed their hand for more than two seconds then this was seen as the end of a bid. If the child reinitiated a bid in less than two seconds then this was not coded as a separate bid. Finally appeals were coded if the child combined a reach with eye contact. The eye contact could be brief and combined with a longer gesture.

Higher level behaviours for initiating behavioural requests included gives and points. A give was coded if the child held an object out towards the researcher or pushed an object towards the researcher. A give could occur with or without eye contact. A point was coded if the child used an extended index finger to show a desired object. Sometimes a reach turned into a point or vice versa. If this was the case then the higher level behaviour was coded (point). Points to request could occur with or without eye contact. Again if any of these behaviours were directed towards the mother then they were coded as a 'bid to caregiver'.

4.5.4. *Parental Questionnaires*

The M-CHAT (Robins, Fein & Barton, 2009) is a parental questionnaire consisting of 20 questions relating to social communication skills. For example: 'Does your child show you things by bringing them to you or holding them up for you to see - not to get help, but just to share?' for each question the parent is required to indicate 'yes' or 'no' in response. The questionnaire can be used as a screening tool for Autism Spectrum Conditions (ASC) but it was explained to parents that researchers were interested in using the questionnaire as they wanted to assess the social communication behaviours of the child and not because they suspected they had a dual diagnosis of ASC.

For all questions except 2, 5 and 12 a response of 'no' indicates ASC risk whereas items 2, 5 and 12 are reverse coded and an answer of yes indicates ASC risk. Therefore a numerical value can be determined by counting how many of the answers provided by the parent relate to risk for ASC. The authors then advise that a total score of 0-2= low risk, 3-7= medium risk and 8-20= high risk.

To measure maternal well-being, parents were given the Hospital Anxiety and Depression Scale (HADS; Zigmond & Snaith, 1983) which consists of 14 questions, seven of which relate to anxiety and seven to depression. For each question there is a four answer Likert scale format for example: 'I feel cheerful – never, not often, sometimes, most of the time'. For each question each answer is numerically scored from 0-3. The scores for each subscale can then be calculated separately and the author advises the following ratings: 0-7 normal, 8-10 borderline abnormal and 11-21 abnormal. The scores from the two subscales can also be combined to give a rating of emotional distress (0-42) with higher scores indicating higher levels of distress.

4.5.5. *Parent-Child Interaction*

At the end of the testing session, a 5-minute parent-child interaction was filmed. For this task, the researcher left the room and watched from an adjacent observation room. The parent and child were left with a selection of toys including a ball, cars, a doll and books. They were informed that they could play on the floor with any of the toys and to try to play normally like they would at home. The interactions were video-recorded so that they could be coded at a later date (the coding scheme used is described in an earlier section 2.5.1).

4.6. *Intervention*

The intervention lasted for 10 weeks. Parents were required to bring their child to the University of Reading once per week for an intervention session with the researcher, which lasted for approximately 20-30 minutes. Parents were given tasks to do at home three times a week for 10 minutes. For some participants who were unable to travel, the researcher conducted their session in the participant's home. The methodology was based on and adapted from Whalen and Schreibman (2003) and focused on improving responding to joint attention. At each session the child was required to reach a criterion so they could move onto the next level the following week. To master a level, the child had to score 3/3 on two trials out of 4. The following 7 levels of responding to joint attention were trained: 1) response to hand on object, 2) response to object being tapped, 3) response to showing of object, 4) eye contact, 5) following a point (within line of vision), 6) following a point in a book and 7) following a point (outside of visual field). Throughout the intervention a hierarchy of prompts were used: 1) calling the child's name such as: 'Tom'; 2) if the child didn't respond, then directive was used: 'Tom looking'; and 3) if the child didn't respond to the directive than a physical prompt was used. This was dependent on the level and involved: gently holding the child's hand on a toy for 5 seconds, using a prompting toy to change the child's focus, physically gently turning the child towards the desired object, or the researcher moving closer to the toy they were pointing at.

Researcher led sessions took place at the University of Reading or in the child's home and lasted for around 20-30 minutes. The child was required to sit on a small chair or on their parent's lap at a table opposite the researcher. The researcher would present 4 trials each including 3 different opportunities for responding. Between each trial, the researcher would provide a reward to reinforce motivation, such as bubbles, and as the child moved through the

levels, this time was also used for ‘maintenance tasks’ (repetition of levels that had been previously ‘mastered’). The researcher live coded each of the child’s responses so that they could document their progress over time. The following codes were used for the child’s response: correct response, prompted response and incorrect response. Correct response was given if the child engaged with the toy for 5 seconds without additional prompts. For example, if the child looked at the toy, attempted to reach for the toy or looked between the researcher and the toy. The child was then given the toy to play with and whilst the child was attending to the toy the researcher would provide language input. They would name the toy and then describe how the child was manipulating it. For example, ‘ball!’, ‘Tom’s bouncing the ball’, ‘bounce, bounce’. If the child required prompts to respond correctly, then this was given the code ‘prompted response’ and did not count towards the mastery of the level. Prompts were applied if the child did not show an interest and did not initially respond to the researcher’s bid. The code ‘incorrect response’ was used if the child discarded the toy straight away or refused to look at the toy regardless of attempts to prompt the child.

4.6.1. *Intervention Levels*

Level 1 required the child to respond to a toy which the researcher placed their hand on. Commonly the toys used for this level included: soft books, spikey balls, Mr Tumble and shakers. However, this varied as the researcher made sure to use toys that the individual child showed preference for in order to increase their motivation. Once the researcher had placed the child’s hand on the toy, if the child engaged with the toy for at least 5 seconds, this was scored as a ‘correct response’. The researcher would then name the toy ‘it’s a shaker’ and let the child play with the toy and provide lots of language input, ‘shake shake’. The researcher kept up this exchange until the child lost interest in the toy and would then select a new toy and repeat the process. If the child did not respond correctly the researcher, would attempt to use prompts as described previously.

For level 2 the protocol was very similar to level 1 except that this time the researcher tapped a toy on the table in front of the child. Commonly the toys used for the level included 'hard' toys which would make a banging sound when tapped on the table: cars, shakers, box toy, etc. After tapping the toy, the researcher would leave the toy within reach of the child so that they were able to obtain it. Again, the child was required to engage with the toy for at least 5 seconds.

For level 3 the procedure was similar to the previous levels except that, in order to get the child's attention the researcher activated a toy in front of them. Toys used for this level included: windup toys, spinning tops and toys that 'talked' or 'sang' when activated. The toy was activated within reach of the child so they were able to retrieve it. The researcher would activate the toy again if the child made such a bid.

Level 4 was a prerequisite for future levels and the aim was to achieve eye contact with the child when requested. For this, the researcher used bubbles or an electronic windmill toy that lit up. The researcher would call the child's name and once they made eye contact would blow bubbles as a reward. Alternatively, the researcher would hold the windmill up to their face and call the child's name; if the child made eye contact the researcher would activate the toy and give it to the child to play with as a reward.

The final three levels centred on the child being able to follow the point of another person. For level 5 the child was required to follow the point (and therefore the focus of attention) of the researcher to a toy which was in the child's line of vision. The researcher selected two toys and put them on the table but out of reach of the child. Initially eye contact was established with the child by calling the child's name and if needed pointing to the researcher's nose. The researcher then pointed to one of the two toys and said: 'Let's play with the ball'. The child was required to follow their point and look at the selected toy. If they

did this correctly, the researcher provided verbal praise before removing the toy that had not been selected and giving the child the correct toy to play with. The researcher then provided lots of language input as the child was manipulating the toy. For example: 'Tom's got the shaker', 'shake shake'. If the child did not look at the correct toy, then the researcher would prompt the child to look at the correct toy and remove the 'distractor' toy.

Level 6 focused on following a point within a book. For this the researcher held a large picture book in front of the child and pointed to various pictures one by one in the book whilst saying the child's name. For a correct response the child needed to follow the researcher's point to look at the selected pictures. Prior to this level parents were asked about which picture books their child particularly enjoyed and in some instances they brought their own books to the session. If the child did not follow the point they were given verbal prompts and a prompt toy was used to try and direct their attention.

The final level (level 7) aimed to get the child to follow a point to an object/toy that was outside of their direct line of vision. Toys were placed around the room to the left, right and behind the child. The researcher would establish eye contact with the child before pointing to the target toy and calling the child's name as they turned to the face the toy. The researcher would then look back between the child and the toy and provide further verbal encouragement if needed. The child had to first make eye contact with the researcher and then follow their point to score a correct response. If they responded correctly the researcher would get the toy and give it to the child so they could play with it as a reward. If the child did not follow the point, further verbal prompts were given and then either the parent would gently move the child to face the target toy or the researcher would gradually move closer to the target toy until the child turned to look.

During the training the researcher provided lots of positive reinforcement and feedback – firstly so the child was congratulated when they responded correctly but also to increase their motivation. Verbal praise was given such as: ‘good girl’, ‘well done’ and the child was able to play with the target toy once they responded correctly. Bubbles were also given in-between each of the four trials and the child was encouraged to watch and ‘pop’ the bubbles.

4.6.2. *Parent Sessions*

Parents were instructed to complete their element of training three times a week for approximately 10 minutes. At the start of the intervention, parents were provided with a manual and then each week they were given instructions to take home as well as a diary so that they could record if they had managed to complete all the training and to provide any comments (see appendix E). Parents were asked to repeat the activity that the researcher had administered in the session. For each training session parents were instructed to find a comfortable place to play with their child with a selection of toys. They were then instructed to make a bid for attention (the exact type of bid depended on what level they were instructed to work on that week) once a suitable opportunity in play occurred, i.e. when the child had finished playing with one toy. For example, they may have placed their child’s hand on a toy. If the child correctly responded to their bid the parent would provide positive feedback whilst letting the child play with the selected toy and providing lots of language input. If the child did not respond or responded incorrectly the parent was advised to provide prompts (as previously described).

4.6.3. *Treatment Fidelity*

To ensure that suitable precautions were taken to achieve treatment fidelity, the National Institute of Health Behaviour Change Consortium checklist (UCDHSC centre for Nursing Research, 2006) was followed. The first step was ‘Fidelity to theory’ – was the

intervention based on relevant theory and did it contain ‘active ingredients’ based on this. The intervention we chose was published in a peer reviewed journal and previous research has identified that responding to joint attention is an important precursor for language development for children with DS (Harris et al., 1999; Mason-Apps, 2013; Mundy et al., 1995). Furthermore, all participants received 10 intervention sessions lasting 20-30 minutes. The intervention activity was documented and completed in each session.

The second step was ‘Provider training’ – was the researcher capable of delivering the planned intervention. Standardised materials and scoring sheets were created for the researcher to use in each session and the researcher received on-going supervision throughout. Initially intervention sessions were vide-recorded and reviewed by the researcher’s supervisors.

The third step was ‘Treatment implementation’ – did the researcher administer the intervention as it was designed. A treatment manual was created and utilised for the duration of the intervention. Weekly instructions were also made for each task with clear step by step instructions. At the end of the intervention a questionnaire was given to parents to obtain their perception of the intervention programme.

The fourth step was ‘Treatment receipt’ – did participants receive the relevant active ingredients of the intervention. An immediate post-test and further follow up assessments were used to see if there had been any changes in responding to joint attention scores. Parents completed a weekly diary to show completion of ‘homework’.

The final step was ‘Treatment enactment’ – did the participant demonstrate the targeted skill in practice. An independent outcome measure was used (the ESCS) to see if changes in responding to joint attention had generalised to different toys/task.

4.7. Post-Test

The post-test assessments occurred a week after the last researcher intervention session as this gave parents time to complete their final three sessions at home. The post-test mostly replicated the pre-test and took approximately one and a half hours to complete, including breaks. The session took place in a clinic room/child friendly lab at the University of Reading or in the participant's home. Direct assessments included: the PLS-4, the MSEL (fine motor, gross motor and visual reception) and the ESCS. Parents were asked to complete a CDI. Parents were also given a questionnaire which asked how satisfied they were with the intervention and gave them the opportunity to give any feedback (see appendix F).

4.8. Six Month Follow Up

The first follow up assessment took place approximately 6 months after the pre-test assessments when the children were 24-29 months old. This was therefore in line with 'time point two' of the control group data. The main focus of this assessment was to see if the intervention effects had continued or had declined for responding to joint attention. However, to follow the pattern of the control group and in order for additional comparisons to be made, all the pre-test assessments were re-administered including: PLS-4, the MSEL and the ESCS. Parents were also asked to complete a copy of the Reading CDI and a five minute parent-child play interaction was filmed.

4.9. One Year Follow Up

The final follow up occurred approximately 1 year after the first assessment (pre-test) when the children were between 30-35 months old. This final assessment focussed on the language outcomes of the child. In line with the control group data participants were tested using the PLS-4 and the MSEL and parents were asked to complete the Reading CDI.

Figure 4.3 below demonstrates the general procedure for the intervention group and the control group:

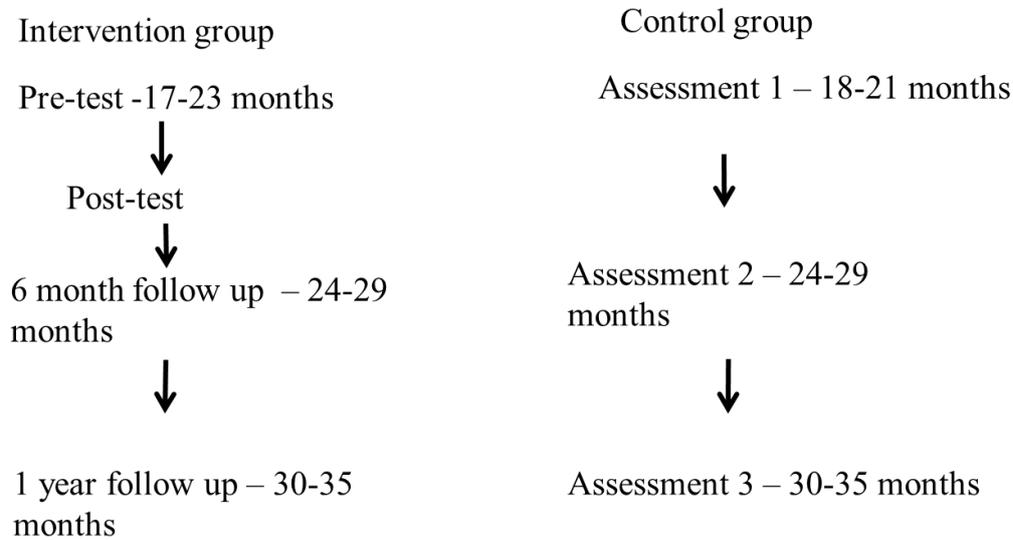


Figure 4.3: Stages of study for the intervention group and the control group

Chapter 5 – Results Time Point One/Pre-test Assessment Comparison of Intervention Group and Control Group

5.1. *Pre-Test Assessment Comparison of the Intervention Group and the Control Group*

The first aim was to compare the intervention and control groups to see if there were any significant differences between the two groups for non-verbal mental age, receptive and expressive language and joint attention. Initially tests of normality were conducted to assess the variables (see appendix G). Mann Whitney U / independent t-tests were then used to assess any differences. All analyses were done using raw scores since the majority of the participants were at floor for the standardised measures and the variability between participants would be lost if standard scores were used.

Non-verbal mental age was calculated by combining the raw scores from the visual reception and fine motor sub-scales from the Mullen Scale of Early Learning. There was no significant differences between the intervention and control groups in terms of non-verbal mental age, fine motor or gross motor skills. There was a marginally significant difference in terms of visual reception. The control group scored significantly higher than the intervention group ($p = .044$) but the effect size was small ($r = -.036$, see table 5.1).

Table 5.1: Means and standard deviations for the intervention group and control group for age and non-verbal measures

	Intervention group ($n=16$)	Control group ($n=14$)	t	U
CA (days)	601.00 (52.29)	594.86 (36.76)		
NVMA	34.63 (2.34)	35.21 (5.55)	-3.7	-
Fine motor	18.69 (0.87)	17.36 (2.84)	-	82
Gross motor	12.25 (1.65)	13.86 (3.53)	-	74.5
Visual reception	15.94 (1.81)*	17.86 (3.08)*	-2.112*	-

Note. CA= chronological age, NVMA= non-verbal mental age, * $p < .05$

No significant differences were found between the two groups for receptive language, total language, receptive vocabulary, expressive vocabulary or number of signs. However, there was a significant difference between expressive language with the control group scoring significantly higher ($Mdn = 19.32$) than the intervention group ($Mdn = 12.16$; $p = .024$, $r = -0.39$), and a small effect size was found (see tables 5.2 and 5.3).

Table 5.2: Descriptive statistics for PLS-4 for the intervention and control group

	Intervention group ($n= 16$)	Control group ($n=14$)	t	U
PLS AC	19.69 (2.06)	19.64 (4.24)	.037	-
PLS EC	18.06 (2.32)*	20.36 (2.98)*	-	58.5
PLS TL	37.75 (3.75)	40.00 (6.85)	-1.135	-

Table 5.3: Means and standard deviations for receptive/expressive language and vocabulary scores for the intervention and control group

	Intervention group ($n= 16$)	Control group ($n=13$)	t	U
EV	4.31 (4.85)	2.23 (3.63)	-	66
RV	68.56 (49.68)	63.31 (55.72)	.268	-
Signs	11.75 (21.35)	9 (15.71)	-	96.5

Note. PLS AC= auditory comprehension, PLS EC= expressive communication, PLS TL= total language, EV= expressive vocabulary, RV= receptive vocabulary, * $p < .05$

Next, joint attention and behavioural requests were compared. No significant differences were found for total responding to joint attention, following proximal points, gaze following, points to the left and right or points behind. No significant differences were found for total initiating joint attention, low-level initiating joint attention or high-level initiating joint attention. No significant differences were found between the groups for: total initiating behavioural requests or low-level initiating behavioural requests. However, a significant difference was found for high-level initiating behavioural requests with the control group scoring significantly higher ($Mdn= 20.64$) than the intervention group ($Mdn= 9.73$; $p < .001$,

$r = -.64$). When looking at the raw data in the intervention group all children scored between 0 and 4 whereas for the control group all children scored between 0 and 6 except for one child who scored 12 (see table 5.4).

Table 5.4: Means and standard deviations for Early Social Communication Scale scores for the intervention group and the control group

	Intervention group ($n = 16$)	Control group ($n = 14$)	t	U
IJA total	14.44 (10.991)	20.5 (12.62)	-	80.5
IJA high	1.25 (3.38)	2.14 (3.98)	-	76.5
IJA low	13.19 (10.08)	17.5 (9.07)	-1.225	-
RJA total	43.6 (22.86)	48.5 (26.84)	-.540	-
Gaze	32.03 (20.9)	33.93 (33.05)	-.185	-
L and R	54.69 (34.42)	53.57 (44.78)	-	110
Behind	9.38 (15.48)	14.29 (30.56)	-	107.5
Proximal point	55.17 (35.83)	63.01 (33.43)	-.622	-
IBR total	10.93 (3.63)	13.64 (6.07)	1.470	-
IBR high	1.13 (1.46)***	4.57 (2.82)***	-	26
IBR low	9.8 (3.57)	8.86 (5.8)	.531	-

Note. IJA= initiating joint attention, RJA= responding to joint attention, L and R= left and right, IBR= initiating behavioural requests, *** $p < .001$

There were no significant differences between the groups for total number of utterances used or strong verbal control. However a significant difference was found for mild verbal control with the mothers from the intervention group using significantly more mild verbal controls than the control group ($p = .002$, $r = .058$), which is a medium sized effect. There were no significant differences between the groups in terms of positive expressed emotion, negative expressed emotion, coercions/intrusions, verbal elaboration, emotional tone, sensitivity or reciprocity (see table 5.5).

Table 5.5: Means and standard deviations for maternal interactive style for the intervention group and the control group

	Intervention group ($n = 13$)	Control group ($n = 12$)	t	U
Total utterances	95.46 (19.72)	85.92 (19.05)	1.229	-
Strong control	16.31 (8.71)	17 (12.31)	-.163	-
Mild control	27.31 (12.72)**	12.58 (6.87)**	3.555	-
PEEM	6.23 (4.94)	4 (3.81)	1.257	-
NEEM	0.46 (0.66)	0.17 (0.39)	-	60
Coercions	1.69 (2.29)	2.25 (3.19)	-	70.5
Verbal Elaboration	2.54 (1.2)	2.75 (0.87)	-	58.5
Emotional tone	3.31 (1.11)	2.92 (1.24)	-	60.5
Sensitivity	2.77 (1.30)	2.83 (1.12)	-.132	-
Reciprocity	3.08 (1.12)	3.25 (1.14)	-	72

Note. PEEM= positive expressed emotion, NEEM= negative expressed emotion, ** $p < .01$

5.2. *The Hospital and Anxiety Depression Scale*

The Hospital and Anxiety Depression Scale (HADS) was used to measure levels of anxiety and depression of the mothers in the intervention group. This was administered as mothers were required to be directly involved in the intervention sessions. The scale recommends that a score of 0-7 for either element is classified as ‘normal’, a score of 8-10 is ‘borderline abnormal’ and a score of 11-21 is ‘abnormal’. For the anxiety subscale 13 mothers had a score of 0-7 and so ‘normal’. The other two mothers had a score of 9 and 10 so were ‘borderline abnormal’.

For the depression subscale twelve mothers scored between 0-7 and so were within the ‘normal’ range. The other three mothers scored 8, 9 and 10 and were therefore within the ‘borderline abnormal’ range (Scores from the HADS are shown in figure 5.1 below).

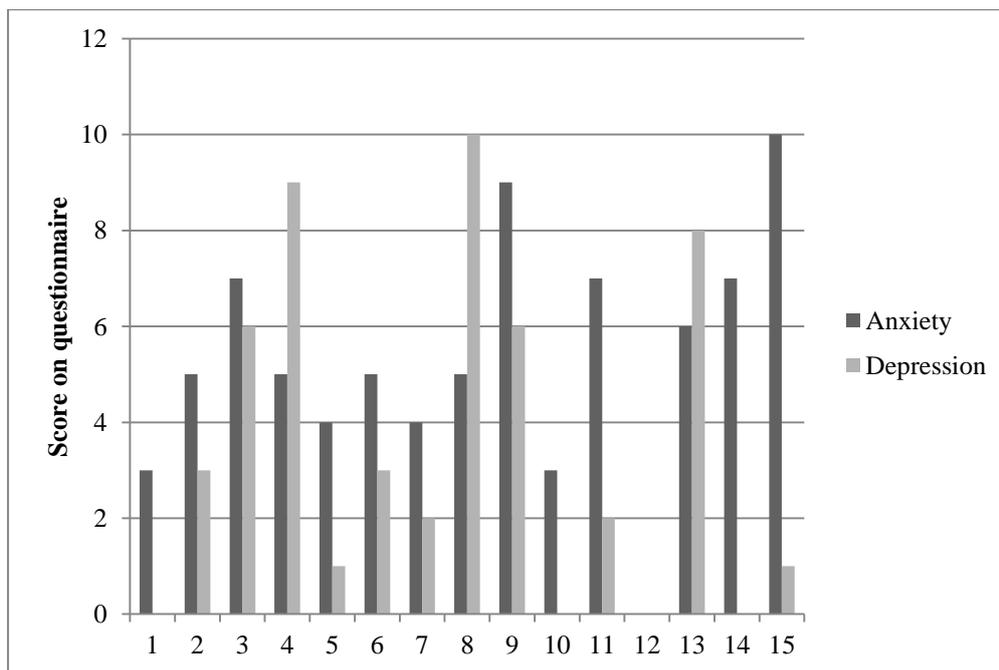


Figure 5.1: Maternal scores for Anxiety and Depression from the HADS

5.3. M-CHAT

The scores on the M-CHAT indicate risk for ASC, with a higher score indicating a higher risk. The scoring guidelines stipulate that a score between 0-2 indicates ‘low risk’ suggesting that no further action is required. A score between 3 to 7 is ‘medium risk’ which means a follow up assessment or observation may be needed. Finally, a score of 8-20 indicates ‘high risk’ and suggests that the child may be eligible for a diagnostic assessment and /or early intervention. It is worth noting that some of the questions which indicate risk of ASC are also characteristic of children with DS of this age. For example: ‘have you ever wondered if your child might be deaf?’ and ‘does your child walk?’ These questions were therefore removed. One child’s parent did not return the questionnaire. Six children scored within the ‘low risk’ criteria, 7 children received a score indicating a medium risk and 2 children scored within the high risk range (see figure 5.2).

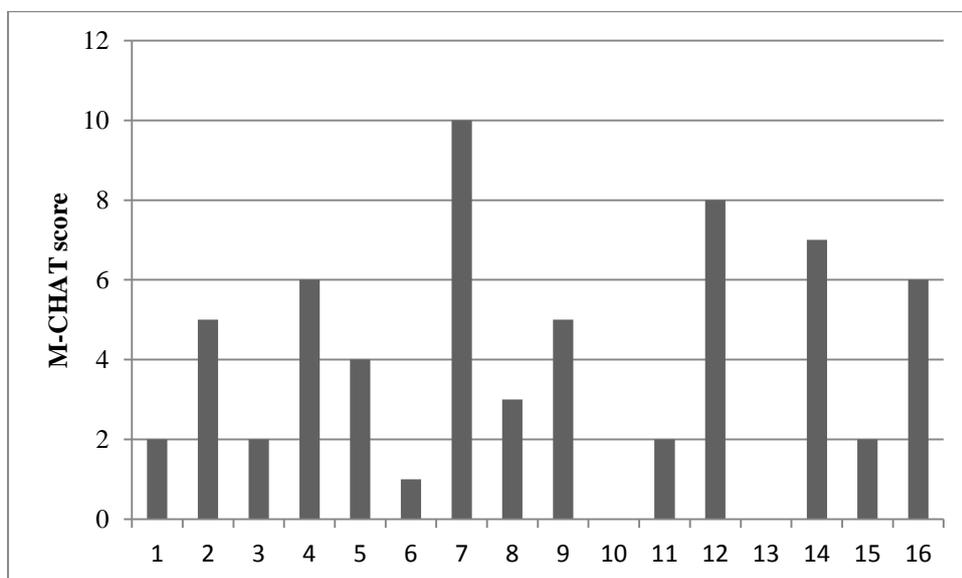


Figure 5.2: Participant scores for ASC risk (items removed)

Chapter 6 - Pre-Post-Test Results

As no control data was available for this time point, repeated measure ANCOVA's were used to determine if there was a significant difference between pre-and post-test results for responding to joint attention, receptive and expressive language and receptive and expressive vocabulary for the intervention group only, whilst controlling for changes in non-verbal mental age (if assumptions were met). At this stage responding to joint attention was the primary outcome measure and language/vocabulary scores were secondary outcome measures.

6.1. *Responding to Joint Attention*

Our first objective was to see if there were significant differences between the mean scores of responding to joint attention pre-and post-intervention for the intervention group only. We looked at the total responding to joint attention score as well as the scores for the individual tasks – the proximal point task (point in a book) and the gaze following task (pointing to posters).

As a first step, in order to assess the normality of the data, one sample Kolmogorov-Smirnov tests were run for the responding to joint attention variables. The pre and post data for total responding to joint attention scores and the gaze following task were normally distributed. However, for the proximal point task, the post test scores were not normally distributed and couldn't be corrected through a log transformation (see appendix G). Hence, a Wilcoxon signed ranks test was used. Performance on the book pointing task was significantly higher at post-test ($Mdn= 83$) than at pre-test ($Mdn= 66.7$), $Z= -2.710$, $p= .007$, $r= -0.48$ suggesting that participants were more likely to attend to the book points after the intervention. The ranks showed that 1 participant had a lower score at post-test (100% - 83%), for 4 participants there was no change and the remaining 11 participants had higher scores at the post-test (see figure 6.1).

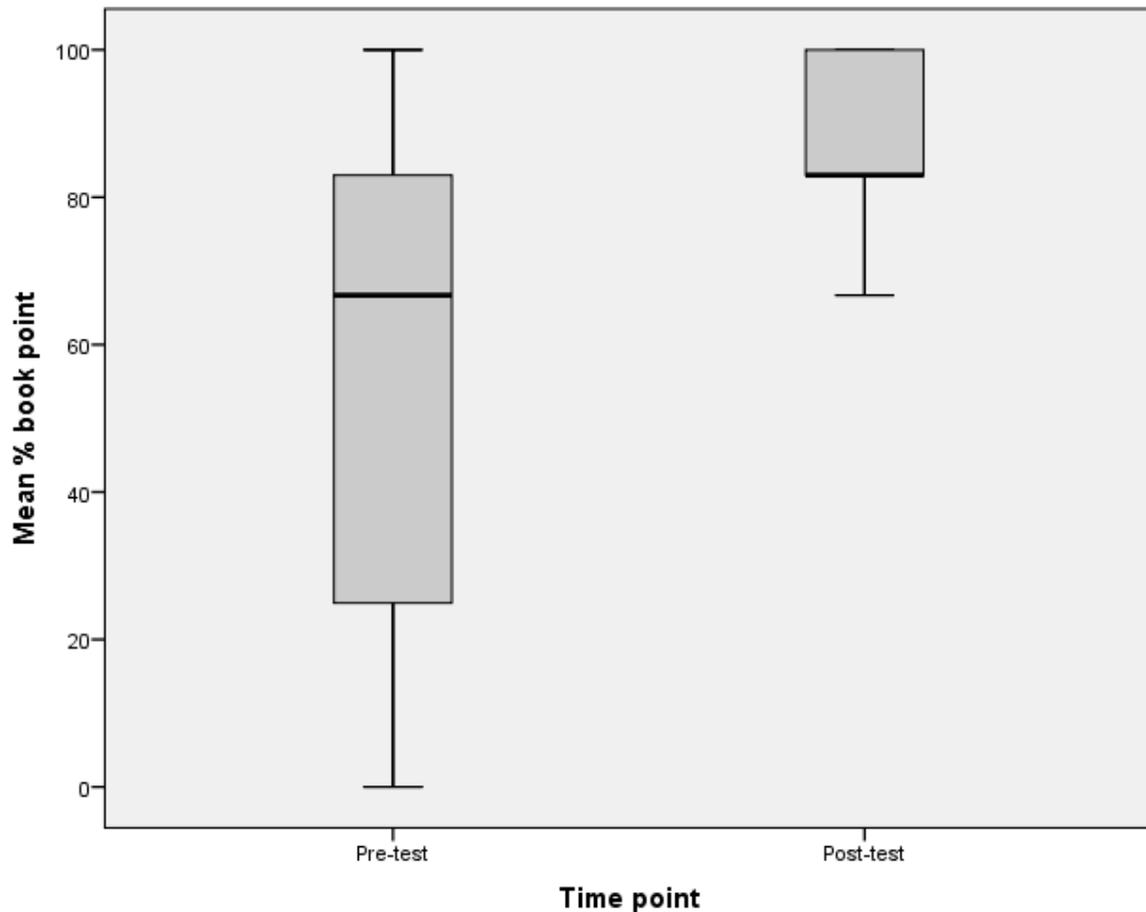


Figure 6.1: Box plot showing mean % scores for the proximal point task pre and post intervention

Next, a repeated measures ANCOVA was used to compare the mean scores pre and post-test for the gaze following task, controlling for changes in non-verbal mental age. Participants obtained significantly higher scores at post-test ($M= 60.16$, $SE= 7.15$) in comparison to the pre-test ($M= 32.03$, $SE= 5.23$), $F(1, 14)= 18.79$, $p= .001$, $d= -1.956$. For this task 15 children's scores improved following the intervention and one child's performance stayed the same (see figure 6.2).

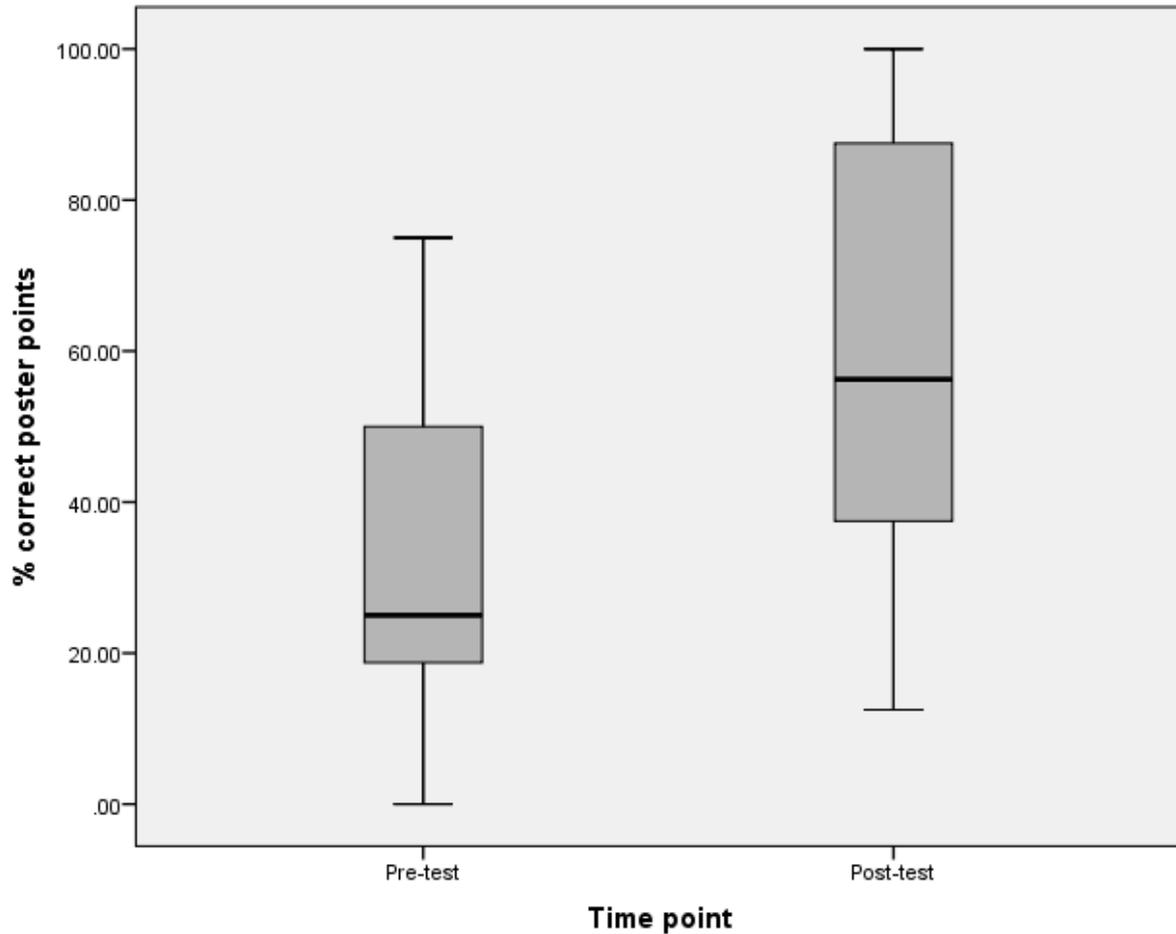


Figure 6.2: Box plot showing mean scores for the gaze following task pre and post intervention

A further, repeated measures ANCOVA was run to compare the mean scores pre and post-test for the total responding to joint attention scores (combining both the proximal points and gaze following task). There was a significant difference between the pre and post-test scores with the group scoring significantly higher at post-test ($M= 73.75$, $SE= 4.51$) in comparison to the pre-test ($M= 43.6$, $SE= 5.71$), $F(1, 14)= 9.845$, $p= .007$, $d= -1.539$. Fifteen children had improved scores following the intervention and one child's score slightly decreased (75% – 72.75%, see figure 6.3).

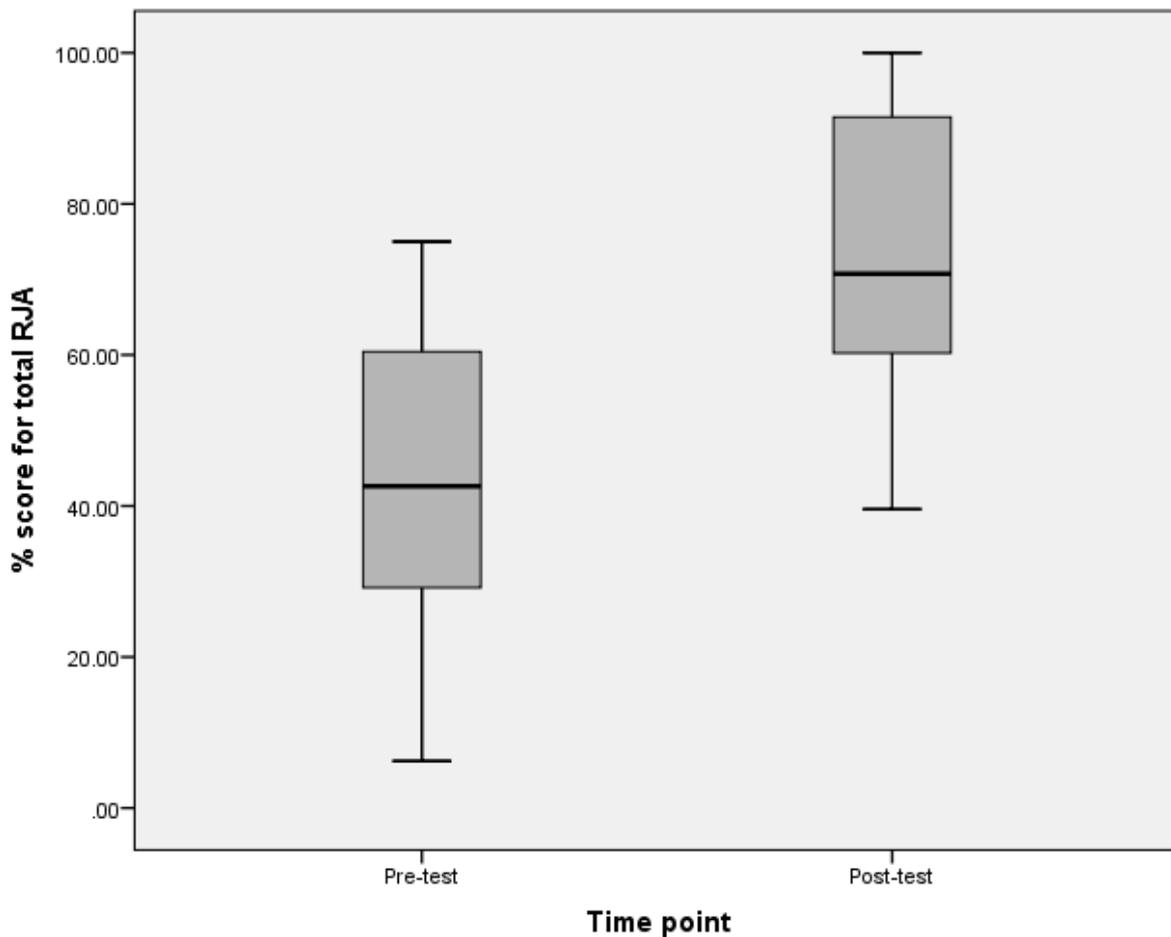


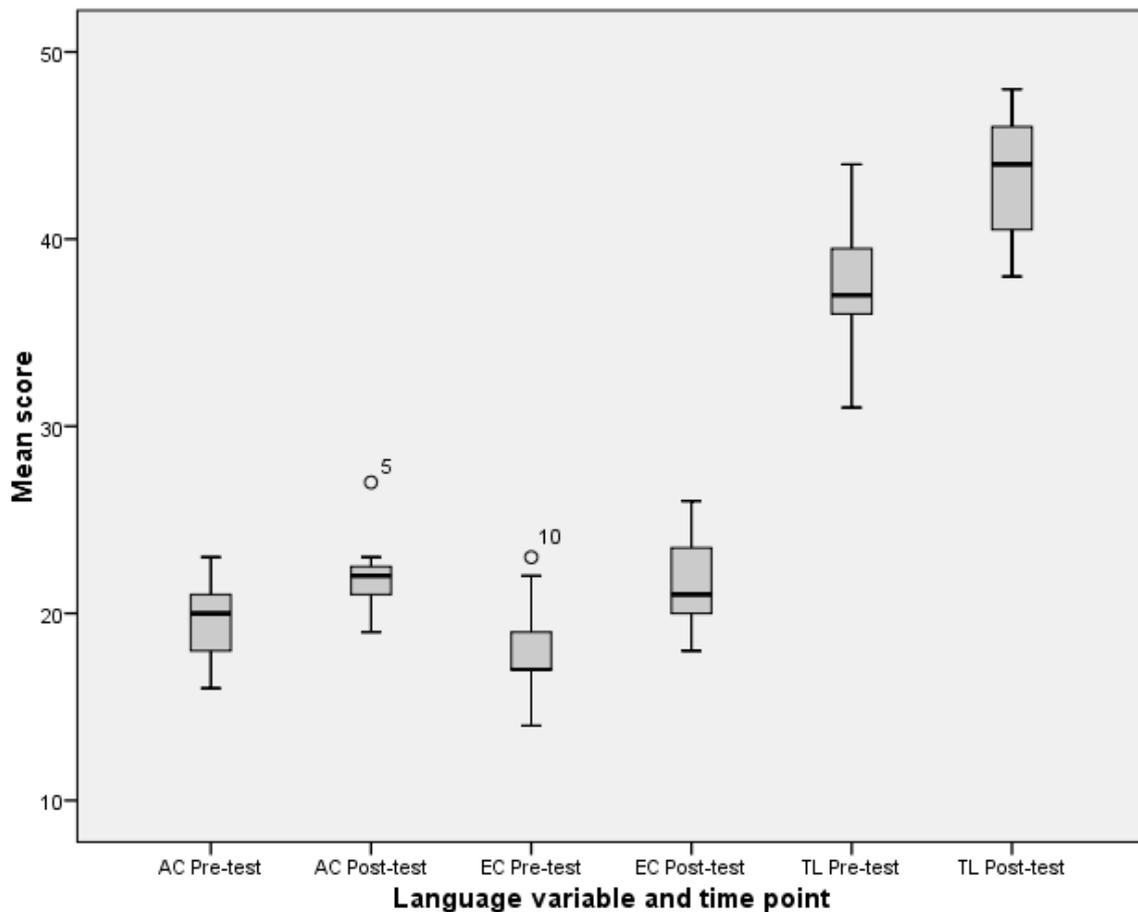
Figure 6.3: Box plot showing mean scores for total responding to joint attention pre and post intervention

6.2. Receptive and Expressive Language (PLS-4)

For receptive language the assumption of normality was violated for the post-test score (see appendix G), hence a Wilcoxon signed ranks test was used. There was a significant difference for receptive language with the group scoring significantly higher at the post-test ($Mdn= 22$) in comparison to the pre-test ($Mdn= 20$), $Z= -3.082$, $p= .002$, $r= -.77$. Twelve children had improved scores and four children did not improve. As can be seen from the box plot there is an outlier identified at the post-test. This participant obtained a score of 27 and all the other participants scored between 19 and 23.

For expressive language, the result from the pre-test was not normally distributed (see appendix G); hence a Wilcoxon signed ranks test was used to compare the pre and post-test results. A significant difference was found between the results with participants scoring significantly higher at the post-test ($Mdn= 21$) in comparison to the pre-test ($Mdn= 17$), $Z= -3.541$, $p< .001$, $r= -.86$. The results showed that all 16 of the children improved from pre to post-test for expressive language. The boxplot shows that there was an outlier at the pre-test assessment. This participant scored 23 whilst the rest of the group obtained a score between 16 and 22.

The pre and post-test scores for total language were normally distributed (see appendix G) and so a repeated measures ANCOVA controlling for non-verbal mental age was used to see if there were any significant differences between the pre and post-test scores. A significant difference was found with participants scoring significantly higher at the post-test ($M= 43.5$, $SE= 0.83$) when compared to the pre-test ($M= 37.75$, $SE= 0.94$), $F(1, 14)= 28.716$, $p< .001$, $d= -1.62$ (see figure 6.4).



Note. AC= auditory comprehension, EC= expressive communication, TL= total language

Figure 6.4: Box plot showing mean scores for receptive, expressive and total language scores pre and post intervention

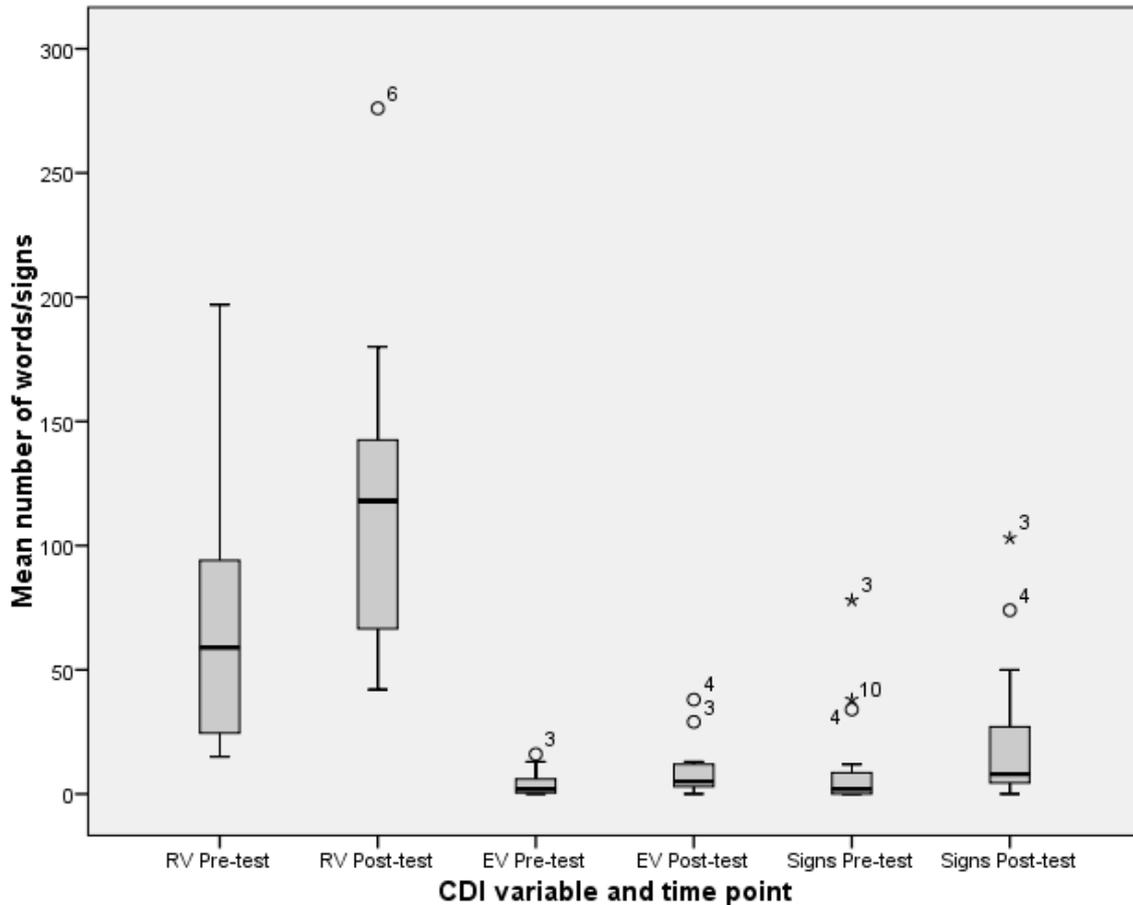
6.3. Vocabulary and Use of Signs

Initially we investigated whether there was a significant difference between the pre and post-test results for receptive vocabulary. As the data were normally distributed, a repeated measures ANCOVA was used. A significant difference was found for receptive vocabulary when comparing the scores with significantly higher results found at the post-test ($M= 115.93$, $SE= 16.28$) in comparison to the pre-test ($M= 65.67$, $SE= 12.91$), $F(1, 13)= 16.127$, $p= .001$, $d= -1.04$. As can be seen from the boxplot, there was an outlier who scored 276 whereas the rest of the sample scored between 42 and 176. All children improved on this

measure from pre to post-test, however, one participant did not return their questionnaire and so does not have a result for the post-test.

Next we looked at the results for expressive vocabulary pre and post-intervention. For this analysis we used a Wilcoxon signed ranks test as the data were not normally distributed. A significant difference was found between the pre and post-intervention results with significantly higher results obtained at the post-test ($Mdn= 5$) in comparison to the pre-test ($Mdn= 2.5$), $Z= -2.805$, $p= .005$, $r= -.72$. Five of the children did not improve between the pre and post-test whereas 10 of the children did. As demonstrated by the boxplot below one outlier was identified at the pre-test who scored 16 whereas the rest of the sample scored between 0 and 13. A further two outliers are present at the post-test who scored 29 and 38 and the rest of the sample fell between 0 and 13.

Finally, we compared the pre and post intervention results for the number of signs understood or used. As this data was not normally distributed a Wilcoxon signed ranks test was used. A significant difference was found between the pre and post-test results with participants using significantly more signs at post-test ($Mdn= 8$) in comparison to at the pre-test ($Mdn= 2$), $Z= -3.061$, $p= .002$, $r= -.79$. The results showed that three participants didn't improve between pre and post-test and the remaining 12 did. Three outliers are identified at the pre-test who scored 34, 38 and 78, whereas the rest of the sample scored between 0 and 13. A further two outliers are shown on the boxplot below for the post-test results scored 103 and 74, the rest of the sample scored between 0 and 50 (see figure 6.5).



Note. RV= receptive vocabulary, EV= expressive vocabulary

Figure 6.5: Boxplot showing mean numbers of words and signs understood and produced pre and post intervention

6.4. Factors Associated with Post-Test Responding to Joint Attention Scores

A correlation analysis was used to investigate if any of the baseline variables were associated with responding to joint attention post-test scores. Baseline non-verbal mental age, joint attention scores, maternal well-being (measured by the Hospital Anxiety and Depression Scale) and M-CHAT scores were investigated. All the variables were normally distributed except the post-test proximal point score (see Appendix H for full correlation matrix).

6.4.1. *Gaze Following Post-Test*

Regarding the gaze following post-test scores, significant correlations were found with gaze following baseline scores $r(12) = .844, p < .001$, total responding to joint attention baseline $r(12) = .586, p = .028$ and M-CHAT scores $r(12) = -.564, p = .036$ when controlling for the child's chronological age. No significant correlations were found between gaze following post-test scores and non-verbal mental age, maternal well-being, proximal point or initiating joint attention. A regression analysis was used to further investigate the significant correlations.

The post-test gaze following score was entered as the dependent variable with age, gaze following baseline and M-CHAT scores entered as the predictor variables. The total responding to joint attention baseline score was omitted from this analysis since it included the gaze following baseline score. All the assumptions were met for this analysis. The first model which included age was not significant $F(1, 13) = .518, p = .484, R^2 = .038$. The addition of the baseline gaze following scores significantly improved the model. The model was significant $R^2 = .724, F(2, 12) = 12.727, p < .001$ and baseline gaze following scores was a significant predictor. Adding the M-CHAT did not significantly improve the model. However, the overall model was significant $R^2 = .736, F(3, 11) = 10.2, p = .002$ but only baseline gaze following was a significant predictor (see table 6.1).

Table 6.1: Regression analysis for post-test gaze following scores

Variable	Model 1			Model 2			Model 3		
	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β
Age	.095	.131	.196	.086	.073	.179	.072	.078	.150
Gaze				1.124	.206	.828***	1.025	.254	.755**
M-CHAT							-1.388	1.986	-.134
<i>R</i> ² change				.686			.012		
<i>F</i> for <i>R</i> ² change				29.789***			.488		

Note. ** - $p < .01$, *** - $p < .001$

6.4.2. Proximal Point Post-Test

The proximal point post-test variable was not normally distributed, and a log transformation was unable to correct this, therefore Spearman's rho was used. Further investigation of the data showed that the majority of children were scoring 80-100% correct on this measure. The gaze following baseline correlation with the proximal point post-test was of borderline significance $r_s(16) = .494$, $p = .052$. The only variable which significantly correlated with the proximal point post-test was the M-CHAT $r_s(15) = -.671$, $p = .006$.

To investigate if the M-CHAT was a significant predictor, it was entered into a regression analysis controlling for the child's age. The first model including just age was not significant $R^2 = .024$, $F(1, 13) = .007$, $p = .933$. The addition of the M-CHAT significantly improved the model. The model was significant $R^2 = .415$, $F(2, 12) = 4.258$, $p = .040$ and the M-CHAT was a significant predictor (see table 6.2).

Table 6.2: Regression analysis for proximal point post-test scores

Variable	Model 1			Model 2		
	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β
Age	-.004	.052	-.024	-.033	.042	-.175
M-CHAT				-2.641	.906	-.661*
<i>R</i> ² change				.415		
<i>F</i> for <i>R</i> ² change				8.505*		

Note. * - $p < .05$

6.4.3. Total Responding to Joint Attention Post-Test

Partial correlations controlling for the child's age were conducted to see if any baseline predictor variables correlated with total responding to joint attention post-test scores. Significant correlations with total responding to joint attention post-test scores were found for gaze following baseline scores $r(12) = .832$, $p < .001$, total responding to joint attention baseline scores $r(12) = .584$, $p = .028$ and the M-CHAT $r(12) = -.649$, $p = .012$. A regression analysis was used to see if any of the significantly correlated variables would be significant predictors of post-test total responding to joint attention. The total responding to joint attention baseline score was omitted from this analysis since such a strong correlation was found with baseline gaze following scores which is incorporated in the total responding to joint attention score. Age was entered into the first block, baseline gaze following into the second and the M-CHAT was added in the final block.

The first model which included just the child's age was not significant $R^2 = .022$, $F(1, 13) = .297$, $p = .595$. The addition of gaze following baseline significantly improved the

model. The model was significant $R^2 = .699$, $F(2, 12) = 13.922$, $p = .001$ and baseline gaze following was a significant predictor. The model was not further improved by adding the M-CHAT. The overall model was significant $R^2 = .747$, $F(3, 11) = 10.83$, $p = .001$ but only baseline gaze following was a significant predictor (see table 6.3).

Table 6.3: Regression analysis showing significant predictors of post-test total responding to joint attention scores

Variable	Model 1			Model 2			Model 3		
	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β
Age	.045	.083	.149	.040	.048	.132	.022	.047	.073
Gaze				.697	.134	.823***	.572	.155	.675**
M-CHAT							-1.756	1.213	-.272
R^2 change					.676		.048		
F for R^2 change					29.594***		2.098		

Note. ** - $p < .01$, *** - $p < .001$

6.5. Parental Questionnaires

At the end of the intervention all parents were given a questionnaire asking how satisfied they were with the intervention and if they felt they had seen any positive effects within their child (see appendix F). Thirteen out of 16 parents returned the questionnaire. One-hundred percent of those who returned the questionnaire reported that they were extremely satisfied with the intervention and 100% felt that their child's responding to joint attention had improved. Seventy-seven percent of parents felt that their child's speech and language had improved as a result of the intervention. Finally, 92% of parents reported that

they had noticed improvement in other areas of development such as sustained attention and that they had changed how they communicated with their child after participating in the intervention (see figure 6.6).

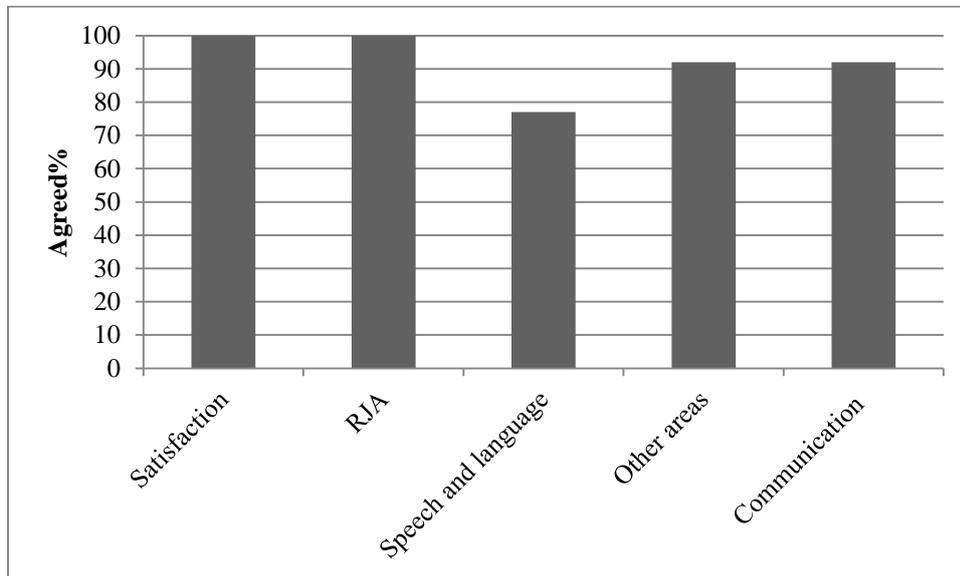


Figure 6.6: Parental satisfaction with intervention

6.6. Parent diary

As mentioned in the methodology chapter (4.6) parents were asked to complete a weekly diary to indicate if they had completed three intervention sessions at home that week. All parents reported that they had completed the obligatory three sessions. Therefore this data was not used in any additional analysis when looking at the intervention outcomes.

6.7. Summary

The results show that participants' responding to joint attention improved from pre to post-test in both the proximal points and the gaze following task. Furthermore, significant gains were observed for receptive and expressive language, vocabulary and use of signs. Results from the parental questionnaire added further support as 100% of parents felt that

their child's responding to joint attention had improved as a result of taking part in the intervention.

Further analysis was then conducted to investigate if any of the baseline variables were associated with post-test scores. Baseline variables included in this analysis were: non-verbal mental age, joint attention, maternal interactive style, maternal well-being and the M-CHAT. Baseline responding to joint attention significantly positively correlated with responding to joint attention post-test scores whereas the M-CHAT significantly negatively correlated with responding to joint attention post-test scores. This shows that those in the sample who had a higher risk of ASC tended to have worse responding to joint attention outcomes. The M-CHAT was a significant predictor of post-test scores on the proximal points task controlling for the child's age but it was not a significant predictor of scores on the gaze following task or total responding to joint attention post-test scores when controlling for the children's age and baseline responding to joint attention.

Chapter 7 - Results – Follow Up One

These results compared the intervention and control group when children were aged 24-30 months. At this stage language/vocabulary scores were the primary outcome measures. The results from the first follow up assessment, which took place 6 months after the initial assessment, were compared to those of the control participants at time point 2. The intervention group mean age was 807.13 days ($SD= 63.85$, range 719-919) and the mean age of the control group was 794.46 ($SD= 49.96$, range 741-899). There were no differences in chronological age between the two groups $U= 93.5$, $Z= -.461$, $p= .650$, $r= -.08$. However, a significant difference was found for non-verbal mental age with the control group scoring significantly higher ($M= 43.58$) than the intervention group ($M= 39.25$) $t(26)= -2.362$, $p= .026$, $d= .87$. Further inspection of the data revealed three outliers in the control group

(participant 2, 9 and 14) whose high scores were driving this difference. These participants were therefore removed for the ANCOVA analysis and there was no longer a significant difference between groups for non-verbal mental age (intervention group $M= 39.25$, control group $M= 40.56$) $t(23)= -.923$, $p= .366$, $d= .40$.

An ANCOVA analysis was used to assess group differences on joint attention and language measures at this time point whilst controlling for the children's baseline performance, chronological age and non-verbal mental age. Initially assumptions of normality, independence of the independent variable and covariate, homogeneity of variances and homogeneity of regression slopes were assessed. If any of the assumptions were violated then a non-parametric ANCOVA was carried out. This was done by using the Quade statistic (1967) in a three step process. Initially the dependent variables and covariates were ranked, ignoring the group variable. Next, a linear regression of the ranks of the dependent variable and the ranks of the covariate was run and the unstandardized residuals were saved. Finally, a one-way ANOVA was computed using the saved residuals from the previous step as the dependent variable and the group variable as the factor. The statistics from this ANOVA is the non-parametric result.

7.1. Responding to Joint Attention (Total)

7.1.1. Assumptions

A one-sample Kolmogorov-Smirnov test revealed that the total responding to joint attention scores at the first follow up were normally distributed in both groups (see appendix G). The independence of the independent variable and covariate assumption was met, for total responding to joint attention baseline $F(1, 25)= .089$, $p= .767$ (from previous analysis this assumption was also met for age and non-verbal mental age). The Levene's test was not significant $F(1, 22)= 2.328$, $p= .141$, indicating that the variances of the two groups were

equal. The homogeneity of regression slopes assumption was also met - group*age $F(2, 17)=.337, p=.718$, group*non-verbal mental age $F(2, 17)=.512, p=.608$ and group*responding to joint attention baseline $F(2, 17)=2.730, p=.094$.

As all the assumptions were met, a one way ANCOVA controlling for baseline performance and age was carried out. The intervention group obtained higher scores ($M=74.68, SD=18.44$) than the control group ($M=61.86, SD=28.44$), however this difference was not significant $F(1, 19)=1.212, p=.285, \eta^2=0.06$ (see figure 7.1).

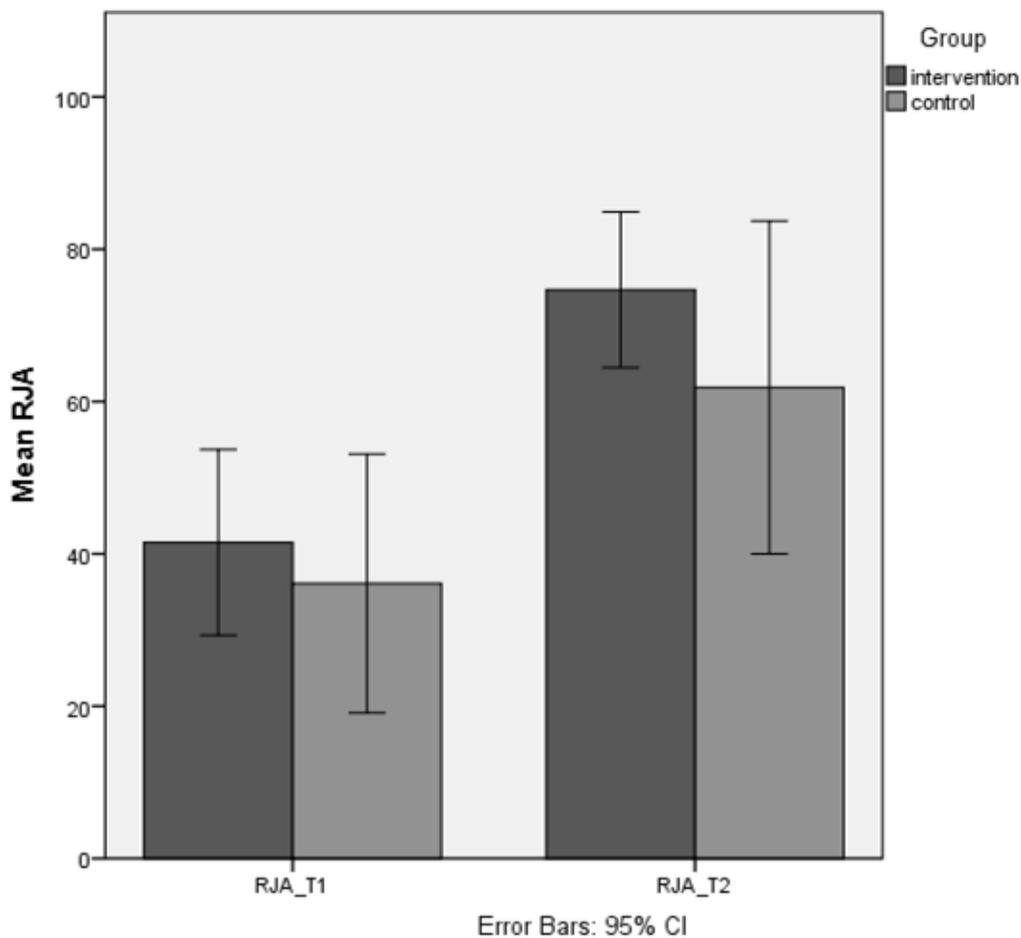


Figure 7.1: Responding to joint attention scores at pre-test and at first follow up for the intervention and control group

7.2. Receptive Language

The receptive language results were normally distributed in the control group but not in the intervention group (see appendix G), and a log transformation was not able to correct this. The independence of the independent variable and covariate was met – receptive language baseline $F(1, 25) = .213, p = .648$ as was the homogeneity of variance $F(1, 23) = .070, p = .793$. However, the homogeneity of regression slopes was violated - group*age- $F(2, 18) = 1.472, p = .256$, group*non-verbal mental age $F(2, 18) = 5.920, p = .011$, group*receptive language baseline $F(2, 18) = 1.442, p = .263$. As some of the assumptions were violated a non-parametric ANCOVA was used controlling for baseline performance and age. The intervention group had slightly higher scores ($M = 23.88, SD = 2.03$) than the control group ($M = 23, SD = 2.69$), but this difference was not significant $F(1, 23) = .812, p = .377, \eta^2 = .034$ (see figure 7.2).

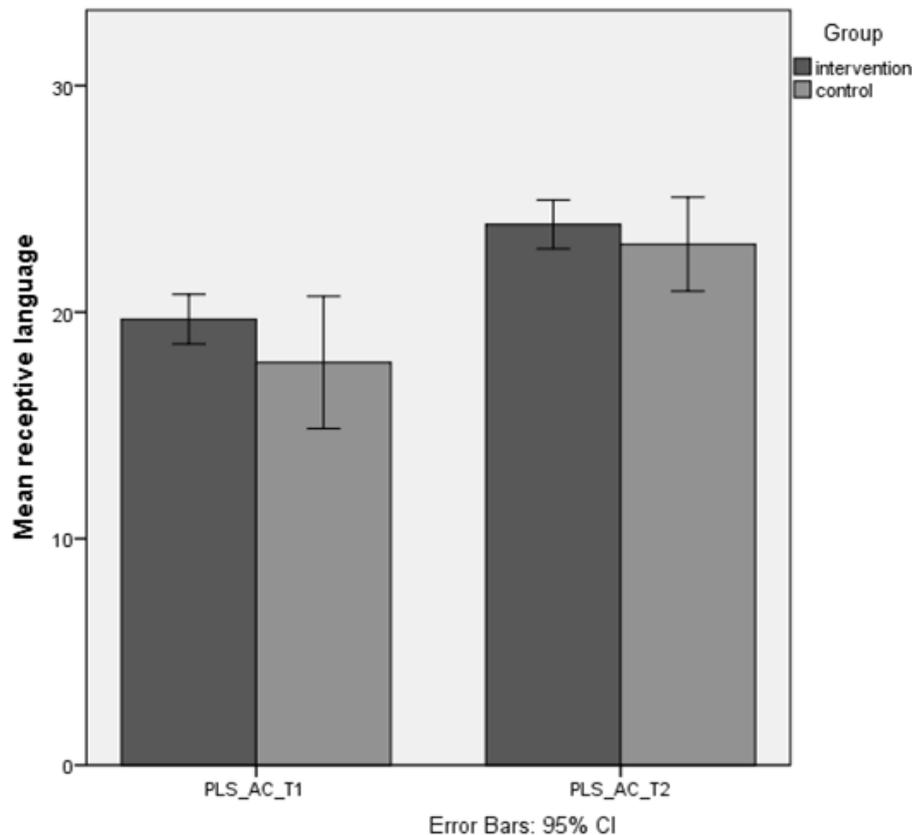


Figure 7.2: Receptive language scores pre-test and at first follow up for the intervention and control group

7.3. Expressive Language

A one-sample Kolmogorov-Smirnov test showed that expressive language scores were normally distributed in both groups (see appendix G). The independence of the independent variable and covariate assumption was met for the expressive language baseline scores $F(1, 25) = 3.381, p = .078$. The homogeneity of variances assumption was met as the Levene's test was not significant $F(1, 23) = .027, p = .871$. The homogeneity of regression slopes was met group*age $F(2, 18) = .478, p = .628$, group*non-verbal mental age $F(2, 18) = .591, p = .564$ and group*expressive language baseline $F(2, 18) = 1.150, p = .339$. The intervention group had slightly higher scores than the control group ($M = 23.75, SD = 1.88, M = 23.56, SD = 1.74$) but this difference was not significant $F(1, 20) = .352, p = .560, \eta^2 = .017$ (see figure 7.3).

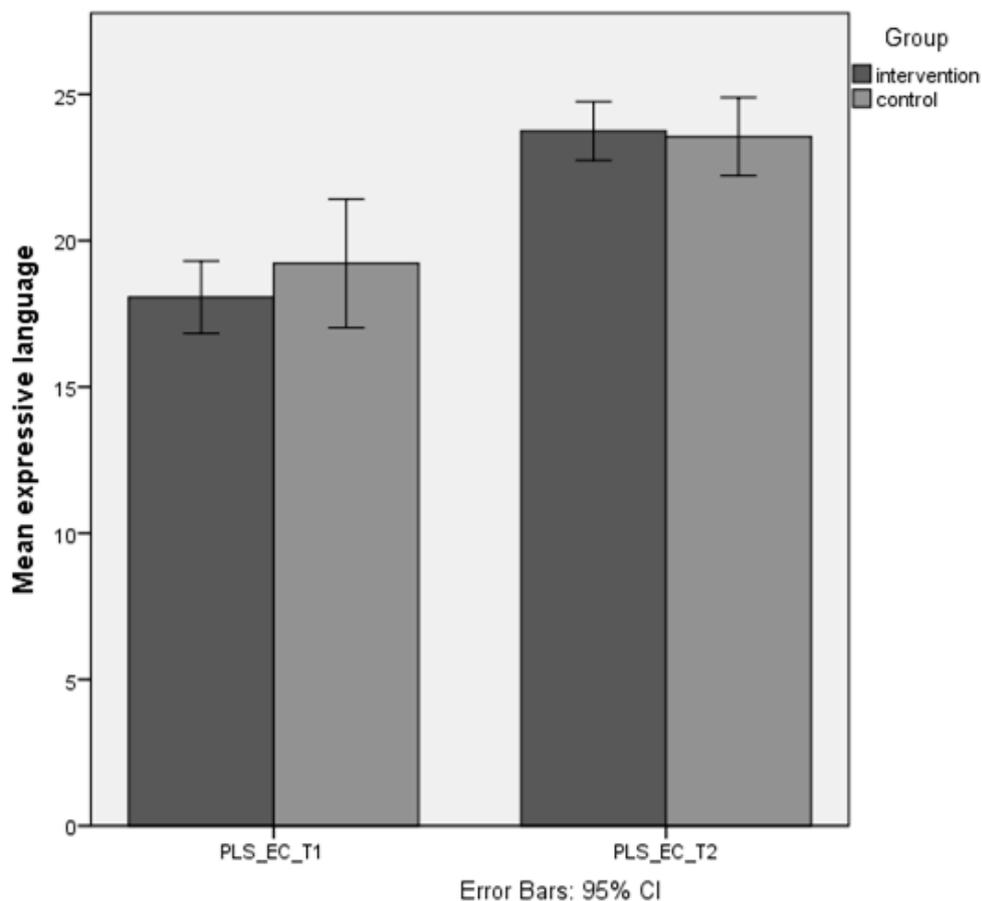


Figure 7.3: Expressive language scores pre-test and at first follow up for the intervention and the control group

7.4. Total Language Scores

The total language scores were normally distributed in both groups (see appendix G). The independence of the independent variable and covariate assumption was met for total language baseline $F(1, 25) = .393, p = .536$, as was the homogeneity of variances $F(1, 23) = .792, p = .383$. The homogeneity of regression slopes was met for age $F(2, 18) = 1.199, p = .324$ and total language baseline $F(2, 18) = 1.026, p = .379$ but was violated for non-verbal mental age $F(2, 18) = 5.958, p = .010$. As one of the assumptions was violated, a non-parametric ANCOVA was carried out. The intervention group had marginally higher scores ($M = 47.63, SD = 2.83$) than the control group ($M = 46.56, SD = 3.81$) however this difference was not significant $F(1, 23) = 2.943, p = .100, \eta^2 = .113$ (see figure 7.4).

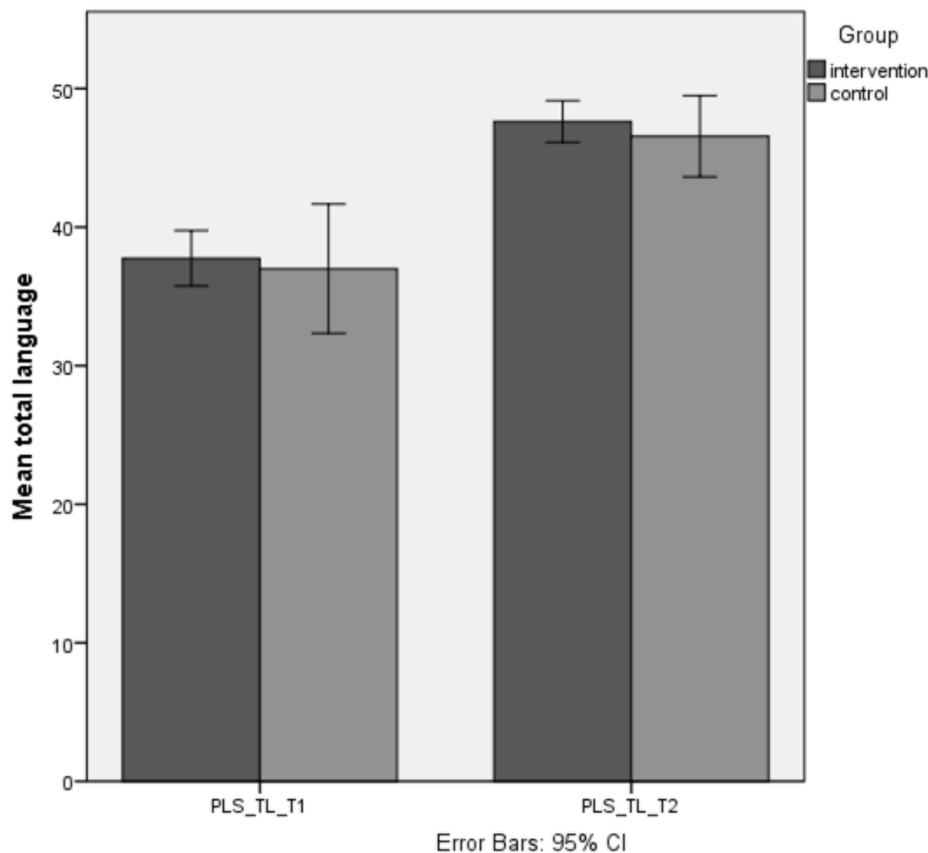


Figure 7.4: Total language scores at pre-test and first follow up for the intervention and control group

7.5. Receptive Vocabulary

Receptive vocabulary was normally distributed for both the intervention and the control group (see appendix G). The independence of the independent variable and covariate assumption was met receptive vocabulary baseline $F(1, 24)= 1.491, p= .234$. As was the homogeneity of variances $F(1, 20)= 2.345, p= .141$. The homogeneity of regression slopes was met for group*age $F(2, 15)= .172, p= .844$, group*non-verbal mental age $F(2, 15)= 1.005, p= .390$ and group*receptive language baseline $F(2, 15)= 3.428, p= .059$. The intervention group were reported to understand more words ($M= 156.8, SD= 73.04$) than the control group ($M= 93, SD= 47.66$), but this difference was not significant $F(1, 17)= 3.515, p= .078, \eta^2= .171$ (see figure 7.5).

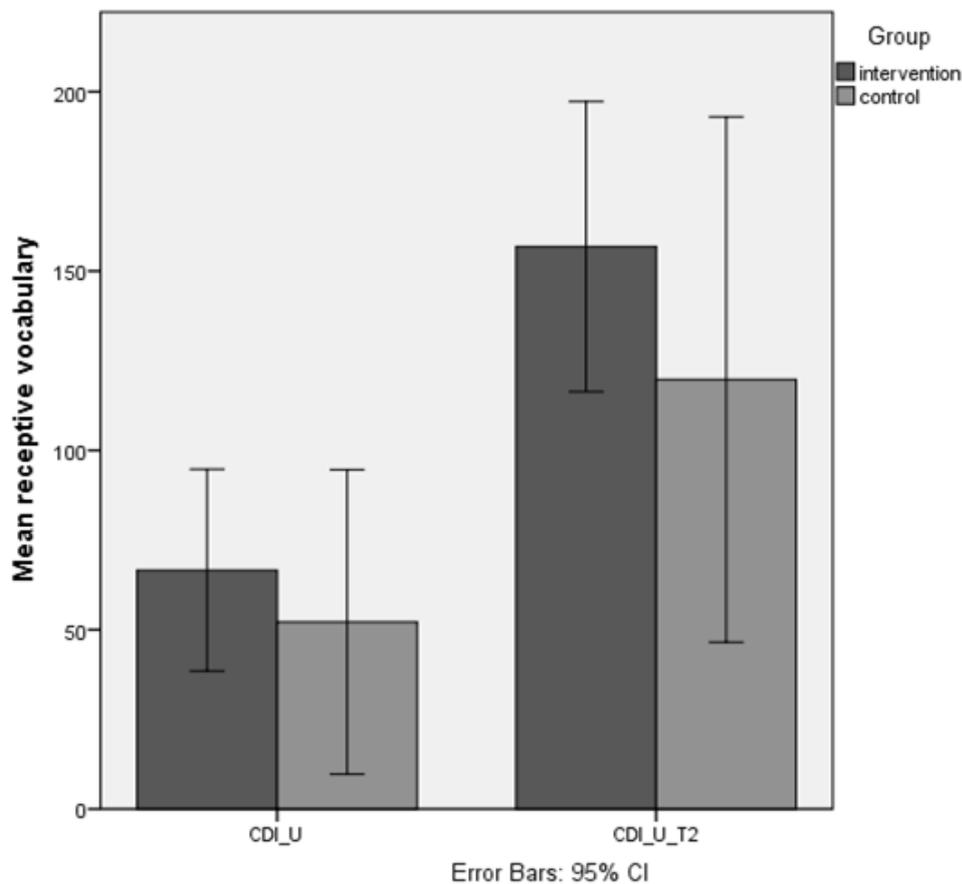


Figure 7.5: Receptive vocabulary scores pre-test and at first follow up for the intervention and control group

7.6. Expressive Vocabulary

A one-sample Kolmogorov-Smirnov test revealed that the expressive vocabulary data violated the assumption of normality in the intervention group, but the control group was normally distributed (see appendix G). This was corrected using a log transformation. The independence of the independent variable and covariate assumption was met for expressive vocabulary $F(1, 24) = 1.753, p = .198$. The Levene's test was of borderline significance $F(1, 18) = 4.399, p = .050$, however further analysis of the data revealed that variance was below the critical value and therefore the assumption was not violated. The homogeneity of regression slopes was met, group*age $F(2, 13) = .030, p = .971$, group*non-verbal mental age $F(2, 13) = .021, p = .979$, and group*expressive vocabulary baseline $F(2, 13) = 1.844, p = .197$. The intervention group were reported to produce more words ($M = 25.07, SD = 26.91$) than the control group ($M = 6.78, SD = 6.89$). However, this difference was not significant $F(1, 15) = 1.404, p = .255, \eta^2 = .086$ (see figure 7.6).

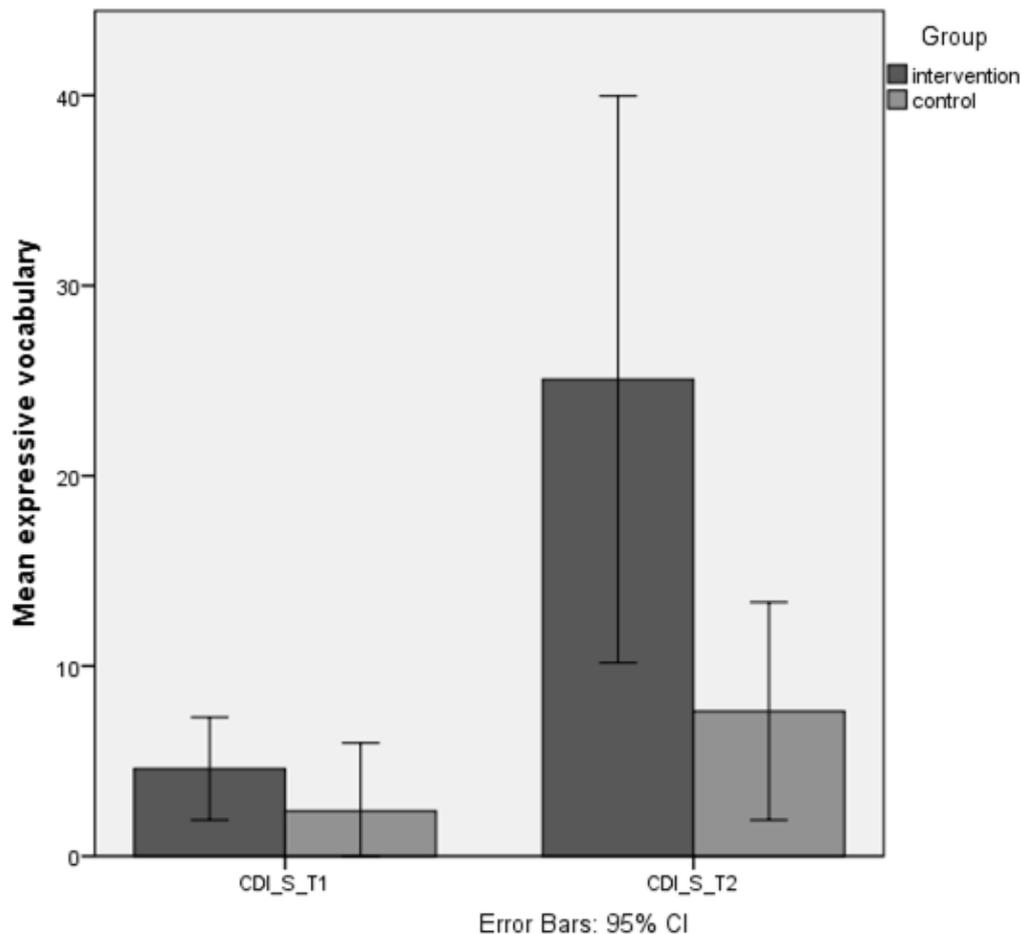


Figure 7.6: Expressive vocabulary scores at pre-test and at first follow up for the intervention and control group

7.7. Use of Sign

The assumption of normality was violated in the intervention group but not in the control group, and a log transformation was not able to correct this. The independence of the independent variable and covariate assumption was met - signs $F(1, 24) = 1.155, p = .293$, as was the homogeneity of variances $F(1, 20) = .221, p = .644$. The homogeneity of regression slopes assumption was met for group*age $F(2, 15) = .899, p = .428$ and group*non-verbal mental age $F(2, 15) = 2.637, p = .104$ but was violated for group*signs baseline $F(2, 15) = 32.757, p < .001$. As some of the assumptions were violated a non-parametric ANCOVA was carried out. The intervention group were reported to understand and use more signs ($M =$

48.13, $SD= 49.5$) than the control group ($M= 30$, $SD= 30.14$). However, this difference was not significant $F(1, 20)= .141$, $p= .711$, $\eta^2= .007$ (see figure 7.7).

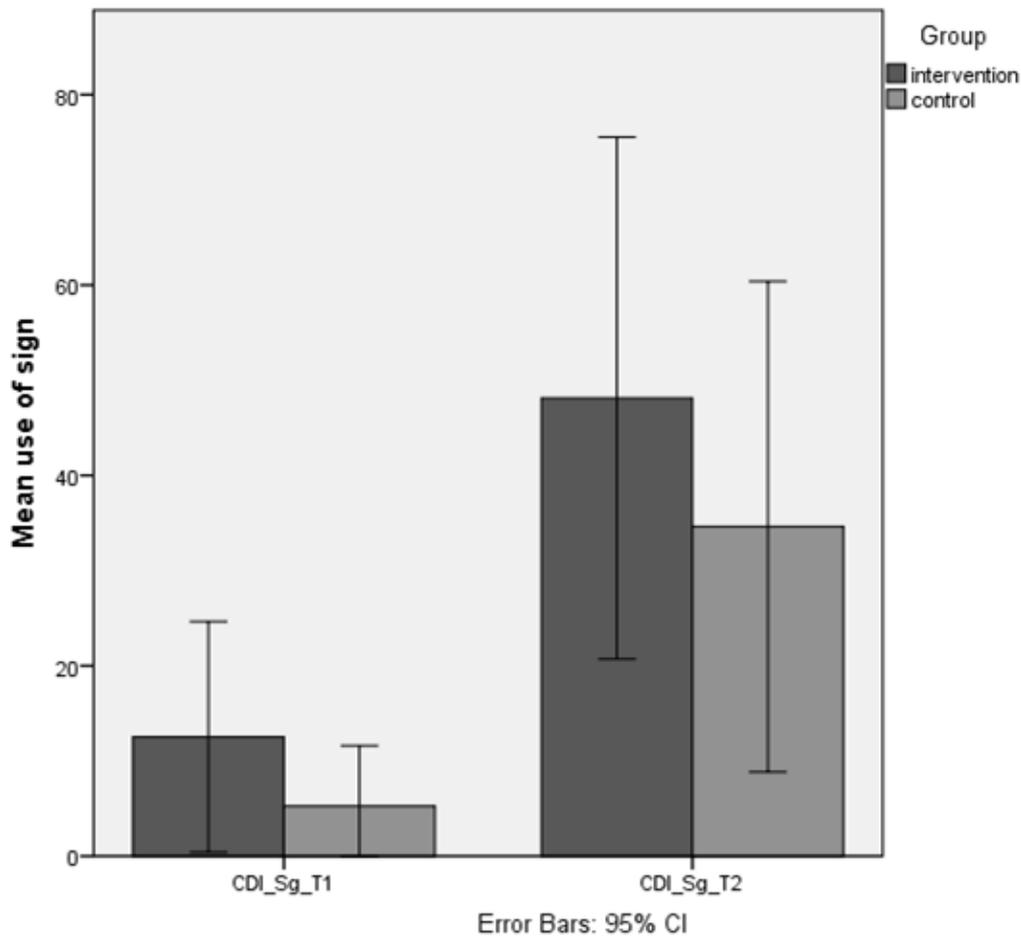


Figure 7.7: Use of signs pre-test to first follow up comparing the intervention and control group

7.8. Intervention Effects

Finally, we wanted to see if the effects of the intervention had been maintained or had declined since the participants were no longer receiving weekly intervention. This analysis focused on the intervention group only. There was a slight increase in responding to joint attention scores between the post-test ($M= 73.75$, $SD= 18.05$) and follow up ($M= 74.68$, $SD= 18.44$) but no significant difference $t(14)= -.246$, $p= .810$, $d= .05$, therefore suggesting that

the effects of the intervention had been maintained. Further investigation of the data found that 4 children obtained the same score at the post test and follow up, 6 children obtained higher scores and for 5 children the scores slightly decreased (see figure 7.8).

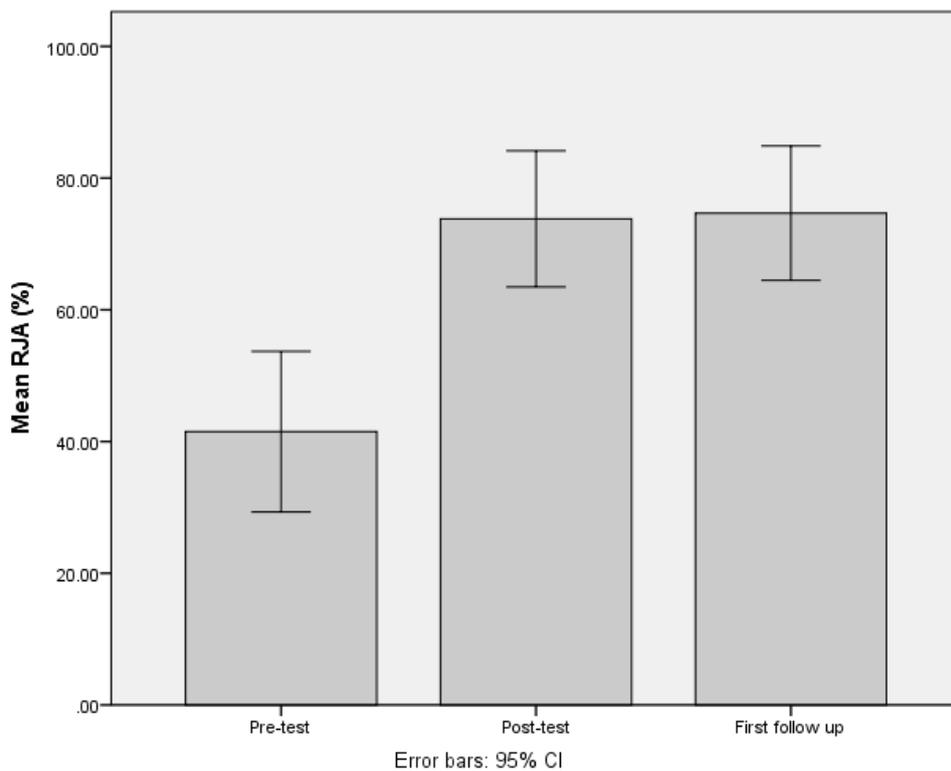


Figure 7.8: Total responding to joint attention scores at pre-test, post-test and first follow up for the intervention group

7.9. Summary

7.9.1. Intervention Results

The intervention effects seem to have been maintained between the post-test and the first follow up as there are no differences between the responding to joint attention scores from the post-test assessment and the first follow up. However, this also means no further progress has been made since the intervention sessions have stopped. This is despite the fact

that only one child out of 16 scored 100%, demonstrating there is still scope for improvement for most children in the group.

In terms of language, no differences were found for receptive or expressive language when measured by the PLS-4, as both groups had very similar results and the variability within groups was small. This is despite the initial difference in expressive language, with the control group having significantly higher scores.

Similarly, no significant differences were found between the two groups for receptive vocabulary, expressive vocabulary or use of sign measured by the Reading CDI. However, a wide variability was evident with this measure. The intervention group were reported to understand more words, produce more words and use more signs than the control group; however at this stage the difference between the intervention and control groups was not significant.

Chapter 8 - Results – Final Follow Up

This section compared the language outcomes of the intervention and control group when aged 30-35 months (which was 12 months post the initial assessment). At this stage language/vocabulary scores were the primary outcome measures. The data used formed the intervention groups' second follow up and the control groups' third assessment session and will therefore be referred to as time point 3. An ANCOVA analysis was used to investigate if there were any differences for receptive/expressive language and vocabulary when controlling for the child's age and baseline scores. Firstly, descriptive statistics were run (see table 8.1).

Table 8.1: Descriptive statistics for age, non-verbal mental age, language and vocabulary at the second follow up

	Means (SD)			
	IG	N	CG	N
Age in days	980 (54.47)	16	991.23 (43.65)	13
NVMA	43 (5.15)	16	46.23 (15.54)	13
PLS AC	26.56 (3.05)	16	30 (4.97)	13
PLS EC	26.13 (2.22)	16	26.46 (1.66)	13
PLS TL	52.69 (4.03)	16	56.46 (6.09)	13
RV	239 (100.05)	15	198.08 (107.73)	12
EV	53.67 (65.69)	15	36.42 (42.88)	12
Signs	84.47 (62.67)	15	68.91 (58.33)	11

Note. IG= intervention group, CG= control group, NVMA= non-verbal mental age, PLS AC= auditory component, PLS EC= expressive component, RV= receptive vocabulary, EV= expressive vocabulary

Initial analyses were carried out to investigate whether there were any significant differences at this stage between the intervention and control group for chronological age or non-verbal mental age. Normality tests revealed that chronological age was normally distributed for both groups, but non-verbal mental age was not normally distributed for the control group (see appendix G). Therefore, an independent t-test was used to analyse chronological age and a Mann Whitney U test for non-verbal mental age. There were no

significant differences between groups for chronological age $t(27) = -.601, p = .553, d = .23$. However, a significant difference was found for non-verbal mental age with the control group scoring significantly higher than the intervention group $U = 54.5, Z = -2.179, p = .028, r = -.4$. Outliers were removed (control group participants 2, 9 and 14) so that there were no significant differences between groups $U = 38.5, Z = -1.907, p = .057, r = -.37$. The independence of the independent variable and covariate assumption was assessed for each covariate in chapter 7 (no significant differences for baseline language, vocabulary scores, age or non-verbal mental age) and so it is not re-stated here.

8.1. *Receptive Language*

To compare the language scores at this stage an ANCOVA analysis was used controlling for the children's age and baseline performance. Initially the results for receptive language were compared. This variable was normally distributed for both groups. The Levene's test was not significant $F(1,23) = .505, p = .484$ indicating that the variance was equal across groups. The homogeneity of the regression slope was met for both age $F(2, 18) = .416, p = .666$, and baseline performance $F(2, 18) = .172, p = .843$, but was violated for non-verbal mental age $F(2, 18) = 4.818, p = .021$. As one of the assumptions was violated a non-parametric ANCOVA was used. The intervention group obtained slightly lower scores ($M = 26.56, SD = 3.05$) than the control group ($M = 28.22, SD = 3.23$), however this difference was not significant $F(1, 23) = .017, p = .897, \eta^2 = .001$ (see figure 8.1).

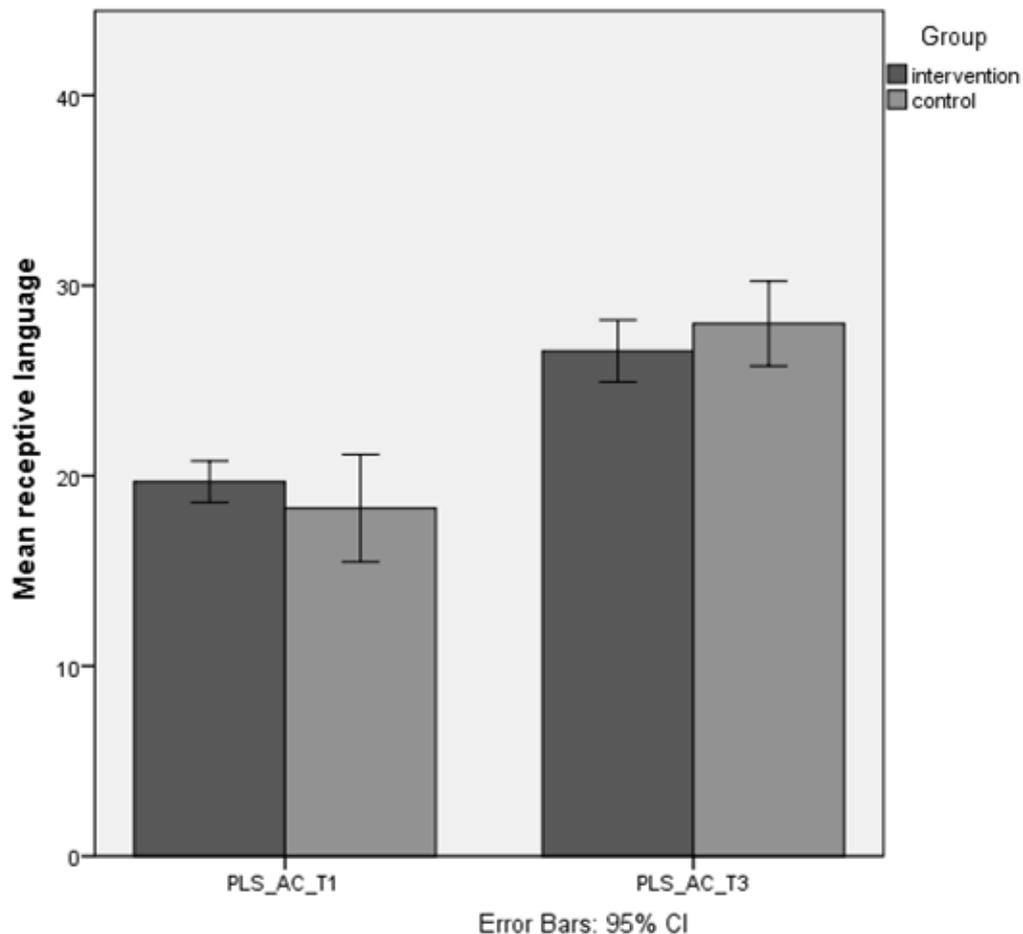


Figure 8.1: Receptive language scores on PLS-4 at the pre-test assessment and at the final follow up comparing the intervention and control group

8.2. Expressive Language

Next the expressive language scores were compared. This variable was normally distributed for both groups. The Levene's test was not significant suggesting that the variance was equal across groups $F(1, 23) = 1.624, p = .215$. The homogeneity of regression slopes was met for age $F(2, 18) = .032, p = .968$, non-verbal mental age $F(2, 18) = .275, p = .763$ and baseline scores $F(2, 18) = 1.170, p = .333$. The intervention group scored slightly higher ($M = 26.13, SD = 2.22$) than the control group ($M = 26, SD = 1.5$) but this difference was not significant $F(1, 20) = .823, p = .375, \eta^2 = .040$ (see figure 8.2).

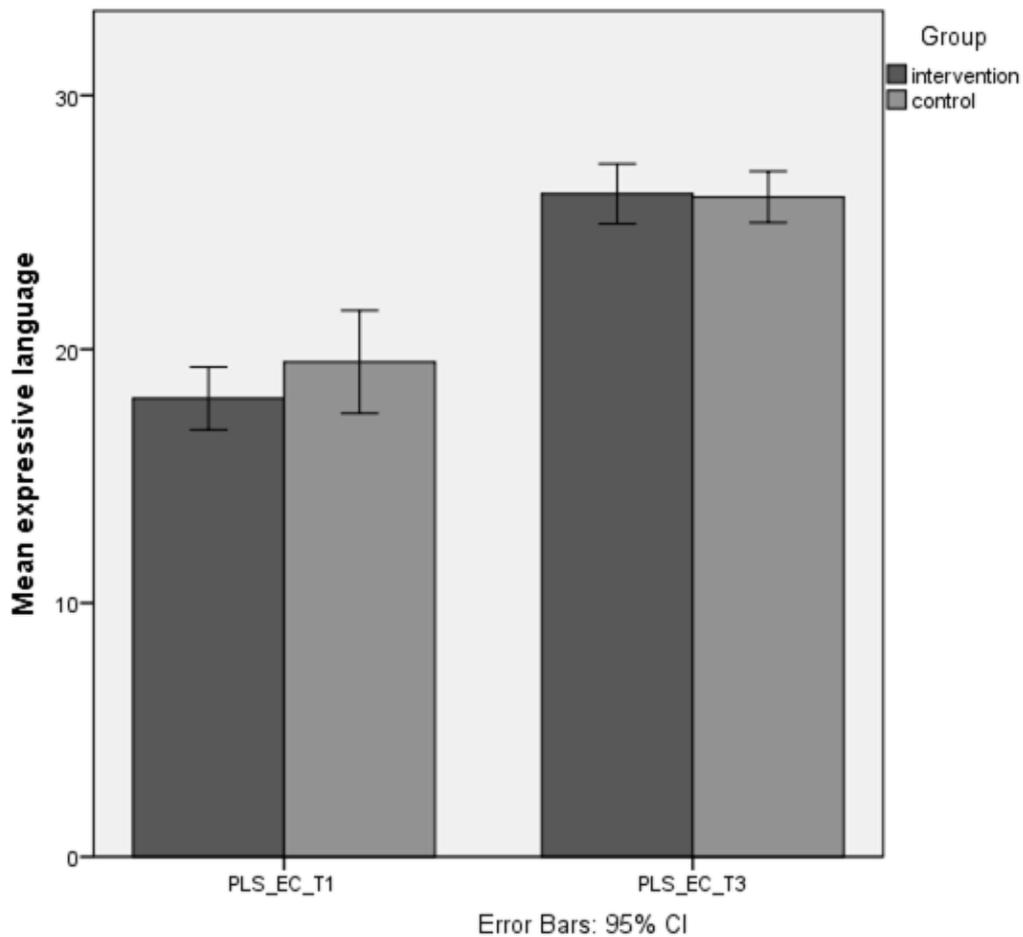


Figure 8.2: Expressive language scores on PLS-4 at the pre-test assessment and at the final follow up comparing the intervention and control group

8.3. Total Language

The total language scores were normally distributed for both groups. The Levene's test was not significant $F(1, 23) = .663, p = .424$. The homogeneity of regression slopes assumption was met for age $F(2, 18) = .218, p = .806$ and for baseline scores $F(2, 18) = 1.025, p = .379$ but not for non-verbal mental age $F(2, 18) = 4.587, p = .025$. As one of the assumptions was violated a non-parametric ANCOVA was used. The control group scored slightly higher ($M = 54.22, SD = 4.02$) than the intervention group ($M = 52.69, SD = 4.03$) but this difference was not significant $F(1, 23) = .071, p = .793, \eta^2 = .003$ (see figure 8.3).

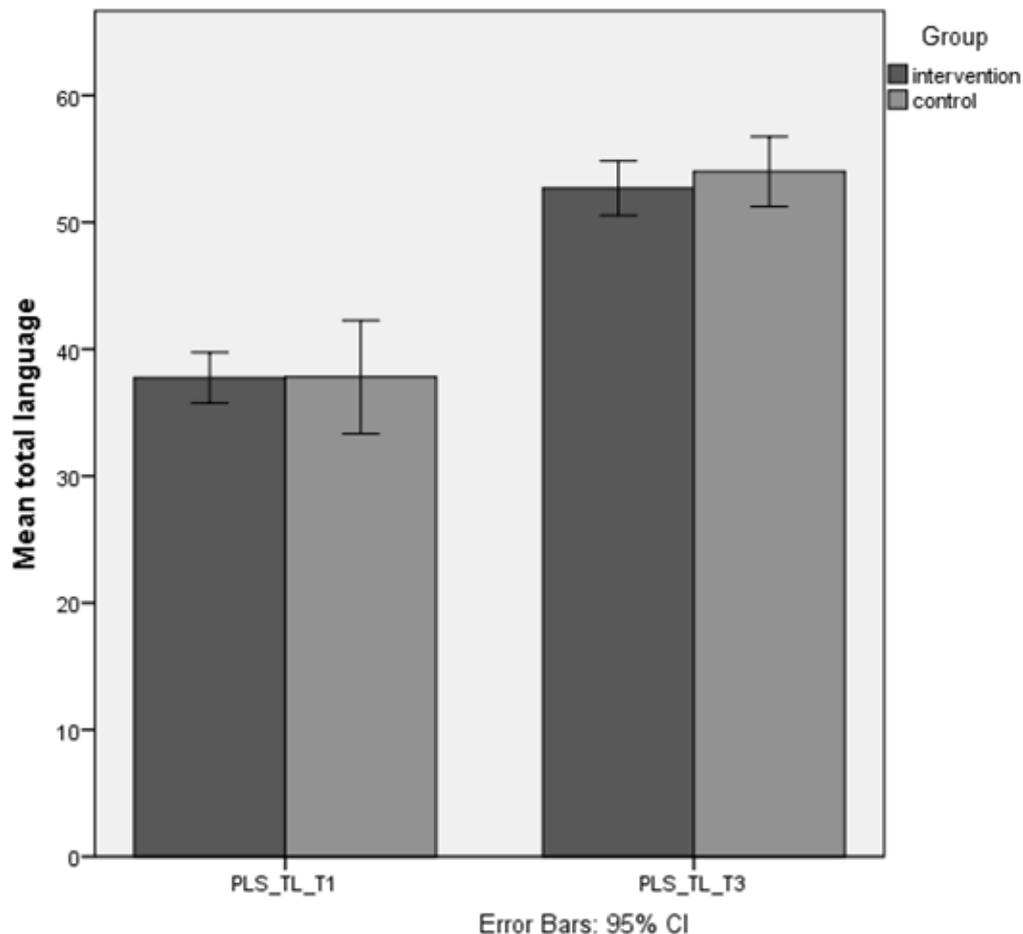


Figure 8.3: Total language scores on PLS-4 at the pre-test assessment and at the final follow up comparing the intervention and control group

8.4. *Receptive Vocabulary*

The scores for receptive vocabulary were normally distributed for both the intervention group and the control group. The Levene's test was not significant indicating that the variance between groups was equal $F(1, 21) = 3.099, p = .093$. The homogeneity of regression slopes was met – group*age $F(2, 16) = .121, p = .887$, group*non-verbal mental age $F(2, 16) = 1.295, p = .301$, and group*receptive vocabulary baseline $F(2, 16) = 2.745, p = .094$. The intervention group was reported to understand more words ($M = 239, SD = 100.05$) than the control group ($M = 133.63, SD = 59.13$) and this difference was significant $F(1, 18) = 5.978, p = .025, \eta^2 = .249$ (see figure 8.4).

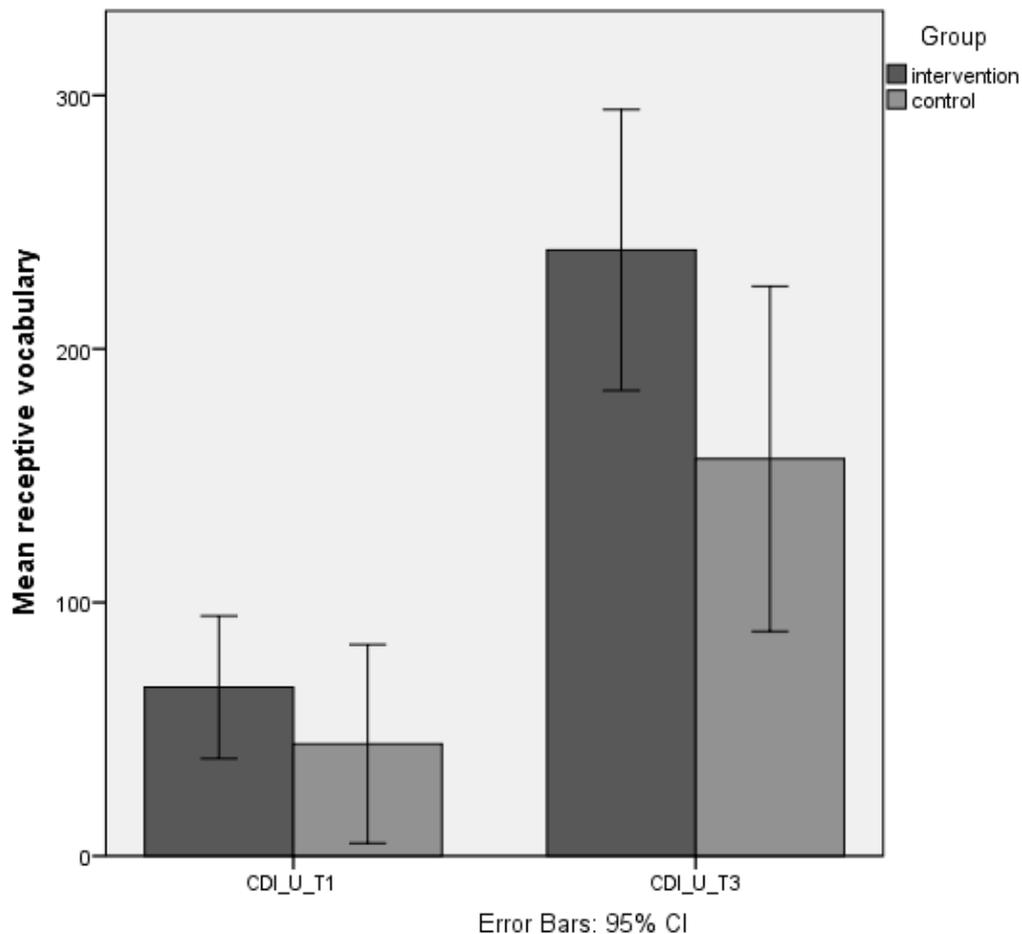


Figure 8.4: Receptive vocabulary scores on the Reading Communicative Development Inventory at the pre-test to the final follow up comparing the intervention and control group

8.5. Expressive Vocabulary

The scores for expressive vocabulary were not normally distributed for the intervention group or the control group; however a log transformation was successful in making the data normally distributed. The Levene's test was not significant $F(1, 19) = .074$, $p = .789$. The homogeneity of regression slopes was met for age*group $F(2, 14) = .373$, $p = .695$, group*non-verbal mental age $F(2, 14) = .725$, $p = .503$, and group*expressive vocabulary baseline $F(2, 14) = 3.635$, $p = .054$. Although the intervention group were reported to produce more words ($M = 53.67$, $SD = 65.69$) than the control group ($M = 23.89$, $SD = 29.7$) this difference was not significant $F(1, 16) = .127$, $p = .726$, $\eta^2 = .008$ (See figure 8.5).

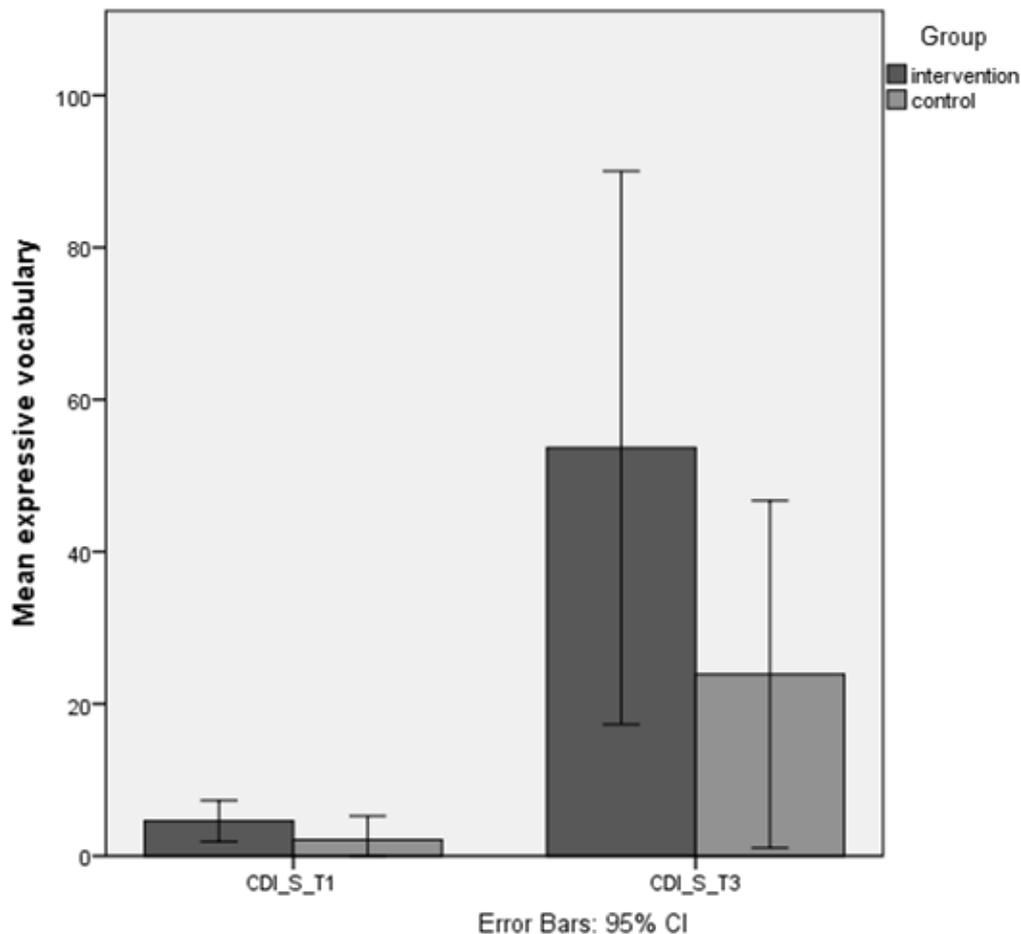


Figure 8.5: Bar chart showing expressive vocabulary from the pre-test to the final follow up comparing the intervention and control group

8.6. Use of Signs

The scores for use of signs were normally distributed for both groups. The Levene's test was not significant $F(1, 20) = 1.331, p = .262$ indicating that variance was equal across groups. The homogeneity of regression slopes was met for age $F(2, 15) = .028, p = .972$, non-verbal mental age $F(2, 15) = 2.260, p = .139$ and baseline scores $F(2, 15) = 1.768, p = .204$. Although the intervention group was reported to use more signs ($M = 84.47, SD = 62.67$) than the control group ($M = 45.86, SD = 47.42$) a significant differences was not found $F(1, 17) = 4.076, p = .06, \eta^2 = .193$ (see figure 8.6).

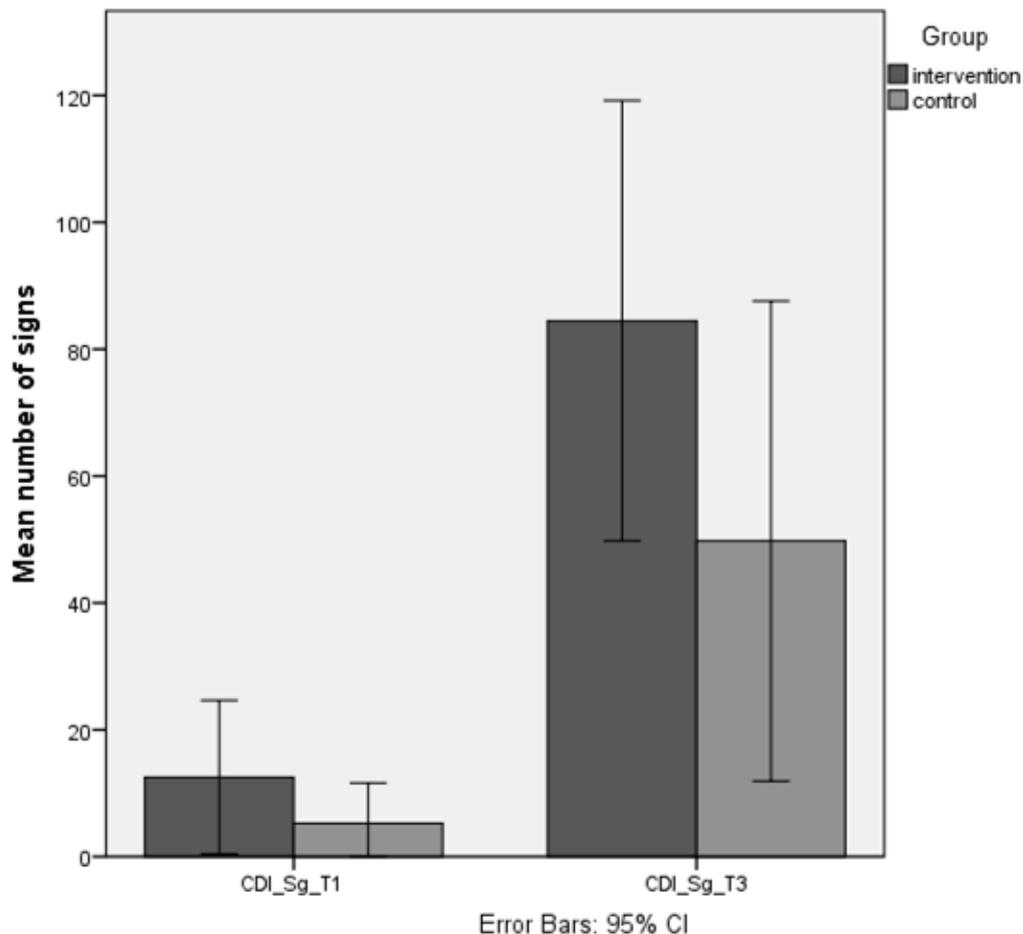


Figure 8.6: Number of signs at the pre-test and at the final follow up comparing the intervention and the control group

8.7. Summary

At the final stage, the participants were aged between 30-35 months. A significant difference was found for receptive vocabulary as measured by the CDI with the intervention group reported to understand significantly more words than the control group at this stage. This may mean that improving responding to joint attention at an earlier stage had a cascading effect on receptive vocabulary skills. There were no significant differences between groups for receptive, expressive or total language scores as measured by the PLS-4 at this time point, and similarly to results from previous chapters, there was little variability at this stage. The intervention group were reported to produce more words and signs on average

than the control group; however this difference was not significant. Similarly to the time point 2 results, there was wide variability on the CDI for both groups. The range varied greatly for how many words the children were reported to produce – in both groups there were children who were not producing any words but also children producing over 100 words. In terms of signs, some parents actively encouraged their child to sign and would use sign to communicate with them whereas other parents would not hence the number of signs are dependent on whether parents support their child's use of signs.

Chapter 9- Discussion

9.1. Gains in Responding to Joint Attention

The intervention focused on improving responding to joint attention skills in children with DS at 17-23 months with a view to improving language outcomes at 30-35 months. Initial results from an immediate post-intervention assessment were analysed to see if any improvements had been made regarding responding to joint attention. Data was only available for the intervention group at this stage of the analysis, as unfortunately there was no comparison data available. Responding to joint attention was assessed using the proximal points and gaze following tasks from the Early Social Communication Scales. Significant gains were found for total responding to joint attention, proximal points and the gaze following task therefore suggesting that the intervention may have accelerated responding to joint attention development. This supports previous research from Whalen & Schreibman (2003) who also found that this type of intervention improved responding to joint attention but for children with ASC. This would suggest that this type of intervention is feasible for individuals with DS aged between 17 and 23 months. One of the reasons why our study may have been successful is because of the specific intervention method used, or because the intervention sessions were delivered in both a clinic setting and at home and by both a

researcher and the child's primary caregiver. It has been noted that it is paramount that the child's caregiver is trained in the intervention method to ensure the targeted skills generalise (Capone, 2010). However, it is not possible to tease apart whether it is more important to have parent sessions or researcher sessions in our study.

Further analysis investigated which baseline factors may have affected which children achieved higher responding to joint attention results at the post-test. Initial non-verbal mental age and joint attention were investigated, as well as the child's score on the M-CHAT. The M-CHAT is a social communication questionnaire designed for toddlers which indicates risk of ASC. Initial maternal measures were also investigated including maternal interactive style (encompassing verbal control and elaboration, expressed emotion, intrusions, emotional tone sensitivity and reciprocity) and finally maternal well-being (anxiety and depression score). Only baseline responding to joint attention scores were found to be a significant predictor of immediate post-test responding to joint attention scores in a regression model accounting for age. However, there was a significant negative correlation between the M-CHAT and immediate post-test responding to joint attention scores, indicating that children who had a higher risk of ASC tended to score lower on the responding to joint attention tasks. Since the M-CHAT assesses early social communication skills (and some of the questions relate to joint attention skills), it is not surprising that those with worse overall social communication skills would make less progress with regards to joint attention. Joint attention is a known deficit for children with ASC (Charman, Swettenham, Baron-Cohen, Cox, Baird & Drew, 1997; Mundy, Sigman & Kasari, 1990), hence the more at risk a child is for ASC; the more likely they are to have difficulties with early social communication skills.

Recent research has identified that the co-morbidity of ASC for children with DS is around 16% (Richards, Jones, Groves, Moss & Oliver, 2015). None of the children in our study had a confirmed diagnosis of ASC, however due to the young age of participants it is

likely that such potential co-morbidities would not be identified until a later stage. This is an important consideration for future research as it is likely that the extent of ASC traits could negatively impact on a child's development in areas such as early communication skills. Two children were identified by the M-CHAT as being in the 'high risk' bracket for ASC, which equates to around 12.5% of the sample, which is in line with the percentage predicted for the population (Richards et al., 2015).

No significant correlations were found between post-test responding to joint attention scores and initial non-verbal mental age, initiating joint attention, maternal interactive style or maternal well-being. With regards to maternal well-being, the majority of mothers were scoring in the 'typical' range for levels of both anxiety and depression. A few parents scored in the 'borderline' range but none of the parents exceeded this cut-off. This is in line with previous research which has found that parents of children with DS tend to have better well-being than parents of children with other additional needs such as ASC (Dabrowska & Pisula, 2010; Eisenhower, Baker & Blacher, 2005) and fragile X syndrome (Abbeduto et al., 2004).

Responding to joint attention was assessed again at the first follow up (6 months post intervention onset) and at this stage the intervention group could be compared to the control group. The intervention group was found to have higher responding to joint attention scores than the control group but this difference was not significant. Further analysis showed no difference between the intervention groups' responding to joint attention score at the immediate post-test assessment and their score at the first follow up. This suggested that the intervention effects had been maintained but that no further progress had been made once intervention sessions had ceased. This is unlike previous studies with other clinical populations which have found that children's joint attention abilities (which were the focus of intervention) decrease once intervention sessions have finished (Whalen & Schreibman, 2003; Whalen, Schreibman & Ingersoll, 2006). This may be because participants in these

studies had ASC and joint attention is a known deficit for individuals with ASC (Charman et al., 1997; Mundy et al., 1990). However, in our study, only one child in the intervention group scored 100% for responding to joint attention at the immediate post-test and first follow up, which means that there was still scope for improvement for the majority of children. Many children were still struggling to attend to objects outside of their visual field as measured through the behind poster points on the Early Social Communication Scale. Parents reported that they were still using some of the techniques they had learnt from the intervention but the extent and quantity of this was not clear.

These findings have important implications for future research. It may be that more intervention studies are needed, or that parents require a strict plan to continue with once one-to-one sessions have finished. The results would suggest that the intervention possibly helped to accelerate responding to joint attention development and that this stage was reached earlier due to the intervention than it would have been otherwise reached. In future research, a control group needs to be included at this stage to ensure acceleration in responding to joint attention is due to the intervention and not to spontaneous development.

9.2. Language and Vocabulary Outcomes 6 months Post Intervention Onset

The language and vocabulary outcomes of the control group and the intervention group were assessed at a follow up assessment which took place 6 months after the start of the intervention. A significant difference for non-verbal mental age was found at this time point with the control group obtaining significantly higher scores than the intervention group. Further investigation revealed that this difference was driven by three outliers who were subsequently removed for the ANCOVA analysis. Age, non-verbal mental age and baseline scores were controlled for in the ANCOVA analysis. There were no significant differences between groups for language (including receptive and expressive language) as measured by the PLS-4. There was very little variability on this measure and the group scores were very

similar. In terms of vocabulary, the intervention group were reported to produce and understand more words and signs as measured by the CDI than the control group. However, this difference was not significant. In contrast to the language scores, there was great variability for all the vocabulary scores. Some children were not producing any words whereas others were producing over 50.

9.3. Language and Vocabulary Outcomes 12 months Post Intervention Onset

Language and vocabulary outcomes were compared at the third-time point for both the intervention and control group, 12 months since the beginning of the intervention. At this stage participants were aged 30-35 months. The intervention group was reported to understand more words than the control group (measured by the CDI). This difference was significant and a small to medium effect size was found. This suggests that accelerating early responding to joint attention abilities may have had a cascading effect on later receptive vocabulary. However, these results must be interpreted with caution since a randomised control design was not used. Future research where participants are randomly allocated to groups is needed to determine whether gains are due to the intervention or spontaneous development.

No significant differences were found between the two groups for receptive language, expressive language or total language scores as measured on the PLS-4. Similar to the first follow up findings, there was little variability on the PLS-4 and the groups' mean raw scores were very similar. The intervention group was reported to produce more words than the control group but this difference was not significant. There were individuals in the intervention group who were not producing any words and others who were producing over 100; this highlights the large variability in language development that is evident for individuals with DS (Buckley, 1993). The intervention group was reported to understand and use more signs at this stage but the difference was not significant.

This finding supports previous research which has found that interventions focusing on improving early joint attention skills can have a knock-on effect on later language development. For example, Whalen, Schreibman & Ingersoll (2006) found that after a joint attention intervention participants' use of spontaneous speech had increased and was at a similar level to that expected of TD children of similar mental age. The idea of targeting early precursor skills with a view to improving areas of weakness such as language has been discussed by researchers (Souza, Souza & Karmiloff-Smith, 2017) and offers support for the Pivotal Skill Hypothesis (Koegel & Frea, 1993). This hypothesis would suggest that responding to joint attention is a pivotal skill and so by targeting this in intervention may lead to broader changes in other areas, and in this case language. However, further research in the field has argued that such pivotal skills may instead mark readiness for change rather than causing a change in other areas. Instead such pivotal skills may be an important marker for individual differences and may be able to explain why specific interventions work for some individuals and not for others (Mundy & Crowson, 1997). This may explain why positive gains were not seen for all individuals who participated in the intervention. Training pivotal skills has been used with other populations, for example to improve the social skills and symbolic play of children with ASC (e.g. Pierce & Schreibman, 1997; Stahmer, 1995)

Part 3 – Early Predictors of Language Development

Chapter 10 – Concurrent and Longitudinal Predictors of Language

This chapter focused on the concurrent and longitudinal predictors of language and vocabulary for children with DS. For this purpose, the two groups were combined to form a group of 30. This increased power and allowed us to analyse the predictors and language skills of a group of 30 children with DS across 1 year when they were aged between 17-35

months. Initially, the predictors of time point 2 language and vocabulary scores will be focused on and then the predictors of time point 3 language and vocabulary scores.

10.1. *Baseline Predictors of Time Point 2 Language*

The focus of this analysis was to investigate whether any of the baseline variables taken at time point 1 when participants were 17-23 months were significant predictors of language scores at time point 2 when children were aged 24-30 months. The following baseline variables were considered – non-verbal mental age, maternal interactive style and joint attention. Initially normality tests were run and as some of the variables were not normally distributed Spearman's rho was used. The first analysis included running a correlation between the baseline language scores at Time Point 1 with language scores from Time Point 2 so that these could be controlled for in later analyses. Significant positive correlations were found between baseline language/vocabulary scores and language/vocabulary scores at time point 2 (see table 10.1).

Table 10.1: Spearman's rho correlations between baseline language scores and time point 2 language scores

	PLS AC	PLS EC	PLS TL	RV	EV	Signs
PLS AC T2	.550**					
PLS EC T2		.441*				
PLS TL T2			.572**			
RV T2				.896***		
EV T2					.569**	
Signs T2						.749***

Note. PLS= Preschool Language Scales, AC= auditory comprehension, EC= expressive communication, TL= total language, RV= receptive vocabulary, EV= expressive vocabulary, T2= time point 2, * $p < .05$, ** $p < .01$, *** $p < .001$

Next, correlations were run focusing on the baseline predictors – non-verbal mental age, joint attention and maternal interactive style. In terms of baseline predictors, receptive language significantly correlated with baseline non-verbal mental age and baseline responding to joint attention. Expressive language correlated with baseline non-verbal mental age only. Total language significantly correlated with baseline non-verbal mental age and baseline responding to joint attention. None of the baseline maternal interactive style variables or initiating joint attention significantly correlated with expressive, receptive or total language scores at this time point.

Receptive vocabulary scores significantly correlated with baseline non-verbal mental age and baseline responding to joint attention. There were no significant correlations with

expressive vocabulary scores. Use of signs significantly correlated with baseline non-verbal mental age only. Neither expressive, receptive vocabulary nor use of signs significantly correlated with baseline maternal interactive style or initiating joint attention (see table 10.2 for significant correlations, see Appendix H for full correlation matrix).

Table 10.2: Significant correlations between language/vocabulary scores at time point 2 and baseline predictors

	NVMA	RJA
PLS AC	.743***	.428*
PLS EC	.404*	-
PLS TL	.767***	.511**
RV	.678***	.437*
Signs	.575**	-

Note. NVMA= non-verbal mental age, RJA= responding to joint attention, PLS= Preschool Language Scales, AC= auditory comprehension, EC= expressive communication, TL= total language, RV= receptive vocabulary, *** - $p < .001$, ** - $p < .01$, * $p < .05$

10.2. Regression Analyses

In order to find out whether non-verbal mental age or baseline responding to joint attention would be significant predictors of receptive language, expressive language, total language or receptive vocabulary at time point 2, regression analyses were run controlling for the child's age and baseline language score.

10.2.1. *Predictor Variables of Receptive, Expressive and Total Language*

In the first analysis receptive language (time point 2) was entered as the dependent variable, age was entered into the first block, followed by baseline receptive language scores and finally non-verbal mental age baseline and responding to joint attention baseline. The first model which included just age was not significant $R^2 = .001$, $F(1,26) = .016$, $p = .009$. The second model was significantly improved by the addition of receptive language (baseline). The model was significant $R^2 = .355$, $F(2, 25) = 6.876$, $p = .004$ and receptive language (baseline) was a significant predictor. Non-verbal mental age and responding to joint attention baseline were added in the third block and this significantly improved the model. The overall model was significant $R^2 = .645$, $F(4, 23) = 10.428$, $p < .001$ and non-verbal mental age baseline was the only significant predictor (see table 10.3).

Table 10.3: Regression analysis showing time point 1 predictors of receptive language

Variable	Model 1			Model 2			Model 3		
	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β
Age	-.001	.010	-.025	.003	.008	.057	-.007	.007	-.141
PLS AC				.599	.162	.601**	.246	.155	.247
NVMA							.462	.112	.637***
RJA							.005	.018	.046
R^2 change				.354			.290		
<i>F</i> for R^2 change				13.728**			9.374**		

Note. PLS AC= Preschool Language Scales auditory comprehension, NVMA= non-verbal mental age, RJA= responding to joint attention, ** - $p < .01$, *** - $p < .001$

In the second analysis expressive language was the dependent variable; age was entered into the first block, baseline expressive language scores in the second and baseline non-verbal mental age in the third. The first model including just age was not significant $R^2 = .022$, $F(1, 26) = .590$, $p = .449$. The addition of baseline scores significantly improved the model. The model was significant $R^2 = .250$, $F(2, 25) = 4.162$, $p = .028$ and expressive language baseline was a significant predictor. Adding baseline non-verbal mental age did not significantly improve the model and the final model was not significant $R^2 = .275$, $F(3, 24) = 3.014$, $p = .050$ (see table 10.4).

Table 10.4: Regression analysis showing significant predictors for expressive language

Variable	Model 1			Model 2			Model 3		
	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β
Age	.005	.006	.149	.006	.006	.192	.005	.006	.149
PLS EC				.316	.115	.479*	.273	.125	.414*
NVMA							.075	.085	.171
R ² change				.228			.024		
F for R ² change				7.586*			.788		

Note. PLS EC= Preschool Language Scales expressive communication, NVMA= non-verbal mental age, * - $p < .05$

The following analysis assessed whether baseline non-verbal mental age or responding to joint attention would be significant predictors of total language scores when controlling for the child's age and baseline scores. Age was entered into the first block,

baseline total language in the second and baseline non-verbal mental age and responding to joint attention in the third. The first model was not significant $R^2 = .002$, $F(1, 26) = .064$, $p = .802$. The addition of baseline scores significantly improved the model. The model was significant $R^2 = .396$, $F(2, 25) = 8.193$, $p = .002$ and total language baseline was a significant predictor. The model was significantly improved by adding baseline non-verbal mental age and responding to joint attention. The model was significant $R^2 = .624$, $F(4, 23) = 9.541$, $p < .001$ and only baseline non-verbal mental age was a significant predictor (see table 10.5).

Table 10.5: Regression analysis showing significant predictors for total language scores

Variable	Model 1			Model 2			Model 3		
	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β
Age	.003	.013	.050	.009	.011	.132	-.003	.010	-.045
PLS TL				.497	.123	.633***	.244	.134	.311
NVMA							.540	.150	.567**
RJA							.007	.026	.045
R^2 change				.393			.228		
<i>F</i> for R^2 change				16.285***			6.973**		

Note. PLS TL= Preschool Language Scales total language, NVMA= non-verbal mental age, RJA= responding to joint attention, ** - $p < .01$, *** - $p < .001$

10.2.2. *Predictor Variables of Receptive Vocabulary and Use of Signs*

In the analysis that follows, receptive vocabulary was the dependent variable and the predictor variables were age, baseline receptive vocabulary, baseline non-verbal mental age and baseline responding to joint attention. The first model was not significant $R^2 = .132$, $F(1, 23) = 3.484$, $p = .075$. The addition of baseline receptive vocabulary significantly improved the model. The model was significant $R^2 = .779$, $F(2, 22) = 38.87$, $p < .001$ and receptive vocabulary baseline was a significant predictor. The final addition of baseline non-verbal mental age and responding to joint attention did not significantly improve the model. However, the model was significant $R^2 = .806$, $F(4, 20) = 20.831$, $p < .001$ but receptive vocabulary baseline was the only significant predictor (see table 10.6).

Table 10.6: Regression analysis showing significant predictors of receptive vocabulary

Variable	Model 1			Model 2			Model 3		
	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β
Age	.487	.261	.363	.120	.142	.090	.086	.141	.064
RV				1.349	.168	.850***	1.076	.245	.678***
NVMA							3.176	2.914	.159
RJA							.472	.376	.139
R^2 change				.648			.027		
<i>F</i> for R^2 change				64.620***			1.395		

Note. RV= receptive vocabulary, NVMA= non-verbal mental age, RJA= responding to joint attention, *** - $p < .001$

The final regression analysis addressed the question of whether baseline non-verbal mental age would be a significant predictor of use of signs at time point 2 controlling for the

child's age and baseline score. Age was entered into the first block, baseline scores into the second and baseline non-verbal mental age into the third. The first model was not significant $R^2 = .025$, $F(1, 24) = .615$, $p = .441$. The addition of baseline scores significantly improved the model. The model was significant $R^2 = .814$, $F(2, 23) = 50.308$, $p < .001$ and use of signs baseline was a significant predictor. Adding baseline non-verbal mental age did not significantly improve the model. However, the model was significant $R^2 = .834$, $F(3, 22) = 36.831$, $p < .001$ but the only significant predictor was baseline use of signs (see table 10.7).

Table 10.7: Regression model showing significant predictors of number of signs used and understood

Variable	Model 1			Model 2			Model 3		
	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β
Age	.119	.152	.158	.026	.068	.034	-.007	.069	-.009
Signs				2.057	.208	.897***	1.934	.215	.843***
NVMA							1.647	1.012	.160
R^2 change				.789			.020		
<i>F</i> for R^2 change				97.527***			2.652		

Note. NVMA= no-verbal mental age, *** - $p < .001$

10.3. Summary – Time Point 1 Predictors of Language Scores at Time Point 2

The correlational analysis showed that baseline language scores, non-verbal mental age and responding to joint attention were significantly associated with language and vocabulary outcomes at time point 2. Regression analyses were therefore used to reveal if

baseline non-verbal mental age or responding to joint attention were significant predictors of language outcomes controlling for baseline language scores and the child's age. Baseline non-verbal mental age was found to be a significant predictor of children's receptive language scores when controlling for age, baseline receptive language and initial responding to joint attention skills. A similar pattern was observed for total language scores where baseline non-verbal mental age emerged as a significant predictor. However, non-verbal mental age was not a significant predictor of expressive language, receptive vocabulary or use of signs. Responding to joint attention was not a significant predictor for any of the variables once age and baseline language scores were controlled for. The results would suggest that early non-verbal cognitive ability is an important factor for language skills – in particular receptive language.

10.4. *Time Point 2: Concurrent Predictors of Language/Vocabulary*

A further analysis was carried out to investigate if there was a concurrent relationship between predictor variables at time point 2 and concurrent language/vocabulary scores. The following predictor variables were used in the analysis – age, non-verbal mental age, joint attention and maternal interactive style. Initially normality tests were run and as many of the variables were not normally distributed Spearman's rho was used.

Receptive, expressive and total language scores correlated with non-verbal mental age only. Receptive vocabulary significantly correlated with non-verbal mental age, responding to joint attention, maternal sensitivity and reciprocity. Expressive vocabulary significantly negative correlated with use of verbal mild control and significantly positively correlated with positive expressed emotion. Finally use of signs correlated with non-verbal mental age only (see table 10.8 for significant correlations, see Appendix H for full correlation matrix).

Table 10.8: Significant correlations between language/vocabulary scores and concurrent predictor variables

	NVMA	RJA	Sen	Recip	Mild VC	PEEM
PLS AC	.688***					
PLS EC	.439*					
PLS TL	.747***					
RV	.599**	.427*	.565*	.475*		
EV					-.478*	.486*
Signs	.508*					

Note. NVMA= non-verbal mental age, RJA= responding to joint attention, Sen= sensitivity, Recip= Reciprocity, VC= verbal control, PEEM= positive expressed emotion, PLS= Preschool Language scales, AC= auditory comprehension, EC= expressive communication, TL= total language, RV= receptive vocabulary, EV= expressive vocabulary, *** - $p < .001$, ** - $p < .01$, * - $p < .05$,

10.4.1. Regression Analysis: Predictors of Language Scores

Next, regression analyses were run controlling for the child's age to see if any of the variables were significant predictors of language/vocabulary outcomes. Initially receptive language at time point 2 was used as the dependent variable, age was entered into the first block and non-verbal mental age into the second. The first model which included just age was not significant $R^2 = .001$, $F(1, 26) = .016$, $p = .900$. The addition of non-verbal mental age significantly improved the model and the second model was significant $R^2 = .577$, $F(2, 25) = 17.056$, $p < .001$. Non-verbal mental age was a significant predictor (see table 10.9).

Table 10.9: Regression model showing significant predictors of receptive language

Variable	Model 1			Model 2		
	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β
Age	-.001	.010	-.025	-.004	.007	-.084
NVMA				.424	.073	.762***
<i>R</i> ² change				.576		
<i>F</i> for <i>R</i> ² change				34.074***		

Note. NVMA= non-verbal mental age, *** - $p < .001$

In the second analysis, expressive language was used as the dependent variable, with age in the first step and non-verbal mental age in the second. The first model including just age was not significant $R^2 = .022$, $F(1, 26) = .590$, $p = .449$. The addition of non-verbal mental age did not significantly improve the model and the second model was not significant $R^2 = .160$, $F(2, 25) = 2.378$, $p = .113$. Neither age nor non-verbal mental age were significant predictors (see table 10.10).

Table 10.10: Regression model showing concurrent predictors of expressive language

Variable	Model 1			Model 2		
	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β
Age	.005	.006	.149	.004	.006	.120
NVMA				.125	.062	.372
<i>R</i> ² change				.138		
<i>F</i> for <i>R</i> ² change				4.095		

Note. NVMA= non-verbal mental age

Next, total language scores were used as the dependent variable. Age was entered into the first block and non-verbal mental age into the second. The first model was not significant $R^2 = .002$, $F(1, 26) = .064$, $p = .802$. The addition of non-verbal mental age significantly improved the model and the final model was significant $R^2 = .562$, $F(2, 25) = 16.039$, $p < .001$. Non-verbal mental age was a significant predictor of total language scores controlling for age (see table 10.11).

Table 10.11: Regression model showing significant predictors of total language score

Variable	Model 1			Model 2		
	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β
Age	.003	.013	.050	-.001	.009	-.009
NVMA				.550	.097	.750***
<i>R</i> ² change				.560		
<i>F</i> for <i>R</i> ² change				31.938***		

Note. NVMA= non-verbal mental age, *** - $p < .001$

10.4.2. Regression Analysis: Predictors of Receptive and Expressive Vocabulary and Use of Signs

Receptive vocabulary scores were entered as the dependent variable, with age in the first block, non-verbal mental age and responding to joint attention in the second and sensitivity and reciprocity in the third. The first model which included just age was not significant $R^2 = .073$, $F(1, 16) = 1.262$, $p = .278$. The model was significantly improved by adding non-verbal mental age and responding to joint attention and non-verbal mental age was a significant predictor. The second model was significant $R^2 = .675$, $F(3, 14) = 9.709$, $p = .001$. The addition of sensitivity and reciprocity did not significantly improve the model. However, the final model was significant $R^2 = .761$, $F(5, 12) = 7.661$, $p = .002$ but only non-verbal mental age was a significant predictor (see table 10.12).

Table 10.12: Regression model showing significant predictors of receptive vocabulary

Variable	Model 1			Model 2			Model 3		
	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β
Age	.440	.392	.270	.216	.252	.133	.358	.248	.220
NVMA				8.648	2.157	.640**	6.991	2.153	.518**
RJA				1.111	.572	.311	.694	.575	.194
Sensitivity							1.616	13.793	.027
Reciprocity							27.069	19.416	.331
<i>R</i> ² change				.602			12.987**		
<i>F</i> for <i>R</i> ² change				.086			2.165		

Note. NVMA= non-verbal mental age, RJA= responding to joint attention, ** - $p < .01$

Next, an analysis was run using expressive vocabulary as the dependent variable; age was entered into the first block followed by mild verbal control and positive expressed emotion in the second. The first model which included just age was not significant $R^2 = .000$, $F(1, 17) = .004$, $p = .948$. The model was significantly improved by the addition of verbal mild control and positive expressed emotion. Mild verbal control was a significant predictor, however the model was not significant $R^2 = .381$, $F(3, 15) = 3.074$, $p = .060$ (see table 10.13).

Table 10.13: Regression model showing significant predictors of expressive vocabulary

Variable	Model 1			Model 2		
	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β
Age	-.005	.077	-.016	.005	.066	.014
Mild VC				-.768	.310	-.504*
PEEM				1.543	.858	.371
R^2 change				.380		
<i>F</i> for R^2 change				4.608*		

Note. VC= verbal control, PEEM= positive expressed emotion, * - $p < .05$

Finally use of signs was considered in a regression model; age was entered into the first block and non-verbal mental age into the second. The first model which included just age was not significant $R^2 = .018$, $F(1, 23) = .423$, $p = .522$. The addition of non-verbal mental age did not significantly improve the model and the final model was not significant $R^2 = .159$, $F(2, 22) = 2.079$, $p = .149$ (see table 10.14).

Table 10.14: Regression model showing predictors of use of sign

Variable	Model 1			Model 2		
	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β
Age	.106	.164	.134	.090	.155	.114
NVMA				3.126	1.628	.376
<i>R</i> ² change				.141		
<i>F</i> for <i>R</i> ² change				3.686		

Note. NVMA= non-verbal mental age

10.5. *Summary - Concurrent Predictors of Language at Time Point 2*

This analysis was used to investigate whether any of the concurrent predictor variables (non-verbal mental age, joint attention and maternal interactive style) were significantly related to language/vocabulary scores. Non-verbal mental age was found to be a significant predictor of both receptive and total language scores, accounting for the child's age. Although non-verbal mental age significantly correlated with expressive language, it was not a significant predictor when entered into a regression model.

Non-verbal mental age, responding to joint attention, maternal sensitivity and reciprocity (during a mother-child interaction) significantly correlated with receptive vocabulary. However, when entered into a regression model only non-verbal mental age was a significant predictor. Positive expressed emotion significantly positively correlated with expressive vocabulary but mild verbal control had a significant negative relationship with non-verbal mental age. When entered into a regression model the addition of positive expressed emotion and mild verbal control significantly improved the model, mild verbal

control was a significant predictor but the model was not significant. Non-verbal mental age significantly correlated with use of signs but was not a significant predictor when entered into a regression model.

10.6. *Baseline Predictors of Time Point 3 Language Scores*

This analysis was done to see if any of the baseline variables would be significantly associated with language/vocabulary outcomes at time point 3 when participants were aged 30-35 months. The following baseline variables were included in the analysis – non-verbal mental age, joint attention, maternal interactive style and language/vocabulary. Spearman's rho was used as some of the variables were not normally distributed. Initial correlations were run to see if the baseline language measures correlated with outcomes at time point 3 so that these could be controlled for in later analyses. All the baseline measures correlated with the respective outcomes at time point 3 (see table 10.15).

Table 10.15: correlation analysis between baseline language scores and language scores at time point 3

	PLS AC	PLS EC	PLS TL	RV	EV	Signs
PLS AC T3	.373*					
PLS EC T3		.495*				
PLS TL T3			.569**			
RV T3				.798***		
EV T3					.463*	
Signs T3						.625**

Note. PLS= Preschool Language Scales, AC= auditory comprehension, EC= expressive communication, TL= total language, RV= receptive vocabulary, EV= expressive vocabulary, T3= time point 3, * - $p < .05$, ** - $p < .01$, *** - $p < .001$

Further correlations were then run to establish whether baseline non-verbal mental age, joint attention or maternal interactive style would significantly correlate with language outcomes. Receptive language significantly correlated with non-verbal mental age. Expressive language significantly correlated with non-verbal mental age and responding to joint attention. Total language significantly correlated with non-verbal mental age, responding to joint attention and verbal elaboration. Receptive vocabulary significantly correlated with non-verbal mental age and responding to joint attention. Expressive vocabulary significantly correlated with non-verbal mental age. Use of signs significantly correlated with non-verbal mental age (see table 10.16, see Appendix H for full matrix).

Table 10.16: Significant correlations between language/vocabulary scores and baseline predictor variables

	NVMA	RJA	Verbal Elaboration
PLS AC	.459*		
PLS EC	.478**	.459*	
PLS TL	.529**	.399*	.482*
RV	.677***	.392*	
EV	.651***		
Signs	.501**		

Note. NVMA= non-verbal mental age, RJA= responding to joint attention, PLS= Preschool Language Scales, AC= auditory comprehension, EC= expressive communication, TL= total language, RV= receptive vocabulary, EV= expressive vocabulary, * - $p < .05$, ** $p < .01$, *** - $p < .001$

10.6.1. *Regression Analysis – Predictors of Expressive and Receptive Language*

Next regression analyses were run for each of the language outcomes to see which variables were significant predictors. The children's age was controlled for in each analysis and the baseline language score if a significant correlation had been found.

In the first analysis receptive language was the dependent variable; age was entered into the first block, receptive language baseline into the second and non-verbal mental age into the third. The first model including the child's age only was not significant $R^2 = .003$, $F(1, 27) = .072$, $p = .790$. The addition of baseline receptive language did not significantly improve the model and the model was not significant $R^2 = .124$, $F(2, 26) = 1.842$, $p = .179$. The model was significantly improved by adding baseline non-verbal mental age. The model was significant $R^2 = .291$, $F(3, 25) = 3.413$, $p = .033$ and non-verbal mental age baseline was a significant predictor (see table 10.17).

Table 10.17: Regression analysis showing significant predictors of receptive language

Variable	Model 1			Model 2			Model 3		
	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β
Age	-.005	.017	-.052	.001	.016	.009	-.009	.016	-.108
PLS AC				.001	.016	.009	.109	.303	.074
NVMA							.515	.213	.497*
R^2 change				.121			.166		
<i>F</i> for R^2 change				3.605			5.865*		

Note. PLS AC= Preschool Language Scales auditory comprehension, NVMA= non-verbal mental age, * - $p < .05$

The next analysis focused on expressive language, age was entered into the first block, expressive language baseline into the second and non-verbal mental age and responding to joint attention into the third. The first model including just age was not significant $R^2 = .015$, $F(1, 27) = .409$, $p = .528$. The second model which added baseline scores was marginally significant $R^2 = .201$, $F(2, 26) = 3.266$, $p = .054$. The final model which added baseline non-verbal mental age and responding to joint attention was not significant $R^2 = (4, 24) = 2.536$, $p = .067$ (see table 10.18).

Table 10.18: Regression analysis showing significant predictors of expressive language

Variable	Model 1			Model 2			Model 3		
	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β
Age	.005	.008	.122	.005	.007	.115	.002	.007	.055
PLS EC				.317	.129	.431*	.192	.149	.261
NVMA							.140	.093	.297
RJA							.009	.017	.106
R^2 change				.186			.096		
<i>F</i> for R^2 change				6.046*			1.629		

Note. PLS EC= Preschool Language Scales expressive communication, NVMA= non-verbal mental age, RJA= responding to joint attention, * - $p < .05$

Next total language was entered as the dependent variable; age was in the first block, total language baseline in the second and non-verbal mental age, responding to joint attention and verbal elaboration in the third. The first model including just age was not significant $R^2 =$

.000, $F(1, 22) = .002$, $p = .966$. The addition of baseline total language significantly improved the model. The model was significant $R^2 = .338$, $F(2, 21) = 5.351$, $p = .013$ and baseline total language was a significant predictor. The final model was not significantly improved by adding non-verbal mental age, responding to joint attention and verbal elaboration. However, the model was significant $R^2 = .483$, $F(5, 18) = 3.358$, $p = .026$ but none of the predictors were significant (see table 10.19).

Table 10.19: Regression analysis showing significant predictors of total language

Variable	Model 1			Model 2			Model 3		
	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β
Age	-.001	.024	-.009	-.003	.020	-.029	-.004	.023	-.037
PLS TL				.701	.214	.581**	.433	.264	.359
NVMA							.417	.286	.300
RJA							.029	.049	.129
VE							1.190	.982	.232
R^2 change				.337			.145		
<i>F</i> for R^2 change				10.699**			1.682		

Note. PLS TL= Preschool Language Scales total language, NVMA= non-verbal mental age, RJA= responding to joint attention, VE= verbal elaboration, ** - $p < .01$

10.6.2. *Regression Analysis – Predictors of Receptive and Expressive Vocabulary and Use of Signs*

Further regression analyses were conducted to see if any of the baseline variables were predictive of vocabulary outcomes at time point 3. Initially receptive vocabulary was entered as the dependent variable; age was put into the first block, baseline receptive vocabulary in the second, followed by non-verbal mental age and responding to joint attention in the third.

The first model including just age was not significant $R^2 = .024$, $F(1, 25) = .613$, $p = .441$. The addition of baseline scores significantly improved the model. The model was significant $R^2 = .613$, $F(2, 24) = 19.035$, $p < .001$ and baseline scores were a significant predictor. The addition of non-verbal mental and responding to joint attention did not significantly improve the model. However, the overall model was significant $R^2 = .634$, $F(4, 22) = 9.508$, $p < .001$ but only receptive vocabulary baseline was a significant predictor (see table 10.20).

Table 10.20: Regression analysis showing significant predictors of receptive vocabulary

Variable	Model 1			Model 2			Model 3		
	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β
Age	.319	.407	.155	-.118	.271	-.057	-.078	.279	-.038
RV BL				1.556	.257	.796***	1.215	.429	.622*
NVMA							5.472	4.974	.227
RJA							-.047	.643	-.011
R^2 change				.589			.020		
F for R^2 change				36.583***			.606		

Note. RV= receptive vocabulary, BL= baseline, NVMA= non-verbal mental age, RJA= responding to joint attention, * - $p < .05$, *** - $p < .001$

Next, expressive vocabulary was the dependent variable; age was put into the first block, baseline expressive vocabulary in the second and non-verbal mental age in the third. The first model which included age only was not significant $R^2 = .026$, $F(1, 25) = .673$, $p = .420$. The next model which added baseline expressive vocabulary was not significant $R^2 = .192$, $F(2, 24) = 2.855$, $p = .077$. Finally, the last model which added non-verbal mental age was also not significant $R^2 = .251$, $F(3, 23) = 2.566$, $p = .079$ (see table 10.21).

Table 10.21: Regression analysis showing significant predictors of expressive vocabulary

Variable	Model 1			Model 2			Model 3		
	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β
Age	.182	.221	.162	.205	.206	.183	.160	.205	.143
EV BL				5.150	2.319	.408*	3.867	2.474	.306
NVMA							3.470	2.588	2.65
R^2 change				.166			.059		
<i>F</i> for R^2 change				4.932*			1.798		

Note. EV= expressive vocabulary, BL= baseline, NVMA= non-verbal mental age, * - $p < .05$

Finally use of signs was entered as the dependent variable; with age in the first block, baseline signs in the second and non-verbal mental age in the third. The first model was not significant $R^2 = .014$, $F(1, 24) = .351$, $p = .559$. The addition of baseline scores significantly improved the model. The model was significant $R^2 = .467$, $F(2, 23) = 10.066$, $p = .001$ and baseline use of signs was a significant predictor. The final addition of non-verbal mental age

did not significantly improve the model. However, the model was significant $R^2 = .518$, $F(3, 22) = 7.893$, $p = .001$ but only baseline signs was a significant predictor (see table 10.22).

Table 10.22: Regression analysis showing significant predictors of number of signs used and understood

Variable	Model 1			Model 2			Model 3		
	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β
Age	.141	.238	.120	.057	.180	.049	.030	.176	.025
Signs BL				2.090	.473	.676***	1.806	.496	.584**
NVMA							3.396	2.212	.247
R^2 change				.452			.052		
<i>F</i> for R^2 change				19.511***			2.358		

Note. BL= baseline, NVMA= non-verbal mental age, ** - $p < .01$, *** - $p < .001$

10.7. Summary – Time Point 1 Predictors of Language Outcomes at Time Point 3

The regression analysis was used to see if any of the baseline variables that significantly correlated with time point 3 language or vocabulary outcomes would be significant predictors of language and vocabulary outcomes at time point 3 controlling for age and baseline scores. For the majority of the outcomes it was found that precursor variables were not significant when accounting for baseline language/vocabulary scores. This was the case for expressive language, total language, receptive vocabulary, expressive vocabulary and use of signs. However, initial non-verbal mental age was a significant predictor of receptive language outcomes at time point 3 in a model accounting for age and

baseline receptive language scores. This suggests that early non-verbal cognitive ability is an important factor for later language development for children with DS.

10.8. *Time Point 2 Predictors of Time Point 3 Outcomes*

A further analysis was completed to see if any of the time point 2 predictor variables (when the children were between 24 and 29 months of age) were associated with time point 3 language/vocabulary outcomes (when the children were between 30 and 35 months). Initially correlations were carried out to see if any of the time point 2 language scores correlated with time point 3 scores so that these could be controlled for in an analysis (see table 10.23).

Table 10.23: Spearman's rho correlations between language/vocabulary scores at time points 2 and 3

	PLSAC2	PLSEC2	PLSTL2	RV2	EV2	Signs2
PLS AC3	.702***					
PLS EC3		.617***				
PLS TL3			.697***			
RV3				.920***		
EV3					.570**	
Signs3						.777***

Note. PLS= Preschool Language Scales, AC= auditory comprehension, EC= expressive communication, TL= total language, RV= receptive vocabulary, EV= expressive vocabulary, 2= time point 2, 3= time point 3, * - $p < .05$, ** - $p < .01$, *** - $p < .001$

Next correlations were run between language/vocabulary outcomes and predictor variables. Spearman's rho was used as many of the variables were not normally distributed. Receptive language significantly correlated with non-verbal mental age and responding to joint attention. Expressive language and total language significantly correlated with non-verbal mental age only. Receptive vocabulary correlated with non-verbal mental age, responding to joint attention and reciprocity. Expressive vocabulary did not correlate with any of the predictor variables. Use of signs significantly correlated with non-verbal mental age (see table 10.24, see Appendix H for full correlation matrix).

Table 10.24: Significant correlations between language/vocabulary scores and time point 2 predictor variables

	NVMA	RJA	Reciprocity
PLS AC	.717***	.409*	
PLS EC	.447*		
PLS TL	.691***		
RV	.473*	.403*	.484*
Signs	.564**		

Note. NVMA= non-verbal mental age, RJA= responding to joint attention, PLS= Preschool Language Scales, AC= auditory comprehension, EC= expressive communication, TL= total language, RV= receptive vocabulary, *** - $p < .001$, ** - $p < .01$, * - $p < .05$

10.8.1. *Regression – Predictors of Receptive and Expressive Language Scores*

Regression analyses were run controlling for the child's age and language/vocabulary score at time point 2. Initially receptive language was entered as the dependent variable; with age in the first block, receptive language time point 2 in the second and non-verbal mental age and responding to joint attention in the third block. The first model including just age was not significant $R^2 = .001$, $F(1, 25) = .014$, $p = .906$. The addition of time point 2 receptive language scores significantly improved the model and the second model was significant $R^2 = .694$, $F(2, 24) = 27.179$, $p < .001$. The model was improved further by adding non-verbal mental age and the final model was significant $R^2 = .777$, $F(4, 22) = 19.155$, $p < .001$. Both time point 2 receptive language and non-verbal mental age were significant predictors (see table 10.25).

Table 10.25: Regression model showing significant time point 2 predictors of receptive language outcomes

Variable	Model 1			Model 2			Model 3		
	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β
Age	-.002	.018	-.024	.003	.010	.032	-.005	.009	-.053
PLS AC				1.259	.171	.834***	.728	.252	.483**
NVMA							.386	.135	.459**
RJA							-.001	.022	-.003
R^2 change				.693			.083		
F for R^2 change				54.313***			4.103*		

Note. PLS AC= Preschool Language Scales auditory comprehension, NVMA= non-verbal mental age, RJA= responding to joint attention, * - $p < .05$, ** - $p < .01$, *** - $p < .001$

Next expressive language was entered as the dependent variable; with age in the first block, time point 2 expressive language in the second and non-verbal mental age in the third. The first model which included just age was not significant $R^2 = .017$, $F(1, 26) = .456$, $p = .506$. The addition of time point 2 expressive language significantly improved the model and the second model was also significant $R^2 = .342$, $F(2, 25) = 6.494$, $p = .005$. The model did not significantly improve when non-verbal mental age was added. The final model was significant $R^2 = .396$, $F(3, 24) = 5.250$, $p = .006$ and only time point 2 expressive language was a significant predictor (see table 10.26).

Table 10.26: Regression model showing significant time point 2 predictors of expressive language outcomes

Variable	Model 1			Model 2			Model 3		
	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β
Age	.005	.008	.131	.003	.007	.077	.002	.007	.051
PLS EC				.653	.186	.572**	.546	.196	.478*
NVMA							.098	.066	.254
R^2 change				.325			.054		
<i>F</i> for R^2 change				12.335**			2.16		

Note. PLS EC= Preschool Language Scales expressive communication, NVMA= non-verbal mental age, * - $p < .05$, ** - $p < .01$

In the next analysis total language scores were entered as the dependent variable; with age in the first block, time point 2 total language in the second and non-verbal mental age in the third. The first model which included just age was not significant $R^2 = .001$, $F(1, 26) =$

.013, $p = .909$. The inclusion of time point 2 total language significantly improved the model and the second model was significant $R^2 = .593$, $F(2, 25) = 18.249$, $p < .001$. The addition of non-verbal mental age further improved the model. The final model was significant $R^2 = .734$, $F(3, 24) = 22.051$, $p < .001$ and both non-verbal mental age and time point 2 total language scores were significant predictors (see table 10.27).

Table 10.27: Regression model showing significant time point 2 predictors of total language outcomes

Variable	Model 1			Model 2			Model 3		
	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β
Age	.002	.022	.023	.003	.014	.029	-.006	.012	-.055
PLS TL				1.091	.181	.770***	.474	.229	.335*
NVMA							.602	.169	.580**
R^2 change				.593			.140		
<i>F</i> for R^2 change				36.466***			12.648**		

Note. PLS TL= Preschool Language Scales total language, NVMA= non-verbal mental age,

* - $p < .05$, ** - $p < .01$, *** - $p < .001$

10.8.2. Regression – Predictors of Receptive Vocabulary and Use of Signs

Initially receptive vocabulary was entered as the dependent variable, with age in the first block, time point 2 receptive vocabulary in the second and non-verbal mental age, responding to joint attention and reciprocity in the third. The first model which included age

was not significant $R^2 = .010$, $F(1, 15) = .150$, $p = .704$. The addition of time point 2 receptive vocabulary significantly improved the model and the second model was significant $R^2 = .914$, $F(2, 14) = 74.756$, $p < .001$. The model was not further improved by the addition of non-verbal mental age, responding to joint attention and reciprocity. The final model was significant $R^2 = .934$, $F(5, 11) = 31.196$, $p < .001$ but only time point 2 receptive vocabulary was significant (see table 10.28).

Table 10.28: Regression model showing significant time point 2 predictors of receptive vocabulary outcomes

Variable	Model 1			Model 2			Model 3		
	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β
Age	.198	.511	.100	-.091	.157	-.046	-.006	.173	-.003
RV				1.142	.094	.962***	.869	.181	.733**
NVMA							1.899	1.909	.118
RJA							.493	.417	.110
Reciprocity							12.399	10.717	.124
R^2 change				.904			.020		
<i>F</i> for R^2 change				147.892***			1.099		

Note. RV= receptive vocabulary, NVMA= non-verbal mental age, RJA= responding to joint attention, ** - $p < .01$, *** - $p < .001$

As none of the time point 2 predictor variables significantly correlated with expressive language outcomes a regression analysis was not run. In the final regression model

use of signs was entered as the dependent variable; age was entered into the first block, time point 2 signs into the second and non-verbal mental age into the third. The first model which incorporated age was not significant $R^2 = .618$, $F(1, 22) = .641$, $p = .432$. The addition of time point 2 use of signs significantly improved the model and the second model was significant $R^2 = .618$, $F(2, 21) = 16.991$, $p < .001$. Adding non-verbal mental age did not improve the model. The final model was significant $R^2 = .659$, $F(3, 20) = 12.909$, $p < .001$ and time point 2 use of signs was a significant predictor (see table 10.29).

Table 10.29: Regression model showing significant time point 2 predictors of use of sign

Variable	Model 1			Model 2			Model 3		
	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β	<i>B</i>	<i>SE</i>	β
Age	.209	.261	.168	.121	.168	.097	.098	.163	.079
Signs				1.052	.185	.771***	.939	.193	.689***
NVMA							2.467	1.582	.221
R^2 change				.590			.041		
F for R^2 change				32.425***			2.430		

Note. NVMA= non-verbal mental age, *** - $p < .001$

10.9. Summary – Time Point 2 Predictors of Language/Vocabulary Outcomes

This analysis investigated if any of the time point 2 predictor variables would be significant predictors of language/vocabulary outcomes when controlling for age and time point 2 scores. Receptive language was found to significantly correlate with non-verbal mental age and responding to joint attention. When entered into a regression model non-

verbal mental age was a significant predictor in a model accounting for age, time point 2 scores and responding to joint attention. Expressive language correlated with non-verbal mental age only but non-verbal mental age was not a significant predictor. Total language also correlated with non-verbal mental age only. In a regression model non-verbal mental age was a significant predictor of total language scores controlling for age and time point 2 scores.

Receptive vocabulary significantly correlated with non-verbal mental age, responding to joint attention and reciprocity and these variables were entered in a regression model. None of the variables were significant predictors. Expressive vocabulary did not significantly correlate with any of the predictor variables and so a regression was not run. Finally, use of signs correlated with non-verbal mental age only but non-verbal mental age was not a significant predictor when entered into a model accounting for age and time point 2 scores.

10.10. *Discussion - Concurrent and longitudinal predictors of language and vocabulary*

10.10.1. *Factors Associated with Language/Vocabulary Development at 23-30 months*

The analysis in part 3 was completed to see if any time point 1 (longitudinal) or time point 2 (concurrent) factors were associated with time point 2 language/vocabulary scores when participants were aged 23-30 months or time point 3 language/vocabulary scores when participants were aged 30-35 months. The whole group of 30 children were included in this analysis. A longitudinal relationship was found between baseline non-verbal mental age, responding to joint attention and language/vocabulary outcomes at time point 2. When entered into a regression model, non-verbal mental age was a significant predictor of receptive language and total language scores in a model controlling for age, baseline

language scores and responding to joint attention. In the same analysis, responding to joint attention was not a significant predictor. This highlights that both early non-verbal cognitive skills and responding to joint attention are important factors for later language development. This is in line with previous research which has also found that responding to joint attention is important for concurrent and longitudinal language outcomes for children with DS (Mason-Apps et al. 2013; Sigman & Ruskin, 1999) and TD children (Morales, Mundy and Rojas, 1998; Mundy & Gomes, 1998).

A concurrent relationship was found between non-verbal mental age and receptive and total language scores. Non-verbal mental age was found to be a significant predictor accounting for the child's age. Non-verbal mental age, responding to joint attention, maternal sensitivity and reciprocity (during a mother-child interaction) significantly correlated with receptive language. However, only non-verbal mental age was a significant predictor when entered into a regression model.

The results highlight that concurrent non-verbal mental age is important for receptive language, receptive vocabulary and use of signs. This is in line with research for TD children which suggests that various early cognitive processes underpin and facilitate language development e.g. auditory processing and working memory (Baddeley, 2003; Deak, 2014). It is also in line with Piaget's (1971) theory of cognitive development which suggests that general cognitive development facilitates a child's language skills. Furthermore, this suggests that for children with DS language and non-verbal mental age may develop in synchrony.

Although there have been cases of reported asynchrony between language and other cognitive abilities (e.g. Van Der Lely, 1997), recent research in syndromic disorders such as DS and Williams syndrome suggests that language is generally in line with non-verbal mental age (Alfieri et al., 2017; Grieco, Pulsifer, Seligsohn, Skotko & Schwartz, 2015).

There has also been research into whether non-verbal mental age is related to language abilities in other developmental disorders such as Autism Spectrum Conditions (ASC) and Developmental Language Disorder (DLD). Studies have shown that increases in non-verbal mental age are associated with an increase in language abilities for infants with ASC (e.g. Luyster, Lopez & Lord, 2007; Luyster, Kadlec, Carter & Tager-Flusberg, 2008), therefore suggesting that non-verbal mental age is important for language development. Furthermore, non-verbal mental age was found to be a significant predictor of concurrent expressive and receptive language in toddlers with ASC aged 18-33 months (Luyster et al., 2008).

There is sometimes a reported discrepancy between non-verbal mental age and language for individuals with ASC, as some individuals have relatively good non-verbal abilities but delays in expressive and receptive language (Mitchell et al., 2006; Paul, Chawarska, Cicchetti & Volkmar, 2008). However, even if that is the case and there are individual differences in cognitive ability, non-verbal mental age has still emerged as the most significant predictor of concurrent language abilities in children with ASC (*Mean age= 30 months*) (Weismer, Lord, & Esler, 2010).

Existing research studies suggest that non-verbal mental age is an especially important predictor of concurrent and longitudinal language abilities during the early stages of development (Thurm Lord, Lee & Newschaffer, 2007; Weismer et al., 2010). Our findings are in line with this trend because non-verbal mental age appeared to be the most important predictor for concurrent and longitudinal language outcomes for infants with DS in our study.

With regards to DLD, the traditional view has been that DLD is a domain specific impairment and that individuals have a language impairment but that all other areas of development are intact (Tomblin, et al. 1997; van der Lely, 1997). However, an increasing

body of research has been suggesting that it is very difficult to distinguish between language and difficulties with non-verbal cognition, particularly in developmental disorders such as DLD (Botting & Marshall, 2017). There is evidence that individuals with DLD may have difficulties with aspects of non-verbal cognition, such as working memory (e.g. Archibald & Gathercole, 2010; Lum, Conti-Ramsden, Page & Ullman, 2012). Vugs, Hendriks, Cuperus, and Verhoeven (2014) in a study of 58 children with DLD aged 4-5 years, reported that the DLD group performed significantly worse than the TD group on measures of working memory, and importantly, the working memory score was predictive of which children had DLD, with a sensitivity of 88% and specificity of 90%. In summary, a growing number of studies highlight that non-verbal mental age may be an important factor for language abilities in other developmental disorders including ASC and DLD. This is in line with our results, which also found that non-verbal mental age is the most significant predictor of concurrent and longitudinal language abilities for infants with DS.

Our results are also supportive of previous research which shows that receptive language generally develops in line with non-verbal mental age for children with DS (Abbeduto, Murphy, Cawthon et al., 2003; Caselli et al., 1998; Fidler & Nadel, 2007; Miller, 1999). This may suggest that for children with DS the language deficit seen is part of the developmental process as opposed to a specific modular deficit (Thomas & Karmiloff-Smith, 2003; further theoretical implications are discussed in chapter 11.1). However, this leads to further questions around the developmental process such as whether poor language skills constrain a child's non-verbal mental age or vice versa (Botting, 2005).

Responding to joint attention, maternal sensitivity and reciprocity were also positively related to receptive vocabulary. Reciprocity was measured by observing how much time mother-child dyads spent in joint attention episodes during a 5-minute play interaction.

Therefore, the results are in line with previous research which has found that joint attention is important for concurrent and longitudinal language development in children with DS (Mundy et al. 1995) as well as research which found that the amount of time spent in joint attention interactions significantly predicted concurrent and longitudinal receptive vocabulary for children with DS (Zampini, Salvi & D’Odorico, 2015). However, responding to joint attention did not account for any unique variance after age and non-verbal mental age were controlled for. Previous research has also reported that sensitivity is important for language development in TD children (Leigh et al., 2013) since maternal sensitivity facilitates reciprocal interactions between the child and the care giver. Furthermore, if the mother follows the child’s lead, this increases the likelihood of the child absorbing information as they do not have to shift their attention (Landry, Smith, Miller-Loncar & Smith, 1997). To our knowledge no previous research has found a relationship between sensitivity and language scores for children with DS, however one study found a relationship between maternal sensitivity and children’s scores on the Bayley Mental Development Index (Crawley & Spiker, 1983) suggesting that maternal sensitivity was positively related to general development scores.

Although only non-verbal mental age was a significant predictor, the correlation analysis highlights that both child factors (non-verbal mental age) and maternal factors (sensitivity and reciprocity) may be important for concurrent language abilities. This is supportive of the transactional model of language development (Sameroff, 1975) which suggests that child behaviours may impact maternal sensitivity, e.g. children with language delays may not reciprocate their mothers’ attempts of interaction which may in turn mean that mother’s attempts to interact with their child decrease (Wheeler, Hatton, Reichardt, & Bailey, 2007). On the other hand, if a child has higher cognitive ability this may lead to more sensitive parenting which in turn may benefit the child’s language development.

Maternal positive expressed emotion significantly positively correlated with expressive vocabulary and mild verbal control had a significant negative relationship. When entered into a regression model controlling for age and baseline scores, the addition of positive expressed emotion and mild verbal control significantly improved the model. Mild verbal control was a significant predictor but the model was not significant. Finally, non-verbal mental age significantly correlated with use of signs but was not a significant predictor when entered into a regression model.

There were no significant predictors of expressive language. However, a positive correlation was found between positive expressed emotion by the mother and children's expressive language. This links to previous research which has found that maternal warmth and praise are linked to language development for TD children (Clarke-Stewart & Apfel, 1979; Landry, Smith, Swank, Assel & Vellet, 2001; Steelman, Assel, Swank, Smith & Landry, 2002) because such maternal behaviours are thought to help create a stimulating social environment whereby children are encouraged to engage in reciprocal interactions and motivated to learn (Bigelow et al., 2010).

A negative relationship was found between mild verbal control and expressive vocabulary. During coding, a distinction was made between 'strong' verbal control, examples of which include 'come here', 'get the ball', and mild verbal control, examples of which include 'would you like to play with the ball'. A possible explanation is that shorter, simpler prompts mean that the child is more likely to understand the message. Previous research has found that during early language development, mothers' speech to their TD children usually consists of short simple utterances with lots of repetition (Snow, 1995). Furthermore, Iverson, Longobardi, Spampinato & Caselli (2006) reported that mothers of children with DS ($M=47.6$ months) simplified utterances to their child more than mothers of TD children with equivalent expressive language skills. The authors argue that this may be a strategy employed

by the mothers to reflect their child's language abilities. Simplifying their utterances may prevent over-loading the child and may increase the likelihood that the child understands the utterance. Therefore, the negative correlation found between mild verbal control and expressive language may be because such utterances tended to be longer in length and may not facilitate language development.

10.10.2. *Factors Associated with Vocabulary/Language Development at 30-35 Months*

Further analyses were completed to see if any of the time point 1 or time point 2 predictor variables were associated with vocabulary/language outcomes at time point 3. Initially the analysis was completed for time point 1 variables. Baseline non-verbal mental age correlated with receptive language and was found to be a significant predictor in a model accounting for age and baseline receptive language. Expressive language significantly correlated with non-verbal mental age and responding to joint attention; however, they were not significant predictors. Total language significantly correlated with initial non-verbal mental age, responding to joint attention and maternal verbal elaboration. Receptive vocabulary significantly correlated with non-verbal mental age and responding to joint attention. Expressive vocabulary and use of signs correlated with non-verbal mental age only.

Next, the analysis was repeated for the predictor variables at time point 2 and language /vocabulary outcomes at time point 3. Receptive language significantly correlated with non-verbal mental age and responding to joint attention. Non-verbal mental age was found to be a significant predictor accounting for age, time point 2 scores and responding to joint attention. Expressive language correlated with non-verbal mental age only but when entered into a regression model non-verbal mental age was not a significant predictor. Total language scores also significantly correlated with non-verbal mental age and non-verbal mental age was a significant predictor accounting for age and time point 2 scores. Receptive

vocabulary correlated with non-verbal mental age, responding to joint attention and reciprocity but none of the variables were significant predictors when entered into a regression model. Expressive vocabulary did not correlate with any of the predictor variables. Finally use of signs correlated with non-verbal mental age only but non-verbal mental age was not a significant predictor in a model including age and time point 2 scores. The results are in line with previous research that has found that responding to joint attention is associated with concurrent and longitudinal language development for children with DS (Mason-app et al. 2013; Mundy et al. 1995; Zampini et al. 2015).

The results suggest that language predictors change over time for children with DS. Our initial study (study 1 of this thesis) showed that responding to joint attention was a significant predictor of concurrent language scores when children were 17-23 months old. At this stage, there was no relationship between maternal interactive style and language. Although non-verbal mental age and responding to joint attention consistently correlated and were predictors of vocabulary/language scores, as the children got older correlations were also found with aspects of maternal interactive style, i.e. sensitivity, reciprocity and positive expressed emotion. A similar pattern has been found for TD children whereby during early development early social communication skills such as joint attention and a positive maternal interactive style have been positively associated with language skills (Colonnesi, Stams, Koster & Noom, 2010; Delgado, Mundy, Crowson, Markus, Yale & Schwartz, 2002; Leigh, Nievar & Nathans, 2011; Markus, Mundy, Morales, Delgado & Yale, 2000; Nozadi et al., 2013; Tomsello & Farrar, 1986). Again this is supportive of the transactional model of development (Sameroff, 1975), as reciprocal social interactions between the child and caregiver promote maternal sensitivity which in turn aids language development.

Furthermore, maternal verbal elaboration at time point 1 significantly correlated with total language scores at time point 3 suggesting that infants whose parents used more verbal

elaboration when infants were 17-22 months had higher total language scores at 30-35 months. However, verbal elaboration was not a significant predictor when entered into a regression model. This may suggest that it is not the amount of verbal input which matters; rather the quality and contingency of the input. Previous research has identified that quality of parental input significantly predicted child's vocabulary skills when controlling for quantity of input (Cartmill et al., 2013; Rowe, 2012).

The concurrent and longitudinal analysis consistently found that non-verbal mental age is an important factor for language development for children with DS. Furthermore, responding to joint attention also emerged as an important factor in concurrent and longitudinal language development. This supports previous research that has found that responding to joint attention is related to concurrent and longitudinal language outcomes for children with DS (Mason-Apps, 2013; Mundy et al. 1995).

Chapter 11 – Discussion

This chapter will discuss the theoretical and practical implications of the thesis. The limitations of the studies and directions for future research will also be identified.

11.1. *Theoretical implications*

The results have important implications for theoretical views of language acquisition. The early intervention results are supportive of a social interactionist view of language development. This view emphasises the importance of the child's early environment and interactions in aiding language development. It is thought that early social communication behaviours such as joint attention enable a child to understand how and why an adult is using language (Tomasello, 2006). Therefore targeting a precursor skill (e.g. responding to joint attention) may enhance later abilities (e.g. receptive vocabulary) as targeting a component skill may help to achieve later milestones (Capone, 2010). Furthermore, the idea that atypical

delays can be addressed through the use of intervention has been explored through the developmental model of risk factors, risk processes and outcomes (Dawson, 2008). This model suggests that an individual is susceptible to a delay/disorder due to genetics and other risk factors; however intervention can alter the child's environment and interactions thus leading to a reduction in symptoms. This model could be applied to the language difficulties exhibited by the majority of individuals with DS. DS is a genetic condition and it is known that many individuals with DS have some form of speech and/or language delay. Early speech and language interventions can be administered to alter the individual's experiences and environment – e.g. change how the parent communicates with the child. This in turn can improve speech and language outcomes.

Furthermore, our analysis of concurrent and longitudinal language predictors found that non-verbal mental age was a significant predictor of language development concurrently and longitudinally for children with DS aged 17-35 months. This goes against the modular view of language, or more recent models such as the neuropsychological view which suggests that language is independent of other aspects of cognitive development and other areas of development. One area can be 'broken' but this will not affect other areas of development (D'Souza & Karmiloff-Smith, 2017). Instead our data and analyses support a neuro-constructivist view of language development that suggests that language and cognitive development are inter-twined. Therefore, if an individual has a language delay then they are also likely to have domain general impairments (D'Souza, D'Souza & Karmiloff-Smith, 2017).

The results of our study found that non-verbal mental age was consistently related to concurrent and longitudinal language skills, suggesting that for children with DS language development is strongly related to cognitive precursors such as non-verbal mental age and responding to joint attention (Stojanovik, 2014). Other early social communication skills such

as initiating joint attention seem to be associated with language development for TD children (see Mason-Apps, 2013). One way to interpret these findings would be to assume that different pre-linguistic precursors seem to be related to language for children with DS in comparison to TD children which suggests that children with DS may be following an atypical trajectory of language development rather than a typical trajectory which is simply delayed (Eilam & Levy, 2013). Another interpretation would be (as noted in chapter one) that the precursors to language in DS are similar to those identified in studies of TD children (responding to joint attention has been related to language for TD children in various studies Crowson, Markus, Yale & Schwartz, 2002; Delgado, Mundy, Colonesi, Stams, Koster & Noom, 2010; Markus, Mundy, Morales, Delgado & Yale, 2000; Tomsello & Farrar, 1986). This may suggest that the language trajectory in DS is delayed. However, the notion of delay would suggest that individuals with DS would eventually reach the same end point in terms of language skills as the TD population (Thomas et al., 2009). From previous research we know that this is not the case and that difficulties with language continue into adolescence and adulthood (e.g. Rondal & Comblain, 1996). This is the case in general as levels of non-verbal mental age rarely reach the typical range (Chapman & Hesketh, 2000). Furthermore, it would be expected that there would be a delay across cognitive domains (Thomas et al., 2009). However, for individuals with DS there is variation across, as well as within domains, e.g. receptive language is in advance of expressive language and sometimes exceeds level expected compared to non-verbal mental age (Abbeduto, Murphy, Cawthon et al., 2003; Caselli et al., 1998; Fidler & Nadel, 2007; Miller, 1999). In our study receptive language was simply in line with non-verbal mental age. Whilst acknowledging the potential lack of power, our data would suggest that the development of language in DS may follow an atypical pathway. Furthermore, in our study in chapter two – when infants with DS were compared to

a TD group of equivalent non-verbal mental age different pre-linguistic precursors were found to be predictors of concurrent language abilities.

Studying a population with a genetic disorder such as DS has allowed us to potentially tease apart the different contribution of various precursor skills which is not possible to do with typical children as development happens in synchrony and it is therefore difficult to tease apart which precursor skills may be more facilitative than others (Thomas et al., 2009). Further longitudinal research is needed to determine this trajectory for children with DS.

11.2. *Practical/clinical implications*

In terms of the intervention, these preliminary results suggest that early responding to joint attention skills may have been accelerated and that this may have had a positive effect on later speech/language, specifically receptive vocabulary.

Furthermore, positive feedback was obtained from all parents who took part in the study and all 16 participants completed the study from start to end. Parents expressed that they were very satisfied with the intervention process and that they had seen changes in their child's responding to joint attention and speech and language. Parents also reported that they had seen positive changes in other areas of development, e.g. sustained attention and that they had changed the way they communicated with their child. All parents attended all sessions and were compliant with regards to 'homework'; they also reported that the intervention moved at the right pace and that the instructions were clear and easy to follow.

In addition, results from the concurrent and longitudinal analysis found that responding to joint attention and non-verbal mental age are important predictors for language development longitudinally and concurrently. This suggests that further research should be done in this area but with a larger sample and using a randomised control trial design. If a larger scale randomised control trial found the intervention to have benefits for early

communication skills for children with DS, it could be used by portage/early intervention workers.

11.3. *Limitations and ideas for future research*

There are a number of limitations to be discussed. Firstly, a small sample size was used due to restraints on time and resources. Each child was seen 14 times by the researcher and an extensive amount of travel occurred as participants lived in various counties across the UK. This meant that we were underpowered for analyses such as ANCOVA and multiple regression which may mean results are inflated, or under-estimated and so should be interpreted with caution.

Furthermore, existing data from a non-randomised pre-existing group were used as the control group. This likely led to the differences in non-verbal mental age as the study developed. This also constrained the analysis we could do since we did not have any control comparison data at the point of the immediate post-test assessment. Future research needs to use a larger scale randomised design. This will help to establish if differences between the groups are due to the intervention as opposed to spontaneous development/chance. Three groups should also be used – a joint attention intervention group, an alternative intervention group and a no-treatment control group. This will ensure whether changes are due to joint attention intervention or repeated contact time.

A further limitation of the intervention was that the quality of parent training sessions was not controlled. Parents were asked to complete a weekly diary documenting if they had been able to do three practise sessions at home. However, the quality and quantity of parental sessions could not be fully controlled. Furthermore, all parents reported each week that they had completed the three training sessions at home. Although this may have been the case there was no other way to check/control for this other than relying on parental report. A way

to overcome this would be to ask parents to film their sessions at home. Future research could have formal parent training sessions where parents are observed by the researcher to ensure they are using techniques appropriately and in a uniformed way. Homework sessions could also be video recorded so that the parent could receive feedback from the researcher.

Future research should also look to control for other interventions/techniques that the child is receiving. As a self-selecting sample was used and since a high level of commitment was required for the study, it may have attracted parents who were keen to learn more and improve their child's speech and language. At the start of the study no children in the intervention or control group were receiving targeted one to one speech and language therapy. However, it is likely that over the year that the study took place parents accessed other forms of support/advice on strategies and techniques. To ensure that this intervention specifically is having some effect future participants would have to agree to forego other projects/interventions for the duration of the study.

A further limitation due to the self-selecting sample is that across both groups a representative socio-economic sample was not obtained. The majority of parents were educated to at least degree level and had professional occupations. Future research should consider widening recruitment strategies to ensure a wider spread of socio-economic status.

A further potential limitation of the study is the measures used to assess language/vocabulary development. Vocabulary was assessed by the parental questionnaire the Reading Communicative Development Inventory. Although it is widely known that parents/caregivers are accurate in reporting their child's language capabilities (e.g. Sachse & Suchodoletz, 2008) it is possible that results may have been inflated. Since parents were aware that they had taken part in a communication intervention those in the intervention group may have felt compelled to report changes over time. On the other hand, since

intelligibility is often poor for children with DS (Kumin, 2006) it is possible that parents did not report all the words their child produced. A further difficulty for parents is accurately reporting what words their child understands.

A standardised language assessment (the PLS-4) was used to assess language. The assessment is designed for TD children and not children with additional needs. There was little variability at each time point for this measure and many children reached a 'ceiling' for expressive language. The PLS-4 however, is structured in a way such that there is a sudden jump from assessing whether a child is producing 5-10 words to assessing whether a child is producing questions and multi-word combinations (e.g. subject, verb, object). It therefore ignores the difference between a child who is producing 5 words and a child who is producing over 100 words but may not be producing questions or multi-word combinations. As participants got older, their challenging behaviour increased and in addition to the PLS-4 and Reading Communicative Development Inventory an unstructured language assessment may have been useful. Research has identified that using standardised assessments to measure language capabilities of children can be problematic since children often talk less when in a new environment and their performance is often affected by factors such as fatigue and lack of familiarity with the assessor (Mervis & Becerra, 2003). Further researchers have identified the difficulty of obtaining reliable scores of receptive and expressive language when assessing children with additional needs in a single assessment (Charman, Drew & Baird, 2003). This may explain partly why we found a difference between groups on the vocabulary questionnaire but not on the direct measure of language (PLS-4). Furthermore, the Communicative Development Inventory has been adapted in Spanish specifically for children with DS and concurrent validity and test re-test reliability has been established (CDI-Downs; Galeote, Checa, Sanchez-Palacios, Sebastian & Soto, 2016). This suggests that such

instruments can specifically be adapted for children with DS and this would be beneficial for future research purposes.

A final limitation is that the child's use of sign language (key word signing) was also monitored through the Reading Communicative Development Inventory and used as an outcome measure. However, some parents reported that they did not use sign with their child whereas other parents consistently used sign to communicate with their child. Therefore in both groups (intervention and control) there were some children who were reported as not understanding/using any signs. This could be due to the capabilities of the child but may also be because this form of communication was not being used and recognised by these parents. Although, key word signing is actively encouraged by professionals (Buckley & Bird, 2001) some of the parents reported that they were concerned that using sign would further delay their child's onset of speech.

11.4. *Conclusion*

The thesis has addressed important gaps in the literature in the area of early language development for infants with DS. Part 1 focused on which precursors are important for early language development for children with DS and supported previous research in the area which has also identified responding to joint attention.

The second part reviewed the existing literature regarding early language interventions for infants with DS. This highlighted that no existing interventions focus on improving joint attention for infants with DS. Further, it highlighted the need for more research in this area.

Finally, the intervention was the first study to focus on improving responding to joint attention for children with DS. Three separate follow up assessments were also completed for all children in the intervention which allowed us to consider the long-term effects. This is rare in the intervention literature and many published studies only include an immediate post-test

assessment. A further benefit is that longitudinal data for 30 infants with DS with multiple data points was collected. This gave further insights into the rate of development of language, joint attention and non-verbal mental age for children with DS aged 17-35 months.

The results of the intervention suggested that responding to joint attention may have been improved and that this could have positive cascading effects on later receptive vocabulary. A further positive finding is that a 10-week parent-researcher intervention is feasible for children with DS at 17-23 months. This warrants further research in this area using a randomised control trial design with three groups – a responding to joint attention intervention group, an alternative intervention group and a control group, as well as completing the research across multiple sites so that a larger sample size could be collected. The inclusion of three groups will allow control over effects obtained due to spontaneous development or repeated contact time.

The language predictor analysis revealed that various precursor skills are important throughout language development including non-verbal mental age, responding to joint attention, reciprocity, maternal sensitivity, maternal positive expressed emotion and maternal verbal elaboration. Non-verbal mental age was consistently a significant predictor of concurrent and longitudinal language abilities and thus supporting social interactionist views of language development. This analysis also highlighted the need for further research in this area. A better understanding of how precursors are related to concurrent and longitudinal language abilities is needed to inform further early intervention studies to improve speech and language outcomes for children with DS.

References

- Abbeduto, L., Murphy, M. M., Cawthon, S. W., Richmond, E. K., Weissman, M. D., Karadottir, S., & O'Brien, A. (2003). Receptive language skills of adolescents and young adults with Down or fragile X syndrome. *American Journal on Mental Retardation, 108*(3), 149-160.
- Abbeduto, L., Seltzer, M. M., Shattuck, P., Krauss, M. W., Orsmond, G., & Murphy, M. M. (2004). Psychological well-being and coping in mothers of youths with autism, down syndrome, or fragile X syndrome. *American Journal on Mental Retardation, 109*(3), 237-254.
- Abbeduto, L., Warren, S. F., & Conners, F. A. (2007). Language development in Down syndrome: From the prelinguistic period to the acquisition of literacy. *Developmental Disabilities Research Reviews, 13*(3), 247-261.
- Adamson, L. B., Bakeman, R., Deckner, D. F., & Ronski, M. (2009). Joint engagement and the emergence of language in children with autism and Down syndrome. *Journal of autism and developmental disorders, 39*(1), 84.
- Akhtar, N., Dunham, F., & Dunham, P. J. (1991). Directive interactions and early vocabulary development: The role of joint attentional focus. *Journal of child language, 18*(1), 41-49.
- Alfieri, P., Menghini, D., Marotta, L., De Peppo, L., Ravà, L., Salvaguardia, F., Varuzza, C. & Vicari, S. (2017). A comparison between linguistic skills and socio-communicative abilities in Williams syndrome. *Journal of Intellectual Disability Research, 61*(9), 866-876.
- Andrade, R. V., & Limongi, S. C. O. (2007). The emergence of expressive communication in

the child with Down syndrome. *Pró-Fono Revista de Atualização Científica*, 19(4), 387-392.

Anlar, B., & Yalaz, K. (1996). *Denver II Geliimsel Tarama Testi Turk Cocuklarma Uyarlamasz ve Standardizasyonu El Kitabz [Turkish Manual for Denver II]*. Ankara, Turkey: Meteksan.

Aparicio, M. T. S. (1989). Effect of observational training of parents in the early stimulation of trisomy-21 babies. *Early child development and care*, 41, 89-101.

Aparicio, S. T. M., & Balana, J. M. (2002). Early language stimulation of Down's syndrome babies: a study on the optimum age to begin. *Early child development and care*, 172, 651-656.

Archibald, L. M., & Gathercole, S. E. (2006). Short- term and working memory in specific language impairment. *International Journal of Language & Communication Disorders*, 41(6), 675-693.

Atkinson, R. L., Atkinson, R. C., Smith, E. E., & Bem, Nolen-Hoeksema, D. J. (1996). *Hilgard's Introduction to Psychology*. New York, USA: Harcourt Brace Company.

Baddeley, A. (2003). Working memory and language: An overview. *Journal of communication disorders*, 36(3), 189-208.

Bakeman, R., & Adamson, L. B. (1984). Coordinating attention to people and objects in mother-infant and peer-infant interaction. *Child development*, 1278-1289.

- Baumwell, L., Tamis-LeMonda, C. S., & Bornstein, M. H. (1997). Maternal verbal sensitivity and child language comprehension. *Infant Behavior and Development, 20*(2), 247-258.
- Bayley, N. (1969). *Bayley scales of infant development*. New York, USA: Psychological Corporation.
- Berger, J., & Cunningham, C. C. (1983). Development of early vocal behaviors and interactions in Down's syndrome and nonhandicapped infant–mother pairs. *Developmental Psychology, 19*(3), 322.
- Berglund, E., Eriksson, M., & Johansson, I. (2001). Parental reports of spoken language skills in children with Down syndrome. *Journal of speech, language, and hearing research, 44*(1), 179-191.
- Bigelow, A. E., MacLean, K., Proctor, J., Myatt, T., Gillis, R., & Power, M. (2010). Maternal sensitivity throughout infancy: Continuity and relation to attachment security. *Infant Behavior & Development, 33*, 50–60.
- Bird, E. K. R., Cleave, P., Trudeau, N., Thordardottir, E., Sutton, A., & Thorpe, A. (2005). The language abilities of bilingual children with Down syndrome. *American Journal of Speech-Language Pathology, 14*(3), 187-199.
- Bittles, A. H., & Glasson, E. J. (2004). Clinical, social, and ethical implications of changing life expectancy in Down syndrome. *Developmental Medicine & Child Neurology, 46*(4), 282-286.
- Bornstein, M.H. (2006). On the significance of social relationships in the development of children's earliest symbolic play: An ecological perspective. In A. Gönçü & S.

- Gaskins (Eds.), *Play and development: Evolutionary, sociocultural and functional perspectives*. Mahwah, NJ: Erlbaum.
- Botting, N. (2005). Non- verbal cognitive development and language impairment. *Journal of child psychology and psychiatry*, 46(3), 317-326.
- Botting, N., & Marshall, C. (2017). Domain- specific and Domain- general Approaches to Developmental Disorders. *The Wiley Handbook of Developmental Psychopathology*, 139-159.
- Braine, M. D. (1994). Is nativism sufficient?[*]. *Journal of Child Language*, 21(1), 9-31.
- Breiner, J., & Forehand, R. (1982). Mother-child interactions: A comparison of a clinic-referred developmentally delayed group and two non-delayed groups. *Applied research in mental retardation*, 3(2), 175-183.
- Brooks, R., & Meltzoff, A. N. (2005). The development of gaze following and its relation to language. *Developmental science*, 8(6), 535-543.
- Brunet, O., & Lézine, I. (1976). *Echelle de Developpement Psychomoteur de la Premiere Enfance (second ed.)*, Paris, France: PUF.
- Buckley, S., & Sacks, B. (1987). *The adolescent with Down's syndrome: Life for the teenager and for the family*. Portsmouth, UK: Portsmouth Polytechnic.
- Buckley, S. (1993). Language development in children with Down syndrome-Reasons for optimism. *Down syndrome research and practice*, 1(1), 3-9.
- Buckley, S., & Bird, G. (2001). *Speech and language development for children with Down syndrome (5-11 years)*. Portsmouth, UK: DSE Enterprises.

Burgoyne, K., Duff, F. J., Clarke, P. J., Buckley, S., Snowling, M. J., & Hulme, C. (2012).

Efficacy of a reading and language intervention for children with Down syndrome: a randomized controlled trial. *Journal of Child Psychology and Psychiatry*, *53*(10), 1044-1053.

Butterworth, G., & Jarrett, N. (1991). What minds have in common is space: Spatial

mechanisms serving joint visual attention in infancy. *British journal of Developmental Psychology*, *9*, 55-72.

Camarata, S., Yoder, P., & Camarata, M. (2006). Simultaneous treatment of grammatical and

speech-comprehensibility deficits in children with Down syndrome. *Down Syndrome Research and Practice*, *11*(1), 9-17.

Capone, G. T., Grados, M. A., Kaufmann, W. E., Bernad-Ripoll, S., & Jewell, A. (2005).

Down syndrome and comorbid autism-spectrum disorder: Characterization using the aberrant behavior checklist. *American journal of medical genetics Part A*, *134*(4), 373-380.

Capone, N.C. (2010). *Language assessment and intervention: a developmental approach*.

Sudbury, MA: Jones and Bartlett Publishers.

Carbone, V. J., Sweeney-Kerwin, E. J., Attanasio, V., & Kasper, T. (2010). Increasing the

vocal responses of children with autism and developmental disabilities using manual sign mand training and prompt delay. *Journal of applied behavior analysis*, *43*(4), 705-709.

Carpenter, M., Nagell, K., Tomasello, M., Butterworth, G., & Moore, C. (1998). Social

cognition, joint attention, and communicative competence from 9 to 15 months of age. *Monographs of the Society for Research in Child Development*, *63*, i-174.

- Cartmill, E. A., Armstrong, B. F., Gleitman, L. R., Goldin-Meadow, S., Medina, T. N., & Trueswell, J. C. (2013). Quality of early parent input predicts child vocabulary 3 years later. *Proceedings of the National Academy of Sciences, 110*(28), 11278-11283.
- Casby, M. W., & McCormack, S. M. (1985). Symbolic play and early communication development in hearing-impaired children. *Journal of Communication Disorders, 18*(1), 67-78.
- Casby, M. W., & Ruder, K. F. (1983). Symbolic play and early language development in normal and mentally retarded children. *Journal of Speech, Language, and Hearing Research, 26*(3), 404-411.
- Caselli, M. C., Vicari, S., Longobardi, E., Lami, L., Pizzoli, C., & Stella, G. (1998). Gestures and words in early development of children with Down syndrome. *Journal of Speech, Language, and Hearing Research, 41*(5), 1125-1135.
- Chapman, R. S., Schwartz, S. E., & Bird, E. K. R. (1991). Language skills of children and adolescents with Down syndrome: I. Comprehension. *Journal of Speech, Language, and Hearing Research, 34*(5), 1106-1120.
- Chapman, R. S., Seung, H. K., Schwartz, S. E., & Bird, E. K. R. (1998). Language skills of children and adolescents with Down syndrome: II. Production deficits. *Journal of Speech, Language, and Hearing Research, 41*(4), 861-873.
- Chapman, R. S., Seung, H. K., Schwartz, S. E., & Bird, E. K. R. (2000). Predicting language production in children and adolescents with Down syndrome: The role of comprehension. *Journal of Speech, Language, and Hearing Research, 43*(2), 340-350.

- Chapman, R. S. & Hesketh, L. J. (2000). Behavioural phenotype of individuals with Down syndrome. *Mental Retardation and Developmental Disabilities Research Reviews*, 6, 84-95.
- Charman, T., Swettenham, J., Baron-Cohen, S., Cox, A., Baird, G., & Drew, A. (1997). Infants with autism: an investigation of empathy, pretend play, joint attention, and imitation. *Developmental psychology*, 33(5), 781.
- Charman, T., Drew, A., Baird, C., & Baird, G. (2003). Measuring early language development in preschool children with autism spectrum disorder using the MacArthur Communicative Development Inventory (Infant Form). *Journal of child language*, 30(1), 213-236.
- Chiat, S., & Roy, P. (2008). Early phonological and sociocognitive skills as predictors of later language and social communication outcomes. *Journal of Child Psychology and Psychiatry*, 49(6), 635-645.
- Chomsky, N. (1976). On the biological basis of language capacities. In *The neuropsychology of language* (pp. 1-24). US: Springer.
- Cielinski, K. L., Vaughn, B. E., Seifer, R., & Contreras, J. (1995). Relations among sustained engagement during play, quality of play, and mother-child interaction in samples of children with Down syndrome and normally developing toddlers. *Infant Behavior and Development*, 18(2), 163-176.
- Clarke-Stewart, K. A., & Apfel, N. (1979). Evaluating parental effects on child development. In L. S. Schulman (Ed.), *Review of research in education*. (pp. 47- 119). Itasca, IL: Peacock.

- Cleland, J., Wood, S., Hardcastle, W., Wishart, J., & Timmins, C. (2010). Relationship between speech, oromotor, language and cognitive abilities in children with Down's syndrome. *International journal of language & communication disorders*, 45(1), 83-95.
- Colonnesi, C., Stams, G. J. J., Koster, I., & Noom, M. J. (2010). The relation between pointing and language development: A meta-analysis. *Developmental Review*, 30(4), 352-366.
- Crawley, S. B., & Spiker, D. (1983). Mother-child interactions involving two-year-olds with Down syndrome: A look at individual differences. *Child development*, 1312-1323.
- Dabrowska, A., & Pisula, E. (2010). Parenting stress and coping styles in mothers and fathers of pre-school children with autism and Down syndrome. *Journal of Intellectual Disability Research*, 54(3), 266-280.
- Davis, A. S. (2008). Children with Down syndrome: Implications for assessment and intervention in the school. *School psychology quarterly*, 23(2), 271.
- Dawson, G. (2008). Early behavioral intervention, brain plasticity, and the prevention of autism spectrum disorder. *Development and psychopathology*, 20(3), 775-803.
- Deák, G. O. (2014). Interrelations of language and cognitive development. In P. Brooks & V. Kampe (Eds.), *Encyclopedia of Language Development*. (pp. 284-291). Thousand Oaks, CA: Sage.
- DeLeon, I. G. & Iwata, B. A. (1996). Evaluation of a multiple-stimulus presentation format for assessing reinforcer preferences. *Journal of Applied Behavior Analysis*, 29(4), 519-533.

- Delgado, C. E., Mundy, P., Crowson, M., Markus, J., Yale, M., & Schwartz, H. (2002). Responding to joint attention and language development: A comparison of target locations. *Journal of Speech Language and Hearing Research, 45*(4), 715-719.
- Dodd, B., Hua, Z., Crosbie, S., & Holm, A. (2002). *Diagnostic Evaluation of Articulation and Phonology*. London: The Psychological Corporation.
- D'Souza, D., D'Souza, H. & Karmiloff-Smith, A. (2017). Precursors to language development in typically and atypically developing infants and toddlers: the importance of embracing complexity. *Journal of Child Language, 44*(3), 591-627.
- D'Souza, H., & Karmiloff-Smith, A. (2017). Neurodevelopmental disorders. *Wiley Interdisciplinary Reviews: Cognitive Science, 8*(1-2).
- Eisenhower, A. S., Baker, B. L., & Blacher, J. (2005). Preschool children with intellectual disability: syndrome specificity, behaviour problems, and maternal well-being. *Journal of Intellectual Disability Research, 49*(9), 657-671.
- Ejiri, K. (1998). Relationship between rhythmic behavior and canonical babbling in infant vocal development. *Phonetica, 55*(4), 226-237.
- Fabbretti, D., Pizzuto, E., Vicari, S., & Volterra, V. (1997). A story description task in children with Down's syndrome: lexical and morphosyntactic abilities. *Journal of Intellectual Disability Research, 41*(2), 165-179.
- Fenson, L., Dale, P. S., Reznick, J. S., Thal, D., Bates, E., Hartung, J. P., Pethick, S. & Reilly, J. S. (1993). *MacArthur Communicative Development Inventory : users guide and technical manual*. San Diego, CA: Singular Publishing Company

- Fewell, R. R., Ogura, T., Notari-Syverson, A., & Wheeden, C. A. (1997). The relationship between play and communication skills in young children with Down syndrome. *Topics in early childhood special education, 17*(1), 103-118.
- Fey, M. E., Warren, S. F., Brady, N., Finestack, L. H., Bredin-Oja, S. L., Fairchild, M., Sokol, S. & Yoder, P. J. (2006). Early effects of responsivity education/prelinguistic milieu teaching for children with developmental delays and their parents. *Journal of Speech, Language, and Hearing Research, 49*(3), 526-547.
- Fidler, D. J. (2006). The emergence of a syndrome-specific personality profile in young children with Down syndrome. *Down syndrome: Neurobehavioural specificity*, 139-152.
- Fidler, D. J. (2005). The emerging Down syndrome behavioral phenotype in early childhood: Implications for practice. *Infants & Young Children, 18*(2), 86-103.
- Fidler, D. J., & Nadel, L. (2007). Education and children with Down syndrome: neuroscience, development, and intervention. *Mental Retardation and Developmental Disabilities Research Reviews, 13*, 262-271.
- Fidler, D. J., Hepburn, S. L., Mankin, G., & Rogers, S. J. (2005). Praxis skills in young children with Down syndrome, other developmental disabilities, and typically developing children. *American Journal of Occupational Therapy, 59*(2), 129-138.
- Fidler, D. J., Philofsky, A., & Hepburn, S. L. (2007). Language phenotypes and intervention planning: Bridging research and practice. *Mental retardation and developmental disabilities research reviews, 13*(1), 47-57.

- Fidler, D. J., Philofsky, A., Hepburn, S. L., & Rogers, S. J. (2005). Nonverbal requesting and problem-solving by toddlers with Down syndrome. *American Journal on Mental Retardation, 110*, 312-322.
- Fodor, J. A. (1983). *The modularity of mind: An essay on faculty psychology*. Massachusetts :MIT press.
- Fowler, A. E. (1990). Language abilities in children with Down syndrome: Evidence for a specific syntactic delay. *Children with Down syndrome: A developmental perspective, 9*, 302-328.
- Fowler, A. E., Gelman, R., & Gleitman, L. R. (1994). The course of language learning in children with Down syndrome: Longitudinal and language level comparisons with young normally developing children. In H. Tager-Flusberg (Eds.), *Constraints on language acquisition* (pp. 91-140). Hillsdale, NJ: Erlbaum.
- Frith, U., & Frith, C. D. (1974). Specific motor disabilities in Downs syndrome. *Journal of Child Psychology and Psychiatry, 15*(4), 293-301.
- Galeote, M., Checa, E., Sánchez-Palacios, C., Sebastián, E., & Soto, P. (2016). Adaptation of the MacArthur-Bates Communicative Development Inventories for Spanish children with Down Syndrome: Validity and reliability data for vocabulary. *American Journal of Speech-Language Pathology, 25*(3), 371-380.
- Ghaziuddin, M., Tsai, L. Y., & Ghaziuddin, N. (1992). Autism in Down's syndrome: presentation and diagnosis. *Journal of Intellectual Disability Research, 36*(5), 449-456.

- Gilmore, L., Cuskelly, M., Jobling, A., & Hayes, A. (2009). Maternal support for autonomy: Relationships with persistence for children with Down syndrome and typically developing children. *Research in developmental disabilities, 30*(5), 1023-1033.
- Girolametto, E. L. (1988). Improving the social-conversational skills of developmentally delayed children: An intervention study. *Journal of Speech and Hearing Disorders, 53*, 156-167.
- Girolametto, L., Weitzman, E., & Clements-Baartman, J. (1998). Vocabulary intervention for children with Down syndrome: Parent training using focused stimulation. *Infant-Toddler Intervention: The Transdisciplinary Journal, 8*(2), 109-25.
- Glenn, S., Dayus, B., Cunningham, C., & Horgan, M. (2001). Mastery motivation in children with Down syndrome. *Down Syndrome Research and Practice, 7*(2), 52-59.
- Grieco, J., Pulsifer, M., Seligsohn, K., Skotko, B., & Schwartz, A. (2015). Down syndrome: Cognitive and behavioral functioning across the lifespan. *American Journal of Medical Genetics Part C: Seminars in Medical Genetics, 169*(2), 135-149.
- Hallers-Haalboom, E. T., Mesman, J., Groeneveld, M. G., Endendijk, J. J., van Berkel, S. R., van der Pol, L. D., & Bakermans-Kranenburg, M. J. (2014). Mothers, fathers, sons and daughters: Parental sensitivity in families with two children. *Journal of Family Psychology, 28*(2), 138.
- Hamilton, A., Plunkett, K., & Schafer, G. (2000). Infant vocabulary development assessed with a British communicative development inventory. *Journal of child language, 27*(3), 689-705.

- Hamouda, H. B., Mnasri, H., Ghanmi, S., Soua, H., Sakli, G., Dogui, M., & Sfar, M. T. (2014). West syndrome associated with Down syndrome: case report and literature review. *Pediatrics Polska*, *89*(3), 203-206.
- Harris, S., Kasari, C., & Sigman, M. D. (1996). Joint attention and language gains in children with Down syndrome. *American journal of mental retardation: AJMR*, *100*(6), 608-619.
- Hayes, A., & Batshaw, M. L. (1993). Down syndrome. *Pediatric Clinics of North America*, *40*(3), 523-535.
- Hines, S., & Bennett, F. (1996). Effectiveness of early intervention for children with Down syndrome. *Developmental Disabilities Research Reviews*, *2*(2), 96-101.
- Hoff-Ginsberg, E. (1987). Topic relations in mother-child conversation. *First Language*, *7*(20), 145-158.
- Iverson, J. M., & Goldin-Meadow, S. (2005). Gesture paves the way for language development. *Psychological science*, *16*(5), 367-371.
- Iverson, J. M., Longobardi, E., Spampinato, K., & Caselli, C. M. (2006). Gesture and speech in maternal input to children with Down's syndrome. *International Journal of Language & Communication Disorders*, *41*(3), 235-251.
- Jarrold, C., & Baddeley, A. D. (1997). Short-term memory for verbal and visuospatial information in Down's syndrome. *Cognitive neuropsychiatry*, *2*(2), 101-122.
- Jarrold, C., Baddeley, A. D., & Hewes, A. K. (1999). Genetically dissociated components of working memory: evidence from Downs and Williams syndrome. *Neuropsychologia*, *37*(6), 637-651.

- Jones, E. A., Carr, E. G., & Feeley, K. M. (2006). Multiple effects of joint attention intervention for children with autism. *Behavior Modification, 30*(6), 782-834.
- Karaaslan, O., & Mahoney, G. (2013). Effectiveness of Responsive Teaching with children with Down syndrome. *Mental Retardation, 51*(6), 458-469.
- Karaaslan, O., Diken, I. H., & Mahoney, G. (2013). A randomized control study of responsive teaching with young Turkish children and their mothers. *Topics in Early Childhood Special Education, 33*(1), 18-27.
- Karmiloff-Smith, A. (1992). *Beyond modularity: A developmental approach to cognitive science*. Cambridge, MA: MIT Press.
- Karmiloff-Smith, A. (1998). Development itself is the key to understanding developmental disorders. *Trends in Cognitive Sciences, 2*, 389–398.
- Karmiloff-Smith, A. (2009). Nativism versus neuroconstructivism: rethinking the study of developmental disorders. *Developmental psychology, 45*(1), 56.
- Kasari, C., Freeman, S., & Paparella, T. (2006). Joint attention and symbolic play in young children with autism: A randomized controlled intervention study. *Journal of Child Psychology and Psychiatry, 47*(6), 611-620.
- Kent, R. D., & Vorperian, H. K. (2013). Speech impairment in Down syndrome: A review. *Journal of Speech, Language, and Hearing Research, 56*(1), 178-210.
- Klee, T., Carson, D. K., Gavin, W. J., Hall, L., Kent, A., & Reece, S. (1998). Concurrent and predictive validity of an early language screening program. *Journal of Speech, Language, and Hearing Research, 41*(3), 627-641.

- Koegel, R. L., & Frea, W. D. (1993). Treatment of social behavior in autism through the modification of pivotal social skills. *Journal of Applied Behavior Analysis*, 26(3), 369-377.
- Kogan, K. L., & Tyler, N. (1973). Mother-child interaction in young physically handicapped children. *American Journal of Mental Deficiency*, 77(5), 492-7.
- Kumin, L. (1994). Intelligibility of speech in children with Down syndrome in natural settings: Parents' perspective. *Perceptual and Motor Skills*, 78(1), 307-313.
- Kumin, L. (2002). Maximizing speech and language in children and adolescents with Down syndrome. *Down syndrome: Visions for the 21st century*, 407-419.
- Kumin, L. (2006). Speech intelligibility and childhood verbal apraxia in children with Down syndrome. *Down Syndrome Research and Practice*, 10(1), 10-22.
- Kumin, L., Council, C., & Goodman, M. (1994). A longitudinal study of the emergence of phonemes in children with Down syndrome. *Journal of Communication Disorders*, 27(4), 293-303.
- Krakow, J. B., & Kopp, C. B. (1983). The effects of developmental delay on sustained attention in young children. *Child Development*, 1143-1155.
- Lai, F., & Williams, R. S. (1989). A prospective study of Alzheimer disease in Down syndrome. *Archives of neurology*, 46(8), 849-853.
- Laing, E., Butterworth, G., Ansari, D., Gsödl, M., Longhi, E., Panagiotaki, G., Paterson, S. & Karmiloff-Smith, A. (2002). Atypical development of language and social communication in toddlers with Williams syndrome. *Developmental Science*, 5(2), 233-246.

- Landry, S. H., Smith, K. E., Miller-Loncar, C. L., & Swank, P. R. (1997). Predicting cognitive-language and social growth curves from early maternal behaviors in children at varying degrees of biological risk. *Developmental psychology, 33*(6), 1040.
- Landry, S. H., Smith, K. E., Swank, P. R., Assel, M. A., & Vellet, S. (2001). Does early responsive parenting have a special importance for children's development or is consistency across early childhood necessary?. *Developmental psychology, 37*(3), 387.
- Lang, R., Machalicek, W., Rispoli, M., & Regester, A. (2009). Training parents to implement communication interventions for children with autism spectrum disorders (ASD): A systematic review. *Evidence-Based Communication Assessment and Intervention, 3*(3), 174-190.
- Laws, G., Byrne, A., & Buckley, S. (2000). Language and memory development in children with Down syndrome at mainstream schools and special schools: a comparison. *Educational Psychology, 20*(4), 447-457.
- Laws, G., & Bishop, D. V. (2004). Verbal deficits in Down's syndrome and specific language impairment: a comparison. *International Journal of Language & Communication Disorders, 39*(4), 423-451.
- Laws, G., & Bishop, D. V. (2004). Pragmatic language impairment and social deficits in Williams syndrome: a comparison with Down's syndrome and specific language impairment. *International Journal of Language & Communication Disorders, 39*(1), 45-64.

- Laws, G., & Hall, A. (2014). Early hearing loss and language abilities in children with Down syndrome. *International journal of language & communication disorders, 49*(3), 333-342.
- Leigh, P., Nievar, M. A., & Nathans, L. (2011). Maternal sensitivity and language in early childhood: A test of the transactional model. *Perceptual and motor skills, 113*(1), 281-299.
- Levy, Y., & Eilam, A. (2013). Pathways to language: a naturalistic study of children with Williams syndrome and children with Down syndrome. *Journal of child language, 40*(1), 106-138.
- Lieven, E., Salomo, D., & Tomasello, M. (2009). Two-year-old children's production of multiword utterances: A usage-based analysis. *Cognitive Linguistics, 20*(3), 481-507.
- Lum, J. A., Conti-Ramsden, G., Page, D., & Ullman, M. T. (2012). Working, declarative and procedural memory in specific language impairment. *cortex, 48*(9), 1138-1154.
- Luyster, R., Lopez, K., & Lord, C. (2007). Characterizing communicative development in children referred for autism spectrum disorders using the MacArthur-Bates Communicative Development Inventory (CDI). *Journal of Child Language, 34*(3), 623-654.
- Luyster, R. J., Kadlec, M. B., Carter, A., & Tager-Flusberg, H. (2008). Language assessment and development in toddlers with autism spectrum disorders. *Journal of autism and developmental disorders, 38*(8), 1426-1438.
- Lynch, M. P., Oller, D. K., Steffens, M. L., & Levine, S. L. (1995). Onset of speech-like vocalizations in infants with Down syndrome. *American Journal on Mental Retardation, 100*, 68-86.

- McCathren, R. B., Yoder, P. J., & Warren, S. F. (1995). The role of directives in early language intervention. *Journal of early intervention, 19*(2), 91-101.
- McCathren, R. B., Warren, S. F., Yoder, P. J., Cole, K. N., Dale, P. S., & Thal, D. J. (1996). Prelinguistic predictors of later language development. *Assessment of communication and language, 6*, 57-75.
- MacDonald, J. D. & Nickols, M. (1978). *Environmental language inventory*. Coloumbus, OH: Charles E Merrill.
- Mahoney, G., Robinson, C., & Powell, A. (1992). Focusing on Parent--Child Interaction: The Bridge to Developmentally Appropriate Practices. *Topics in early childhood special education, 12*(1), 105-120.
- Mahoney, G., & Wheeden, C. A. (1998). Effects of teacher style on the engagement of preschool aged children with special learning needs. *Journal of Developmental and Learning Disorders, 2*, 293-315.
- Mahoney, G. (1999). *Maternal Behavior Rating Scale*. Tallmadge, OH: Family Child Learning Center.
- Manolson, A. (1992). *It takes two to talk: A parent's guide to helping children communicate*. Ontario, Canada: The Hanen Centre.
- Marfo, K. (1990). Maternal directiveness in interactions with mentally handicapped children: An analytical commentary. *Journal of Child Psychology and Psychiatry, 31*(4), 531-549.

- Markus, J., Mundy, P., Morales, M., Delgado, C. E., & Yale, M. (2000). Individual differences in infant skills as predictors of child-caregiver joint attention and language. *Social Development, 9*(3), 302-315.
- Martin, G. E., Klusek, J., Estigarribia, B., & Roberts, J. E. (2009). Language characteristics of individuals with Down syndrome. *Topics in Language Disorders, 29*, 112-132.
- Mason-Apps, E. (2013) *Early predictors of language development in typically developing infants and infants with Down syndrome*. University of Reading. Unpublished PhD thesis.
- Meindl, J. N., & Cannella-Malone, H. I. (2011). Initiating and responding to joint attention bids in children with autism: A review of the literature. *Research in developmental disabilities, 32*(5), 1441-1454.
- Mervis, C. B., & Robinson, B. F. (2000). Expressive vocabulary ability of toddlers with Williams syndrome or Down syndrome: A comparison. *Developmental Neuropsychology, 17*(1), 111-126.
- Mervis, C. B., Becerra, A. M., Rondal, J. A., & Buckley, S. (2003). Lexical development and intervention. *Speech and language intervention in Down syndrome, 63-85*.
- Miller, J. F. (1999). Profiles of language development in children with Down syndrome. *Improving the communication of people with Down syndrome, 11-39*.
- Miller, J.F., Leddy, M.G. & Leavitt, L.A. (1999) *Improving the Communication of People with Down Syndrome*. Baltimore, MD: Paul H. Brookes Publishing.

- Miller, J. F., Miolo, G., Sedey, A., Pierce, K., & Rosin, M. (1989). Predicting lexical growth in children with Down syndrome. In *Poster presented at the Annual Meeting of the American Speech-Language-Hearing Association, St. Louis, MO.*
- Mitchell, S., Brian, J., Zwaigenbaum, L., Roberts, W., Szatmari, P., Smith, I., & Bryson, S. (2006). Early language and communication development of infants later diagnosed with autism spectrum disorder. *Journal of Developmental & Behavioral Pediatrics, 27*(2), 69-78.
- Morales, M., Mundy, P., & Rojas, J. (1998). Following the direction of gaze and language development in 6-month-olds. *Infant Behavior and Development, 21*(2), 373-377.
- Morales, M., Mundy, P., Delgado, C., Yale, M., Messinger, D., Neal, R., & Schwartz, H. K. (2000). Responding to joint attention across the 6- through 24-month age period and early language acquisition. *Journal of Applied Developmental Psychology, 21*, 283-298.
- Mullen, E. M. (1995). *Mullen scales of early learning* (AGS ed.). San Antonio, TX: Pearson.
- Mundy, P., Sigman, M., Kasari, C., & Yirmiya, N. (1988). Nonverbal communication skills in Down syndrome children. *Child development, 235-249.*
- Mundy, P., Sigman, M., & Kasari, C. (1990). A longitudinal study of joint attention and language development in autistic children. *Journal of Autism and developmental Disorders, 20*(1), 115-128.
- Mundy, P., Kasari, C., Sigman, M., & Ruskin, E. (1995). Nonverbal communication and early language acquisition in children with Down syndrome and in normally developmental children. *Journal of Speech, Language, and Hearing Research, 38*, 157-168.

- Mundy, P., Hogan, A., & Doehring, P. (1996). A Preliminary Manual for the Abridged Early Social Communication Scale (ESCS). Unpublished manuscript.
- Mundy, P., & Crowson, M. (1997). Joint attention and early social communication: Implications for research on intervention with autism. *Journal of Autism and Developmental disorders*, 27(6), 653-676.
- Mundy, P., & Gomes, A. (1998). Individual differences in joint attention skill development in the second year. *Infant Behaviour and Development*, 21, 469-482.
- Mundy, P., Fox, N., & Card, J. (2003). EEG coherence, joint attention and language development in the second year. *Developmental Science*, 6(1), 48-54.
- Mundy, P., Block, J., Delgado, C., Pomares, Y., Van Hecke, A. V., & Parlade, M. V. (2007). Individual differences and the development of joint attention in infancy. *Child development*, 78(3), 938-954.
- Murray, L. & Karpf, J. (2000). *Coding scheme for structured mother-infant play interaction at 12 months*, University of Reading.
- Murray, L., Fiori-Cowley, A., Hooper, R., & Cooper, P. (1996). The impact of postnatal depression and associated adversity on early mother-infant interactions and later infant outcome. *Child development*, 67(5), 2512-2526.
- Mutton, D., Alberman, E., & Hook, E. B. (1996). Cytogenetic and epidemiological findings in Down syndrome, England and Wales 1989 to 1993. National Down Syndrome Cytogenetic Register and the Association of Clinical Cytogeneticists. *Journal of medical genetics*, 33(5), 387-394.

- Nozadi, S. S., Spinrad, T. L., Eisenberg, N., Bolnick, R., Eggum-Wilkens, N. D., Smith, C. L., Gaertner, A. K., & Sallquist, J. (2013). Prediction of toddlers' expressive language from maternal sensitivity and toddlers' anger expressions: A developmental perspective. *Infant Behavior and Development, 36*(4), 650-661.
- O'Toole, C., & Chiat, S. (2006). Symbolic functioning and language development in children with Down syndrome. *International journal of language & communication disorders, 41*(2), 155-171.
- Oliver, B., & Buckley, S. (1994). The language development of children with Down syndrome: First words to two-word phrases. *Down Syndrome Research and Practice, 2*(2), 71-75.
- Palisano, R. J., Walter, S. D., Russell, D. J., Rosenbaum, P. L., Gémus, M., Galuppi, B. E., & Cunningham, L. (2001). Gross motor function of children with Down syndrome: creation of motor growth curves. *Archives of Physical Medicine and Rehabilitation, 82*(4), 494-500.
- Park, A. H., Wilson, M. A., Stevens, P. T., Harward, R., & Hohler, N. (2012). Identification of hearing loss in pediatric patients with Down syndrome. *Otolaryngology--Head and Neck Surgery, 146*(1), 135-140.
- Pary, R. J., & Hurley, A. D. (2002). Down syndrome and autistic disorder. *Mental Health Aspects of Developmental Disabilities, 5*, 64-65.
- Paul, R., Chawarska, K., Cicchetti, D., & Volkmar, F. (2008). Language outcomes of toddlers with autism spectrum disorders: A two year follow- up. *Autism Research, 1*(2), 97-107.
- Piaget, J. (1962). *Play, Dreams and Imitation in Childhood*. New York: Norton.

- Piaget, J. (1971). The theory of stages in cognitive development. In D. R. Green, M. P. Ford & G. B. Flamer (Eds.), *Measurement and Piaget*, (pp. 1-111). New York: McGrall Hill.
- Pierce, K., & Schreibman, L. (1997). Multiple peer use of pivotal response training to increase social behaviors of classmates with autism: Results from trained and untrained peers. *Journal of applied behavior analysis*, 30(1), 157-160.
- Pinker, S. (1994). *The language instinct*. London: Penguin.
- Presson, A. P., Partyka, G., Jensen, K. M., Devine, O. J., Rasmussen, S. A., McCabe, L. L., & McCabe, E. R. (2013). Current estimate of Down syndrome population prevalence in the United States. *The Journal of pediatrics*, 163(4), 1163-1168.
- Pueschel, S. M., & Pueschel, J. K. (1994). Biomedical Concerns in Persons with Down Syndrome. *Pediatric Physical Therapy*, 6(4), 222.
- Reynell, J., & Gruber, C. (1990). *Reynell Developmental Language Scales: US edition*. LA, California: Western Psychological Services.
- Rice, M. L., Warren, S. F., & Betz, S. K. (2005). Language symptoms of developmental language disorders: An overview of autism, Down syndrome, fragile X, specific language impairment, and Williams syndrome. *Applied psycholinguistics*, 26(1), 7-27.
- Richards, C., Jones, C., Groves, L., Moss, J., & Oliver, C. (2015). Prevalence of autism spectrum disorder phenomenology in genetic disorders: a systematic review and meta-analysis. *The Lancet Psychiatry*, 2(10), 909-916.
- Roberts, J., Long, S. H., Malkin, C., Barnes, E., Skinner, M., Hennon, E. A., & Anderson, K. (2005). A comparison of phonological skills of boys with fragile X syndrome and

- Down syndrome. *Journal of Speech, Language, and Hearing Research*, 48(5), 980-995.
- Roberts, J. E., Price, J., & Malkin, C. (2007). Language and communication development in Down syndrome. *Mental retardation and developmental disabilities research reviews*, 13(1), 26-35.
- Robins, D. L., Fein, D., & Barton, M. L. (2009). *The Modified Checklist for Autism in Toddlers (M-CHAT) self-published*. Storrs, CT.
- Rocha, M. L., Schreibman, L., & Stahmer, A. C. (2007). Effectiveness of training parents to teach joint attention in children with autism. *Journal of Early Intervention*, 29(2), 154-172.
- Rondal, J., & Comblain, A. (1996). Language in adults with Down syndrome. *Down Syndrome Research and Practice*, 4(1), 3-14.
- Rondal, J. (1998). Cases of exceptional language in mental retardation and Down syndrome: Explanatory perspectives. *Down Syndrome Research and Practice*, 5(1), 1-15.
- Rowe, M. L. (2012). A longitudinal investigation of the role of quantity and quality of child-directed speech in vocabulary development. *Child development*, 83(5), 1762-1774.
- Sameroff, A. (1975). Transactional models in early social relations. *Human development*, 18(1-2), 65-79.
- Sanz, M. T. (1996). A comparison of vicarious and written training techniques applied to early stimulation by parents of their Down syndrome babies. *Early child development and care*, 125, 111-119.

- Sanz, T., Menendez, J., & Rosique, T. (2010). Study of different social rewards used in Down's syndrome children's early stimulation. *Early child development and care, 181*(4), 487-492.
- Sachse, S., & Von Suchodoletz, W. (2008). Early identification of language delay by direct language assessment or parent report? *Journal of Developmental & Behavioral Pediatrics, 29*(1), 34-41.
- Savasir, I., Sezgin, N., & Erol, N. (2005). *Ankara Developmental Screening Inventory [Ankara Gelişim Tarama Envanteri-AGTE] (Jrd ed.)*. Ankara: Turkish Psychology Association.
- Scaife, M., & Bruner, J. S. (1975). The capacity for joint visual attention in the infant. *Nature, 253*, 265-266.
- Sigman, M., & Ruskin, E. (1999). Continuity and change in social competence of children with autism, Down syndrome and developmental delays. *Monographs of the Society for Research in Child Development, 64*, 115-130.
- Singer Harris, N. G., Bellugi, U., Bates, E., Jones, W., & Rossen, M. (1997). Contrasting profiles of language development in children with Williams and Down syndromes. *Developmental Neuropsychology, 13*(3), 345-370.
- Slonims, V., & McConachie, H. (2006). Analysis of mother–infant interaction in infants with Down syndrome and typically developing infants. *American journal on mental retardation, 111*(4), 273-289.
- Smith, D. S. (2001). Health care management of adults with Down syndrome. *American family physician, 64*(6), 1031-1044.

- Snow, C. S. (1977). The development of conversation between mothers and babies. *Journal of Child Language*, 4, 1-22.
- Snow, C. (1995). Issues in the study of input: Fine tuning, universality, individual and developmental differences, and necessary causes. In P. Fletcher & B. MacWhinney (Eds.), *The handbook of child language* (pp. 180-193). Oxford, UK: Basil Blackwell.
- Sparrow S.S., Balla, D.A., & Cicchetti, D.V. (1984). *Vineland Adaptive Behaviour Scales: Interview edition (survey form)*. American Guidance Services: Circle Pines, MN.
- Stahmer, A. C. (1995). Teaching symbolic play skills to children with autism using pivotal response training. *Journal of autism and developmental disorders*, 25(2), 123-141.
- Steele, A., Scerif, G., Cornish, K., & Karmiloff-Smith, A. (2013). Learning to read in Williams syndrome and Down syndrome: syndrome-specific precursors and developmental trajectories. *Journal of Child Psychology and Psychiatry*, 54(7), 754-762.
- Steelman, L. M., Assel, M. A., Swank, P. R., Smith, K. E., & Landry, S. H. (2002). Early maternal warm responsiveness as a predictor of child social skills: Direct and indirect paths of influence over time. *Journal of Applied Developmental Psychology*, 23(2), 135-156.
- Sterling, A., & Warren, S. F. (2014). Maternal responsivity in mothers of young children with Down syndrome. *Developmental neurorehabilitation*, 17(5), 306-317.
- Stoel-Gammon, C. (2001). Down syndrome phonology: Developmental patterns and intervention strategies. *Down Syndrome Research and Practice*, 7, 93-100.

- Stojanovik, V. (2014) Language in genetic syndromes and cognitive modularity. In: Cummings, L. (Eds.), *The Cambridge handbook of communication disorders* (pp. 541-558). Cambridge: Cambridge University Press.
- Stoneman, Z. (2007). Examining the Down syndrome advantage: Mothers and fathers of young children with disabilities. *Journal of Intellectual Disability Research*, 51(12), 1006-1017.
- Tamis-LeMonda, C. S., Bornstein, M. H., & Baumwell, L. (2001). Maternal responsiveness and children's achievement of language milestones. *Child development*, 72(3), 748-767.
- Tannock, R. (1988). Mothers' directiveness in their interactions with their children with and without Down syndrome. *American Journal on Mental Retardation*, 93, 154-165.
- Thomas, M. S., & Karmiloff-Smith, A. (2003). Modeling language acquisition in atypical phenotypes. *Psychological review*, 110(4), 647.
- Thomas, M. S., Annaz, D., Ansari, D., Scerif, G., Jarrold, C., & Karmiloff-Smith, A. (2009). Using developmental trajectories to understand developmental disorders. *Journal of speech, language, and hearing research*, 52(2), 336-358.
- Thomas, M. S., & Thomas, M. (2010). Language acquisition in developmental disorders. *Language acquisition across linguistic and cognitive systems*, 67-87.
- Thurm, A., Lord, C., Lee, L. C., & Newschaffer, C. (2007). Predictors of language acquisition in preschool children with autism spectrum disorders. *Journal of Autism and Developmental Disorders*, 37(9), 1721-1734.

- Tomasello, M. (1988). The role of joint attentional processes in early language development. *Language Sciences*, 10(1), 69-88.
- Tomasello, M., & Todd, J. (1983). Joint attention and lexical acquisition style. *First language*, 4(12), 197-211.
- Tomasello, M., & Farrar, M. J. (1986). Joint attention and early language. *Child development*, 1454-1463.
- Tomasello, M. (1995). Joint attention as social cognition. *Joint attention: Its origins and role in development*, 103-130.
- Tomasello, M. (2003). *Constructing a language: A usage-based account of language acquisition*. Cambridge, MA: Harvard University Press.
- Tomblin, J. B., Records, N. L., Buckwalter, P., Zhang, X., Smith, E., & O'Brien, M. (1997). Prevalence of specific language impairment in kindergarten children. *Journal of speech, language, and hearing research*, 40(6), 1245-1260.
- UNICEF, (1982). *Curriculum de estimulación Precoz*. Guatemala: Instituto de la Salud.
- UCDHSC Center for Nursing Research, (2006). Enhancing treatment fidelity in health behavior change studies: best practices and recommendations from the NIH Behavior Change Consortium. *Health Psychology*, 23, 443-451.
- Van der Lely, H. K. (1997). Language and cognitive development in a grammatical SLI boy: Modularity and innateness. *Journal of Neurolinguistics*, 10(2), 75-107.
- Vilaseca, R. M., & Del Rio, M. J. (2004). Language acquisition by children with Down syndrome: A naturalistic approach to assisting language acquisition. *Child Language Teaching and Therapy*, 20(2), 163-180.

- Venuti, P., De Falco, S., Esposito, G., & Bornstein, M. H. (2009). Mother–child play: children with Down syndrome and typical development. *American journal on intellectual and developmental disabilities, 114*(4), 274-288.
- Vugs, B., Hendriks, M., Cuperus, J., & Verhoeven, L. (2014). Working memory performance and executive function behaviors in young children with SLI. *Research in Developmental Disabilities, 35*(1), 62-74.
- Warren, S. F., & Brady, N. C. (2007). The role of maternal responsivity in the development of children with intellectual disabilities. *Developmental Disabilities Research Reviews, 13*(4), 330-338.
- Weismer, S. E., Lord, C., & Esler, A. (2010). Early language patterns of toddlers on the autism spectrum compared to toddlers with developmental delay. *Journal of autism and developmental disorders, 40*(10), 1259-1273.
- Weller, E. L., & Mahoney, G. J. (1983). A comparison of oral and total communication modalities on the language training of young mentally handicapped children. *Education and Training of the Mentally Retarded, 103-110*.
- Wetherby, A. M., Woods, J., Allen, L., Cleary, J., Dickinson, H., & Lord, C. (2004). Early indicators of autism spectrum disorders in the second year of life. *Journal of autism and developmental disorders, 34*(5), 473-493.
- Werner, H., & Kaplan, B. (1963). *Symbol formation: An organismic-developmental approach to language and the expression of thought*. New York: Wiley.
- Whalen, C., & Schreibman, L. (2003). Joint attention training for children with autism using behavior modification procedures. *Journal of Child psychology and psychiatry, 44*(3), 456-468.

- Whalen, C., Schreibman, L., & Ingersoll, B. (2006). The collateral effects of joint attention training on social initiations, positive affect, imitation, and spontaneous speech for young children with autism. *Journal of autism and developmental disorders*, 36(5), 655-664.
- Wheeler, A., Hatton, D., Reichardt, A., & Bailey, D. (2007). Correlates of maternal behaviours in mothers of children with fragile X syndrome. *Journal of Intellectual Disability Research*, 51(6), 447-462.
- White, P. J., O'Reilly, M., Streusand, W., Levine, A., Sigafoos, J., Lancioni, G., Fragale, C., Pierce, N., & Aguilar, J. (2011). Best practices for teaching joint attention: A systematic review of the intervention literature. *Research in Autism Spectrum Disorders*, 5(4), 1283-1295.
- Whitehurst, G. J., Smith, M., Fischel, J. E., Arnold, D. S., & Lonigan, C. J. (1991). The continuity of babble and speech in children with specific expressive language delay. *Journal of Speech, Language, and Hearing Research*, 34(5), 1121-1129.
- Wright, C. A., Kaiser, A. P., Reikowsky, D. I., & Roberts, M. Y. (2013). Effects of a naturalistic sign intervention on expressive language of toddlers with Down syndrome. *Journal of Speech, Language, and Hearing Research*, 56(3), 994-1008.
- Yoder, P. J., & Warren, S. F. (2002). Effects of prelinguistic milieu teaching and parent responsivity education on dyads involving children with intellectual disabilities. *Journal of Speech, Language, and Hearing Research*, 45(6), 1158-1174.
- Yoder, P., Woynaroski, T., Fey, M., & Warren, S. (2014). Effects of dose frequency of early communication intervention in young children with and without Down syndrome. *American Journal on Intellectual and Developmental Disabilities*, 119(1), 17-32.

- Zampini, L., & D'Odorico, L. (2011). Gesture production and language development: A longitudinal study of children with Down syndrome. *Gesture, 11*(2), 174.
- Zampini, L., Salvi, A., & D'odorico, L. (2015). Joint attention behaviours and vocabulary development in children with Down syndrome. *Journal of intellectual disability research, 59*(10), 891-901.
- Zigmond, A. S., & Snaith, R. P. (1983). The hospital anxiety and depression scale. *Acta psychiatrica scandinavica, 67*(6), 361-370.
- Zimmerman, I. L., Steiner, V. G., & Pond, R. E. (2002). *Preschool language scales* (4th ed.). San Antonio, TX: The Psychological Corporation

Appendix A: Information letters and consent forms

A.1 Recruitment poster for intervention study



Early Language Intervention for Infants with Down Syndrome aged 18-21 months

- Previous research by members of our team has found that responding to joint attention (following the point/gaze of another person) at 18-21 months of age in infants with Down syndrome is positively related to language outcomes at 30-35 months of age
- Researchers at the University of Reading are offering a free intervention programme which will target responding to joint attention in infants with Down syndrome at 18-21 months
- The intervention will occur over 10 weeks – sessions by a researcher and the parent at both the University and at home

If your child is aged between 18-21 months (or will be before the 31st May 2016) and you are interested in taking part or would like more information please contact Emily Seager:

E.Seager@pgr.reading.ac.uk

This study has been reviewed according to procedures specified by the University Research Ethics Committee and has been given a favourable opinion for conduct
www.reading.ac.uk/internal/res/ResearchEthics

A.2 Information sheet for intervention study



School of Psychology and Clinical Language Sciences

1. Contact address:

School of Psychology and Clinical Language Sciences

University of Reading

Earley Gate

Reading

RG6 7BE

Researcher: Emily Seager

Email: E.Seager@pgr.reading.ac.uk

Researcher (Supervisor): Dr. Vesna Stojanovik

Email: V.Stojanovik@reading.ac.uk

Background

Joint attention involves a shared focus between two people and an object or event; for example a mother may point and comment at an aeroplane in the sky, if her child was to follow her point and also look at the aeroplane they have responded to her initiation of joint attention.

Previous research by members of our team has found that responding to joint attention at 18 months was the most significant predictor of language at 30-35 months in a group of children with Down syndrome. Therefore the aim of this study is to target response to joint attention between 18 and 21 months of age with a view to improving language outcomes at 30-35 months.

Why are we doing this study?

The majority of children with Down syndrome have some form of language difficulty and many start school with only a few words in their vocabulary (Buckley & LePrevost, 2002). Research has highlighted that early intervention leads to better language outcomes (Aparicio and Balana, 2002) but currently very few preschool aged children with Down syndrome receive targeted language interventions. Hence we would like to find out if this intervention at age 18-21 months would lead to better language outcomes at 30-35 months.

What is the purpose of the study?

This study aims to improve language outcomes for children with Down syndrome at 30-35 months by targeting responding to joint attention.

Who would we like, is eligible, to participate in the study?

Children with Down syndrome aged 18-21 months

Do I have to take part?

It is up to you whether you wish your son/daughter to take part; you are advised to discuss any queries you may have with the researchers.

If you do choose to take part you are free to withdraw from the study at any time without having to provide a reason and without this having any effect on any future educational provision or speech and language therapy your child may get.

What will be involved if you take part?

If you choose to take part, you and your child will be invited to come to the University for some initial assessments of language, joint attention and non-verbal abilities. These will include your child being engaged with toys and pictures. We will also ask you to engage in a 5 minute play session with your child. All these assessments will be video recorded but only for the purpose of data analysis.

After this the intervention itself will start, which will take place over 10 weeks. Researchers will fully train parents and provide them with any materials they may need. Parents will be asked to visit the University with their child for one intervention session per week over the 10 weeks. These sessions will take no more than half an hour and will involve games with a variety of toys that look to facilitate responding to joint attention in your child. Some of these sessions will be video recorded but only for the purpose of providing the parent with feedback.

The rest of the intervention will be carried out by parents in their homes and should occur 3 times a week for the course of the intervention.

Once the intervention has finished parents will be asked to come to the University to have their child assessed again for language, joint attention and non-verbal skills.

Three months later parents will be asked to visit the University so that their child's joint attention can be assessed (once the child is aged between 24-29 months).

Finally parents will be asked to bring their child to the University once they are 30-35 months old so that their language can be assessed again.

Confidentiality, storage and disposal of information

All the data collected will be strictly confidential and will be used for data analysis only. Once all the data for your child has been collected this will then be anonymised. The data will be securely stored in a locked cabinet in a locked room of the University.

Are there any benefits/risks to taking part [e.g. health]?

There are no risks to taking part however we are hoping that the intervention will have benefits for you and your child's language development

What expenses and/or payment or equivalent be made for participation in the study?

We will pay travel expenses from your home address to the University of Reading at 0.45p per mile (or rail fares) when you bring your child to the University of Reading.

What will the results of the study be used for?

The data that we collect will be written up as part of a doctoral thesis and may be published in academic journals and presented at conferences. When this is done no reference will be made to any personal information and the results will not be linked to any specific individual. If you would like a copy of the results we will be happy to provide one.

Who has reviewed the study?

This project has been reviewed by the University of Reading Research Ethics Committee and has been given a favourable opinion for conduct. All researchers involved in the study have a valid DBS check from the University of Reading.

Contact details for further questions, or in the event of a complaint

If you have any further questions please contact:
E.seager@pgr.reading.ac.uk

Thank you for your help.
Emily Seager

A.3 Consent form



**University of
Reading**

Consent Form

1. I have read and had explained to me by Emily Seager the accompanying Information Sheet relating to the project on:

Early language intervention for infants with DS
2. I have had explained to me the purposes of the project and what will be required of me, and any questions I have had have been answered to my satisfaction. I agree to the arrangements described in the Information Sheet in so far as they relate to my participation.
3. I understand that participation is entirely voluntary and that I have the right to withdraw from the project any time, and that this will be without detriment.
4. I agree to the interview/session being videotaped.
5. This application has been reviewed by the University Research Ethics Committee and has been given a favourable ethical opinion for conduct.
6. I have received a copy of this Consent Form and of the accompanying Information Sheet.

Name:

Name of child:

Date of birth:

Signed:

Date:

Appendix B: Maternal interaction style coding scheme

Coding Scheme for Structured Mother-Infant Play Interaction at 12 Months.

Play Manual for play sessions carried out according to instructions set out in Stein, Woolley, Cooper & Fairburn (1994). Where indicated, the play manual has been modified and adapted from Stein, Woolley, Cooper & Fairburn (1994)¹, Wolke, Skuse & Mathiasen (1990)², Skuse, Wolke & Reilly (1992)³, and Hinde & Tamplin (1983)⁴. New items have been included and others excluded.

Structured Play Interaction Scales - Overview:

(Event Count (EC) and Rating Scale (RS))

Infant Measures:

Vocalisations % (RS/EC) ^{2&3}

Inhibition (RS)

Emotional Tone (RS) ^{2&3}

Self-Regulation (RS)

Mother Measures:

Verbal Control (EC) ⁴

Positive Expressed Emotion (EC) ¹

Negative Expressed Emotion (EC) ¹

Maternal Coercions/Intrusions (EC) ¹

Maternal Verbal Elaboration (RS)

Maternal Emotional Tone (RS) ^{2&3}

Sensitivity (RS) ^{2&3}

Joint Measures:

General Atmosphere (RS) ^{2&3}

Reciprocity (RS) ^{2&3}

Lynne Murray and Janne C.
Karpf (2000)
The Winnicott Research Unit
Department of Psychology
University of Reading
3 Earley Gate
Reading, RG6 6AL

Infant Vocalisations:

Refers to non-crying utterances or to recognisable utterances embedded in crying. These may be cooing, babbling, consonant sounds or words. Crying, per se, no matter how varied, does not qualify. NOTE: for this item, also record how many (and which) actual words/word approximations the infant elicits.

1. Definitely quiet, no, or hardly any vocalisations. Guide: 0-2 vocalisations.
2. Few vocalisations of short duration. Guide: 3-4 vocalisations.
3. Vocalisations occur as part of activities, but too intermittent to constitute vocal excitement, chatter or the like. Guide: 5-7 vocalisations.
4. Vocalisations constitute an obvious part of the infant's activity, infant vocalises for the sake of vocalising. Guide: 8-9 vocalisations.
5. Infant vocalises for most of interaction. Guide: at least 10 vocalisations.

Infant Inhibition:

Refers to how inhibited the infant seems in play and how participatory and comfortable in the situation. The very inhibited infant will show recurrent signs of wariness of toy, camera, Experimenter (whilst playing with the mother), and seek proximity with mother, as well as minimal motor movement / intensity when playing with toy as well as not explore toy or environment much. Thus this child may seem placid. The very comfortable infant will not show signs of inhibition whilst playing with the mother, and feels comfortable enough with the Experimenter not to have relapses of shyness/ coyness/ proximity seeking between toys.

1. Very Inhibited. Hardly any variation in intensity when playing with toy. Recurrent signs of wariness of toys, their function, the camera, Experimenter or other environmental circumstances. Repeated proximity seeking with mother or hardly any exploring. Overall impression is either very shy or inhibited most of the time, perhaps to the extent that warming up to the toy may not happen at all or only towards the end of the 2 ½ minutes.
2. Inhibited. Seems reluctant to initiate engagement over toy or may be preoccupied with other more familiar environmental issues (in the room or outside) and thus seems easily affected or distracted by outside factors (not as a result of poor attention, but perhaps more because this is what is familiar to the child and it is a way to shut out other unfamiliar objects/situations). It may well take time for the child to warm up to the toy on each occasion, but he or she will play with the toy, albeit usually in a quiet placid way. Variations in the intensity with which the infant plays with the toy may not be very discriminable. This child will repeatedly check the camera or Experimenter, often with a concerned look or frown,
3. Moderately Relaxed. This infant seems sociable or otherwise engaged with the environment about half the time. Thus, will show sociable/engaged behaviour on and off with moderate or more than just very brief instances of shyness, inhibition or wariness. There is detectable variation in the intensity of play with toy.
4. Relaxed. This infant does not seem affected by the situation most of the time, and seems sociable or engaged with the toy, mother, environment or even Experimenter most of the time. There may be instances of brief coyness, wariness or reassurance seeking behaviour. There is a detectable variation in the intensity in the manner he or she engages with things.
5. Very Relaxed. This child does not seem affected by the situation at all and seems content playing with the toy or engaging with mother, environment or even Experimenter. This is not to say the infant will be loud and bold, but rather that he or she gets on with the playing or has the ability to explore other aspects in the room without appearing inhibited in his/her actions, and without this appearing to be a distraction mechanism (so the child is likely to share this experience with others in the room). The infant, who does not physically move around a lot, can still be engaging with the

environment in a relaxed or natural way. Detectable variation in intensity in the manner he or she engages with things.

Infant Emotional Tone:

Refers to how happy or unhappy and fussy the infant is during the session. Take into account positive or negative verbal as well as non-verbal signs of happiness or frustration. Those infants who become very absorbed but occasionally 'let out' a positive signal will tend towards being more happy than not happy.

1. Very Unhappy. Infant seems very unhappy during the whole session, gets upset, cries and fusses for most of the session, strong protest, may wail.
2. Unhappy. At times rather unhappy and whining, fussy, short verbal protest, but responds happily to encouragement. There may be some non-verbal evidence of frustration (such as frowning).
3. Moderately Happy. Content (smiles and vocalises positively) half the time, may become briefly upset, equal mix of positive and negative affect (verbal and non-verbal). (An infant who appears neutral in tone receives a code of 3N).
4. Happy. Appears to be in a happy state more than half the time; smiles and happy vocalisations dominate, may have one brief period of negative affect or short periods of neutral mood.
5. Very Happy. Radiates happiness, highly excited, nothing is upsetting (never becomes upset), animated expressive, smiling, gleeful.

Infant Self-Regulation:

Refers to how well regulated (emotionally and physically) the infant appears generally and in response to positive, negative or neutral events during the play (including toy changes, and in particular during the last three toy sessions which are supposed to be more difficult and likely to elicit more frustration). Note the number of state-changes the infant may cycle through, as well as whether strategies adopted are of a self-soothing (e.g. sucking or fiddling) or self-distracting (e.g. focusing on alternative objects or reorienting in relation to mother/toy) nature. Thus, this is the overall impression of how emotionally and physically well regulated the infant *expresses* him or herself, or how '*contained*' they seem in the situation. It is the responses emanating from the child that are of importance. A well-regulated infant will show a *range of well-modulated responses to likes and dislikes*, whereas a dysregulated infant will resort to a limited and often extreme (e.g. marked hyper- or over activity or 'stillness' / emotional 'flatness') type of response. Incomplete or sudden bursts of movement or action and stereotypies all contribute to disorganisation when they do not make sense in the context of the infant's activity. Strange vocalisations (screeching quality, odd crying or breathing) and fearful facial expressions (which may be momentary) are also indicators of disorganisation. Play quality is likely to be affected with dysregulation. Thus, a child who is dysregulated may not engage in constructive, enjoyable play (e.g. may instead be throwing toy around randomly or just handling it without exploring).

1. Very dysregulated. Physically in terms of being hyper-, over- or under-active a most of the time. Emotionally in terms of throwing tantrums with squirming and crying and/or screaming for longer periods, or marked change between tantrums and over-excitement. Alternatively, this infant may appear emotionally very flat or apathetic. The play session may sometimes have to be cut short. Also, disorganised behaviours, such as stilling or stereotypies may occur.
2. Dysregulated. Hyper-, over- or under-active a lot of the time but may have moments of less (or more) active or normal activity level. Brief instances of fussing, crying or screaming which may be recurrent. There may be an instance of disorganised behaviour.
3. Moderately Regulated. Normal activity level more than half the time, and responds appropriately on an emotional level about half the time or more. No instances of crying or screaming although fussing or squirming may be seen. No disorganised behaviour.

4. Well Regulated. Normal activity level most of the time and mostly responds appropriately on an emotional level. Seems ‘contained’ most of the time, but may have brief moments of fussing. No disorganised behaviour, and quality of play is good.
5. Very Well Regulated. Normal activity level most of the time, and responds appropriately on an emotional level throughout the session. Although there may be very transient moments of fussing, the infant very quickly resumes play, and seems very ‘contained’ in the situation. No disorganised behaviour apparent.

Maternal Verbal Control Behaviour:

Rate Strong and Mild Control, utterances counted as separate if 3 seconds apart. Take tone of voice into account. If two statements are the same (and they follow each other), they are counted as only one statement. Take care not to code the maternal style, for example, the quietly spoken mother is not necessarily less verbally controlling than the loudly spoken mother is.

Strong Control (commands, which are often imperatives)

Commands (‘Come here!’, ‘Bring the...!’)

Strong Request (‘Look here!’)

Inhibition (‘That won’t work’)

Forbids (‘No, don’t do...!’)

Cautioning (‘I will take it away...’)

Correcting (No, you have to do...’)

Mild Control (some attempt to influence)

Suggests (‘How about doing...’)

Prompts (‘The circle goes in here...’, ‘Where does this go?’, ‘Who is it?’)

Gentle Requests (‘Would you like to...’, ‘Do you want...’, ‘Can you give...’)

Joint Suggestions (‘Shall we do...’, ‘How about if we...’)

Guides (Information accompanied by practical help)

The total number of utterances of Strong and Mild Verbal Control from the mother is added up, to give a score from zero to the actual count (i.e. 0 → actual count of event).

In addition, the total number of utterances by the mother is recorded.

Maternal Positive Expressed Emotion:

Any positive, affectionate or complimentary comment directed *at the infant*. Include here statements that are fundamentally neutral but said in positive tone of voice (e.g. ‘That’s cheating’, in an affectionate manner). Include also clapping with vocalisation if directed at infant, not necessarily with name but in response to infant’s action.

Maternal Negative Expressed Emotion:

Any critical, negative or denigratory expressions directed *at the infant*. Include here statements that are fundamentally neutral but said in negative tone of voice (e.g. ‘That’s cheating!’, while laughing with an edge to it). Note: ordinary limit setting without denigratory tone does not count as Negative Expressed Emotion (e.g. ‘Don’t do that’ could be limit setting / safety issues). Thus, negative expressed emotion are primarily based on negative (angry, critical or rejecting) comments (i.e. the content rather than tone) directed at the infant.

Maternal Coercions/Intrusions:

This refers to the number of times the mother is coercive or intrusive in play. Coercions are manifested as a forceful positioning of the infant or as forceful guidance to make the infant achieve the play task. This is the case when physically making the child post the correct shape into the sorter or hammer the ball into the hole when there is no apparent volition in the infant to do so. Coercing an infant is very different from guiding or jointly showing how. Intrusive actions inappropriately cut across, take over or disrupt the infant's activity. Intrusive physical proximity or actions that constantly distract and cut across the infant's play are counted. Very forceful verbal instructions that have a cutting across effect and are inappropriately disruptive in impact are counted as intrusions but usually intrusions have a physical intervention component.

Maternal Verbal Elaboration:

This dimension codes the extent to which a mother elaborates verbally on the toy with which she is playing with the infant. This dimension *does not* attempt to rate the effectiveness or appropriateness of the style (this is picked up by sensitivity, coercion/intrusion, verbal control and interaction measures). When rating this dimension, take into account elaborative strategies (or the absence of them) when they are directed at the infant, but do *not* take into account aspects of the infant's behaviour:

- Elaborating and absence of elaboration

Elaborating is a measure of the verbal expansion of use of the toy or information about the toy, which adds to the infant's experience. Verbal elaboration includes comments on form or function (e.g. commenting on the shapes and colours of the balls or shapes (even if this seems beyond the infant's grasp)) or relating it to other more familiar toys/objects the infant may already possess or be familiar with or people the infant knows (e.g. 'daddy' while playing with the telephone, when this goes further than just 'naming' the picture, i.e. 'daddy's calling' or 'hello daddy'). The form of the comment can be instructional ('shall we build a castle' or 'the blue one goes in there') or explanatory ('that's where the ball comes out' or 'the telephone is ringing'). Do not count general comments about infant likes and dislikes. A mother may also comment or elaborate on vocalisation made by the infant.

At the lower end of the scale of maternal verbal elaboration are mothers who never or hardly ever demonstrate verbal elaboration during the play. When they do, elaborations are very simple, only very occasional, and include only *naming* of toy, colour, shape or picture (on phone) or they might verbalise the noise the toy makes, e.g. 'bang, bang' or 'ring, ring'. At the higher end of the scale are mothers who demonstrate frequent, 'simpler' elaborations (again, mentioning of colour, noise) or other flexible/imaginative uses of the toy (e.g. 'shape sorter as a drum' or 'bricks in tower', or attempt to engage the infant in familiar games using the toy such as 'peek-a-booh') or, usually less frequent, higher quality, or 'educational' elaborations (e.g. by counting aloud number of items or comparing toy or function to another familiar object or concept, for example 'red, green, like your traffic light!' or 'daddy is calling from work'). Do not necessarily count *stating* the number of items (e.g. 'two balls') as an instance of 'higher quality counting' (e.g. 'one, two, three.....').

1. No verbal elaboration. This mother never elaborates verbally on form, shape, function, colour or count and never likens the infant's experience to something familiar.
2. Little verbal elaboration. This mother elaborates verbally a few times (guide: 1-5 times), but never uses more sophisticated ways of relating the toy to more familiar aspects of the infant's world. The types of elaborations include simple naming of toy, colour, shape or noise. [If there are signs of a flexible style of elaboration, but the total number of elaborations is small, forcing a rating of 2 is possible].
3. Moderate verbal elaboration. This mother's elaborations are frequent (guide: 5+ times) but not sophisticated. Although she may show some flexibility in referring to the toy (e.g. by likening the shape sorter to a drum), she never counts or extends the concept to other familiar concepts.

Her elaborations are kept simple. [If there are signs of a higher quality style of elaboration, but the total number of elaborations is very small, or the mother uses this higher quality style once or twice while not making use of any simpler elaborations, forcing a rating of 3 is possible].

4. Good verbal elaboration. To get this score, the mother must make use of at least one higher quality and at least a few other 'simpler' elaborations. [Alternatively, she could be using higher quality elaborations on a number of occasions, while making no, or little use of simpler elaborations].
5. Very good verbal elaboration. This mother frequently makes use of both high quality and 'simpler' elaborations. To get this score, she must have at least two high quality and a 'fair few' simpler elaborations. [Alternatively, she could be using higher quality elaborations *extensively*, while making no, or little use of simpler elaborations].

Maternal Emotional Tone:

Refers to how happy or unhappy and fussy the mother is during the play. Some mother's may laugh for no apparent reason, this is not necessarily a sign of happiness, but could indicate that the mother is nervous or uncomfortable – if it is obviously nervous don't count it as happy. Equally, a mother who seems quiet may not be unhappy, but if she seems distant or unfocused or drifts off for periods she is probably not very happy.

2. Very Unhappy. This mother seems very unhappy or distant during the whole play session or for long periods during it. She is either upset and fusses for most of the session or very distant for longer periods. Her verbal protest may be strong.
3. Unhappy. This mother seems rather unhappy and fussy (short verbal protest) or distant more than half the time.
4. Moderately Happy. This mother seems content (smiles and vocalises positively) half the time, may become very briefly upset or distant, mainly neutral mood. (If constantly neutral, code of 3N is given)
5. Happy. This mother appears to be in a predominantly happy state more than half the time, smiles and vocalises positively, some periods of neutral mood occur.
6. Very Happy. This mother seems very happy throughout the session (never becomes upset), is animated, expressive and smiley.

Maternal Sensitivity:

Refers to how sensitively the mother is attuned and responds to the infant's signals. When rating sensitivity during play, attention should be paid to the positioning of the mother and infant (distance between the mother and infant, eye to eye contact possible, freedom of movement possible, a highly sensitive mother may reposition herself rather than her infant when repositioning is necessary). Note: it must be remembered, that the mother has also been instructed to try to keep the infant from *facing away* from the camera). Attention should also be paid to the method used to attain or regain interest in a toy, as well as to the picking up cues from the infant (such as 'asking' for help or wanting to be left to explore alone) and responding to them in a temporally contingent manner. Furthermore, comments and feedback on infant behaviour and accomplishments as well as variation in stimulation during play all contribute to this dimension.

1. Highly Insensitive. The extremely insensitive mother seems geared almost exclusively to her own wishes, moods and activity. I.e. mother's interventions and initiations of interaction are *prompted or shaped largely by signals within herself*; if they mesh with the infant's signals, this is often no more than coincidence. This is not to say that the mother never responds to the infant's signals, as sometimes she does if these signals are intense enough, prolonged enough or often repeated enough. The delay in response is in itself insensitive. Furthermore, since there is usually a disparity between

the mother's own wishes and activity and infant's signals, the mother, who is largely geared to her own signals, routinely ignores or distorts the meaning of the infant's behaviour. Thus, when the mother responds to her infant's signals, *her response is characteristically inappropriate in kind, or fragmented and incomplete.*

2. **Insensitive.** The mother frequently fails to respond to infant's communications appropriately and/or promptly, although she may on some occasions show capacity for sensitivity in her responses to and interactions with her infant. Her insensitivity seems *linked to an inability to see things from the infant's point of view.* She may be too frequently preoccupied with other things and therefore inaccessible to the infant's signals and communications. She may also misperceive signals and interpret them inaccurately because of her own wishes or defences. Furthermore, she may know well enough what the infant is communicating but be disinclined to give him/her what he/she wants – because it is inconvenient or she is not in the mood for it, or because she is determined not to 'spoil' him/her. *She may delay an otherwise appropriate response to such an extent that it is no longer contingent to her infant's signal, and indeed perhaps is no longer appropriate to his state, mood or activity.* This mother may also respond with seeming appropriateness to infant's communications but break off the transactions before the infant is satisfied, so that their interactions seem fragmented and incomplete or her responses perfunctory, half-hearted or impatient. Despite such clear evidence of insensitivity, however, this mother is not as consistently or pervasively insensitive as the mother with even lower ratings. This mother *can modify her own behaviour and goals and can show some sensitivity in her handling of the infant, either when the infant's wishes, moods and activity are not too deviant from the mother's wishes and moods, or when the infant is truly distressed or otherwise communicating very forcefully and compellingly.*
3. **Inconsistently Sensitive.** Although this mother can be quite sensitive on occasion, there are some periods in which she is insensitive to her infant's communication. The mother's inconsistent sensitivity may occur for any one of several reasons, but the outcome is that she seems to have lacunae in regard to her sensitive dealings with the infant – being *sensitive at some times or in respect to some aspects of the infant's experience, but not in others.* Her awareness of the infant may be intermittent and often fairly keen, but sometimes imperious. Her perception of the infant's behaviour may also be distorted in regard to one or two aspects although it is accurate in others. She may be *prompt and appropriate in response to the infant's communications at some times and in most respects, but either inappropriate or slow at times in other respects.* On the whole, however, she is *more frequently sensitive than insensitive.* What is striking is that a mother, who can be as sensitive as she is on so many occasions, can be so insensitive on other occasions.
4. **Sensitive.** This mother also *interprets infant's communications accurately and responds to them promptly and appropriately* – but with less sensitivity than mothers with higher ratings. She may be *less attuned to infant's more subtle behaviours* compared with the highly sensitive mother. Or, perhaps because she is less skilful in dividing her attention between infant and competing demands, *she may sometimes 'miss her cue'.* The infant's *clear and definite signals are, however, neither missed nor misinterpreted.* This mother empathises with the infant and sees things from the infant's point of view. Her perceptions of the infant's behaviour are not distorted. Perhaps because her perception is less sensitive than that of other mothers with a higher rating, her responses are not as consistently prompt or as finely appropriate. *Although there may be occasional little 'mismatches', the mother's interventions are never seriously out of tune with infant's tempo, state and communications.*
5. **Highly Sensitive.** This mother is exquisitely attuned to infant's signals and responds to them promptly and appropriately. *She is able to see things from the infant's point of view; her perceptions of signals and communications are not distorted by her own needs and defences.* She 'reads' the child's signals and communications skilfully, and knows what the meaning is of even subtle, minimal and understated cues. She nearly always gives the infant what he/she indicates he/she wants, although not invariably so. When she feels that it is best not to comply with the demand – if,

for example, the infant is too excited, over-imperious, or wants something he/she should not have – she is tactful in acknowledging his communication and in offering an acceptable alternative. *She has ‘well-rounded’ interactions with the infant, so that the transaction is smoothly completed and both she and the infant feel satisfied. Finally, she makes her responses temporally contingent upon the infant’s signals and communications.*

General Atmosphere of the Interaction:

This refers to the extent of how harmonious or disharmonious (discordant and conflictual) the overall interaction between the mother and infant is. *Consider the infant’s expression in particular (to be discordant, the infant may either adopt a strategy of resisting, ignoring or avoiding).*

1. Very much discord and conflict.
2. Generally negative and/or conflictual; may be occasionally positive.
3. Neutral or bland; overall the atmosphere seems neither positive nor negative.
4. Generally positive and friendly, hardly any instant of conflict or negative feelings may be expressed.
5. Very harmonious, agreeable and peaceful, no conflict or negative feelings expressed.

Reciprocity:

This refers to the extent of mutual interchange between mother and infant. The infant’s input is received and responded to by the mother and vice versa. In particular, the degree of joint orientation and co-ordination of the actions between the mother and the infant in achieving a task goal is rated. The shared co-ordination and turn taking makes a very important contribution to this dimension. Referencing would be expected in higher reciprocal interaction, although the dyad may be so absorbed in sharing actions that there is no need for sharing the gaze as well.

1. No Reciprocity. Hardly any reciprocity is observed; there is no turn taking or communication, the mother and infant engage in different things at a given time and shared co-ordination is hardly ever observed.
2. Little Reciprocity. Reciprocal interaction rarely occurs, and only occasionally do the mother and infant incorporate the other’s suggestions. Turn taking is very rare. It is rare that the mother and infant are manipulating the same aspects of a task or that they are actually co-ordinating their efforts around the toy in focus and the interaction around the toy is tuned in.
3. Moderate Reciprocity. The session is half the time characterised by reciprocal interaction.
4. Much Reciprocity. Much of the session is characterised by reciprocal interaction, most of the exchanges are mutual and characterised by turn taking, and there is joint engagement.
5. Very Much Reciprocity. The entire session is characterised by reciprocal interaction, exchanges are mutual, finely tuned, co-ordinated and smooth.

Appendix C: Demographic questionnaire

ID:

Time point:

Demographic Questionnaire:Date of birth: (DDMMYYYY)

--	--	--	--	--	--	--	--

Gender:

male

female

Medical history (please circle y=yes or n=no):

- Was your child more than three weeks premature? (If 'yes' please give details below) Y / N
- Do you have any concerns about your child's vision? Y / N
- Does your child have any diagnosed mental, physical or emotional disabilities? Y / N

If you responded 'yes' to any of the above please explain or describe here:

-

Hearing status:

- Do you have any concerns about your child's hearing? Y / N

If you have answered 'yes' please continue with the questions below. If you answered 'no' please go on to the next section.

- Does your child have a history of ear infections? Y / N
- How many? _____ When was the most recent? _____
- Has your child had a hearing test? Y / N
- If 'yes' when? _____

Any further information:

Language and childcare:

- What language(s) does your child hear being spoken fluently at home?

- What is the main language that your child hears at home?

- How much time does your child spend in childcare per week?

- Type of childcare (please tick):

Family member

Child minder

Nursery

Nanny/Au pair

Other

- What languages do they hear spoken fluently whilst in childcare?

Other children:

- Do you have any other children? Y / N

- If 'yes' please provide their date of birth

Support services:

- Have you received or are currently receiving portage? Y / N

- Are you receiving support/therapy from speech and language services? Y / N

- If 'yes' please give details:

-

Mother:Date of birth:

--	--	--	--	--	--	--	--

Occupational status: (please tick)Employed (full time) Unemployed Employed (part time) Employed (on maternity leave) Self-employed

If employed what is your full job title? _____

Highest level of education (please tick):

None	
GCSEs/O-levels or equivalent	
A-level or equivalent	
NVQ, HND or equivalent	
Degree	
Post-graduate degree	
Other (please give details)	

Were you born in the United Kingdom? Y / N

If not, how long have you been living in the United Kingdom? _____

First language spoken: _____

Any other languages spoken: _____

Spouse or partner (if living with family):Date of birth:

--	--	--	--	--	--	--	--

Gender:Occupational status: (please tick)Employed (full time) Unemployed Employed (part time) Employed (on maternity leave) Self-employed

If employed what is your full job title? _____

Highest level of education (please tick):

None	
GCSEs/O-levels or equivalent	
A-level or equivalent	
NVQ, HND or equivalent	
Degree	
Post-graduate degree	
Other (please give details)	

Were you born in the United Kingdom? Y / N

If not, how long have you been living in the United Kingdom? _____

First language spoken: _____

Any other languages spoken: _____

Appendix D: Intervention materials used by researcher

Participant number: Intervention session: Date: (/ /) Level: –
 following a point

Trial number	1	2	3	Total % correct
Response T1				
Response T2				
Response T3				
Response T4				
Maintenance L1 (hand on)				
Maintenance L2 (tapping)				

Maintenance L3 (showing)				
Maintenance L5 (choosing toys)				
Maintenance L6 (book)				

Comments:

Appendix E: Parent materials for intervention

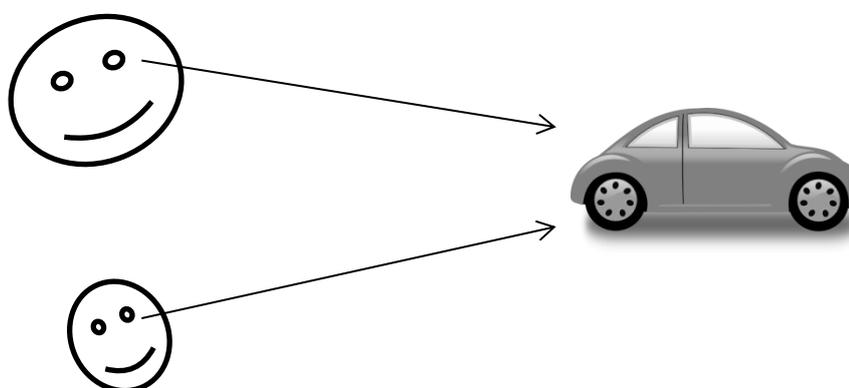
E.1 Intervention manual and weekly instructions for parents

Manual for parents: Improving responding to joint attention



What is joint attention?

Joint attention involves a shared focus between two people and an object/event. For example a parent may point out of the window and say to their child 'look at the aeroplane' – if the child follows the adult's gaze and also looks at the aeroplane then they have responded to the adult's bid for joint attention. As the child gets older their responses will become more sophisticated – they may look at the aeroplane and then back at the adult.



Why focus on joint attention to improve language?

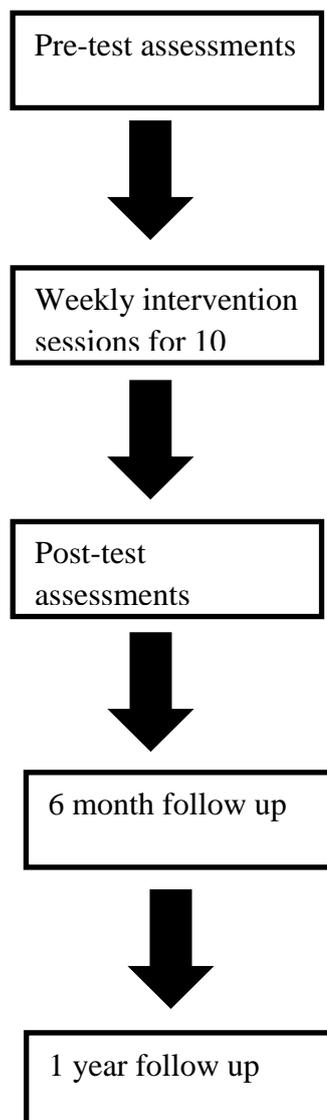
The aim of this intervention study is to improve a child's language – so why have we chosen to focus on responding to joint attention? Joint attention has been identified as a 'precursor' to language development for both children with Down syndrome and typically developing children. This means that it is a skill that develops before children start to develop language and is also related to later language development. When a parent and child have a shared focus e.g. a toy, it gives the adult an opportunity to talk to the child about that object and provide lots of language input.

Previous research from members of our team at the University of Reading found that for children with Down syndrome, responding to joint attention at 18-21 months was the most significant predictor of expressive (production/speech) and receptive (understanding) language at 30-35 months – those who were better at responding to joint attention at 18-21 months had higher language scores when they were 30-35 months.

Therefore improving the child's responding to joint attention at 18-21 months, may result in better language outcomes for the child at 30-35 months.

Intervention plan (overview):

- Aim: to improve responding to joint attention
- Duration of intervention: 10 weeks
- Researcher led sessions: 1 session per week with researchers in University clinic – consisting of structured and unstructured tasks (should take approximately 30 minutes)
- Parent led sessions: 3 sessions per week at home each lasting around 10 minutes practising activities from researcher sessions. Focus on obtaining child's attention and then naming object
- Child will move through levels of increasing difficulty
- Throughout the intervention, positive reinforcement will be used – child will be rewarded when they respond correctly



Parent sessions

Parent training

For the parent training, play with your child in a comfortable environment (e.g. on the floor) for around 10 minutes with a selection of preselected toys– this should be done 3 times a week.

The training is designed to promote generalisation of the skills learnt in the researcher-led sessions to normal day to day activities. This is why we ask you to use some of your child’s own toys in this part of the training. Suitable toys include: picture books, toy cars, dolls, wind-up toys, any ‘interactive’ toys, (e.g. if you press buttons they make a sound or light up).

For this part of the training you should continue to work on the level suggested by the researcher as well as any levels your child has already ‘passed’. For example, if you are currently working on level 3 (responding to showing), then also include opportunities for your child to respond to previous levels (responding to hand on object and responding to object being tapped).

As this element of training is designed to be ‘unstructured’, the play session should be child led – focus on toys that your child wants to play with/shows an interest in. When a suitable opportunity in play arises (for example a child loses interest in the toy they have been playing with or throws away a toy), present your child with a new toy (tapping/showing, depending on the level you are working at). When the child engages with the toy, make sure you name it (e.g. it’s a truck or it’s a dolly). Try and aim to do this 5 times during the session. Reward your child if they respond correctly by blowing bubbles or letting them play with the target toy. **The training proceeds through a number of levels (1-7). Each level is explained below.**

Parent training: level 1 – hand on object

Parent training should take place **three** times a week with **each** session lasting **10 minutes**.

Play with your child in a comfortable environment (e.g. on the floor), with a selection of preselected toys (provided by the researcher).

As this element of training is designed to be ‘unstructured’, the play session should be child led – focus on toys that your child wants to play with/shows an interest in.

When a suitable opportunity in play arises (e.g. the child has thrown away a toy they have been playing with or has lost interest in the toy they have been playing with), present your child with a new toy by gently placing their hand onto a toy. When the child engages with the toy (keeps their hand on the toy, looks at the toy or holds the toy and looks at you), make sure you name it (e.g. ‘it’s a rattle’). You can then describe what the child does with it e.g. ‘Tom is shaking the rattle’ ‘shake shake’. The child is required to ‘engage’ with the toy for at least 5 seconds.

Try and aim to do this at least 5 times during the play session. Reward your child if they respond correctly (i.e. if they play with or look at the target toy for 5 seconds), by blowing bubbles or letting them play with the target toy.

If your child does not respond or responds incorrectly, firstly use verbal prompts: call their name e.g. 'Tom looking' and then gently hold their hand on the toy for 5 seconds.

Repeat the process with a different toy.

Step by step:

1. Play on floor with child
2. Every 2-3 minutes (approximately) place your child's hand on a toy
3. If/when child engages, name the toy and provide positive feedback 'well done'
4. If the child does not engage, gently hold their hand on toy for 5 seconds
5. Play with the selected toy and provide lots of language input
6. Repeat with a different toy

Parent training: level 2 – tapping an object

When a suitable opportunity in play arises, present your child with a new toy by tapping an object in front of them. When the child engages with the toy (reaches for the toy, looks at the toy or holds the toy and looks at you), make sure you name it (e.g. 'it's a car'). You can then describe what the child does with it e.g. 'Tom is pushing the car, brum brum'. The child is required to 'engage' with the toy for at least 5 seconds.

Try and aim to do this at least 5 times during the play session. Reward your child if they respond correctly (i.e. if they play with or look at the target toy for 5 seconds), by blowing bubbles or letting them play with the target toy.

If your child does not respond or responds incorrectly, firstly use verbal prompts: call their name e.g. 'Tom looking' and then gently hold their hand on the toy for 5 seconds.

Step by step:

1. Play on floor with child
2. Tap a toy in front of your child
3. If/when child engages name the toy and provide positive feedback 'well done'
4. If the child does not engage, gently hold their hand on toy for 5 seconds

5. Play with the selected toy and provide lots of language input
6. Repeat with a different toy

Parent training: level 3 – showing an object

Play with your child in a comfortable environment (e.g. on the floor) with a selection of toys including some which you can activate, for example: wind up toys and toys that ‘talk/sing’ when you press a button.

When a suitable opportunity in play arises, ‘activate’ a new toy in front of your child to get their attention – for example wind up a toy so that it moves towards your child. Once you have activated the toy, alternate your eye gaze between the child and the toy. When the child engages with the toy (e.g. touches the toy, looks at the toy or holds the toy and looks at you), make sure you name it (e.g. ‘it’s a ball’). You can then describe what the child does with it ‘Tom is bouncing the ball’, ‘bounce bounce’. The child is required to ‘engage’ with the toy for at least 5 seconds.

Try and aim to do this at least 5 times during the play session. Reward your child if they respond correctly (i.e. if they play with or look at the target toy for 5 seconds), by using, for example, verbal praise (‘good girl/boy’) or by letting them play with the target toy.

If your child does not respond or responds incorrectly, firstly use verbal prompts: call their name e.g. ‘Tom looking’ and secondly provide a physical prompt such as gently holding their hand on the toy for 5 seconds.

Step by step:

1. Play on floor with child
2. Select a new toy and activate it in front of your child
3. If/when child engages name the toy and provide positive feedback ‘well done’
4. If the child does not engage gently hold their hand on toy for 5 seconds
5. Play with the selected toy and provide lots of language input
6. Repeat with a different toy

Parent training: level 4 – eye contact

This week the goal is to achieve eye contact with your child as it is necessary for them to be able to do this confidently for the next level.

Hold a toy in front of your child and call their name. If they make eye contact with you, give them verbal praise ('well done!' or 'good looking') and then give them the toy to play with for a few seconds.

Try and aim to do this at least 5 times during the play session. If your child does not respond or responds incorrectly, use a further series of verbal prompts 'Tom looking' and point to or bring the toy to your face.

Also continue to work on previous levels within the 10 minutes so within each session make sure you give your child an opportunity to respond by placing their hand on a toy, tapping a toy and activating a toy

Step by step:

1. Play on floor with child
2. Every minute (approximately) try to make eye contact with your by prompting them to look at you by calling their name
3. If/when your child makes eye contact with you give positive feedback 'well done'
4. If the child does not make eye contact straight away provide prompts
5. Carry on 'normal' play

Parent training: level 5 – following a point

This week the goal is for your child to follow your line of regard by following your point. For example, if you look and point at an object your child should also look at and focus on the same object.

Put two toys in front of your child. Call your child's name so that they make eye contact with you. Then point to one of the toys and say: 'Let's play with the ball' (for example). Ideally your child will follow your point and look at the selected toy. You should then name it and let them play with it until they are ready to play with a new toy. Then repeat the process again.

If your child responds incorrectly and does not engage with the selected toy, then you should use prompts. For example you could gently turn their head towards the target toy or pick up the target toy.

You can alternate who chooses which toy to play with so another time you could just put two toys in front of your child and ask them to choose.

Also continue to work on previous levels within the 10 minutes so within each session make sure you give your child an opportunity to respond by placing their hand on a toy, tapping a toy and activating a toy

Step by step:

1. Play on floor with child
2. Put two toys in front of your child and point to the one you want to play with
3. If/when your child starts to play with the toy name it and provide lots of language input
4. If the child does not make eye contact straight away provide prompts
5. Play with the selected toy and provide lots of language input
6. Repeat with a different toys

Parent training: level 6 – following a point in a book

This week the goal is for your child to follow your line of regard by following your point in a book.

Sit with your child and look through a book together. Point to different pictures and describe them to your child. 'Look at the cow!' 'What noise does a cow make? Moo'

You can alternate who points to pictures so you can let your child point to some too and when they do make sure you describe the picture they pointed to.

Also continue to work on previous levels within the 10 minutes so within each session make sure you give your child an opportunity to respond by placing their hand on a toy, tapping a toy and activating a toy

Step by step:

1. Play on floor with child
2. Open a picture book and point to things you think your child will be interested in
3. If/when your child looks at the picture describe it
4. If the child does not make eye contact straight away provide prompts

Parent training level 7 – following a point

Parent training should take place **three** times a week with **each** session lasting **10 minutes**.

This week the goal is for your child to follow your line of regard by following your point to something that is outside of their visual field (e.g. behind them or something they have to look up or down for).

Place toys/pictures around the room you are playing in, such as for example, behind the child or to the left/right of them.

Make eye contact with your child and then point to one of the toys and say 'Look, there's your dolly'. Encourage your child to turn and follow your point. Once they have followed your point, give them verbal praise (e.g. 'well done' , 'good boy/girl') and get the toy for them as a reward and let them

play with it. As they play with the toy provide lots of language input: 'look at dolly's pretty dress' 'dolly is waving hello'; 'you wave hello to dolly' etc.

If your child does not turn to follow your point gently turn their face towards the direction of the toy.

This level can also be practised in other day to day activities, such as for example, you could point to things out of a car window or objects when you are playing outside.

Also continue to work on previous levels within the 10 minutes so within each session make sure you give your child an opportunity to respond by placing their hand on a toy, tapping a toy and activating a toy

Step by step:

1. Play on floor with child
2. Point to a toy that you have placed around the room
3. If/when your child follows your point, get the toy and give it to the child so they can play with it
4. Repeat with a different toy

E.2 Intervention diary for parents

Intervention diary: week Level

Session	Completed?	Comments
1	Yes / No	
2	Yes / No	
3	Yes / No	

Appendix F: Parent questionnaire – satisfaction of interventionQuestionnaire: Early intervention for infants with Down syndrome

1. How satisfied are you generally with the intervention programme? (please circle)

Very satisfied Fairly satisfied Neutral Fairly dissatisfied Very dissatisfied

2. Do you think your child's responding to joint attention has improved since starting the intervention?

Yes

No

Comments: _____

3. Do you think any improvement in joint attention has generalised to your child's speech/language/communication skills?

Yes

No

Comments: _____

4. Have you noticed any improvement in other areas of your child's development?

Yes

No

Comments: _____

5. Have you changed anything about your communication with your child since the start of the intervention?

Yes

No

Comments: _____

6. Any general feedback would be greatly appreciated – what worked well, what could be improved, why are you satisfied/dissatisfied with the programme?

Appendix G: Normality tests

G.1 Results of one-sample Kolmogorov-Smirnov test for the DS group and TD group

	DS	TD
Age	.089	.200
NVMA	.056	<.001
Strong VC	.200	.011
Mild VC	.165	.200
PEEM	.036	.200
NEEM	<.001	<.001
Intrusions	<.001	<.001
Verbal elaboration	.007	.001
Emotional tone	.068	.013
Sensitivity	.066	.012
Reciprocity	<.001	.054
IJA	.200	.002
RJA	.119	.135
PLS AC	.200	.003
PLS EC	.004	<.001
PLS TL	.031	.095

Note. NVMA= Non-verbal mental age, VC= verbal control, PEEM= positive expressed emotion, NEEM= negative expressed emotion, IJA= initiating joint attention, RJA= responding to joint attention, PLS= Preschool Language Scales, AC= auditory comprehension, EC= expressive communication, TL total language

G.2 Results of one-sample Kolmogorov-Smirnov test for the intervention group and the control group at time point 1, 2 and 3

	IG 1	CG1	IG2	CG2	IG3	CG3
Age	.200	.200	.038	.148	.200	.090
NVMA	.200	.200	.200	.200	.200	.200
RJA	.200	.200	.200	.200	-	-
PLS AC	.200	.200	.024	.200	.110	.200
PLS EC	<.001	.113	.148	.139	.200	.200
PLS TL	.073	.200	.139	.200	.200	.200
RV	.200	.104	.200	.200	.200	.155
EV	.126	<.001	.002	.074	.010	.073
Signs	<.001	<.001	.038	.177	.200	.200

Note. IG= intervention group, CG= control group, NVMA= non-verbal mental age, RJA= responding to joint attention, PLS= Preschool Language Scales, AC= auditory comprehension, EC= expressive communication, TL total language, RV= receptive vocabulary, EV= expressive vocabulary

G.3 Results of one-sample Kolmogorov-Smirnov test for the intervention group at the post-test assessment

	Intervention group
Age	.038
NVMA	.200
M-CHAT	.199
HADS	.200
RJA total	.200
Proximal points	.000
Gaze following	.200
PLS AC	.024
PLS EC	.148
PLS TL	.139
RV	.200
EV	.002
Signs	.038

Note. NVMA= Non-verbal mental age, HADS= Hospital Anxiety and Depression Scale, RJA= responding to joint attention, PLS= Preschool Language Scales, AC= auditory comprehension, EC= expressive communication, TL total language, RV= receptive vocabulary, EV= expressive vocabulary

G.4 Results of one-sample Kolmogorov-Smirnov for whole group analysis

	Time point 1	Time point 2	Time point 3
Age	.200	.012	.039
NVMA	.200	.002	.024
IJA	.200	.017	-
RJA	.200	.086	-
Strong VC	.200	.008	-
Mild VC	.200	.200	-
PEEM	.018	.038	-
NEEM	<.001	<.001	-
Intrusions	.002	<.001	-
VE	<.001	.005	-
ET	.032	<.001	-
Sensitivity	.118	.007	-
Reciprocity	.001	.021	-
PLS AC	.041	.182	.005
PLS EC	.018	.010	.200
PLS TL	.200	.149	.200
RV	.027	.200	.200
EV	.001	<.001	.001
Signs	<.001	.008	.070

Note. NVMA= Non-verbal mental age, VC= verbal control, PEEM= positive expressed emotion, NEEM= negative expressed emotion, IJA= initiating joint attention, RJA= responding to joint attention, VE= verbal elaboration, ET= emotional tone, PLS= Preschool Language Scales, AC= auditory comprehension, EC= expressive communication, TL total language, RV= receptive vocabulary, EV= expressive vocabulary

Appendix H: Full Correlation Matrices

H.1: Correlation matrix – language scores and predictor variables for the TD and DS group

			Correlations															
			Age	NVMA	Strong	Mild	PEE	NEE	Intrusions	Verbal_elab	Emo_tone	Sensitivity	Reciprocity	IJA	RJA	PLS_AC	PLS_EC	PLS_TL
Spearman's rho	Age	Correlation Coefficient	1.000	.112	-.196	.162	.159	-.196	.169	-.161	.117	.083	.177	-.136	-.121	.170	.376*	-.319
		Sig. (2-tailed)	.	.556	.349	.439	.449	.347	.421	.441	.578	.693	.398	.483	.525	.369	.041	.086
		N	30	30	25	25	25	25	25	25	25	25	25	25	29	30	30	30
	NVMA	Correlation Coefficient	.112	1.000	.072	-.001	.014	.048	.003	-.255	-.152	-.134	-.138	.012	-.145	.218	.033	.115
		Sig. (2-tailed)	.556	.	.732	.995	.946	.819	.987	.218	.467	.522	.512	.952	.444	.247	.861	.546
		N	30	30	25	25	25	25	25	25	25	25	25	25	29	30	30	30
	Strong	Correlation Coefficient	-.196	.072	1.000	.262	.184	.336	.291	.106	-.221	-.296	-.242	-.086	-.099	.178	.063	.145
		Sig. (2-tailed)	.349	.732	.	.206	.380	.101	.158	.615	.288	.151	.244	.683	.636	.395	.767	.489
		N	25	25	25	25	25	25	25	25	25	25	25	25	25	25	25	25
	Mild	Correlation Coefficient	.162	-.001	.262	1.000	.376	-.141	.101	.151	.410*	.348	.395	-.108	.323	.107	.178	.193
		Sig. (2-tailed)	.439	.995	.206	.	.064	.501	.630	.471	.042	.088	.050	.608	.115	.611	.394	.354
		N	25	25	25	25	25	25	25	25	25	25	25	25	25	25	25	25
	PEE	Correlation Coefficient	.159	.014	.184	.376	1.000	.104	-.048	-.210	.115	.117	.198	-.254	.253	.460*	.607**	.643**
		Sig. (2-tailed)	.449	.946	.380	.064	.	.622	.821	.313	.586	.579	.342	.221	.223	.021	.001	.001
		N	25	25	25	25	25	25	25	25	25	25	25	25	25	25	25	25
	NEE	Correlation Coefficient	-.196	.048	.336	-.141	.104	1.000	.541**	-.142	-.581**	-.547**	-.559**	-.205	-.132	.047	.099	.100
		Sig. (2-tailed)	.347	.819	.101	.501	.622	.	.005	.498	.002	.005	.004	.326	.529	.823	.638	.635
		N	25	25	25	25	25	25	25	25	25	25	25	25	25	25	25	25
	Intrusions	Correlation Coefficient	.169	.003	.291	.101	-.048	.541**	1.000	.135	-.281	-.518**	-.534**	-.017	-.091	.058	.190	.214
		Sig. (2-tailed)	.421	.987	.158	.630	.821	.005	.	.519	.173	.008	.006	.936	.664	.781	.362	.304
N		25	25	25	25	25	25	25	25	25	25	25	25	25	25	25	25	
Verbal_elab	Correlation Coefficient	-.161	-.255	.106	.151	-.210	-.142	.135	1.000	.437*	.307	.202	.225	.139	-.473*	-.017	-.123	
	Sig. (2-tailed)	.441	.218	.615	.471	.313	.498	.519	.	.029	.136	.332	.280	.506	.017	.936	.557	
	N	25	25	25	25	25	25	25	25	25	25	25	25	25	25	25	25	
Emo_tone	Correlation Coefficient	.117	-.152	-.221	.410*	.115	-.581**	-.281	.437*	1.000	.867**	.762**	.369	.183	-.141	.187	.106	
	Sig. (2-tailed)	.578	.467	.288	.042	.586	.002	.173	.029	.	.000	.000	.070	.381	.502	.371	.615	
	N	25	25	25	25	25	25	25	25	25	25	25	25	25	25	25	25	
Sensitivity	Correlation Coefficient	.083	-.134	-.296	.348	.117	-.547**	-.518**	.307	.867**	1.000	.850**	.315	.052	-.165	.101	.011	
	Sig. (2-tailed)	.693	.522	.151	.088	.579	.005	.008	.136	.000	.	.000	.125	.804	.430	.632	.958	
	N	25	25	25	25	25	25	25	25	25	25	25	25	25	25	25	25	
Reciprocity	Correlation Coefficient	.177	-.138	-.242	.395	.198	-.559**	-.534**	.202	.762**	.850**	1.000	.279	.081	-.238	.177	.057	
	Sig. (2-tailed)	.398	.512	.244	.050	.342	.004	.006	.332	.000	.000	.	.177	.700	.253	.398	.788	
	N	25	25	25	25	25	25	25	25	25	25	25	25	25	25	25	25	
IJA	Correlation Coefficient	-.136	.012	-.086	-.108	-.254	-.205	-.017	.225	.369	.315	.279	1.000	-.308	.003	-.020	.018	
	Sig. (2-tailed)	.483	.952	.683	.608	.221	.326	.936	.280	.070	.125	.177	.	.105	.989	.917	.926	
	N	29	29	25	25	25	25	25	25	25	25	25	29	29	29	29	29	
RJA	Correlation Coefficient	-.121	-.145	-.099	.323	.253	-.132	-.091	.139	.183	.052	.081	-.308	1.000	-.022	.182	.131	
	Sig. (2-tailed)	.525	.444	.636	.115	.223	.529	.664	.506	.381	.804	.700	.105	.	.910	.335	.489	
	N	30	30	25	25	25	25	25	25	25	25	25	29	30	30	30	30	
PLS_AC	Correlation Coefficient	.170	.218	.178	.107	.460*	.047	.058	-.473*	-.141	-.165	-.238	.003	-.022	1.000	.449	.704**	
	Sig. (2-tailed)	.369	.247	.395	.611	.021	.823	.781	.017	.502	.430	.253	.989	.910	.	.013	.000	
	N	30	30	25	25	25	25	25	25	25	25	25	29	30	30	30	30	
PLS_EC	Correlation Coefficient	.376*	.033	.063	.178	.607**	.099	.190	-.017	.187	.101	.177	-.020	.182	.449*	1.000	.927**	
	Sig. (2-tailed)	.041	.861	.767	.394	.001	.638	.362	.936	.371	.632	.398	.917	.335	.013	.	.000	
	N	30	30	25	25	25	25	25	25	25	25	25	29	30	30	30	30	
PLS_TL	Correlation Coefficient	.319	.115	.145	.193	.643**	.100	.214	-.123	.106	.011	.057	.018	.131	.704**	.927**	1.000	
	Sig. (2-tailed)	.086	.546	.489	.354	.001	.635	.304	.557	.615	.958	.788	.926	.489	.000	.000	.	
	N	30	30	25	25	25	25	25	25	25	25	25	25	29	30	30	30	

*. Correlation is significant at the 0.05 level (2-tailed).

** . Correlation is significant at the 0.01 level (2-tailed).

Correlations

		Age	NVMA	Strong	Mild	PEE	NEE	Intrusions	Verbal_elab	Emo_tone	Sensitivity	Reciprocity	IJA	RJA	PLS_AC	PLS_EC	PLS_TL
Age	Pearson Correlation	1	.308	-.302	.011	-.180	-.008	-.084	-.456*	-.136	-.255	-.163	.174	.092	.131	-.090	.019
	Sig. (2-tailed)		.134	.183	.963	.435	.971	.718	.038	.557	.264	.479	.438	.662	.532	.668	.928
	N	25	25	21	21	21	21	21	21	21	21	21	22	25	25	25	25
NVMA	Pearson Correlation	.308	1	-.102	-.004	.060	.164	.108	-.161	-.086	.056	.055	.108	.313	.526**	.167	.386
	Sig. (2-tailed)	.134		.660	.988	.795	.477	.642	.486	.710	.809	.814	.633	.127	.007	.426	.057
	N	25	25	21	21	21	21	21	21	21	21	21	22	25	25	25	25
Strong	Pearson Correlation	-.302	-.102	1	-.295	.039	.185	.304	-.189	-.157	-.292	-.163	.155	.004	-.182	.098	-.042
	Sig. (2-tailed)	.183	.660		.195	.865	.422	.181	.412	.497	.199	.479	.503	.985	.430	.672	.857
	N	21	21	21	21	21	21	21	21	21	21	21	21	21	21	21	21
Mild	Pearson Correlation	.011	-.004	-.295	1	.226	-.066	-.147	.385	.288	.402	.230	-.318	.094	-.025	-.205	-.134
	Sig. (2-tailed)	.963	.988	.195		.326	.775	.525	.085	.206	.071	.316	.160	.686	.915	.373	.563
	N	21	21	21	21	21	21	21	21	21	21	21	21	21	21	21	21
PEE	Pearson Correlation	-.180	.060	.039	.226	1	-.043	-.113	.072	.679**	.400	.525*	-.312	.005	-.225	-.030	-.140
	Sig. (2-tailed)	.435	.795	.865	.326		.853	.627	.757	.001	.072	.015	.169	.983	.327	.898	.544
	N	21	21	21	21	21	21	21	21	21	21	21	21	21	21	21	21
NEE	Pearson Correlation	-.008	.164	.185	-.066	-.043	1	.177	-.407	-.269	-.408	-.290	-.334	.220	.115	.078	.109
	Sig. (2-tailed)	.971	.477	.422	.775	.853		.444	.067	.238	.066	.203	.139	.338	.619	.737	.639
	N	21	21	21	21	21	21	21	21	21	21	21	21	21	21	21	21
Intrusions	Pearson Correlation	-.084	.108	.304	-.147	-.113	.177	1	.244	-.010	-.022	.100	.351	.511*	.219	.208	.241
	Sig. (2-tailed)	.718	.642	.181	.525	.627	.444		.287	.966	.925	.667	.118	.018	.341	.366	.292
	N	21	21	21	21	21	21	21	21	21	21	21	21	21	21	21	21
Verbal_elab	Pearson Correlation	-.456*	-.161	-.189	.385	.072	-.407	.244	1	.380	.577**	.468*	.020	.138	.077	-.001	.042
	Sig. (2-tailed)	.038	.486	.412	.085	.757	.067	.287		.089	.006	.032	.931	.552	.741	.998	.858
	N	21	21	21	21	21	21	21	21	21	21	21	21	21	21	21	21
Emo_tone	Pearson Correlation	-.136	-.086	-.157	.288	.679**	-.269	-.010	.380	1	.819**	.826**	.013	.154	-.166	.187	.019
	Sig. (2-tailed)	.557	.710	.497	.206	.001	.238	.966	.089		.000	.000	.956	.506	.471	.417	.935
	N	21	21	21	21	21	21	21	21	21	21	21	21	21	21	21	21
Sensitivity	Pearson Correlation	-.255	.056	-.292	.402	.400	-.408	-.022	.577**	.819**	1	.839**	.074	.104	-.019	.255	.140
	Sig. (2-tailed)	.264	.809	.199	.071	.072	.066	.925	.006	.000		.000	.750	.653	.936	.264	.546
	N	21	21	21	21	21	21	21	21	21	21	21	21	21	21	21	21
Reciprocity	Pearson Correlation	-.163	.055	-.163	.230	.525*	-.290	.100	.468*	.826**	.839**	1	-.028	.305	.026	.289	.184
	Sig. (2-tailed)	.479	.814	.479	.316	.015	.203	.667	.032	.000	.000		.906	.179	.911	.204	.426
	N	21	21	21	21	21	21	21	21	21	21	21	21	21	21	21	21
IJA	Pearson Correlation	.174	.108	.155	-.318	-.312	-.334	.351	.020	.013	.074	-.028	1	.170	.200	.339	.307
	Sig. (2-tailed)	.438	.633	.503	.160	.169	.139	.118	.931	.956	.750	.906		.449	.373	.122	.164
	N	22	22	21	21	21	21	21	21	21	21	21	22	22	22	22	22
RJA	Pearson Correlation	.092	.313	.004	.094	.005	.220	.511*	.138	.154	.104	.305	.170	1	.558**	.549**	.627**
	Sig. (2-tailed)	.662	.127	.985	.686	.983	.338	.018	.552	.506	.653	.179	.449		.004	.004	.001
	N	25	25	21	21	21	21	21	21	21	21	21	22	25	25	25	25
PLS_AC	Pearson Correlation	.131	.526**	-.182	-.025	-.225	.115	.219	.077	-.166	-.019	.026	.200	.558**	1	.557**	.874**
	Sig. (2-tailed)	.532	.007	.430	.915	.327	.619	.341	.741	.471	.936	.911	.373	.004		.004	.000
	N	25	25	21	21	21	21	21	21	21	21	21	22	25	25	25	25
PLS_EC	Pearson Correlation	-.090	.167	.098	-.205	-.030	.078	.208	-.001	.187	.255	.289	.339	.549**	.557**	1	.890**
	Sig. (2-tailed)	.668	.426	.672	.373	.898	.737	.366	.998	.417	.264	.204	.122	.004	.004		.000
	N	25	25	21	21	21	21	21	21	21	21	21	22	25	25	25	25
PLS_TL	Pearson Correlation	.019	.386	-.042	-.134	-.140	.109	.241	.042	.019	.140	.184	.307	.627**	.874**	.890**	1
	Sig. (2-tailed)	.928	.057	.857	.563	.544	.639	.292	.858	.935	.546	.426	.164	.001	.000	.000	
	N	25	25	21	21	21	21	21	21	21	21	21	22	25	25	25	25

*. Correlation is significant at the 0.05 level (2-tailed).

**. Correlation is significant at the 0.01 level (2-tailed).

H.2: Correlation matrix for post-test responding to joint attention score and baseline variables

Correlations

		NVMA_BL	HADS_total	MCHAT	Behind_BL	L_R_BL	Poster_BL	Book_Point_BL	Total_RJA_B L	IJA_BL	Book_point_P T	L_R_PT	Behind_PT	Poster_PT	Total_RJA_P T
NVMA_BL	Pearson Correlation	1	-.109	-.558*	.163	.168	.199	.249	.286	.641**	.279	.160	.314	.289	.317
	Sig. (2-tailed)		.698	.031	.546	.534	.460	.352	.282	.007	.295	.554	.236	.279	.232
	N	16	15	15	16	16	16	16	16	16	16	16	16	16	16
HADS_total	Pearson Correlation	-.109	1	-.101	-.355	-.040	-.160	.183	.069	-.095	-.132	.028	-.168	-.106	-.125
	Sig. (2-tailed)	.698		.720	.194	.887	.569	.514	.808	.736	.640	.921	.549	.708	.656
	N	15	15	15	15	15	15	15	15	15	15	15	15	15	15
MCHAT	Pearson Correlation	-.558*	-.101	1	-.163	-.595*	-.549*	-.588*	-.708**	-.487	-.621*	-.587*	-.470	-.583*	-.659**
	Sig. (2-tailed)	.031	.720		.561	.019	.034	.021	.003	.066	.013	.022	.077	.023	.008
	N	15	15	15	15	15	15	15	15	15	15	15	15	15	15
Behind_BL	Pearson Correlation	.163	-.355	-.163	1	.303	.620*	-.143	.171	.121	.242	.269	.584*	.524*	.491
	Sig. (2-tailed)	.546	.194	.561		.254	.010	.597	.526	.655	.367	.313	.018	.037	.053
	N	16	15	15	16	16	16	16	16	16	16	16	16	16	16
L_R_BL	Pearson Correlation	.168	-.040	-.595*	.303	1	.936**	.363	.712**	.320	.511*	.738**	.649**	.774**	.775**
	Sig. (2-tailed)	.534	.887	.019	.254	.000	.000	.167	.002	.227	.043	.001	.007	.000	.000
	N	16	15	15	16	16	16	16	16	16	16	16	16	16	16
Poster_BL	Pearson Correlation	.199	-.160	-.549*	.620*	.936**	1	.246	.650**	.309	.511*	.708**	.751**	.831**	.820**
	Sig. (2-tailed)	.460	.569	.034	.010	.000		.359	.006	.245	.043	.002	.001	.000	.000
	N	16	15	15	16	16	16	16	16	16	16	16	16	16	16
Book_Point_BL	Pearson Correlation	.249	.183	-.588*	-.143	.363	.246	1	.896**	.189	.127	.364	.083	.217	.212
	Sig. (2-tailed)	.352	.514	.021	.597	.167	.359	.000	.000	.482	.640	.166	.760	.420	.430
	N	16	15	15	16	16	16	16	16	16	16	16	16	16	16
Total_RJA_BL	Pearson Correlation	.286	.069	-.708**	.171	.712**	.650**	.896**	1	.290	.333	.609*	.408	.550*	.541*
	Sig. (2-tailed)	.282	.808	.003	.526	.002	.006	.000		.277	.208	.012	.116	.027	.030
	N	16	15	15	16	16	16	16	16	16	16	16	16	16	16
IJA_BL	Pearson Correlation	.641**	-.095	-.487	.121	.320	.309	.189	.290	1	.189	.256	.256	.290	.290
	Sig. (2-tailed)	.007	.736	.066	.655	.227	.245	.482	.277		.483	.338	.339	.276	.277
	N	16	15	15	16	16	16	16	16	16	16	16	16	16	16
Book_point_PT	Pearson Correlation	.279	-.132	-.621*	.242	.511*	.511*	.127	.333	.189	1	.679**	.353	.542*	.746**
	Sig. (2-tailed)	.295	.640	.013	.367	.043	.043	.640	.208	.483		.004	.180	.030	.001
	N	16	15	15	16	16	16	16	16	16	16	16	16	16	16
L_R_PT	Pearson Correlation	.160	.028	-.587*	.269	.738**	.708**	.364	.609*	.256	.679**	1	.534*	.809**	.856**
	Sig. (2-tailed)	.554	.921	.022	.313	.001	.002	.166	.012	.338	.004		.033	.000	.000
	N	16	15	15	16	16	16	16	16	16	16	16	16	16	16
Behind_PT	Pearson Correlation	.314	-.168	-.470	.584*	.649**	.751**	.083	.408	.256	.353	.534*	1	.929**	.848*
	Sig. (2-tailed)	.236	.549	.077	.018	.007	.001	.760	.116	.339	.180	.033		.000	.000
	N	16	15	15	16	16	16	16	16	16	16	16	16	16	16
Poster_PT	Pearson Correlation	.289	-.106	-.583*	.524*	.774**	.831**	.217	.550**	.290	.542*	.809**	.929**	1	.964**
	Sig. (2-tailed)	.279	.708	.023	.037	.000	.000	.420	.027	.276	.030	.000	.000		.000
	N	16	15	15	16	16	16	16	16	16	16	16	16	16	16
Total_RJA_PT	Pearson Correlation	.317	-.125	-.659**	.491	.775**	.820**	.212	.541*	.290	.746**	.856**	.848**	.964**	1
	Sig. (2-tailed)	.232	.656	.008	.053	.000	.000	.430	.030	.277	.001	.000	.000	.000	
	N	16	15	15	16	16	16	16	16	16	16	16	16	16	16

*. Correlation is significant at the 0.05 level (2-tailed).

**. Correlation is significant at the 0.01 level (2-tailed).

H.4: Correlation matrix showing concurrent predictors of time point 2 language/vocabulary scores

Correlations

			NVMA_T2	RJA_T2	IJA_T2	Total_U_T2	Strong_T2	Mild_T2	PEE_T2	NEE_T2	Coercions_T2	VE_T2	EmoT_T2	Sen_T2	Recip_T2	PLS_AC_T2	PLS_EC_T2	PLS_TL_T2	CDI_U_T2	CDI_S_T2	CDI_Sg_T2	
Spearman's rho	NVMA_T2	Correlation Coefficient	1.000	.273	.234	.315	.203	.341	-.160	.296	-.030	.356	.150	.198	.220	.688**	.439*	.747**	.599**	.175	.508**	
		Sig. (2-tailed)	.	.169	.283	.164	.378	.130	.488	.192	.896	.114	.517	.390	.339	.000	.020	.000	.002	.402	.010	
		N	28	27	23	21	21	21	21	21	21	21	21	21	21	21	28	28	28	24	25	25
	RJA_T2	Correlation Coefficient	.273	1.000	.259	.342	.257	.209	.100	.334	.174	.171	.191	.422	.387	.351	.058	.300	.427*	.064	.265	
		Sig. (2-tailed)	.169	.	.233	.130	.261	.362	.667	.139	.450	.459	.408	.057	.083	.072	.772	.128	.042	.765	.210	
		N	27	27	23	21	21	21	21	21	21	21	21	21	21	27	27	27	23	24	24	
	IJA_T2	Correlation Coefficient	.234	.259	1.000	.041	.430	-.047	-.084	-.352	.197	.141	.064	-.037	-.139	.273	.059	.159	.003	-.103	.164	
		Sig. (2-tailed)	.283	.233	.	.870	.075	.853	.741	.153	.434	.577	.801	.885	.582	.208	.788	.468	.991	.865	.490	
		N	23	23	23	18	18	18	18	18	18	18	18	18	18	23	23	23	19	20	20	
	Total_U_T2	Correlation Coefficient	.315	.342	.041	1.000	.118	.764**	.075	.185	.158	.441*	.437*	.300	.548*	.174	-.203	.132	.204	-.230	.125	
		Sig. (2-tailed)	.164	.130	.870	.	.610	.000	.747	.423	.493	.045	.047	.187	.010	.452	.378	.567	.416	.344	.611	
		N	21	21	18	21	21	21	21	21	21	21	21	21	21	21	21	21	18	19	19	
	Strong_T2	Correlation Coefficient	.203	.257	.430	.118	1.000	.048	-.116	.315	.741**	.253	-.396	-.187	-.273	.167	.059	.134	-.031	-.198	-.149	
		Sig. (2-tailed)	.378	.261	.075	.610	.	.838	.618	.165	.000	.268	.076	.416	.230	.470	.798	.563	.903	.416	.544	
		N	21	21	18	21	21	21	21	21	21	21	21	21	21	21	21	21	18	19	19	
	Mild_T2	Correlation Coefficient	.341	.209	-.047	.764**	.048	1.000	.077	.111	-.073	.500*	.452*	.412	.649**	.232	-.119	.226	.294	-.478*	.077	
		Sig. (2-tailed)	.130	.362	.853	.000	.838	.	.740	.632	.754	.021	.040	.063	.001	.312	.607	.325	.237	.039	.753	
		N	21	21	18	21	21	21	21	21	21	21	21	21	21	21	21	21	18	19	19	
	PEE_T2	Correlation Coefficient	-.160	.100	-.084	.075	-.116	.077	1.000	-.354	-.266	-.130	.328	.396	.086	-.064	.368	.095	-.047	.486*	.071	
		Sig. (2-tailed)	.488	.667	.741	.747	.618	.740	.	.116	.245	.573	.147	.075	.710	.784	.101	.682	.854	.035	.771	
		N	21	21	18	21	21	21	21	21	21	21	21	21	21	21	21	21	18	19	19	
	NEE_T2	Correlation Coefficient	.296	.334	-.352	.185	.315	.111	-.354	1.000	.464*	.271	-.272	-.252	.000	.299	-.019	.261	.	-.173	-.259	
Sig. (2-tailed)		.192	.139	.153	.423	.165	.632	.116	.	.034	.235	.233	.270	1.000	.188	.935	.254	.	.480	.285		
	N	21	21	18	21	21	21	21	21	21	21	21	21	21	21	21	21	18	19	19		
Coercions_T2	Correlation Coefficient	-.030	.174	.197	.158	.741**	-.073	-.266	.464*	1.000	.028	-.386	-.382	-.240	.084	-.165	-.005	-.400	-.255	-.236		
	Sig. (2-tailed)	.896	.450	.434	.493	.000	.754	.245	.034	.	.903	.084	.088	.294	.717	.476	.983	.100	.291	.331		
	N	21	21	18	21	21	21	21	21	21	21	21	21	21	21	21	21	18	19	19		
VE_T2	Correlation Coefficient	.356	.171	.141	.441*	.253	.500*	-.130	.271	.028	1.000	.216	.297	.422	.188	-.112	.101	.102	-.421	.089		
	Sig. (2-tailed)	.114	.459	.577	.045	.268	.021	.573	.235	.903	.	.348	.192	.057	.415	.630	.663	.688	.072	.717		
	N	21	21	18	21	21	21	21	21	21	21	21	21	21	21	21	21	18	19	19		
EmoT_T2	Correlation Coefficient	.150	.191	.064	.437*	-.396	.452*	.328	-.272	-.386	.216	1.000	.782**	.691**	.053	-.153	.039	.318	.195	.210		
	Sig. (2-tailed)	.517	.408	.801	.047	.076	.040	.147	.233	.084	.348	.	.000	.001	.821	.509	.866	.198	.423	.388		
	N	21	21	18	21	21	21	21	21	21	21	21	21	21	21	21	21	18	19	19		
Sen_T2	Correlation Coefficient	.198	.422	-.037	.300	-.187	.412	.396	-.252	-.382	.297	.782**	1.000	.780**	.116	.044	.154	.565*	.204	.132		
	Sig. (2-tailed)	.390	.057	.885	.187	.416	.063	.075	.270	.088	.192	.000	.	.000	.617	.850	.506	.015	.403	.590		
	N	21	21	18	21	21	21	21	21	21	21	21	21	21	21	21	21	18	19	19		
Recip_T2	Correlation Coefficient	.220	.387	-.139	.548*	-.273	.649**	.086	.000	-.240	.422	.691**	.780**	1.000	.154	-.031	.145	.475*	-.183	.037		
	Sig. (2-tailed)	.339	.083	.582	.010	.230	.001	.710	1.000	.294	.057	.001	.000	.	.505	.892	.532	.046	.454	.881		
	N	21	21	18	21	21	21	21	21	21	21	21	21	21	21	21	21	18	19	19		
PLS_AC_T2	Correlation Coefficient	.688**	.351	.273	.174	.167	.232	-.064	-.299	.084	.188	.053	.116	.154	1.000	.372	.910**	.472*	.064	.495*		
	Sig. (2-tailed)	.000	.072	.208	.452	.470	.312	.784	.188	.717	.415	.821	.617	.505	.	.051	.000	.020	.760	.012		
	N	28	27	23	21	21	21	21	21	21	21	21	21	21	28	28	28	24	25	25		
PLS_EC_T2	Correlation Coefficient	.439*	.058	.059	-.203	.059	-.119	.368	-.019	-.165	-.112	-.153	.044	-.031	.372	1.000	.668**	.261	.668**	.459*		
	Sig. (2-tailed)	.020	.772	.788	.378	.798	.607	.101	.935	.476	.630	.509	.850	.892	.051	.	.000	.217	.000	.021		
	N	28	27	23	21	21	21	21	21	21	21	21	21	21	28	28	28	24	25	25		
PLS_TL_T2	Correlation Coefficient	.747**	.300	.159	.132	.134	.226	.095	.261	-.005	.101	.039	.154	.145	.910**	.668**	1.000	.512*	.305	.571**		
	Sig. (2-tailed)	.000	.128	.468	.567	.563	.325	.682	.254	.983	.663	.866	.506	.532	.000	.000	.	.011	.138	.003		
	N	28	27	23	21	21	21	21	21	21	21	21	21	21	28	28	28	24	25	25		
CDI_U_T2	Correlation Coefficient	.599**	.427*	.003	.204	-.031	.294	-.047	.	-.400	.102	.318	.565*	.475*	.472*	.261	.512*	1.000	.223	.414*		
	Sig. (2-tailed)	.002	.042	.991	.416	.903	.237	.854	.	.100	.688	.198	.015	.046	.020	.217	.011	.	.285	.044		
	N	24	23	19	18	18	18	18	18	18	18	18	18	18	24	24	24	25	25	24		
CDI_S_T2	Correlation Coefficient	.175	.064	-.103	-.230	-.198	-.478*	.486*	-.173	-.255	-.421	.195	.204	-.183	.064	.668**	.305	.223	1.000	.407*		
	Sig. (2-tailed)	.402	.765	.665	.344	.416	.039	.035	.480	.291	.072	.423	.403	.454	.760	.000	.138	.285	.	.043		
	N	25	24	20	19	19	19	19	19	19	19	19	19	19	25	25	25	25	26	25		
CDI_Sg_T2	Correlation Coefficient	.508**	-.265	.164	.125	-.149	.077	.071	-.259	-.236	.089	.210	.132	.037	.495*	.459*	.571**	.414*	.407*	1.000		
	Sig. (2-tailed)	.010	.210	.490	.611	.544	.753	.771	.285	.331	.717	.388	.590	.881	.012	.021	.003	.044	.043	.		
	N	25	24	20	19	19	19	19	19	19	19	19	19	19	25	25	25	24	25	26		

** . Correlation is significant at the 0.01 level (2-tailed).

* . Correlation is significant at the 0.05 level (2-tailed).

H.5: Correlation matrix showing baseline predictors of time point 3 language/vocabulary scores

Correlations

			NVMA_BL	RJA_BL	IJA_BL	Strong_BL	Mild_BL	PEE_BL	NEE_BL	Coercions_B L	VE_BL	EmoT_BL	Sens_BL	Recip_BL	PLS_AC_T3	PLS_EC_T3	PLS_TL_T3	CDI_U_T3	CDI_S_T3	CDI_Sg_T3	
Spearman's rho	NVMA_BL	Correlation Coefficient	1.000	.410*	.259	-.042	-.037	-.008	.024	.037	.014	-.290	-.135	-.205	.459*	.478**	.529**	.677**	.651**	.501**	
		Sig. (2-tailed)	.	.027	.175	.846	.864	.970	.913	.863	.950	.169	.530	.336	.012	.009	.003	.000	.000	.000	.009
		N	29	29	29	24	24	24	24	24	24	24	24	24	24	29	29	29	27	27	26
RJA_BL	RJA_BL	Correlation Coefficient	.410*	1.000	.192	.188	.112	-.014	.079	.514*	.198	-.030	-.032	.147	.367	.459*	.399*	.392*	.361	.383	
		Sig. (2-tailed)	.027	.	.319	.378	.601	.947	.715	.010	.354	.891	.881	.493	.050	.012	.032	.043	.064	.053	
		N	29	29	29	24	24	24	24	24	24	24	24	24	29	29	29	27	27	26	
IJA_BL	IJA_BL	Correlation Coefficient	.259	.192	1.000	.235	-.280	-.289	-.423*	.325	.197	.053	.185	-.014	.249	.276	.287	.024	.183	.141	
		Sig. (2-tailed)	.175	.319	.	.269	.186	.171	.040	.121	.356	.805	.388	.948	.194	.147	.132	.907	.361	.493	
		N	29	29	29	24	24	24	24	24	24	24	24	24	29	29	29	27	27	26	
Strong_BL	Strong_BL	Correlation Coefficient	-.042	.188	.235	1.000	-.154	.007	.102	.408*	-.027	-.098	-.250	-.042	-.156	.022	-.153	-.282	-.012	-.083	
		Sig. (2-tailed)	.846	.378	.269	.	.473	.974	.634	.048	.899	.647	.240	.844	.465	.920	.476	.203	.958	.722	
		N	24	24	24	24	24	24	24	24	24	24	24	24	24	24	24	22	22	21	
Mild_BL	Mild_BL	Correlation Coefficient	-.037	.112	-.280	-.154	1.000	.400	-.006	.019	.199	.328	.244	.302	-.170	-.261	-.160	.347	-.175	.095	
		Sig. (2-tailed)	.864	.601	.186	.473	.	.053	.978	.929	.351	.117	.251	.152	.427	.218	.456	.114	.436	.682	
		N	24	24	24	24	24	24	24	24	24	24	24	24	24	24	24	22	22	21	
PEE_BL	PEE_BL	Correlation Coefficient	-.008	-.014	-.289	.007	.400	1.000	.126	-.148	.182	.636**	.396	.525**	.228	-.054	.185	.082	.010	.107	
		Sig. (2-tailed)	.970	.947	.171	.974	.053	.	.559	.490	.395	.001	.055	.008	.284	.802	.388	.717	.966	.644	
		N	24	24	24	24	24	24	24	24	24	24	24	24	24	24	24	22	22	21	
NEE_BL	NEE_BL	Correlation Coefficient	.024	.079	-.423*	.102	-.006	.126	1.000	-.101	-.408*	.393	.040	.050	.291	.042	.069	-.051	.224	.209	
		Sig. (2-tailed)	.913	.715	.040	.634	.978	.559	.	.639	.048	.206	.157	.298	.846	.747	.812	.315	.298	.363	
		N	24	24	24	24	24	24	24	24	24	24	24	24	24	24	24	22	22	21	
Coercions_BL	Coercions_BL	Correlation Coefficient	.037	.514*	.325	.408*	.019	-.148	-.101	1.000	.393	.040	.050	.291	.204	.327	.304	-.162	.112	-.025	
		Sig. (2-tailed)	.863	.010	.121	.048	.929	.490	.639	.	.057	.852	.818	.167	.339	.119	.148	.470	.619	.914	
		N	24	24	24	24	24	24	24	24	24	24	24	24	24	24	24	22	22	21	
VE_BL	VE_BL	Correlation Coefficient	.014	.198	.197	-.027	.199	.182	-.408*	.393	1.000	.437	.553**	.587**	.339	.290	.482*	.138	-.015	-.090	
		Sig. (2-tailed)	.950	.354	.356	.899	.351	.395	.048	.057	.033	.005	.003	.003	.105	.169	.017	.539	.946	.697	
		N	24	24	24	24	24	24	24	24	24	24	24	24	24	24	24	22	22	21	
EmoT_BL	EmoT_BL	Correlation Coefficient	-.290	-.030	.053	-.098	.328	.636**	-.268	.040	.437	1.000	.853**	.862**	.137	-.245	.059	-.205	-.158	-.118	
		Sig. (2-tailed)	.169	.891	.805	.647	.117	.001	.206	.852	.033	.	.000	.000	.523	.248	.784	.359	.483	.610	
		N	24	24	24	24	24	24	24	24	24	24	24	24	24	24	24	22	22	21	
Sens_BL	Sens_BL	Correlation Coefficient	-.135	-.032	.185	-.250	.244	.396	-.298	.050	.553**	.853**	1.000	.864**	.293	-.123	.236	-.123	-.112	-.077	
		Sig. (2-tailed)	.530	.881	.388	.240	.251	.055	.157	.818	.005	.000	.	.000	.164	.568	.267	.585	.620	.741	
		N	24	24	24	24	24	24	24	24	24	24	24	24	24	24	24	22	22	21	
Recip_BL	Recip_BL	Correlation Coefficient	-.205	.147	-.014	-.042	.302	.525**	-.222	.291	.587**	.862**	.864**	1.000	.317	-.072	.281	-.187	-.132	-.144	
		Sig. (2-tailed)	.336	.493	.948	.844	.152	.008	.298	.167	.003	.000	.000	.000	.131	.738	.183	.404	.559	.534	
		N	24	24	24	24	24	24	24	24	24	24	24	24	24	24	24	22	22	21	
PLS_AC_T3	PLS_AC_T3	Correlation Coefficient	.459*	.367	.249	-.156	-.170	.228	-.042	.204	.339	.137	.293	.317	1.000	.420*	.931**	.491**	.324	.478*	
		Sig. (2-tailed)	.012	.050	.194	.465	.427	.284	.846	.339	.105	.523	.164	.131	.	.023	.000	.009	.099	.013	
		N	29	29	29	24	24	24	24	24	24	24	24	24	29	29	29	27	27	26	
PLS_EC_T3	PLS_EC_T3	Correlation Coefficient	.478**	.459*	.276	.022	-.261	-.054	.069	.327	.290	-.245	-.123	-.072	.420*	1.000	.677**	.562**	.538**	.431*	
		Sig. (2-tailed)	.009	.012	.147	.920	.218	.802	.747	.119	.119	.169	.248	.568	.738	.023	.	.000	.002	.004	.028
		N	29	29	29	24	24	24	24	24	24	24	24	24	29	29	29	27	27	26	
PLS_TL_T3	PLS_TL_T3	Correlation Coefficient	.529**	.399*	.287	-.153	-.160	.185	-.051	.304	.482*	.059	.236	.281	.931**	.677**	1.000	.604**	.458*	.544*	
		Sig. (2-tailed)	.003	.032	.132	.476	.456	.388	.812	.148	.017	.784	.267	.183	.000	.000	.	.001	.016	.004	
		N	29	29	29	24	24	24	24	24	24	24	24	24	29	29	29	27	27	26	
CDI_U_T3	CDI_U_T3	Correlation Coefficient	.677**	.392*	.024	-.282	.347	.082	.224	-.162	.138	-.205	-.123	-.187	.491**	.562**	.604**	1.000	.514**	.669**	
		Sig. (2-tailed)	.000	.043	.907	.203	.114	.717	.315	.470	.539	.359	.585	.404	.009	.002	.001	.	.006	.000	
		N	27	27	27	22	22	22	22	22	22	22	22	22	27	27	27	27	27	26	
CDI_S_T3	CDI_S_T3	Correlation Coefficient	.651**	.361	.183	-.012	-.175	.010	.232	.112	-.015	-.158	-.112	-.132	.324	.538**	.458*	.514**	1.000	.474*	
		Sig. (2-tailed)	.000	.064	.361	.958	.436	.966	.298	.619	.946	.483	.620	.559	.099	.004	.016	.006	.	.014	
		N	27	27	27	22	22	22	22	22	22	22	22	22	27	27	27	27	27	26	
CDI_Sg_T3	CDI_Sg_T3	Correlation Coefficient	.501**	.383	.141	-.083	.095	.107	.209	-.025	-.090	-.118	-.077	-.144	.478*	.431*	.544**	.669**	.474*	1.000	
		Sig. (2-tailed)	.009	.053	.493	.722	.682	.644	.363	.914	.697	.610	.741	.534	.013	.028	.004	.000	.014	.	
		N	26	26	26	21	21	21	21	21	21	21	21	21	21	26	26	26	26	26	

*. Correlation is significant at the 0.05 level (2-tailed).

** Correlation is significant at the 0.01 level (2-tailed).

H.6: Correlation matrix showing time point 2 predictors of time point 3 language/vocabulary scores

			Correlations																	
			NVMA_T2	RJA_T2	IJA_T2	Strong_T2	Mild_T2	PEE_T2	NEE_T2	Coercions_T2	VE_T2	EmoT_T2	Sen_T2	Recip_T2	PLS_AC_T3	PLS_EC_T3	PLS_TL_T3	CDI_U_T3	CDI_S_T3	CDI_Sg_T3
Spearman's rho	NVMA_T2	Correlation Coefficient	1.000	.273	.234	.203	.341	-.160	.296	-.030	.356	.150	.198	.220	.717**	.447*	.691**	.473*	.332	.564**
		Sig. (2-tailed)	.	.169	.283	.378	.130	.488	.192	.896	.114	.517	.390	.339	.000	.017	.000	.015	.097	.003
		N	28	27	23	21	21	21	21	21	21	21	21	21	28	28	28	26	26	25
RJA_T2		Correlation Coefficient	.273	1.000	.259	.257	.209	.100	.334	.174	.171	.191	.422	.387	.409*	.284	.365	.403*	.167	.059
		Sig. (2-tailed)	.169	.	.233	.261	.362	.667	.139	.450	.459	.408	.057	.083	.034	.150	.061	.046	.425	.783
		N	27	27	23	21	21	21	21	21	21	21	21	21	27	27	27	25	25	24
IJA_T2		Correlation Coefficient	.234	.259	1.000	.430	-.047	-.084	-.352	.197	.141	.064	-.037	-.139	.081	-.066	.002	.040	.001	.190
		Sig. (2-tailed)	.283	.233	.	.075	.853	.741	.153	.434	.577	.801	.885	.582	.713	.766	.994	.862	.998	.422
		N	23	23	23	18	18	18	18	18	18	18	18	18	23	23	23	21	21	20
Strong_T2		Correlation Coefficient	.203	.257	.430	1.000	.048	-.116	.315	.741**	.253	-.396	-.187	-.273	-.051	.016	-.102	.040	-.193	-.059
		Sig. (2-tailed)	.378	.261	.075	.	.838	.618	.165	.000	.268	.076	.416	.230	.826	.946	.661	.872	.428	.816
		N	21	21	18	21	21	21	21	21	21	21	21	21	21	21	21	19	19	18
Mild_T2		Correlation Coefficient	.341	.209	-.047	.048	1.000	.077	.111	-.073	.500*	.452*	.412	.649**	.387	.177	.393	.311	.002	.293
		Sig. (2-tailed)	.130	.362	.853	.838	.	.740	.632	.754	.021	.040	.063	.001	.083	.442	.078	.195	.994	.239
		N	21	21	18	21	21	21	21	21	21	21	21	21	21	21	21	19	19	18
PEE_T2		Correlation Coefficient	-.160	.100	-.084	-.116	.077	1.000	-.354	-.266	-.130	.328	.396	.086	-.022	-.055	-.036	-.101	.000	-.197
		Sig. (2-tailed)	.488	.667	.741	.618	.740	.	.116	.245	.573	.147	.075	.710	.924	.813	.878	.682	.999	.434
		N	21	21	18	21	21	21	21	21	21	21	21	21	21	21	21	21	19	18
NEE_T2		Correlation Coefficient	.296	.334	-.352	.315	.111	-.354	1.000	.464*	.271	-.272	-.252	.000	.260	.112	.241	.215	.000	.070
		Sig. (2-tailed)	.192	.139	.153	.165	.632	.116	.	.034	.235	.233	.270	1.000	.255	.628	.293	.376	1.000	.782
		N	21	21	18	21	21	21	21	21	21	21	21	21	21	21	21	19	19	18
Coercions_T2		Correlation Coefficient	-.030	.174	.197	.741**	-.073	-.266	.464*	1.000	.028	-.386	-.240	-.125	-.397	-.268	-.161	-.288	-.093	
		Sig. (2-tailed)	.896	.450	.434	.000	.754	.245	.034	.	.903	.084	.088	.294	.588	.075	.240	.511	.232	.714
		N	21	21	18	21	21	21	21	21	21	21	21	21	21	21	21	19	19	18
VE_T2		Correlation Coefficient	.356	.171	.141	.253	.500*	-.130	.271	.028	1.000	.216	.297	.422	.375	.257	.379	.189	.028	.314
		Sig. (2-tailed)	.114	.459	.577	.268	.021	.573	.235	.903	.	.348	.192	.057	.094	.261	.090	.439	.910	.204
		N	21	21	18	21	21	21	21	21	21	21	21	21	21	21	21	19	19	18
EmoT_T2		Correlation Coefficient	.150	.191	.064	-.396	.452*	.328	-.272	-.386	.216	1.000	.782**	.691**	.224	.029	.229	.230	.265	.261
		Sig. (2-tailed)	.517	.408	.801	.076	.040	.147	.233	.084	.348	.	.000	.001	.329	.900	.317	.344	.274	.296
		N	21	21	18	21	21	21	21	21	21	21	21	21	21	21	21	19	19	18
Sen_T2		Correlation Coefficient	.198	.422	-.037	-.187	.412	.396	-.252	-.382	.297	.782**	1.000	.780**	.204	.322	.285	.386	.186	.265
		Sig. (2-tailed)	.390	.057	.885	.416	.063	.075	.270	.088	.192	.000	.	.000	.375	.155	.211	.103	.446	.288
		N	21	21	18	21	21	21	21	21	21	21	21	21	21	21	21	19	19	18
Recip_T2		Correlation Coefficient	.220	.387	-.139	-.273	.649**	.086	.000	-.240	.422	.691**	.780**	1.000	.356	.269	.427	.484*	-.004	.320
		Sig. (2-tailed)	.339	.083	.582	.230	.001	.710	1.000	.294	.057	.001	.000	.	.114	.238	.050	.036	.987	.196
		N	21	21	18	21	21	21	21	21	21	21	21	21	21	21	21	19	19	18
PLS_AC_T3		Correlation Coefficient	.717**	.409*	.081	-.051	.387	-.022	.260	-.125	.375	.224	.204	.356	1.000	.420*	.931**	.491**	.324	.478*
		Sig. (2-tailed)	.000	.034	.713	.826	.083	.924	.255	.588	.094	.329	.375	.114	.	.023	.000	.009	.099	.013
		N	28	27	23	21	21	21	21	21	21	21	21	21	29	29	29	27	27	26
PLS_EC_T3		Correlation Coefficient	.447*	.284	-.066	.016	.177	-.055	.112	-.397	.257	.029	.322	.269	.420*	1.000	.677**	.562**	.538**	.431*
		Sig. (2-tailed)	.017	.150	.766	.946	.442	.813	.628	.075	.261	.900	.155	.238	.023	.	.000	.002	.004	.028
		N	28	27	23	21	21	21	21	21	21	21	21	21	29	29	29	27	27	26
PLS_TL_T3		Correlation Coefficient	.691**	.365	.002	-.102	.393	-.036	.241	-.268	.379	.229	.285	.427	.931**	.677**	1.000	.604**	.458*	.544**
		Sig. (2-tailed)	.000	.061	.994	.661	.078	.878	.293	.240	.090	.317	.211	.054	.000	.000	.	.001	.016	.004
		N	28	27	23	21	21	21	21	21	21	21	21	21	29	29	29	27	27	26
CDI_U_T3		Correlation Coefficient	.473*	.403*	.040	.040	.311	-.101	.215	-.161	.189	.230	.386	.484*	.491**	.562**	.604**	1.000	.514**	.669**
		Sig. (2-tailed)	.015	.046	.862	.872	.195	.682	.376	.511	.439	.344	.103	.036	.009	.002	.001	.	.006	.000
		N	26	25	21	19	19	19	19	19	19	19	19	19	19	27	27	27	27	26
CDI_S_T3		Correlation Coefficient	.332	.167	.001	-.193	.002	.000	.000	-.288	.028	.265	.186	-.004	.324	.538**	.458*	.514**	1.000	.474*
		Sig. (2-tailed)	.097	.425	.998	.428	.994	.999	1.000	.232	.910	.274	.446	.987	.099	.004	.016	.006	.	.014
		N	26	25	21	19	19	19	19	19	19	19	19	19	27	27	27	27	27	26
CDI_Sg_T3		Correlation Coefficient	.564**	.059	.190	-.059	.293	-.197	.070	-.093	.314	.261	.265	.320	.478*	.431*	.544**	.669**	.474*	1.000
		Sig. (2-tailed)	.003	.783	.422	.816	.239	.434	.782	.714	.204	.296	.288	.196	.013	.028	.004	.000	.014	.
		N	25	24	20	18	18	18	18	18	18	18	18	18	26	26	26	26	26	26

**. Correlation is significant at the 0.01 level (2-tailed).

*. Correlation is significant at the 0.05 level (2-tailed).