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An effectiveness study of a parent-child interaction therapy with children with Down syndrome

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for the degree of Master of Research

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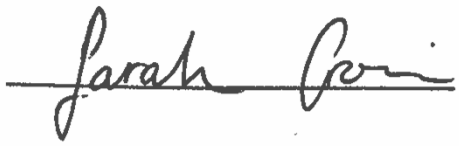
Declaration

This is to certify that the work I am submitting is my own and has not been submitted for another degree, either at University College Cork or elsewhere. All external references and sources are clearly acknowledged and identified within the contents. I have read and understood the regulations of University College Cork concerning plagiarism.

Signed: 

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Abstract

Background: Parents of children with Down syndrome (DS) often demonstrate directive parenting styles which can impede on their child's communication development. For that reason, parent-child interaction therapies have shown to be an effective form of early intervention for children with DS as it facilitates parent coaching while also addressing the specific communication needs of children with DS. This study aimed to explore the effectiveness of the PELD (Promotion of Early Language Development) intervention offered by a speech and language therapy (SLT) service for individuals with DS. The study aimed to explore the impact this programme had on the language development and communicative interactions of children with DS, while also exploring the change in the interaction and communication strategies employed by their parents.

Methodology: A single-subject multiple-baseline design was employed to evaluate the effectiveness of the PELD intervention. Seven child participants and their mothers took part in the study. All participants were aged between 10-17 months at the time of entry. Three terms of the intervention were offered over a 10 month period and families had the option of completing all or some of the terms. Data was collected over three to five time points depending on when the child commenced the intervention. Standardised assessments, parental report and observational measures were used to capture change for both the parent and child.

Results: Improvements in receptive vocabulary, use of key word signs, gesture use and a child's ability to respond to joint attention were noted in the majority of child participants. Children who attended all three terms of the intervention seemed to benefit the most from the PELD programme as they demonstrated a wide range of gestures, understood the most words and used the most Lámh signs post-intervention as reported by their parents. With regards to parent outcomes, all parents were successful in adapting their parenting style and a notable increase in each parents' ability to follow their child's lead, join in and play and incorporate a time delay into parent-child interactions was observed. Parents also used language that was developmentally appropriate for their children and increased their use of labelling and repetition of key words post-intervention.

Conclusions: The PELD programme is the first parent-child interaction therapy to be tailored specifically to children with DS who are of a very young age. There was some indication that the PELD intervention can support the development of early

language skills and the communicative intentions of young children with DS while also upskilling their parents in specific communication and interaction strategies that promote the language development of their child.

Chapter One: Down Syndrome

1.1 Introduction

This first chapter aims to describe Down syndrome (DS) with a particular emphasis on the speech, language and communication development of infants with DS in the early years of life. Parent-child interaction in DS and current parent-child interaction therapies (PCITs) will be discussed in Chapter Two. The term 'Down syndrome' was first identified by John Langton Down in 1866 (Kyle, 2012) and describes a congenital disorder that arises from a chromosomal defect. The syndrome is characterised by trademark physical and intellectual deviances and is currently the most prevailing cause of intellectual disability resulting from a chromosomal abnormality (Abbeduto, Warren & Conners, 2007; Cohen, Nadel & Madnick, 2003). DS is a genetic disorder that predominantly arises from the presence of an extra copy of chromosome 21, Trisomy 21, and to a lesser extent manifests as a result of translocation or mosaicism (Cleland, Wood, Hardcastle, Wishart & Timmins, 2010; Roberts, Price & Malkin, 2007). Translocation describes the attachment of chromosome 21 to another chromosome, whereas mosaicism is the mixture of trisomin and unaffected cells (Rogers, Roizen & Capone, 1996). Advancing maternal age, particularly in mothers over the age of 35 years, is defined as the most common risk factor for having a child with DS (de Graaf et al., 2010; Kyle, 2012; Ypsilanti & Grouios, 2008). Although this is the most recognised risk factor, young maternal age has also been linked with the syndrome (Egan et al., 2011; Mary, Gothandam & Mathew, 2010). In particular, a recent study by Corona-Rivera et al. (2019) found that maternal and paternal age of 19 years and younger may be a predisposing factor for having a child with DS. In addition to this, altered recombination during the cell meiosis has also been associated with the syndrome (Coppedè, 2016; Ghosh, Feingold & Dey, 2009; Lamb et al., 1996).

The prevalence of DS in Ireland is high in comparison to its European neighbours. In Ireland, this prevalence is estimated to be 13.1 per 10,000 live births (de Graaf et al., 2011) with a prevalence of between 9.0-11.8 per 10,000 of infants born with the syndrome in Europe (El-Hady, El-Azim & El-Talawy, 2018). A possible reason for a higher incidence of DS in Ireland may be related to the fact that the termination of pregnancies was not legalised until 2018. Down syndrome may be

diagnosed prenatally by examining fetal chromosomes through tests such as chorionic villus sampling (administered between 10 and 13 weeks of pregnancy) or amniocentesis (administered after 15 weeks of pregnancy) (Benacerraf, 2005; Mayo Clinic, n.d.). However, these tests may not always detect the presence of DS (Benacerraf, 2005), therefore some infants may be diagnosed after birth. Postnatal diagnosis of DS is initially based on the infant's physical appearance with a chromosomal karyotype test being carried out to confirm the presence of a chromosomal anomaly (Mayo Clinic, n.d.). With improvements in healthcare and medical advances over the last forty years, the life expectancy of DS has risen from mid-30s in 1982 (Barnhart & Connolly, 2007) to 60 years and beyond today (Bittles, Bower, Hussain, & Glasson, 2007).

1.1.1 Overview of Down Syndrome.

Down syndrome is a lifelong diagnosis that has a myriad of syndrome specific characteristics. With regards to the physical phenotype, people with DS present with dysmorphic facial features that include a flattened facial profile with a depressed nasal bridge, upward slanting eyes, thickened lips with a broad head and short wide neck (Ypsilanti & Grouios, 2008). The oral cavity of individuals with DS is reduced thus making the tongue appear large for the oral cavity and often protrudes (Roberts et al., 2007). A short stature, hyper flexible joints and hypotonia are also common in individuals, with the latter being linked to gastrointestinal disorders and gastroesophageal reflux experienced by this population (El-Hady et al., 2018; Leshin, 2003). The physiological composition of the brain of these individuals is also a smaller volume when compared to the typical brain with the hippocampus, prefrontal cortex and the cerebellum being of notably smaller size (Pinter, Eliez, Schmitt, Capone & Reiss, 2001; Roberts et al., 2007). Individuals with DS may also present with feeding, eating, drinking and swallowing (FEDS) difficulties (Meyer, Theodoros & Hickson, 2016), visual deficits, particularly issues surrounding visual acuity and contrast (Abou-Elhamd, ElToukhy & Al-Wadaani, 2014) and hearing loss (Roizen, 2002).

In terms of the behavioural phenotype of individuals with DS, past literature has noted a distinctive profile within the syndrome that differs from other neurodevelopmental disorders (Abbeduto et al., 2007). It is important to note that

although a specific profile has been documented, individuals with DS are described as heterogenous in nature, with huge variability noted within the syndrome (Karmiloff-Smith et al., 2016). Challenges for people with DS include cognitive delays, speech impairments and language difficulties (Patel, Wolter-Warmerdam, Leifer & Hickey, 2018). There is a broad range of cognitive impairment within this population with the majority of IQ scores ranging from 30-70 (mean score of 50) (Chapman, 1999) with some individuals demonstrating IQ scores within the normal range (Epstein, 1989). The IQ scores of individuals usually deteriorate with age and in some cases are related to early onset Alzheimer's Disease (Bush & Beail, 2004; Pennington, Moon, Edgin, Stedron & Nadel, 2003). Most individuals with DS experience some cognitive difficulties with particular deficits evident in attention, memory and auditory processing skills (Patterson, Rapsey & Glue, 2013). Although deficits in memory have been reported, the visual memory skills of people with DS are typically stronger than their verbal memory skills (Hick, Botting & Conti-Ramsden, 2005; Jarrold, Baddeley & Phillips, 2007). Furthermore, people with DS may present with additional comorbidities such as Autism Spectrum Disorder (ASD). ASD is becoming more commonly diagnosed in DS with recent studies indicating that the prevalence of ASD and DS co-occurring can be as high as 16-18% (Channell et al., 2015; Richards, Jones, Groves, Moss & Oliver, 2015). Despite the relatively high comorbidity rate, it is difficult to disentangle features of DS and ASD from each other given that intellectual disability (ID) is common in both diagnoses and future research it warranted to explore the presentation and features of ASD within Down syndrome.

With regards to speech development, people with DS differ on an anatomical level with a reduced oral cavity being noted (Roberts et al., 2007). With this in mind, misarticulations can be expected as the larger tongue and irregular dentition may reduce the precision of speech sound movements (Cleland et al., 2010). However, given the heterogenous nature of the syndrome, a range of speech abilities are evident and individuals with DS may present with delayed (Barnes et al., 2009; Meyer et al., 2016) and/or disordered speech (Cleland et al., 2010; Wong, Bebner, McCormack & Butcher, 2015). People with DS also present with reduced motor control and programming which may also contribute to the precision of speech sound movements (Kumin, 1994; Wong et al., 2015). Such inconsistencies are suggestive of dyspraxia although it is rarely diagnosed in people with DS due to the

involvement of neuromuscular deficits such as hypotonicity (Cleland et al., 2010; Kent & Vorperian, 2013). In addition to this, a recent review by Kent and Vorperian (2013) found that people with DS may also present with stuttering and/or cluttering and have reported a prevalence of between 10-45% within this cohort.

In terms of language, the deficits in language noted in DS are not in line with the cognitive profile of the individuals (Chapman, 1997; Cleland et al., 2010; Roberts et al., 2007). There is some inconsistency noted throughout the domains of language with expressive language abilities showing greater deficits in comparison to the receptive language abilities of children with Down syndrome (Cleland et al., 2010; Laws & Hall, 2014). This uneven profile is characteristic of the syndrome (Cleland et al., 2010). In particular, vocabulary comprehension often remains intact and has been shown to follow typical development in keeping with nonverbal mental age in the early years (Fowler, 1995; Laws & Bishop, 2003). On the other hand, people with DS may present with deficits in terms of syntax (the grammar aspect of language that includes word order and rules for sentence formulation) and morphology (the structure of individual words) (Kumin, 1996; Laws & Hall, 2014). Poor narrative skills and pragmatic language and literacy skills are also general characteristic of the syndrome (Abbeduto et al., 2007; Hulme et al., 2012; Frank & Esbensen, 2015; Patton & Hutton, 2016; Smith, Næss & Jarrold, 2017; Ypsilanti & Grouios, 2008). Despite poor literacy skills noted within the syndrome, recent research by Laws and Gunn (2012) and Kay-Raining Bird and Chapman (2011) have reported that some children with DS (around 10%) can achieve decoding skills that are in line with chronological age.

As outlined above, people with DS present with a range of strengths and weaknesses and although current research describes a typical profile, it is important to highlight the immense variability that has been noted within this population (Karmiloff-Smith et al., 2016). In addition to this, the typical profile of DS may not be evident at very young ages (Roberts & Richmond, 2015), therefore early intervention can help to support some of the aspects of difficulty that emerge later on. As the current study explores infants under the age of three years, the remainder of the chapter aims to explore the typical speech, language and communication development of infants with DS in the early years of life.

1.2 The Development of Language in Children with DS under 3 years

In the early stages of the infant with Down syndrome's life, there is a period where language and communication development are in keeping with their typically developing (TD) peers, as their mental age may be in line with their chronological age at this point (Maltese, Rappo, Scifo & Pepi, 2012). As they progress into their second year of life, infants with DS have been noted to spend longer in the prelinguistic stage of communication when compared to their TD peers (Abbeduto et al., 2007). This next section will first outline the differences in prelinguistic communication that is evident in infants with DS when compared to their TD peers and the implications this can have for their language development. Following this, early receptive and expressive language skills in DS is discussed in detail and comparisons to typical development is outlined. Finally, additional factors that may affect the language development of children with DS in the early years is reviewed.

1.2.1 Prelinguistic development.

1.2.1.1 Social interaction.

Firstly, communicative intentions are an important aspect in the prelinguistic stage of early language development. Communicative intentions, such as smiling, laughing, looking, pointing, reaching, emerge in early infancy in children with DS and follow the same developmental path as TD children (Chapman, 1997). However, it has been noted that such intentions tend to be less spontaneous and are instead elicited in imitation or response to the parents' signal of such actions (Chapman, 1997). Interestingly, infants with DS tend to be fixated on their social partner with little interest in objects being noted during parent-child interactions (Chapman, 1997; D'Souza, D'Souza, Johnson & Karmiloff-Smith, 2015; Wright, Kaiser, Reikowsky & Roberts, 2013). This increased interest in a social partner is characteristic of individuals with DS whose social skills are relatively intact when compared to other areas of their development (Down Syndrome Education International, 2020). Baron-Cohen, Leslie and Frith in 1985 reported that the social abilities of children with DS are a relative strength of this population as they found children with DS demonstrate social interest in others and are described as amiable and friendly in manner (Fidler, Hepburn & Rogers, 2006; Freeman & Kasari, 2002). Children with DS exhibit such social behaviours in early infancy with increased smiling and looking noted when

compared to TD peers as reported by Kasari, Mundy, Yirmiya and Sigman (1990), thus, showing that such social behaviours are innate in children with DS from birth. However, recent research by Wishart, Cebula, Willis and Pitcairn (2007) found that although children with DS may demonstrate better social skills than children of other developmental difficulties when matched on non-verbal IQ, some areas of social competence are affected such as emotional regulation and emotional recognition with regards to facial expressions (Kasari, Freeman & Hughes, 2001; Wishart et al., 2007). In addition to this, although children with DS show interest in their caregivers in the early years of life, they are less likely to initiate and maintain such interactions when compared to TD children of the same age (Jones, 1977). Research recommends that the social strengths of children with DS should be maximised and incorporated into interventions to promote further social and cognitive development (Iacob & Musuroi, 2013).

1.2.1.2 Joint attention.

Joint attention describes an individual's ability to engage with an object/activity of interest with another social partner with both parties showing an awareness that the attention is shared (Baldwin, 1995). In TD children, joint attention emerges between 9 and 12 months, with the number of episodes of joint attention substantially increasing between 12 to 18 months (Carpenter, Nagell, Tomasello, Butterworth & Moore, 1998; Bakeman & Adamson, 1984; Abbeduto et al., 2007; Ypsilanti & Grouios, 2008). This skill plays an important role in a child's social, cognitive and language development, in particular the emergence of vocabulary and pragmatic abilities (Baldwin, 1995; Bruner, 1975). In both typically developing children and children with DS, a child's ability to respond to joint attention has shown to be a predictor of early vocabulary development and developmental delays (Zampini, Salvi & D'odorico, 2015; Delgado et al., 2002). A recent study by Mason-Apps, Stojanovik, Houston-Price and Buckley (2018) explored joint attention in 14 infants with DS and compared them to 35 typically developing infants who were matched on non-verbal mental age. The aim of the study was to see if joint attention could predict language outcomes in children with DS. Interestingly, they found that nonverbal mental age and responsiveness to joint attention before the age of 24 months was a predictor for later language development

in the DS group. Similarly, a study by Zampini et al. (2015) (n = 18) who explored the attention abilities of children with DS aged 24 months, reported similar results with the children in this study also demonstrating long periods of joint attention during a play session with their caregivers. Zampini and colleagues also suggest that engaging in joint attention can encourage the vocabulary development of children with DS and should be incorporated into early language interventions (Zampini et al., 2015).

With regard to infants with DS, there are contradictory findings within current literature regarding children with DS's joint attention abilities. On one hand, researchers have found joint attention to be an area of difficulty for infants with DS (Abbeduto et al., 2007; Berger & Cunningham, 1983; Kasari, Freeman, Mundy & Sigman, 1995; Ypsilanti & Grouios, 2008) while other authors take the view that joint attention is a strength of children with DS in comparison to children with other developmental disorders (Adamson, Bakeman, Deckner & Romski, 2009; Hahn, Loveall, Savoy, Neumann & Ikuta, 2018). In a recent metanalysis by Hahn et al. (2018), the joint attention skills of children with DS were compared to that of TD children, children with other developmental disabilities (DD) and children with ASD, all of who were matched on mental age. Hahn and colleagues report that children with DS showed similar joint attention to TD peers of similar mental age and higher joint attention abilities than children with other DDs or ASD (Hahn, et al., 2018).

Although Hahn et al. (2018) describe joint attention as an area of strength for children with DS in comparison to children with other DDs, many researchers are of the belief that the development of joint attention is an area of perceived difficulty for this population. Firstly, Berger and Cunningham (1983) report that children with DS tend to respond to their social partner with inappropriate eye contact that is longer in duration in comparison to TD peers who matched on chronological age. In addition to this, Kasari et al. (1995) found that infants with DS (age 13-42 months) noted a difficulty in shifting their attention between objects and people when compared to TD children of the same mental age. Abbeduto et al. (2007) and Ypsilanti and Grouios (2008) reiterate such findings and report that the deficits evident in joint attention and sustained attention are a contributory factor to the linguistic deficits experienced by individuals with DS in later infancy. This may be due to the fact that infants with DS take longer to disengage from people/faces and demonstrate a

preference for people/faces in comparison to object engagement (Chapman, 1997; Hahn, 2016; Wright et al., 2013). Object play is an important prerequisite skill needed for the development of language and is an area that should be targeted in order to promote language development (Kasari, Gulsrud, Freeman, Paparelle & Helleman, 2012). It has been noted that along with a delay in the engagement of objects, toddlers with DS do not engage with symbols which is again an important milestone for children to overcome in an effort to promote the development of language skills (O'Toole & Chiat, 2006; Wright et al., 2013).

Although these children are fixed on their parents face for longer than expected, the children with DS who spend an increased amount of time engaged with faces had greater vocabularies than the children with DS who engaged with faces to a lesser extent (D'Souza et al., 2015). It was also found that the duration and amount a parent engaged in joint attention with their child was related to the receptive language development of the child (Harris, Kasari & Sigman, 1996; Zampini et al., 2015). Considering the impact joint attention can have on the language development of children with DS, it is important for early language interventions to promote and foster the development of this prelinguistic skill and should capitalise on the joint attention capabilities of children with DS in order to promote the development of other language processes.

1.2.1.3 Gesture.

The link between gesture and language development had been widely researched in a range of clinical populations. In typically developing children, the use of gestures has been linked with the acquisition of spoken language (Colonnesi, Stams, Koster, & Noom, 2010). In particular, deictic gestures (such as pointing and reaching) are an important prerequisite skill for language development (Colonnesi et al., 2010) and the use of deictic gestures can predict later vocabulary size and diversity (Rowe, Özçalışkan & Goldin-Meadow, 2008). In TD children it has been noted that the earlier a child points or reaches for an object, the earlier this word is likely to be produced expressively by the child (Iverson & Goldin-Meadow, 2005). This may be due to the fact that as a child starts to use such gestures, for example when reaching for an object, they are exposed to more language as parents often label the object in response. Deictic, iconic (which are representational gestures for

actions) and conventional gestures (for example waving ‘bye bye’) are the first gestures to be produced by TD infants and typically occur between 9-12 months of age (Carpenter et al., 1998).

Gesture use is a strength of infants with DS and the acquisition of gestures is often comparable to TD peers in terms of age of acquisition and gesture types (Iverson, Longobardi & Caselli, 2003; Zampini & D'Odorico, 2011). In addition to this, Franco and Wishart (1995) and Caselli et al. (1998) report that infants with DS produced nearly twice as many gestures as their typically developing peers to compensate for their delay in expressive language. Kaat-van den Os, Jongmans, Volman and Lauteslager (2015) conducted a systematic review exploring the relationship between gestures and language development in children with DS. Results from the review reiterate the findings noted by Franco and Wishart (1995) and Caselli et al. (1998) with an additional finding that the use of deictic gestures can also predict the language capabilities of a child with DS. In addition, they found that children with DS used deictic gestures such as pointing, showing and giving, in a similar manner to their TD peers (who were matched on mental age) but unlike TD children they extended the use of these gesture types when requesting an object (Kaat-van den Os et al., 2015). However, it is important to note that although infants with DS, use deictic gestures to request objects, their overall non-verbal requesting ability is delayed in comparison to TD peers which in itself has been found to be linked to later expressive language skills (Mundy, Kasari, Sigman & Ruskin, 1995). Another gesture that is evident in young children with DS is the use of conventional gestures. A study by Chan and Iacono (2001) found that conventional gestures were the most prevalent gestures in children with DS aged between 17-19 months (n = 3) with deictic gestures being the second most prevalent gestures used by the children. With regards to iconic gestures, they were found to be the most commonly used gestures by older children with DS aged between 3;8 to 8;3 years in a study by Stefanini et al. (2007). This may be partially related to the fact that key word signing, which is discussed later in the chapter, is commonly used with children with DS to compensate for the spoken language deficits noted by this population.

Not only is gesture use an area of relative strength for children with DS, it is also important for the development of language (Dimitrova et al., 2016). Previous research has also shown that gesture use can predict the language abilities of children with DS at 24 and 36 months of age (Kaat-van den Os et al., 2015; Zampini &

D'Odorico, 2011). Therefore, the development and promotion of gestures should be incorporated into early language interventions where possible to promote the language development of these children.

1.2.1.4 Babbling.

Babbling is an important precursor for the development of speech and expressive language abilities (Nyman & Lohmander, 2018). Before children can communicate verbally using language, all children learn to communicate through gestures and vocalisations (Roberts et al., 2007). In TD infants, early vocalisations such as cooing, laughing and other vegetative sounds emerge from birth to six months (Olswang, Stoel-Gammon, Coggins & Carpenter, 1987). Babbling, which is defined as any vowel and consonant combination (Olswang et al., 1987), emerges between six and 10 months in TD infants (Oller, Eilers, Neal & Cobo-Lewis, 1998). Similarly, babbling in TD children consists of consonants (such as stops, nasals and glides) and syllable shapes that are of a similar composition of the first words elicited by children (Stoel-Gammon, 1985).

For children with DS, there is contradictory evidence regarding the onset of babbling within this population (Kent & Vorperian, 2013). A review by Kent and Vorperian (2013) report that some researchers are of the view that children with DS develop vocalisations and babbling in a similar manner to their TD peers (Dodd, 1972; Steffens, Oller, Lynch, & Urbano, 1992; Smith & Oller, 1981). On the contrary, others report that although early vocalisations follow typical development (Berger & Cunningham, 1983), the emergence of canonical babbling (consonant and vowel combination) is delayed by approximately two months (Cobo-Lewis, Oller, Lynch & Levine, 1996; Lynch, Oller, Steffens & Levine, 1995) and often continues into the second year of life (Stoel-Gammon, 1997). Although the canonical babble is delayed, Smith and Oller (1981) report that the emergence of the reduplicated babble is in line with TD peers. Berger and Cunningham (1983) also found that infants with DS presented with very low levels of vocalisations in the first three to four months of life in comparison to TD infants of the same chronological age. A possible reason for this may be due to the hearing difficulties associated with DS (Berger & Cunningham, 1983). The conflicting findings within the literature may in part be attributed to the different procedural methods employed by the studies (Kent &

Vorperian, 2013). A study by Thiemann-Bourque, Warren, Brady, Gilkerson and Richards (2014) explored early vocal development of nine children with DS (age 9-54 months) to nine TD peers of a similar chronological age. The researchers used the a vocal analysis software known as Language Environment Analysis System (LENA) to measure change in the DS group. Following the analysis, Thiemann-Bourque and colleagues found no significant differences between the early vocalisations of the DS and TD group at the age of 12 months (Thiemann-Bourque et al., 2014). They did however note that the expressive language of children with DS develops at a slower pace after 12 months with a delay in expressive language evident at 24 months of age. Findings by Thiemann-Bourque et al. (2014) state that this delay in expressive language is evident despite the parents of the children providing optimum language input in the early years and that future interventions should explore alternative means of promoting such development in the children with DS at 24 months. Therefore although children with DS develop babbling in a similar way to their TD peers, interventions should focus on creating awareness of the sound shapes and syllable formation that are characteristic of early speech in order to promote and facilitate the transition from babbling to first words given the shared phonotactic proprieties between the two (Stoel-Gammon, 2001).

1.2.2 Early language development.

1.2.2.1 Receptive vocabulary.

It has been noted that for infants with DS, comprehension is a relative strength at this young age despite a delay in expressive language evident from infancy (Ypsilanti & Grouios, 2008). In particular, receptive vocabulary has shown to be relatively intact in young children with DS when compared to the expressive language deficits (for example, the poor morpho-syntactic abilities) noted in this population (Abbeduto et al., 2007; Næss, Lyster, Hulme & Melby-Lervåg, 2011). Although receptive vocabulary remains somewhat intact, this skill develops at a slower rate when compared to TD peers when matched on nonverbal mental age (Cuskelly, Povey & Jobling, 2016). Fast-mapping, which describes the learning of a word after a few exposures, is also a relative strength within this population and is postulated to be related to the strong comprehension abilities demonstrated in children with DS (Chapman, 1997). A study by Mosse and Jarrold (2011) found that individuals with DS can continue to learn new vocabulary through repetition.

Considering this and the fact that the fast-mapping abilities of individual's with DS are relatively intact, vocabulary expansion and development is often a goal for intervention for children in the early years of life, throughout childhood and up to adolescence.

With regards to the types of words used by children with DS, a higher prevalence of nouns is reported in the vocabulary of these individuals which suggests that nouns are more easily acquired than verbs for children with DS population (Chapman, 1997). This may partially be explained by the difficulties in bootstrapping evident in this population which describes the learning of morphological rules and structure of a language through exposure. Chapman (1997) noted a deficit with bootstrapping as children with DS demonstrate difficulty in terms of the understanding of verb modifications with an inability to draw on information about a verb within a given syntactic frame being noted (Chapman, 1997). Even though verb usage is impaired, people with DS have been noted to produce irregular past tense verbs more accurately than regular past tense verbs. Pinker (1991) suggested that irregular past tense verbs are stored differently in the brain and that they may be retrieved from the memory as vocabulary items rather than words to which a morphological rule has been applied.

1.2.2.2 First spoken words.

In typically developing children, first words are usually elicited by their first birthday (Bates, Bretherton & Snyder, 1991). However, for children with DS, first words tend to emerge at the later age of 18-36 months (Roberts et al., 2007; Roizen, 2002), thus demonstrating a delay in expressive language. Expressive vocabulary growth increases but at a slower rate and individuals with DS do not demonstrate the expressive 'vocabulary explosion' that has been noted in typically developing children (Abbeduto et al., 2007; Caselli et al., 1998; Ypsilanti & Grouios, 2008). The gap between expressive and receptive language widens as the infant develops, with researchers believing that language follows typical development that is delayed rather than deviant (Polišenská & Kapalková, 2014). By the age of two years, TD children demonstrate an expressive vocabulary of approximately 250 words. Although there is huge variability noted within children with DS, most do not demonstrate such competence in early vocabulary at two years, thus again

highlighting the expressive language delay. Buckley (2000) outlined the typical vocabulary growth of children with DS and found that children with DS used approximately 28 words at two years of age, a mean of 116 words at three years, a mean of 248 words at four years, 272 at five years and finally 330 words at 6 years (Stoel-Gammon, 2001). Similarly, in a study by Oliver and Buckley (1994) results showed that children with DS started to combine two-word phrases at the age of 36.9 months however, huge variability was again noted among the children. As the child with DS advances from infancy into childhood, specific impairments in syntax and morphology become evident (Sanoudaki & Varlokosta, 2014; Ypsilanti & Grouios, 2008) which suggests children with DS follow a different development trajectory in terms of expressive language development (Chapman et al., 1998). As deficits in expressive language are evident from the first year of life, key-word signing is commonly employed with infants with DS in order to promote expressive communication.

1.2.2.3 Key word signing.

Key word signing consists of both signing and speaking a word aloud as a means of total communication (Mirenda & Iacona, 2009). In Ireland, the key word signing that is used is known as Lámh. Lámh is based on Irish Sign Language and is similar to the Makaton vocabulary (Grove & Walker, 1990) used in the UK and other countries. Lámh is employed by children and adults with intellectual disability as an alternative means of communication (Glacken et al., 2019). The manual signs used in Lámh represent the function of the object/action. Key word signing has been shown to boost early language development in children through signs combined with speech (Launonen, 1996; Miller, 1992). In 2016, Öçalişkan, Adamson, Dimitrova, Bailey and Schmuck compared the use of gestures and baby signs (described as an iconic gestures deliberately taught to the child) of 23 children with DS (age 20-40 months) to 23 TD children (age 18 months). Following the analysis, Öçalişkan and colleagues found that although gesture use by TD children predicted spoken vocabulary one year later, baby sign was more sensitive to predicting the spoken vocabulary of children with DS one year later (Öçalişkan et al., 2016). The authors postulated that this may be due to the fact that deictic gestures are produced in response to an object/activity within a child's immediate environment and deictic

gestures can refer to many different objects. However, as baby signs are used when referring to a specific object and are repeated every time the object is used in interactions, the repetition of the signs may provide an increased opportunity for scaffolding a symbol to an object (Öçalişkan, 2016). Such repetition is important for children with DS given the short-term verbal memory deficits noted in children with DS, which is discussed later in the chapter. It is thought that this symbol to object mapping will eventually encourage the development of spoken word to object mapping as the child's speech develops (Öçalişkan, 2016). For this reason, Lámh is often incorporated into early interventions for children with DS as a means of developing their expressive language (Launonen, 1996; Powell, 1996). Children with DS use key word signs until they can express their intentions verbally (Miller, 1992). Children with DS often plateau at 2-3 word sign combinations and speech tends to emerge around this time (Layton, 2004).

1.3 Additional factors that can affect Language Development

1.3.1 Hearing loss.

Hearing loss in early infancy can have detrimental effects for all children's speech and language development as it restricts a child's input and exposure to a given language (Tomblin, Oleson, Ambrose, Walker & Moeller, 2014). Hearing loss is highly prevalent in infants with DS as a result of the anatomical differences associated with the syndrome, with two-thirds of infants experiencing conductive and/or sensorineural hearing loss in early years (Roizen, 2002). Otitis media, which is an inflammatory disease of the middle ear, is one of the main causes of conductive hearing loss and is common within this population due to narrow auditory canals, anatomical facial differences and immune deficiencies (Roizen, 2002; Mitchell, Call & Kelly, 2003). If otitis media is accompanied with fluid in the middle ear, known as otitis media with effusion (OME), this can lead to fluctuating hearing loss which may contribute to the language difficulties experienced by this population (Mitchell et al., 2003; Roberts et al., 2007). A study by Laws and Hall (2014) adopted the use of retrospective audiological data to explore hearing loss and its effect on the language skills of 41 children between the age 24-48 months. They found that children who experienced recurrent hearing loss throughout this time scored lower on all language measures when compared to children with DS who did not

experience hearing loss. This shows that hearing loss can have a direct impact on the speech and language development of children with DS. Hearing is also vital for the development of social, cognitive and behavioural abilities of all children and children with Down syndrome are at risk in these areas if hearing loss is present (Nightengale, Yoon, Wolter-Warmerdam, Daniels & Hickey 2017; Tedeschi et al., 2015).

1.3.2 Memory.

People with DS often present with difficulties with verbal short-term memory while visual short-term memory is often preserved (Hick, Botting & Conti-Ramsden, 2005; Jarrold, Baddeley & Phillips, 2007). Difficulties with verbal memory are often evident and are suspected to be as a result of reduced brain volume and hippocampal dysfunction (Carlesimo, Marotta & Vicari, 1997). For people with DS, such deficits in verbal short-term memory (VSTM) are independent of the hearing difficulties noted by this population (Brock & Jarrold, 2005; Faught, Conners, Barber & Price, 2016; Jarrold, Baddeley & Phillips, 2002). With regards to the working memory model described by Baddeley and Hitch (1974), it has been reported that deficits in the phonological loop may be a cause for the poor VSTM evident in people with DS (Jarrold et al., 2007). Although VSTM is an area of deficit for the group, Jarrold and Baddeley (1997) postulated that the short-term visuospatial abilities of children with DS were on par with children of a matched vocabulary mental age. This finding was based on a VSTM task (digit span) and a visuospatial task (Corsi span). The findings suggest that visuospatial skills of individuals with Down syndrome is an area of relative strength (Jarrold & Baddeley, 1997) which has been corroborated by numerous authors (Brock & Jarrold, 2005; Jarrold et al., 2007). Considering this, it is important for early interventions to use visual modalities where possible (Iacob & Musuroi, 2013).

1.3.3 Associated medical conditions in early infancy.

In the early years of life, infants with DS may present with other medical conditions after their birth with some resulting in lengthy hospitalisations. Congenital heart malformations, thyroid dysfunction, ophthalmic and Ear Nose and Throat (ENT) issues, hypotonia and gastrointestinal abnormalities are prominent in

the early years of life for these individuals (Kyle, 2012; Roberts et al., 2007). These infants also have a higher risk of developing leukemia (McGuire & Miller, 2017) and seizure disorders, in particular infantile spasms, in comparison to the general population (Leshin, 2003). Congenital heart defects are one of the main reasons for infant hospitalisations with heart malformations, in particular atrioventricular septal defect (AVSD), being evident in 40-60% of the population at birth (Cullum & Liebman, 1969). This high percentage often means that some of these children spend their early life in hospital with recurrent admissions which can affect a child's social, cognitive and communication development along with the quality and quantity of parent-child interactions in the early years of life (Minde, 2002).

1.4 Summary

To summarise, children with Down syndrome present with a range of communication difficulties with huge variability noted across this population. As previously stated, the language abilities of children with DS are not in line with their cognitive abilities and expressive spoken language is notably more impaired than their receptive abilities. Given that areas of deficits have been established, researchers recommend that children with DS should receive early intervention in the hope of reducing the impact that such communication difficulties can have on the person's quality of life (Clunies-Ross, 1979; Faught et al., 2016; Roberts et al., 2007). As previously described, young children present with a range of strengths and weaknesses and it is important for interventions to target the deficits while capitalising on the established strengths of these children. Given the poor short-term verbal memory noted in children with DS, interventions should target their strong visuo-spatial skills by incorporating gestures, key word signing and visuals where possible into potential interventions. Interventions should focus on developing and improving pre-linguistic skills such as joint attention, gesture use and babbling given the link such abilities have on receptive and expressive vocabulary development. Although it is important to understand potential external factors that can impede on the language development of children with DS, such as hearing loss, memory and lengthy hospitalisations, one additional factor that has an immense impact on the language development of children is the quality of parent-child interactions. Parent-

child interactions and different parent-child interaction therapies will be reviewed in the proceeding chapter.

Chapter Two: Parent-Child Interaction Therapy

2.1 Introduction

Early intervention for children with disabilities has been a topic of interest in research since early 1970s (Roberts & Kaiser, 2012; Mahoney et al., 1999). As stated in the preceding chapter, people with Down syndrome exhibit difficulties in terms of speech, language and communication with such deficits being evident from the early years of life. As a result, early intervention is pivotal for this population (Faught et al., 2016) in order to target such deficits and maximise the potential of each child. Researchers have found that brain plasticity is maximal in the early years of life and interventions that target these weaknesses, while optimising the strengths of infants with DS, will promote neural pathway development and strengthen connections within the brain (Regis, Lima, Almeida, Alves & Delgado, 2018). With this in mind, Næss et al. (2011) state that early interventions should focus on the strengths of individual's with DS and be specifically tailored to each individual's needs. Similarly, work by Iacob and Musuroi (2013) have suggested integrating the strong visual spatial memory that is evident in individuals with DS into all therapeutic interventions. These authors also suggest adapting therapy to incorporate the strong social skills evident in people with DS and using each individual's specific interest to drive therapy and motivation (Iacob & Musuroi, 2013). These strengths also include adopting the use of gestures and signing to promote language development in the early stages of life (Wright et al., 2013).

Although the difficulties with speech and language are one of the main characteristics in the profile of Down syndrome, research has also shown that parents of children with Down syndrome adopt a different interaction style in comparison to parents of TD peers (Slonims & McConachie, 2006). As parents play such a pivotal role in the speech and language development of their children, early interventions often incorporate parental goals that aim to upskill parents in communication strategies. This chapter will first explore the different parenting interactions of parents of children with Down syndrome. This will then be followed by a review of current parent-child interaction therapies (PCIT) and the aims of the current study are discussed.

2.2 Parent-Child Interaction in Down Syndrome

Parents are described as a child's first language teacher and hold a significant role in their child's communication development (Kaiser & Roberts, 2013). Parents of children with developmental difficulties often demonstrate directive parenting styles (where parents attempt to control a child's behaviour or activity), which can impede on a child's communication development (Cable & Domsch, 2015; Roberts & Kaiser, 2011). Parenting styles in parents of children with DS has received much attention within the literature throughout the years. Early work by Berger and Cunningham (1983) suggests that parenting styles and parent interaction in DS is different to parents of TD children. The authors have noted poor conversational turn-taking between parents and their children with DS, with 'vocal clashes' evident during parent-child interactions (Berger & Cunningham, 1983; Lynch & Eilers, 1991). Parents of children with DS are often described as being more directive in nature (Roach, Barratt, Miller & Leavitt, 1998; Tannock, 1988), which can further impede on the child's language development (Girolametto, Verbey, & Tannock, 1994). During directive and instructive interactions, parents often reduce the complexity of a task and as a result children are not given the opportunity to explore and develop pivotal cognitive skills and processes (Bibok, Carpendale & Müller, 2009).

A recent study by Schworer, Fidler, Lunkenheimer and Daunhauer (2019) reported that parents are more likely to adopt a more directive parenting style after the child has partially completed an action or task as the parents follow up with the next step of the given task. In addition to this, parents of children with DS have been reported to use increased negative behaviours, which Blacher, Baker and Kaladjian (2013) describe as overall negative affect, hostile verbal communication (tone of voice) and negative nonverbal behaviour (disapproving facial expression) during structured and unstructured tasks, when compared to parents of children with other developmental disorders and TD children. Although directiveness is typically discussed in a negative light when parenting a child with an intellectual disability, research by Hauser-Cram, Howell-Moneta and Young (2012) suggest that maternal directiveness that is non-intrusive and expands on a child's object of interest may be a supportive strategy in promoting child language development. However, as the differences between positive and negative directiveness are subtle, parents often

require training and coaching in such interaction strategies in order for them to be appropriately applied to their interactions with their child (Blacher et al., 2013).

With regards to language use, parents of children with DS have demonstrated to use syntactically complex language which may be too advanced for their child's cognitive level. A study by Johnson-Glenberg and Chapman (2004) found that parents of children with DS use higher mean lengths of utterances (MLUs) with their children when compared to parents of TD children (whose children were younger than the DS group) but were matched for MLU. The authors also report that parents of children with DS used a greater number of different words than that of TD parents and suggested that the children may not have sufficient time to consolidate the meaning of one word before others are introduced when vocabulary is too varied (Johnson-Glenberg & Chapman, 2004).

On the other hand, some authors report contradictory findings and state that parents of children with DS demonstrate similar parenting styles and interactions to that of parents of TD children. A recent study by Seager et al. (2018) aimed to compare a group of twenty-five parents of children with DS (aged 17-23 months) to thirty parents of TD children (aged 9-11 months) whose children were matched in terms of non-verbal mental age. Seager and colleagues found no significant differences in terms of parental sensitivity, which describes a parent's ability to respond to their child's initiations, between the two groups (Seager et al., 2018). These conflicting findings may be as a result of the parenting styles of children with DS evolving throughout the years as more information may be available to parents in recent times (Seager et al., 2018; Sterling & Warren, 2014). Another potential reason for this discrepancy may be due to the fact that, similarly to parents of TD children, a wide range of parenting interactions and styles can be observed for parents of children with DS. In additional to this, a study by Sterling and Warren (2014) compared children with DS (age 23-63 months) to TD children and they found that parents of older children with DS can adapt their parenting style to meet their child's needs. The authors report the parents of older children with DS adopted a facilitated learning style (that is, adjusted their language and interaction in response to their child) when compared to parents of younger children with DS and parents of TD children. A potential reason for the difference between the younger DS group and the older DS group may be related to the fact that the older children with DS demonstrated increased communicative acts and behaviours thus meaning the

parents could follow their child's initiations and facilitate communication in this manner (Sterling & Warren, 2014). This suggests that parents can adapt their parenting style to meet their child's needs.

Regardless of the discrepancies within the literature, these studies clearly highlight variability in parents' comprehension of their child's abilities and suggests that parent training and coaching should be components of early interventions (Phillips, Conners & Curtner-Smith, 2017). Given the pivotal role parents play their child's language development (Kaiser and Roberts, 2013), and the potential for interactional difficulties between children with DS and their parents, it is important for interventions to not only address the areas of deficits for children with DS, but to also upskill and train parents in interaction and communication strategies.

2.3 Parent-Child Interaction Therapy

Parent-child interaction therapy (PCIT), also referred to as parent-mediated intervention, describes an intervention that involves upskilling and training parents in communication strategies that will ultimately promote their child's speech, language and communication development (O'Toole, Lee, Gibbon, van Bysterveldt & Harte, 2018). PCIT is a triadic intervention as it involves three different parties; the clinician, the parent and the child (Roberts & Kaiser, 2012). PCIT follows a cascading model as the clinician must effectively train the parent in the use of the strategies. The parent must then implement these strategies with their child with sufficient accuracy and consistency in order to improve their child's speech, language and communication development (Roberts, Kaiser, Wolfe, Bryant & Spidalieri, 2014). This form of parent education and training should be a collaborative process between clinicians and parents in order to maximise child outcomes (Mahoney et al., 1999). The aim of PCIT is to teach parents how to recognise and respond appropriately to their child's verbal and non-verbal behaviours in the hope of stimulating an increase in these behaviours in the child (Warren et al., 2008). The past decade has seen a rapid development in different parent-child interaction therapies which have been researched in a variety of clinical groups. A recent study by Roberts, Curtis, Sone and Hampton (2019) conducted a systematic review and a meta-analysis of 76 PCIT studies of children with developmental disorders under the age of six years. Findings from the metanalysis

showed a positive correlation between parent training and the communicative outcomes of the children (Roberts et al., 2019). Roberts and colleagues recommend that parent training should be a pivotal goal of early interventions for children with developmental disorders (Roberts et al., 2019).

Higher level research such as Cochrane reviews, have explored PCIT in clinical groups such as Autism Spectrum Disorder (ASD) (Oono, Honey & McConachie, 2013), non-progressive motor speech disorders (Pennington, Akor, Laws & Goldbart, 2018), primary language delay and/or disorder (Law, Garrett & Nye, 2003) and children with Down syndrome (O'Toole et al., 2018). Firstly, the study by Oono et al. (2013) conducted a systematic review of randomised controlled trials (RCTs) of PCITs with children with ASD and their parents. Seventeen studies were included in the review, 10 of which were compared using a metanalysis. Results from the review showed that PCITs in ASD were effective in promoting change in the parents while also promoting the receptive vocabulary of the child as reported by their parents (Oono et al., 2013). However, no change was noted in the overall language development of the child nor in the levels of stress reported by the parents (Oono et al., 2013). Results from the study showed that children with intellectual disability and ASD require more intensive therapy (Oono et al., 2013).

Pennington et al. (2018) conducted a systematic review of studies of children up to five years of age with non-progressive motor speech disorders. The authors reviewed two studies, one implemented the use of The Hanen Parent Program – *It Takes Two to Talk* ® (Manolson, 1992) and the other used a relationship focused intervention. No change was noted in the language outcomes of the children or the attention and initiation of the children (Pennington et al., 2018). In terms of the adult outcomes, it was noted that parents increased their responsiveness, however, no change was noted in parental directiveness (Pennington et al., 2018). Nonetheless, following the analysis, Pennington et al. (2018) concluded that both studies were of low quality with risk of bias in relation to the outcomes in both studies. Similarly, Law et al. (2003) conducted a meta-analysis of 25 studies of children with primary language delay and/or disorder, three of which included studies of PCITs. Results showed no change in child language based on standardised tests regardless of whether the intervention was implemented by the therapist or the parent (Law et al., 2003). However, Law and colleagues reported a high degree of variability within the

25 studies which may have attributed to the lack of difference noted between parent and therapist led interventions (Law et al., 2003).

2.4 Parent-Child Interaction Therapies in Down Syndrome

With regards to children with Down syndrome, PCIT aims to specifically target the interaction and communication difficulties evident in this population while also targeting the parents' interaction with their children. Numerous PCITs have been reported within the literature and although they are referred to using different terms, the primary goals of such interventions are similar in nature. Current systematic reviews that have explored PCIT with children with Down syndrome include studies by Roberts and Kaiser (2011), Te Kaat-van den Os, Jongmans, Volman and Lauteslager (2017) and O'Toole et al. (2018). Firstly, in 2011 Roberts and Kaiser conducted a meta-analysis of PCITs that were implemented with parents of children who had language impairments. Eighteen studies, that included a range of study designs, were included in the review and the children were aged between 18-60 months with some, but not all, having a diagnosis of Down syndrome. The general results of the study found that PCIT increased the receptive and expressive vocabulary of the participating children along with an increase in expressive morphosyntax also being noted (Roberts & Kaiser, 2011). The effect size of child language measures ranged from between -0.15 to 0.82 (dependent on comparison group and outcomes measures) thus showing PCIT had a mostly positive impact on children who present with and without ID (Roberts & Kaiser, 2011). Although this improvement was noted in general for all children in the study, the children who had intellectual disabilities did not improve as much as the children who presented with language impairment alone. As the results suggest that this PCIT can promote the language development of children with language impairments, and to some extent, improve the language abilities of children with ID, specific analysis of children with DS was not conducted. It was also noted that parental responsiveness increased following the intervention (Roberts & Kaiser, 2011).

Secondly, Te Kaat-van den Os et al. (2017) conducted a systematic review of studies of children aged between 12-60 months with developmental delays (n = 228). The authors only included randomised controlled trials (RCTs) with a total of seven studies being included in the review. Although the review was not specific to

children with DS, 93 children with DS were included in the analysis. The studies implemented different PCITs such as the Hanen Parent Program, Enhanced Milieu Teaching (EMT) and Responsivity Education/ Prelinguistic Milieu Teaching (RE/PMT). Five of the seven studies reported an improvement in the parental responsivity, intentional communicative acts of the child (effect size of 0.68), child verbal turns (effect size of 1.2) and vocabulary diversity (effect size of 1.3) (Te Kaat-van den Os et al., 2017). No improvement in the expressive vocabulary of the children was noted in any of the interventions (Te Kaat-van den Os et al., 2017).

With respect to children with Down syndrome, O'Toole et al. (2018) conducted a Cochrane review where the authors systematically explored the impact of PCIT in children with DS alone. Studies were included in the review based on the following inclusion criteria; studies must be RCTs or quasi-RCTs (meaning the method of allocation is known and participants are randomly chosen within this context); children must have a diagnosis of DS and be aged between birth and 6 years of age; participants must be monolingual but could speak any language; and finally, interventions had to consist of parent-mediated interventions that specifically targeted the language and communication development of the children. Three studies fulfilled the inclusion criteria and they were the studies by Girolametto, Weitzman and Clements-Baartman (1998), Kaiser and Roberts (2013) and Karaaslan and Mahoney (2013). The methodology, results and implications of these studies will be discussed in greater detail later in the chapter. The children in this review ranged from 29 months to 6 years of age and all studies were either RCTs or quasi-RCTs. All studies implemented a different intervention with different intensities and measured outcomes using a variety of different measures, therefore a meta-analysis could not be conducted. The authors concluded that although PCIT has been shown to improve the way parents interact with their children, the authors rated all studies in the review as low quality (graded as very low certainty with regards to effect estimates) and there is currently insufficient evidence showing the effect of PCIT has on the receptive and expressive language skills of children with DS (O'Toole et al., 2018).

Current PCITs that have been implemented with children with DS include; Responsivity Education/Prelinguistic Milieu Teaching (RE/PMT), Enhanced Milieu Teaching (EMT), Responsive Teaching, the Hanen Program for Parents and other adapted therapies. The general goals of these interventions aim to increase parental

responsiveness and decrease parental directiveness during parent-child interactions. These goals are addressed by training parents to recognise their child's nonverbal communicative acts, encouraging parents to incorporate time delays into play while also showing parents how they can follow their child's lead and expand on play which in turn will provide a language rich environment for the child. Although these interventions share common overarching goals, they differ in many other aspects such as intervention frequency, intervention duration and delivery of intervention (group versus individual sessions). This next section aims to review the above mentioned PCITs while also highlighting the strengths and weaknesses of each intervention. A summary table of the main outcomes of all studies can be found in Appendix A.

2.4.1 The Hanen Program for Parents.

The first study included in the review by O'Toole et al. (2018) was by Girolametto et al. (1998) who used aspects of The Hanen Parent Program – *It Takes Two to Talk* ® (Manolson, 1992). The study by Girolametto et al. (1998) is one of the most renowned studies of PCITs in children with DS and the strategies are commonly used in clinical practice today. This approach is a family centered method of developing child language. The aims of the programme are to promote responsive interaction strategies such as responding to child orientated behaviours, implementing interaction-promoting strategies and to teach the parent language modeling strategies (Manolson, 1992). The programme consists of a pre-intervention assessment and video recording of the parent-child play interaction, followed by 16 hours of parent group training. This is then followed by three individual feedback sessions that involve video recording the parents using the taught strategies followed by a discussion. The approach of the intervention is naturalistic in nature as it aims to equip parents with strategies that can be implemented throughout their daily routines.

Girolametto et al. (1998) adapted this programme in their study so that it followed a focused stimulation model which allowed parents to choose 10 target words from a group of 20 that they felt would motivate their child the most. This study consisted of 12 children who were randomly allocated to the intervention and control group. The intervention group consisted of six children aged between 29 and

44 months of age. The control group consisted of six children aged between 32 and 41 months of age who continued to received speech and language therapy (SLT) from their current service provider. The intervention consisted of nine 2.5 hour group sessions followed by four individual sessions, however, the overall duration of the intervention was not reported. Results from the study found that although children did not use more spoken words following the intervention, parents reported that the child did use more of the targeted words in free-play interactions (Girolametto et al.,1998). It was also noted that the parents of these children used more focused stimulation techniques and target words when compared to the control group (Girolametto et al.,1998). However, these results should be interpreted with caution as the researchers were not blind to the intervention groups therefore increasing the risk of outcome bias. Fidelity was also only measured in terms of parental attendance and frequency/duration of intervention. Intervention adherence was not measured therefore the quality and consistency in terms of the delivery of the intervention is unknown.

2.4.2 Enhanced Milieu Teaching.

The second study included in the review by O'Toole et al. (2018) was a study by Kaiser and Roberts (2013) who implemented a PCIT known as Enhanced Milieu Teaching (EMT). EMT adopts a behavioural focus and is described by Kaiser (1993) as a naturalistic model of early intervention that involves training a parent in strategies that will promote their child's communication development using the child's interests and initiations as opportunities for reciprocal interaction. The strategies include responsive interaction strategies (e.g. language modeling and expanding, response contingency) and behavioural strategies (e.g. targets that match child's ability, responding to the child's initiations and environmental stimuli) (Roberts & Kaiser, 2012).

In 2013, Kaiser and Roberts compared the effect of EMT that was administered by therapists alone to a group who received EMT by therapists and parents. The language skills of 77 preschool aged children with intellectual disabilities were then measured. Of the participants, eight children with DS received therapist-led EMT intervention and 10 participants with DS received therapist and parent-led intervention. O'Toole et al. (2018) conducted a secondary analysis on the

DS group alone (who were between 30 and 54 months of age). They found no receptive and expressive language differences between the groups based on norm-referenced standardised tests and parental report at all time points (immediately post-intervention, 6 months post-intervention and at the 12 month follow up). This may be due to the fact that both groups were receiving intensive therapy and that the change that would be noticed between the two groups may not have been identified using formal testing. There was, however, an increase in the trained child language targets in the DS therapist and parent-led intervention group immediately after intervention, at six-month follow up but not at the 12-month follow up assessment (O'Toole et al., 2018). No difference was observed between the groups on untrained child language targets.

Although little to no long-term changes were evident in the child outcomes, the parents in the intervention group demonstrated a significant increase in the number of EMT strategies both noted immediately, 6 months post-intervention and 12 months post-intervention on trained and untrained activities (O'Toole et al., 2018). Increases in language modelling strategies were also noted in trained and untrained activities 6 months post-intervention, but not noted at the 12 month post-intervention follow up (O'Toole et al., 2018). This shows that parents can be taught long-term strategies that can promote their child's communication and language development using the EMT intervention but that a refresher session/course may be necessary as the parents discontinued the use of these strategies over time. Although Kaiser and Roberts (2013) did not compare outcomes within the different groups, it was found that in general children who have intellectual disability (ID) benefit from parent and therapist-led intervention. Increases in the number of different words used, lengths of utterances produced and the child's use of therapy targets were also evident in their outcomes (Kaiser & Roberts, 2013). Although the researchers who completed the norm-referenced standardised test were not blind to the experimental condition, the coders of the observational measures were blind thus increasing the validity of the results.

In addition to this, Enhanced Milieu Teaching (EMT) was recently explored by Wright and Kaiser (2017). The authors conducted a study that consisted of four infants with DS aged between 28-33 months and their three mothers and one father (Wright & Kaiser, 2017). One of the main aims of the study was to explore the effect of EMT with words and signs using a Teach-Model-Coach-Review protocol

(TMCR). The TMCR had previously been used by Kaiser and Roberts (2013) and describes different adult learning strategies that show positive results for training parents. The first of the four elements is the *Teach* strategy. This involved the therapists introducing the strategies and explaining the rationale behind the strategies to parents. The *Teach* element is first introduced at an initial workshop and then reintroduced at the start of each therapy session. The second element is the *Model* strategy which involves the therapist working directly with the child while the parent observes. This allows the parent to see how the strategies are implemented (Kaiser & Roberts, 2013). The third element is the *Coach* strategy where the parent takes the lead and tries to incorporate the strategies into their interactions with their child. The therapist observes and provides positive verbal coaching. The final element is *Review* where the parent discusses the strategies with the therapist in order to consolidate skills, receive feedback and answer questions (Kaiser & Roberts, 2013).

Wright and Kaiser (2017) incorporated the TMCR protocol in their single-case multiple-baseline study of four infants and their parents. Parent and child change were captured by recording a 10-minute video for each dyad, which were later transcribed and coded using the Systematic Analysis of Language Transcripts (SALT) software (Miller & Iglesias, 2008). Parents use of the EMT strategies were analysed and child utterances were coded for spontaneous, imitated and prompted signs and words by coders who were blind to time. Treatment fidelity was also measured with 25% of all intervention sessions coded and the authors report that fidelity was above criterion levels for therapist implementation of the EMT strategies and above criterion level for implementation of the TMCR protocol (Wright & Kaiser, 2017). As previous studies have failed to include a comprehensive description of parent training and coaching in DS, the results of the study by Wright and Kaiser (2017) have addressed this paucity of research and have found the TMCR protocol to be effective in training parents in the use of EMT strategies which have been implemented with a high degree of fidelity (Wright & Kaiser, 2017). Thus, suggesting that the TMCR protocol may be an appropriate protocol to be used when training parents of children with DS.

As it was previously noted by Caselli et al. (1998) the prelinguistic stages for infants with DS lasts longer than the typical 12-18 months period. These infants rely on gestures and show a preference for gesture use through early childhood. With this in mind, current PCITs have included gestures and sign language into therapy

programmes as a means of making use of the child's strengths to support communication. Wright, Kaiser, Reikowsky and Roberts (2013) conducted a study that aimed to explore how training the parents using naturalistic sign intervention can promote the expressive language skills of four infants with DS. The infants were aged between 23-29 months at the initial baseline assessment. The authors adopted a single-subject multiple-baseline design with each participant acting as their own control. This intervention included EMT combined with an intervention known as JASPER (joint attention, symbolic play and emotional regulation) that aimed to develop these skills of the child. Prior to commencing the intervention, three to five baselines were collected in order to obtain a visual trend of the children's abilities. Once a trend was noted, the children commenced the intervention. The intervention consisted of 20 sessions (lasting 20-30 minutes) that were delivered twice weekly. Observational measures were used to capture parent and child change by recording a parent-child interactive play activity in the homes of the participants. Each child's use of sign and words (spontaneous, imitated and prompted) were coded and the parent's total number of signs were also recorded. Measures for joint engagement were also captured using an adapted coding system by Bakeman and Adamson (1984) in which four types of joint attention are recorded. Interobserver agreement was conducted on 33% of the baseline, intervention and home observations, all of which were randomly selected prior to the commencement of the study. Wright et al. (2013) reported interobserver agreement for child language measures as 91% agreement, parent language measures during generalisation sessions as 94% agreement, and overall total agreement for joint attention was reported to be 87%. Twenty-five percent of the baseline and intervention sessions were coded for treatment fidelity of both the EMT strategies and JASPER strategies. Sessions that were coded for fidelity were randomly selected prior to the study and interventionists were blind to which sessions were coded for fidelity. Wright et al. (2013) reported overall agreement for the EMT strategies as 98%, with 96% agreement reported for the JASPER strategies.

The study reported that the children acquired between 10-21 new signs and three to nine new spoken words during the intervention (Wright et al., 2013). They also generalised their use of signs to a new environment and used these signs with an untrained communication partner. A modest increase in joint attention and symbol-infused joint engagement (i.e. words with referent-specific gestures) was noted,

however, data for the above were only collected for half the sessions, therefore a true analysis of these variables requires further exploration. To summarise, the results in this study support the use of the EMT/JASPER Words and Signs intervention as a means of supporting the development of young children with DS' expressive language skills. The use of a single-case multiple-baseline design allowed individual change and variability to be captured which is important given the heterogenous nature of children with DS (Karmiloff-Smith et al., 2016). Although the results are promising, long-term effects of this intervention are unknown as no post-intervention/follow up data was collected. Similarly, the true effects this intervention had on the joint engagement of the children are unknown as this was only collected for half of the sessions.

2.4.3 Responsive Teaching.

The third and final study included in the review by O'Toole et al. (2018) was a study by Karaaslan and Mahoney (2013). Responsivity Teaching (RT) involves training parents in strategies that will promote their child's communication development by adapting their responses to match their child's level of functioning while also decreasing the directive parenting style (Mahoney, 1988; Mahoney, Perales, Wiggers & Herman, 2006). The intervention orientates around a child's pivotal behaviours that are necessary for communication development (Karaaslan, Diken & Mahoney, 2011; Mahoney & MacDonald, 2007). These behaviours are described by the authors as social play, joint attention, intentionality, initiation of exploration, cooperation and self-regulation (Mahoney et al., 2006; Karaaslan et al., 2011). Mahoney and Perales (2003) and Mahoney and Perales (2005) explored the effects of RT in young children and their parents between 12-54 months who presented with developmental delays or ASD. The authors found that as parents increased their responsivity, development and social emotional gains were evident in the children (Mahoney & Perales, 2005). However, this study was not specific to children with DS as it included young children with a range of developmental delays. In 2011, Karaaslan and colleagues explored the feasibility of the intervention with two five-year-old children with cognitive impairment and their mothers. One had a diagnosis of ASD and the second child had a diagnosis of Down syndrome. The authors used the Pivotal Behaviour Rating Scale and the Maternal Behaviour

Rating Scale (MBRS) as they are sensitive for detecting change in parent-child interaction therapies (Mahoney & Perales, 2003, 2005). Based on the results from the Turkish Version of the MBRS, although both mothers had a high pre-intervention directiveness score (on the 5-point Likert scale), it decreased by the end of the intervention. Similarly, scores for responsiveness that were relatively low preintervention, increased by the end of the intervention (Karaaslan et al., 2011).

As the results of the study by Karaaslan et al. (2011) could not be generalised given the small sample size and nature of the study, this motivated Karaaslan and Mahoney (2013) to conduct a randomised control trial (RCT) in order to establish the effectiveness of RT intervention. The study consisted of 15 Turkish children with DS aged between 2-6 years and their mothers. The participants were randomly assigned to the intervention group (n=7) and control group (n=8). Both groups continued to receive their standard services throughout the 6-month study while the intervention group received the RT intervention weekly each session lasting 1.5 to 2 hours in duration. The Turkish version of the Maternal Behaviour Rating Scale (MBRS), the Pivotal Behaviour Rating Scale (child outcomes) and the Turkish version of the Denver Developmental Screening Test-II were used to capture change. Firstly, the increase in responsiveness (effect size 0.62) and decrease in directiveness following the intervention was in keeping with the results first noted by Karaaslan et al. (2011) according to the MBRS. Secondly, children in the intervention group increased their attention (effect size 0.63) by 57% and their initiation (effect size 0.68) by 54% when compared to the control group who increased by 11% and 7% for these two measures as noted by the Pivotal behaviour rating scale. In addition to this, the study showed a significant positive correlation ($r = 0.87$, $p = <0.001$) between the increases noted in child engagement (attention and initiation) and each mother's level of responsiveness. Secondary results of the study also indicate that the parents of children with DS, who have a higher mental age, tend to use a less directive parenting style. This may be because the parents may feel they do not have to compensate as much when compared to children with DS who may have a lower mental age and who are less likely to actively engage (Karaaslan & Mahoney, 2013). Although the authors found a correlation between parent responsivity and improvements in child engagement, they did not explore how responsiveness and child engagement (following the training) impacted the child's development. It was noted by O'Toole et al. (2018), that the study by Karaaslan and Mahoney (2013) had

the largest cumulative intensity out of the three studies included in the Cochrane review (Karaaslan and Mahoney's study consisted of approximately 48 hours of intervention; Girolametto et al. (1998) consisted of approximately 26.5 hours; Kaiser and Roberts (2013) consider of approximately 19 hours intervention) with RT also showing the largest effect on the language outcomes of the children. Although these outcomes were only measured immediately post-intervention, the results show some promise for high intensity interventions with this population (Karaaslan & Mahoney, 2013; O'Toole et al., 2018).

In 2015, Karaaslan and Mahoney conducted a mediational analysis (which is an analysis that aims to explore potential underlying causes of a known relationship between two variables) of the Karaaslan and Mahoney (2013) study, and another small RCT by Karaaslan, Diken and Mahoney (2013). Both studies explored the effect of RT in preschool children with a range of disabilities not specific to DS. The study found that RT was highly effective in increasing maternal responsiveness and style of interaction (effect size of 0.82, respectively) and these increases were corelated ($r = 0.84$, $p < .001$) to the positive changes noted in the child's pivotal behaviours which describes the child's overall engagement in terms of initiation and attention (effect size of 0.79). It was also interesting to note that the child's language ($r = 0.93$, $p < .001$) and social developmental ($r = 0.57$, $p < .01$) were affected by the changes in the child's pivotal behaviour and not affected by the mother's responsiveness (Karaaslan & Mahoney, 2015). Despite these promising results, these findings are specific to children with DS aged between 2-6 years and the impact of this intervention in a younger demographic is as yet unknown.

2.4.4 Responsivity Education/ Prelinguistic Milieu Teaching.

Responsivity education/ prelinguistic milieu teaching (RE/PMT) is based on the transactional model of social communication development which follows the belief that early social and communication development are facilitated through reciprocal interactions (McLean & Synder-McLean, 1978), which fosters the development of spontaneous communication (Warren et al., 2008). RE/PMT has similar principles and goals of the previously described EMT, but focuses on pre-linguistic communication. The RE aspect of the intervention involves clinicians coaching parents and encouraging appropriate responses to their child's non-verbal

communication. This is achieved by following their child's lead, building on social play routines, encouraging turn-taking and encouraging the child to communicate and request for objects/actions through time delays (Warren et al., 2008). The clinicians then work directly with the children using the PMT approach. This was based on research that found that PMT resulted in better child outcomes when their parents are more responsive to their communication attempts and intentions (McLean & Synder-McLean, 1978). It is thought that through this combined RE/PMT intervention, this will facilitate a transactional, bidirectional relationship between parent and child, with a particular focus on those that are non-verbal which will in turn promote their language development.

Fey et al. (2006) implemented RE/PMT with 51 children aged between 24-33 months who had developmental disabilities of known and unknown etiologies, 26 of which had DS. This randomised control trial expanded on a study that was previously conducted by Yoder and Warren (2002) who first established this intervention approach and found it to be effective with children who presented with low rates of canonical vocalisations pre-intervention when compared to a control group. For the group of individuals with DS who received intervention, there was an increase in the number of imperative, declarative and total communication acts following therapy (Fey et al., 2006). Although this increase was not statistically significant, the study does show some promise for the positive effects that RE/PMT may have on individuals with DS. One unanticipated finding in the study was that there was no difference between the control group and intervention group in relation to the way parents of children with DS responded. Fey et al. (2006) suggested that this may be due to these parents already demonstrating a relatively high level of responsivity to their children when compared to the parents of children of other etiologies. It is also suggestive that training alone (through RE) may not be sufficient in teaching parents such strategies and that parents require coaching and modelling with their child for interventions to be effective.

In light of the positive, yet modest results of the study by Fey et al. (2006), Warren et al. (2008) conducted a follow-up analysis of the 51 participants in order to determine the long-term effects of this intervention programme. The authors used the same measures as the previous study with the exception of the Communication and Symbolic Behavior Scales (CSBS) as they felt it was developmentally inappropriate given the age of the participants at the time of the follow up assessments. Data for

this study was collected at two additional follow-up points; 6 months (Time 3) and 12 months (Time 4) after the conclusion of the intervention by Fey et al. (2006). Both the intervention and the control group, received a low intensity booster of the RE/PMT intervention in order to ensure the children received a similar intervention intensity within Time 3 and Time 4 time-frame. However, following statistical analysis of the data, findings suggest no long-term benefits of the intervention. Warren et al. (2008) hypothesised that these results may be explained by a number of factors such as the discontinuing of the CSBS as a measure as well as the authors describing their other measures as being 'conservative' which may not have identified all the long-term results of the intervention. Again, findings from both studies are suggestive that it is not enough to train parents alone in RE and that coaching should be incorporated into the sessions in order for change to occur in the parents.

2.4.5 Milieu Communication Teaching.

Finally, in 2014, Yoder, Woynaroski, Fey and Warren carried out a re-analysis of data previously reported on by Fey, Yoder, Warren and Bredin-Oja, (2013) who measured the effect of different dose frequencies of Milieu Communication Teaching (MCT) on children with Down syndrome and other disabilities. MCT is similar to the RE/PMT intervention although they differ in terms of intervention frequency. Another difference includes the fact that all parents, independent of the group, received nine sessions of RE for the first 3 months of MCT. A total of 64 participants were randomly assigned to a low dose frequency group (LDF) (n=31, 16 of which has DS) and a high dose frequency group (HDF) (n=33, 19 of which had DS). The LDF received one hour of MCT a week for nine months compared to the high frequency group who received five hours of MCT a week for nine months. Yoder et al. (2014) found that once the intellectual ability of children with DS was controlled for, the HDF group responded better to the MCT intervention with a greater increase in their growth of vocabulary noted. Results also showed that the children who had functional play skills benefited more from the intervention than those who did not (Yoder et al., 2014). In addition to this, the authors suggest that the therapist and child in the HDF group would have a stronger relationship and that this may also have an impact on the child outcomes (Yoder et

al., 2014). Although, the results suggest positive outcomes following the intervention, the parents were not blind to the intervention which increases the risk of performance bias.

2.4.6 Adapted Parent-Child Interaction Therapies.

Meadan, Angell, Stoner and Daczewitz (2014) conducted a within-subject multiple-baseline study with children with DS and their parents to explore the feasibility of a social pragmatic intervention and the impact it had on a child's communication skills. In their study (n=5), the authors promoted the social communication skills of children with DS aged between 2-5 years by coaching and training their parents using naturalistic and visual teaching strategies as part of a larger three year Parent-Implemented Communication Strategies (PiCS) project. The parents in the study first received a one-to-one individualised training session in their home focusing on naturalistic training strategies which lasted for between 45-60 minutes in duration. The children were not present for this training. Following this, the parents were coached in each of the naturalistic strategies 2-3 times a week for an average of four months, the duration of these sessions were not specified. The authors used a variety of measures to capture change including standardised language assessment, in home observations, along with researcher-developed surveys to assess the child's social communication skills and the parents use of naturalistic teaching strategies.

Following the intervention, results showed that all parents increased their use of naturalistic teaching strategies (as noted by the in-home observations) following the coaching sessions when compared to the number of strategies used following the initial parent training sessions. Therefore highlighting that a 'one time' training session is insufficient and that clinicians should aim to coach parents until they are confident in the implementation of the strategies and that multiple training sessions are inefficient without parent coaching (Meadan et al., 2014). The authors also found that as parents reduced their use of modelling strategies, this showed that their children increased their number of responses according to the in-home observations. Similarly, it was noted that for the parents who implemented a time delay before responding, their children increased their frequencies of initiations post-intervention. A note of caution is warranted in terms of generalisability of the results due to the nature of measures used and the method of data collection. Nonetheless, the findings

from the current study suggest that parents require frequent coaching to learn and effectively implement new teaching strategies with their children and future research should aim to incorporate such coaching into PCITs (Meadan et al., 2014).

With regards to qualitative research, O'Loughlin, Carroll, and Caulfield (2010) explored the effectiveness of a PCIT known as 'Little Owls' from a parents perspective. This is one of the only studies that has explored PCIT within an Irish context. This programme consists of group sessions and is offered to parents of children with DS aged between 12 and 36 months of age. Although only a limited description of the programme is provided, the goals are similar to that outlined by previous PCITs and aims to enhances the communication skills of young children with DS. The programmes targets attention and listening skills, key-word signing, oral motor skills and sensory awareness (O'Loughlin et al., 2010). Group sessions were offered once every fortnight and lasted for one hour in duration. The number of group sessions offered was not specified. Following the group intervention, families received one home visit to facilitate generalisation and carry over of therapy targets.

The study consisted of five semi-structed interviews over three months with five parents who had completed the 'Little Owls' early intervention programme. A range of themes emerged with most parents reporting they enjoyed the group aspect of the intervention as it allowed them to meet other parents of children with DS, share experiences and improve coping strategies (O'Loughlin et al., 2010). In addition to this, huge variability was noted in terms of parental effectiveness and feasibility of the programme. Most parents reported an overall positive experience and found the intervention to be beneficial as it provided them with strategies and knowledge through meeting other parents and clinical professionals. Other parents found the group stressful and rationalised this feeling by stating it was difficult to attend appointments at times. This raises the issue of programme acceptability and the need for all early interventions to fit in within a family's daily routine/activities (O'Loughlin et al., 2010).

2.5 The PELD intervention

Another PCIT within the Irish context is offered by a service who provide speech and language therapy (SLT) for people with Down syndrome. This intervention, referred to as the PELD (Promotion of Early Language Development)

programme to uphold anonymity, is an early intervention programme for children with DS aged between 10-36 months and their parents. The PELD programme consists of both individual and group therapy sessions and strives to promote the pre-linguistic and early language skills of children with DS while also coaching their parents in communication and interaction strategies. The PELD programme is completed over a 10 month period and consists of three terms of the intervention. A total of 18 fortnightly sessions are offered during this period and parents are encouraged to attend all three terms of the intervention. This intervention shares many of the same principles and primary goals of the previously described PCITs which are to target the interaction and communication difficulties evident in this population while also targeting the parents' interaction with their children (Fey et al., 2006; Girolametto et al., 1998; Kaiser & Roberts, 2013; Karaaslan & Mahoney, 2013; Meadan et al., 2014; O'Loughlin et al., 2010; Wright & Kaiser, 2017; Wright et al., 2013; Yoder et al., 2014). Although current research has reported the benefits of PCITs in children with DS, all intervention programmes are designed to be used with a range of developmental disorders, not specific to DS. The PELD intervention programme differs from previous PCITs as it is specifically tailored to children with DS from 10-36 months of age and goals are based on the specific areas of strengths and weaknesses evident in children with DS during these early years of life. The PELD intervention has been guided by previous research and other PCITs and the programme has been designed based on the current empirical evidence base. The PELD intervention differs from other PCITs in the following ways: it is offered over a protracted period of time (10 months), parents are coached on an ongoing basis throughout the sessions (both group and individual) in order to facilitate parent change, the intervention is offered to children as young as 10 months of age and it is specifically tailored to children with DS. As the PELD programme has only been recently developed within the clinical setting, no evaluations of the PELD programme have been completed. A detailed description of the PELD intervention programme is outlined in Chapter Three, section 3.4.

2.6 Summary

To summarise, some parents of children with DS may adopt directive parenting styles which can impede their child's communication development, while

other parents have demonstrated an ability to adapt their parenting style to meet their child's needs. However, given the variety of parenting styles noted among parents of children with DS, it is important for PCITs to be incorporated into early communication interventions for these children so that parents can receive the relevant support, training and coaching during their infant's early years of life. Such training is important as it aims to equip and upskill parents with specific communication strategies with the aim of maxmising the communication outcomes of their child. There are many PCITs documented within the current literature relating to DS and all share similar fundamental goals that aim to increase parental responsiveness and use of parental language with the hope of promoting the language development of children with DS. See Appendix A for a table summary of studies. Previous research has explored the impact these parental strategies have on the expressive vocabulary of children and the use of sign (Wright et al., 2013). Other authors have strived to define the correlation between parental responsiveness and its impact on children's communicative intentions (Karaaslan & Mahoney, 2013). Although there is much research, there is huge variability among the studies with regards to age of participants with the study by Wright et al. (2013) reporting the youngest infants with DS aged 23-29 months at baseline. In addition to this, many studies have failed to provide a detailed report of how the parents were trained and it was not until Wright and Kaiser (2017) conducted their study and suggested that the TMCR protocol was effective for this cohort. In addition to this, most studies have reported positive parental change following a PCIT intervention but that little change is noted in the children. Some authors suggest that perhaps this lack of change may be due to a lack of sensitivity in the outcome measures used (O'Toole et al., 2018) while other others suggest that the intervention needs to be in place for a minimum of six months before change in the child is observed (Karaaslan & Mahoney, 2015; O'Toole et al., 2018). Similarly, only half of the studies measured treatment fidelity and many studies did not implement blinding therefore there is also a risk of outcome bias among the studies.

Current studies are of very low quality with respect to the level of evidence for these interventions and improvements in child outcomes are only evident in lower level research (O'Toole et al., 2018). Based on the review carried out by O'Toole and colleagues a number of recommendations have been made with respect to future interventions. Future research should endeavour to implement treatment

fidelity, include a detailed case history of the children, while also including sufficient information regarding the demographic of the parents and relevant information regarding the clinicians experience (O'Toole et al., 2018). Future therapies should be implemented for longer than 6 months in order to see a change in child outcomes, should ensure measures of blinding are addressed in order to reduce the risk of bias and studies should also provide a detailed description of the procedures used while using appropriate and reliable outcome measures where possible (O'Toole et al., 2018).

2.7 Aims of the study

This study aims to explore the effectiveness of the PELD early intervention programme that is offered to children aged between 10-36 months by an SLT service for people with Down syndrome. This study aims to address previous methodological issues and incorporates the recommendations from past literature throughout the evaluation.

This study aims to address the following research questions:

1. Does the PELD intervention programme encourage the development of language in infants with DS?
2. Does the PELD intervention programme affect the communicative interaction of infants with DS?
3. Does the PELD intervention programme change the way parents interact and communicate with their infants with DS?

Chapter Three: Methodology

3.1 Research Design

A single-subject multiple-baseline design was employed to evaluate the effectiveness of the PELD programme. This design was deemed the most appropriate as the intervention is in its developmental phase and the intervention needs to be refined before higher level research, such as randomized control trails, can be completed (Medical Research Council, 2000). The collection of baseline measures ensured a level of control for each participant and was an effective way of measuring change throughout the programme (Gast & Ledford, 2010).

3.2 Recruitment

Ethical clearance was granted by the Clinical Research Ethics Committee (CREC) prior to the commencement of the study (See Appendix B). The participants were recruited by the SLTs working in an SLT service for individuals with Down syndrome. This service offers the first strand of an early intervention programme to children between the ages of 10 and 18 months. This early intervention will be referred to using the PELD acronym (promoting early language development) as a means of maintaining anonymity. The families that registered for the 18-week programme were approached by the SLTs from this service and informed of the proposed study that aimed to evaluate the effectiveness of the PELD intervention. The parents of the children were given written information sheets regarding the study along with consent forms (See Appendix C and D). Child assent forms were not included due to the age of these children. The written information sheets outlined the following; purpose of the study, what the study would entail, why the participants had been asked to participate, what information would be collected, confidentiality, anonymisation and data storage, what would happen to the results and possible disadvantages of taking part. Parents were assured that they would receive the intervention whether or not they consented to taking part in the study and that all data collected would be anonymised. They were also informed that they could withdraw their consent at any time throughout the study and up to two weeks after the final intervention session.

3.3 Participants

All parents consented to the study which meant a total of seven child participants (four males, three females) and their parents were included. In order to partake in the study, each child participant had to meet the following inclusion criteria: 1) participants must have a diagnosis of Down Syndrome, 2) all participants must be between 10-18 months old, 3) it must be the parents first experience of the PELD programme and 4) English must be the child's first language. Demographic information was documented from each participant's file and additional information was gathered at the first home visit. Each child was allocated a pseudonym to uphold confidentiality. The occupations of the parents were categorised using the Standard Occupation Classification (SOC) (2010). The demographic information for each child and parent participant is described in detail below. A summary of the main characteristics of each parent participant is illustrated in Table 3.1 and a summary of each child participant is presented in Table 3.2. All children and parents in the study attended a different number of terms and sessions. Table 3.3 presents a summary of participant attendance.

3.3.1 Sophie.

Sophie was 13 months and 15 days old at the time of the initial assessment. She is the youngest in her family and has an older sibling that was three years old at the time. Both her parents attended third level education and are in the 40-50 years age range. The occupations of the parents were classified as Professional Occupations (Sophie's father) and Administrative and Secretarial Occupations (Sophie's mother) as per the Standard Occupation Classification (SOC) (2010). At the initial baseline assessment, Sophie's mother was working full-time and her father was on sabbatical leave. Both parents initially intended to take part in the PELD intervention so initial baseline measures were collected for both. However, before the intervention commenced Sophie's father returned to work resulting in Sophie's mother being the primary parent participant in the study. She also switched to part-time work (a four day week) during the intervention. The parents reported no additional caring responsibilities (e.g. an unwell grandparent/other children with additional needs).

Sophie was exposed to two languages at home; English and an additional European language. English is Sophie's first language (L1) and the European language is her second language (L2). Sophie's mother and sibling speak English in the home and Sophie's father predominantly converses using the L2 with some English. Activities at home, such as songs and books, are in both L2 and English depending on which parent is interacting with the child. All other family members and friends speak English. Sophie attends day care five days a week where English is also generally spoken.

Sophie was awaiting an eye examination at the time of the initial baseline assessment and there was also a query of moderate hearing loss following a test that was carried out one month prior to the baseline observations. She had no surgeries prior to the intervention, however, following her birth she had an overnight stay in the intensive care unit (ICU) as a result of Jaundice. She was also admitted to hospital for bronchitis at two months for a period of seven days and has since spent a further overnight stay in hospital for pneumonia. Sophie was diagnosed with an underactive thyroid and during the intervention was taking medication to counteract the effect. Sophie attended physiotherapy and aquatherapy once a month for a duration of one hour each. During the first term of the PELD intervention, Sophie's mother partially attended an additional intervention for children with communication disorders known as the *It Takes Two to Talk- The Hanen Program* which is described in the introduction chapters. It consists of 6-8 group sessions with three individual sessions that promote parent strategies for developing their child's communication and is run by SLTs. Sophie's mother attended two group sessions and one individual session of this course. During the second term of the PELD programme, Sophie's mother completed an additional Lámh course, known as Little Lámh, which is a two and a half hour introductory course to Lámh that teaches 26 signs. During the third term of the intervention, Sophie's mother attended Family Lámh (which consisted of two four hour sessions teaching a total of 150 signs) and Little Skippers (a sing and sign group that teaches songs with Lámh signs to children under two years). Sophie presented with moderate cognitive impairment at initial baseline according to the cognitive subtest of the Bayley Scales of Infant and Toddler Development III (Bayley, 2006).

3.3.2 Jack.

Jack was 11 months and 28 days old at the initial baseline assessment. Jack is an only child and lives with his two parents in a monolingual, English-speaking household. Both parents are within the 30-40 year age range and both had attended third level education (i.e. higher education institutions such as universities or colleges). The occupations of the parents were classified as Professional Occupations (Jack's father) and Associate Professional and Technical Occupations (Jack's mother). Jack's mother was the parent who participated in the PELD intervention as she was availing of carers leave at the time of the study. The only additional caring responsibility that the mother reported was that she was due to go for surgery shortly after the PELD intervention commenced and that her mobility may be affected during the first term of the PELD programme.

Jack was diagnosed with mild hearing loss and was awaiting an Ear Nose and Throat (ENT) appointment at the time of the baseline assessment. Jack was long-sightedness, therefore, glasses were to be worn throughout the intervention. Following his birth, he spent an extended stay in ICU for six days and at three months he underwent surgery to repair an atrioventricular septal defect (AVSD) where he spent ten days in hospital. A pacemaker was also inserted at this time. It was also noted that his thyroid stimulating hormone (TSH) levels were elevated however medication was not required to alter these levels. Jack attended physiotherapy once a month and was also seen by a clinical nurse specialist once a month. Regarding SLT involvement, Jack was seen once by an SLT for feeding, eating, drinking and swallowing (FEDS) concerns and his mother also attended a Family Lámh course prior to the commencement of PELD programme. During the intervention, Jack and his mother attended two SLT sessions offered by a private SLT service focusing on communication development. Jack presented with mild cognitive impairment at initial baseline according to the cognitive subtest of the Bayley Scales of Infant and Toddler Development III (Bayley, 2006).

3.3.3 Conor.

Conor was 13 months and 15 days old at the first baseline assessment. He lives in a monolingual household with his two parents and infant brother who was born during the second term of the PELD intervention. Both parents attended third

level education and their occupations are classified as Professional Occupations. Conor's mother also completed a PhD within her professional field. She was taking an extended maternity leave at the time of the initial baseline assessment. Conor's mother was the parent partaking in the study. His parents reported no additional caring responsibilities at the initial assessment.

Conor's sight was reported to be within normal limits and he was awaiting a hearing review after he presented with moderate hearing loss related to fluid in the ear. Following his birth, he spent two weeks in ICU as a result of Jaundice, breathing difficulties and heart complications. At four and a half months, Conor spent nine days in hospital after he underwent open heart surgery that repaired an atrioventricular septal defect. He was due to go for another heart surgery one year from the initial baseline assessment. Conor attended joint sessions of occupational therapy (OT) and physiotherapy once every fortnight for 6 weeks with a 6 week break in between therapy terms. He has received no SLT input prior to the PELD intervention, however, Conor's mother reported to use some Lámh signs with him that she learnt from a Lámh DVD. During the intervention, Conor and his mother attended 10 one hour sessions of a Sing and Sign group offered by a private service. Conor presented with mild cognitive impairment at initial baseline according to the cognitive subtest of the Bayley Scales of Infant and Toddler Development III.

3.3.4 Daniel.

Daniel was 11 months 2 days at the initial baseline assessment. He lives in a monolingual household with his two parents and his older sibling of two years of age. Both parents attended third level education and their occupations are classified as Sales and Customer Service Occupations. Daniel's mother works on a part-time basis and was the parent who participated in the PELD intervention. The parents reported no additional caring responsibilities.

Daniel's last audiology exam indicated satisfactory/mild hearing loss and was waiting a follow up examination. He also wore glasses for long-sightedness. Following his birth, Daniel spent six weeks in the neonatal unit as a result of Jaundice and a clot in his liver. He was tube fed for four weeks at this time. He was diagnosed with pulmonary sternosis and an atrioventricular septal defect which did not require surgery. With regards to SLT involvement, Daniel was seen by an SLT

while in hospital for FEDS concerns and his parents completed a Little Lámh course prior to the PELD intervention. During the intervention, the family also completed a Sing and Sign group, Little Skippers and Family Lámh. He also attended physiotherapy once a month. Daniel presented with moderate cognitive impairment at initial baseline according to the cognitive subtest of the Bayley Scales of Infant and Toddler Development III.

3.3.5 Ellie.

Ellie was 10 months and 25 days old at the initial baseline assessment. She is the youngest in her family and has two older siblings aged five and three years. Both parents attended third level education. The occupations of the parents were classified as Professional Occupations. Ellie's mother was on carers leave at the time of the initial baseline assessment and was the parent taking part in the study. The primary language spoken at home is English. However, as Ellie's siblings attend a Gaelscoil, her mother reported that she would have been exposed to some Irish phrases or counting in Irish but the family did not converse in the language at home. No additional caring responsibilities were reported.

Ellie's sight had yet to be assessed at the time of the initial baseline and her hearing ability was evaluated with the presence of fluid in the ear warranting a follow-up in six months' time. During the intervention, Ellie presented with query mild to moderate hearing loss relating to fluid in the ear. At six weeks of age she had surgery to repair an atrioventricular septal defect. She was in the intensive care unit pre-surgery for elevated temperatures and rhinovirus and was in ICU post-surgery for pneumonia. A nasogastric tube (NG) tube was inserted during this time and she received SLT while in hospital for FEDS difficulties. Ellie's mother reported that she had been in hospital sporadically and spent approximately half of her first eight months of life in hospital. During the intervention, Ellie spent an additional seven days in hospital as a result of a vomiting bug. In terms of other therapy services, Ellie attends physiotherapy fortnightly. During the intervention, Ellie's mother attended the Family Lámh course. Ellie presented with mild cognitive impairment at initial baseline according to the cognitive subtest of the Bayley Scales of Infant and Toddler Development III.

3.3.6 Grace.

Grace joined the intervention between intervention term one and term two. Due to late enrolment, only one baseline assessment was collected. She was 17 months and 5 days old at the initial baseline assessment. Grace's parents attended third level education and their occupations are classified as Professional Occupations. Grace's siblings were aged three and five at initial baseline and attend a Gaelscoil. Grace's mother was the parent participant who took part in the study and was on carers leave at the time of the initial baseline assessment. No additional caring responsibilities were reported.

Grace's first language is English. However, as her siblings attend a Gaelscoil, Grace's mother reports that she is exposed to Irish at times within the home environment. Grace's mother reported that herself and Grace's older siblings only speak in English to her and her father speaks both English and Irish to Grace. All activities such as sharing books and songs are through English. Although English was reported to be the predominantly used language in the household at initial baseline, it became apparent during the course of the intervention that the family had started to us more Irish with Grace. At the final data collection, Grace's mother estimated that she was exposed to English sixty percent of the time and Irish 40 percent of the time in the home environment.

Grace's hearing and sight were examined prior to the commencement of the study. She was diagnosed with astigmatism and will require glasses at two years of age to correct this. One month before the baseline assessment, Grace was diagnosed with mild hearing loss in both ears related to fluid the ear. Her mother reported that she has been referred to ENT and was hoping to have grommets inserted before two years of age and will then receive a hearing aid. Grace underwent surgery at five months old to repair an atrioventricular septal defect and spent one week in hospital post-surgery. In relation to SLT input prior to the PELD intervention, Grace had attended three SLT sessions which focused on communication development. Her mother had also attended two one hour sessions of Little Skippers and also reported that she was taught some Lámh signs by the clinical nurse specialist and other parents. During the intervention period, Grace and her mother attended one SLT session with their service provider which again focused on communication development. Grace had also received six joint physiotherapy and OT sessions prior to the intervention. Grace presented with moderate cognitive impairment at initial

baseline according to the cognitive subtest of the Bayley Scales of Infant and Toddler Development III.

3.3.7 Luke.

Luke also joined the programme between intervention term one and term two. Due to late enrolment in the programme, only one baseline assessment was collected. He was 12 months and 24 days old at the initial baseline assessment. Luke lives in a monolingual household with his parents and two older siblings aged six and four years. Both parents attended third level education and were in the 30-40 year age range. His parent's occupations are classified as Professional Occupations (Luke's father) and Associate Professional and Technical Occupations (Luke's mother). Luke and his siblings were looked after by a childminder in their own home on the days the mother worked. Luke's mother was the parent who took part in the study. No additional caring responsibilities were reported.

Luke's hearing and sight were tested prior to the intervention and were found to be within normal limits. At 11 months, he spent five days in hospital for bronchitis, which was the only time spent in hospital to date. In terms of previous SLT involvement, Luke's mother attended Little Skippers sing and sign group for four one hour sessions and completed a two hour 'Introduction to Lámh' course. During the intervention, Luke's mother attended a Family Lámh course. He also attends OT and physiotherapy sessions for one hour once a month. Luke presented with moderate cognitive impairment at initial baseline according to the cognitive subtest of the Bayley Scales of Infant and Toddler Development III.

Table 3.1

Summary of demographic information of each parent participant completing the PELD programme

	Sophie's Mo	Jack's Mo	Conor's Mo	Daniel's Mo	Ellie's Mo	Grace's Mo	Luke's Mo
Age range (yrs)	40-50	30-40	30-40	30-40	30-40	30-40	30-40
Gender	F	F	F	F	F	F	F
Ethnicity	white Irish	white Irish	white Irish	white Irish	white Irish	white Irish	white Irish
Relationship to child	Mo	Mo	Mo	Mo	Mo	Mo	Mo
Total no. of children	2	1	2	2	3	3	3
Ages of other children	3yrs	n/a	Born midway through inter.	2yrs	3yrs & 5yrs	3yrs & 5yrs	4yrs & 6yrs
Occupation (Mo)*	Level 4	Level 3	Level 2	Level 7	Level 2	Level 2	Level 3
Occupation (Fo)*	Level 2	Level 2	Level 2	Level 7	Level 2	Level 2	Level 2
Current working status of participating parent	Full-time to part-time	Carers leave	Extended mat. leave	Part-time	Carers leave	Carers leave	Part-time
Education level (Mo)	University	University	University	University	University	University	University
Education level (Fo)	University	University	University	University	University	University	University
Other caring respon.	no	yes	no	no	no	no	no

Notes. Yrs = years; F = female; Mo = Mother; Fo = Father; mat. = maternity; respon. = responsibilities; inter. = intervention; level 2 = Professional occupations; level 3 = Associate professional and technical occupations; level 4 = Administrative and secretarial occupations; level 7 = Sales and customer service occupations
*= Levels of occupations as per the Standard Occupation Classification (SOC) (2010)

Table 3.2

Summary of demographic information of each child participant completing the PELD programme

	Sophie	Jack	Conor	Daniel	Ellie	Grace	Luke
Age (m; d)	13m 15d	11m 28d	13m 15d	11m 2d	10m 25d	17m 7d	12m 24d
Gender	F	M	M	M	F	F	M
Participating child's place in family	youngest	n/a	eldest	youngest	youngest	youngest	youngest
Languages spoken at home	Eng/European	Eng	Eng	Eng	Eng	Eng/Irish	Eng
Hearing	mod?	mild	mod	mild	mild/mod?	mild	WNL
Sight	Waiting exam	Long-sighted	WNL	Long-sighted	WNL	astigmatism	WNL
Previous SLT services	n/a	yes	n/a	yes	yes (FEDS)	yes	yes
Additional SLT *	yes	yes	yes	yes	yes	yes	yes
Bayley Cognitive ax							
Scale score	3	6	5	3	5	2	3
Composite score	65	80	75	65	75	60	65
SD	-2	-1	-1	-2	-1	-2	-2
Attends day care	5 days	n/a	n/a	n/a	n/a	n/a	3 days
Hospital duration	9 days	16 days	23 days	42 days	4 months	7 days	5 days

Notes. m; d = age in months and days at initial baseline; ? = query; F = female; M = male; eng = English; WNL = within normal limits; FEDS = feeding, eating, drinking, swallowing management; ax = assessment; SD = standard deviation from the mean' n/a = does not attend day care

* = additional SLT services are programmes/SLT services that were completed during the PELD intervention

Table 3.3

Total number of sessions attended by all participants per intervention term

	Sophie	Jack	Conor	Daniel	Ellie	Grace	Luke
Term 1							
Group	3/4	3/4	3/4	-	4/4	-	-
Ind	2/2	1/2	2/2	-	1/2	-	-
Term 2							
Group	3/4	-	-	2/4	3/4	3/4	3/4
Ind	1/2	-	-	2/2	2/2	2/2	2/2
Term 3							
Group*	2/3	2/3**	-	3/3	3/3	2/3	2/3
Ind	2/2	2/2**	-	2/2	2/2	2/2	2/2
No. of terms	3	2	1	2	3	2	2
Total sessions	13/17	8/11	5/6	9/10	15/16	9/10	9/10
Total sessions attended by parent in study	13/17	6/11	5/6	9/10	15/16	9/10	9/10

Notes. Ind = individual session; no. = number; - = participant did not sign up for a term of intervention.

*= a group session was cancelled by the service resulting in three group sessions offered for this term

**= the father (parent not participating in study) attended some sessions. Note the discrepancy evident in total sessions for Jack's parent is due to the father attending the intervention instead of the mother for two sessions.

3.4 The PELD Intervention

The PELD intervention is a parent-child interaction therapy implemented with young children with Down syndrome. The goal of the programme is to up-skill parents with the aim of improving their child's speech, language, communication and interaction skills. The PELD intervention is offered to children between 10-36 months of age and consists of two different strands of the programme. A child is allocated to a particular strand depending on their age. For the purpose of the current study, only the first strand of the PELD intervention is evaluated as it ensured it was

the parents first experience of the PELD intervention. All children in this strand ranged from 10-18 months at entry. Three terms of the intervention were offered for each strand and families had the option of completing all or some of the terms. Each term consisted of a different themes with the five primary intervention strategies targeted in each theme (all of which are outlined in the proceeding paragraph). The three terms were rolled out over 10 months and consisted of bi-monthly intervention sessions.

Each intervention term consisted of four group sessions and two individual sessions. Before the first term commenced, parents were invited to attend a one-hour workshop where the goals and strategies of the PELD programme were explained. Group sessions were fortnightly and lasted 45 minutes in duration. Individual sessions were 30 minutes in duration and scheduled halfway through an intervention term (after two group sessions) and at the end of an intervention term. Group sessions consisted of a maximum of seven children and their parents. Two speech and language therapists (SLTs) implemented the intervention while a third SLT recorded the notes for each session. During the group sessions, five early intervention strategies were targeted. These included greetings, listening and attention skills, interaction strategies (such as following your child's lead, *Observe Wait and Listen (OWL*) by the Hanen Early Intervention Programme, 2011), Lámh and speech sound play/practise. A number of parent and child goals were targeted for each strategy (see Table 3.4 for a summary of the goals within each strategy). These five strategies were targeted in each session using five core themes. All themes included functional activities that could be incorporated into everyday routines of the family. The targeted themes included; mealtimes, morning routine and dressing, playtime and books, outdoors, home and family life.

Clinicians adopted aspects of the Teach-Model-Coach-Review (TMCR) protocol that was previously described by Kaiser and Roberts (2013). Clinicians followed the general framework of the TMCR protocol however not every aspect was addressed in each session. As previously mentioned, the clinicians explained the strategies to the parents at the workshop before the first term of the intervention commenced. Goals were re-introduced at the start of every session thus following the *Teach* aspect of the protocol. The *Model* and *Coach* aspects of the protocol were implemented throughout the five goals within the sessions and clinicians would take turns to work with the parents both collectively and individually. Thirty-five percent

(6/17 sessions) of these group sessions were recorded (the first session of each theme) to ensure fidelity to the planned sessions was adhered to. Treatment fidelity will be described in detail later in the chapter. Finally, the fourth and final aspect of the protocol, *Review*, was addressed within the individual sessions. During the individual sessions the clinician and parent chose a specific goal to discuss during this session. The individual sessions created an opportunity for all three parties (clinician, parent and child) to consolidate skills and techniques covered during the group sessions. It also provided an opportunity for parents to ask questions or voice concerns that they may not feel comfortable sharing in the group setting.

Table 3.4

Parent and child goals for the five intervention strategies

Strategy	Parent	Child
Greetings		
Goal	To facilitate child's attention and participation in greetings routine	To attend to participate in greetings routine
Strategies	• Be positioned at child's eye level (Face to face) • Sing song slowly • Use key word signs while singing (e.g. name/where/you) • Wait for child's response • Imitate any gesture/action/vocalisations/word • Encourage child to participate e.g. pull their photo from the bag, use hand over hand (HOH) • Point/name and draw attention to picture • Wait for child's response	• Attention: attend to parent singing the song/photo • Point • Participate in singing • Attempt/produce Lámh sign • Attempt/produce word/vocalisations
Listening & Attention		
Goal	To facilitate child's listening, attention and participation	To listen, attend and identify sounds
Strategies	• Face to face interaction • Encourage child to listen/attend • Use key word signs (wait/listen) • Wait for child's response • Alert child to correction object if they do not respond after 5 seconds • Name sound for child while they are attending to the object • Imitate any gesture/action/vocalisations/word	• Participate in activity by alerting to the sound • Identify sound with reach/eye gaze/ head turn towards the related object • Attempt/produce Lámh sign • Attempt/produce word/vocalisations

Interaction		
Goal	To observe, wait and listen (OWL) and follow child's lead during the specific themed activity e.g. outdoor play	To initiate communication and interaction
Strategies	• Face to face • Use simple sound play and target words/signs • Wait for child to take their turn, observe child's body language and communicative attempts, respond to child communication • Cue child to take a turn with expectant waiting • Imitate any gesture/action/vocalisations/word	• Attend to parent in activity • Participate in activities • Imitate sound/signs/words
Lámh		
Goal	To learn a pre-specified number of Lámh signs and support children to attend to their own hands	To tolerate hand over hand manipulation and attend to Lámh signs with a view to producing Lámh signs in this context.
Strategies	• Face to face • Massage child's hands • Imitate the actions modeled by therapist while saying each word as they sign • Produce each sign 5 times before using HOH with the child • Sing while singing the group song • Continue to use signs to facilitate child's understanding even if child appears not to respond	• Attend to parent in activity • Attend and comply with HOH light massage to fingers and hand • Participate in activity by moving to the music of the song • Tolerate with HOH manipulation • Imitate sounds/signs/ words
Speech sound play		
Goal	To encourage their child to engage in speech sound play	To attend to and imitate speech sound productions of parent
Strategies	• Sing song slowly and cue child to point to body part using HOH • Produce certain sounds clearly while face to face with the child • Wait for child's response • Imitate any gesture/action/vocalisations/word • Make the specific sounds and cue the child to imitate	• Attend to parent's speech productions • Participate in the activity by moving to the music of the song/taking a turn to communicate 'more' • Imitate sounds/signs/words

Notes. All of the strategies were targeted through the five different themes.

3.5 Methods and Materials

The measures employed to evaluate the effectiveness of PELD intervention for each child and parent participant included a range of standardised assessments, parental checklists and observational measures.

3.5.1 Standardised assessments.

The Bayley Scales of Infant and Toddler Development III (Bayley, 2006) was employed to assess the cognitive abilities of the children at the start of the study. This standardised assessment evaluates the development of infants and toddlers aged between one and 42 months through a series of developmental play tasks (Bayley, 2006). These early developmental play tasks can include activities that require the child to search for a fallen object; attend to familiar and unfamiliar objects; and to engage in relational, representation and pretend play. This tool highlights the strengths and weaknesses of a child with a suspected developmental delay and is commonly used when assessing children with DS or children with other intellectual disabilities (Milne, McDonald & Comino, 2011; Russell, van Heerden, van Vuuren, Venter & Joubert, 2016). With regards to the psychometric properties of this assessment, Bayley (2006) report a score of 0.91 for the reliability coefficient for the Cognitive subtest of the Bayley indicating a high reliability. Bayley (2006) report all psychometric properties of the Bayley-III meet the minimal criteria, however the reliability for children under 6 months could be improved. Scale scores, composite scores and standard deviations (SD) were included in this study for the purpose of quantifying the cognitive impairment of each participant. The scores obtained must be viewed with caution as this assessment is not standardised on children with intellectual disabilities therefore the information obtained is used for descriptive purposes only (Glenn, Cunningham & Dayus, 2001). This assessment was administered at the first baseline session only and was administered in the middle of each baseline session so that the child had some time to become familiar with the clinician who completed the assessment. The purpose of administration was to assess the cognitive abilities of each participant, as cognitive deficits may have an impact on the effectiveness of the intervention.

The Preschool Language Scales Fifth Edition (PLS-5) (Zimmerman, Steiner & Pond, 2011) was employed to assess change in each child's language. The PLS-5

is a standardised language assessment that assesses both the receptive and expressive language abilities of children from birth to 7;11 years (Zimmerman et al., 2011). The PLS-5 has been modified from the previous edition (PLS-4) to include the assessment of a wider variety of early play behaviours such as turn-taking, interactive play, functional play and relational play (Zimmerman et al., 2011). The PLS-5 consists of test items that provide information about the child's early expressive communicative intentions and language (for example eye contact, pointing, gesturing, babbling, types of consonants and vowels within the child's repertoire; first words) and early receptive communication acts (for example responding to their name; searching for fallen objects; searching for person who is talking; anticipating what will happen next during play activities; demonstrating different types of play). Items up to the age of 2;0 years can be scored for observations and/or caregiver responses for the elicitation of a task. It was deemed the most appropriate language assessment for the participants in this study given the age and the developmental delay of the participants. In addition to this, if the child did not demonstrate a certain behaviour/task during the assessment, marks were awarded for certain test items if their caregiver could provide an example of a time their child exhibited a behaviour or skill. Only test items up to the age of 2;0 were allowed for caregiver report and test items proceeding this point could only be marked correct if the skill was exhibited by the child at the time of assessment. Although the PLS-5 is not standardised on children with intellectual disability, previous studies have employed this assessment as a means of measuring language change and scale scores have typically been reported (Wright et al., 2013). Scale scores are included as they control for maturation which is a limitation of raw scores (Ebbels, 2017). The current study reported both scale scores and raw scores of the participants to capture change. Given the chronological age and cognitive impairment of the participants, all assessments began at the 0:0-0:2 age starting point. In line with the standardised instructions of the assessment, certain test items were administered by the parents following the SLTs instructions if it was noted that the children were most responsive to a familiar adult. The PLS-was administered at two different time points; the initial baseline assessment and at the post intervention assessment. The PLS-5 was administered directly after the Bayley cognitive screen at the initial baseline session for all participants.

3.5.2 Parental report.

Vocabulary growth was measured using the Vocabulary Checklist 1- First 120 Words (Down Syndrome Education International, 2012). This checklist outlines the typical first 124 words that are acquired by children and was completed at the initial baseline visit, each midway assessment that was completed between terms and post-intervention. The checklist consists of a variety of different word types and divides the vocabulary into nouns, verbs, adjectives, prepositions, pronouns and other functional words. The checklist is designed to be completed by parents as a means of tracking their child's vocabulary development. Parents recorded their child's vocabulary under the following headings: *Understands; Understands and Signs; Says word in imitation; Uses word Spontaneously;* and *Understood by unfamiliar listener*. A child *understands* a word when they can identify the object/action when another option is present. *Understands and signs* is scored when a parent reports that their child consistently uses a sign that is also contextually appropriate. *Says word in imitation* is scored when a child is reported to say a word in response to the adult modelling the word in the preceding act. *Uses word spontaneously* is scored when a child is reported to use a word without requiring an adult prompt. *Understood by a familiar listener* is scored when a child uses a word that can be understood by people outside of the child's immediate family.

Parental stress was also measured in this study. Parental stress has been shown to have an impact on many parent-implemented therapy outcomes with the Parental Stress Index (PSI) being an outcome measure that is widely used with parents of children with cognitive impairment (Kaiser & Roberts, 2011). Although parental stress and its impact on PCITs was not a goal of the current study, it was deemed important for this study to gain some insight with regards to the potential stress experienced by the parents. In order to gain this insight, parents were asked to provide a numerical value for stress by answering the following question; *'On a scale of one to 10, how stressed do you feel parenting a child with Down Syndrome?'*. A score of one indicated that the parents never felt stressed and a score of ten reflected a high level of stress.

3.5.3 Observational measures.

A parent-child play interaction was video recorded at each assessment session and was later coded by the researcher using the Pivotal Behavior Rating Scale (Mahoney & Wheeden, 1998) and an adapted parent language and interaction rating scale. The Pivotal Behavior Rating Scale was employed as it has been effective in capturing change in children with DS in previous studies (Karaaslan et al., 2011). In addition to this, the parent interaction and language rating scale was compiled using aspects of the *'Teacher Interaction and Language Rating Scale'* by Girolametto, Weitzman and Greenberg (2000). This rating has been used as a means of measuring how teachers implement strategies from the Hanen Programme with their pupils. A detailed description of both rating scales is discussed later in the chapter (see Chapter 3, section 3.5.3.2). As the PELD intervention also trains and coaches parents in communication strategies in a group setting, it was deemed appropriate to adapt this published checklist and make it specific to parents of children with DS. Both rating scales were double coded for agreement by an external rater who was blind to time and the goals of the intervention.

3.5.3.1 Parent-child interaction.

A parent-child play interaction was video recorded using an iPad in order to further evaluate the programme. This assessment followed the procedure outlined by Mahoney, Kim and Lin (2007). The video took part at the end of each assessment session and lasted approximately seven minutes in duration as children who are less than the 12-month developmental age can typically only sustain attention for this long (Mahoney, Kim & Lin, 2007). In principle, only the child and the parent should be in the room where the video recording took place, however, as many parents had other young children this was not always feasible. Therefore, although siblings of the children were not captured on the video recording, they may have been present elsewhere in the room at the time of the recording. The parents were instructed to play with their child as they typically would using a standard set of preselected toys. The toys chosen consisted of a mixture of cause-and-effect toys, constructive and manipulative toys and toys that elicited pretend play (Karaaslan & Mahoney, 2015; Wright et al., 2013). The toys consisted of the following; a xylophone, a drum, three books, a doll, a blanket, a red cloth bag, shakers, a bell, a rattle, microphone, pop up

wooden blocks, stacking cups, three small cars, a cup, spoon, building blocks, plastic ball, a phone and a sippy cup. Parents were allowed to use as many or as few of the toys as they liked. Each parent was also instructed to introduce a toy that is familiar to the child (a favourite toy) at the six-minute mark in order to see the impact of familiar versus unfamiliar toys on the child's behaviour. The researcher informed the parent when the six minutes had passed so that all parents introduced their child's favourite toy at the same time. If a child became unsettled or fussy during the interactive play, the video recording was paused and continued once the child had settled (Mahoney, Kim & Lin, 2007). After each session, the video was uploaded to a password protected computer and deleted from the iPad. It is important to note that only the minutes from two to seven (a total duration of five minutes) were analysed. This protocol has been reported in previous studies as it allowed the parent and child to settle in to the play activity (Mahoney & Perales, 2005).

3.5.3.2 Rating scales for measuring parent-child interaction.

The Pivotal Behavior Rating Scale (Mahoney & Wheeden, 1998) measured skills such as attention and initiation (see Appendix E). The attention component is divided into the child's general ability to attend to an activity, their level of persistence within an activity and their involvement in the activity. Ratings of one to five are used across these variables with a score of one meaning the child presented with very low attention and a score of five meaning the child had very high attention. The initiation component of the scale measured the child's ability to initiate activities, the child's ability to initiate interaction with adults and affect which was characterised by the child's emotional state during the interaction. These three variables are graded using a five-point scale with a score of one meaning the child shows very low initiation and a score of five demonstrating a high level of initiation.

The Parent Interaction and Language rating scale was compiled using aspects of the *'Teacher Interaction and Language Rating Scale'* by Girolametto et al. (2000). This measure consisted of a seven-point Likert scale that scored a parent's language and interaction strategies across nine different items. The measure scored a parent's ability to wait and listen, follow their child's lead, encourage turn-taking, use of gesture and Lámh, imitation of their child's actions and finally, the parents use of a variety of labels (See Appendix F). The researcher graded each of the above

items using the five-minute video of the parent-child interactive play observation. A score of one was awarded if the parent *'almost never'* used the strategy and a score of seven was awarded in the parent used the strategy *'consistently'*.

3.6 Procedure

Data for this study was collected over a period of 10 months and consisted of five different assessment sessions. The sessions consisted of two pre-intervention sessions where two baselines were obtained followed by three additional assessment sessions that took place after each term of the PELD programme. These assessment sessions were completed by a researcher who was blind to the purpose, aims, goals and content of the PELD intervention. This researcher has a background in speech and language therapy and worked at the centre where the PELD intervention was offered one day a week while conducting the research. The research was not involved in the PELD intervention during the research and only liaised with the families involved in the research at the home assessment sessions. Each assessment session took place in the family homes of the participants. Sessions took place in the living room or play room of the participants. The majority of sessions took place in a quiet environment with just the child and parent present. As the sessions were in the homes of the participants, additional family members (other parent or siblings) would occasionally be present in the room. In line with ethical procedures, data storage and anonymity were maintained. Figure 3.1 summarises the assessment and intervention process. Assessment measures were taken regardless of whether the participants withdrew from the intervention. Two participants joined the study at a later date (between intervention one and intervention two) so their initial baseline assessments were collected between week 15-16 of 42 weeks. One home assessment could not be completed due to personal circumstances of the family (Jack's family) so there is missing data at the 'Intervention 2' time point for this participant. Table 3.5 presents the age of each participant at the time of the assessment sessions.

The Assessment and Intervention Process

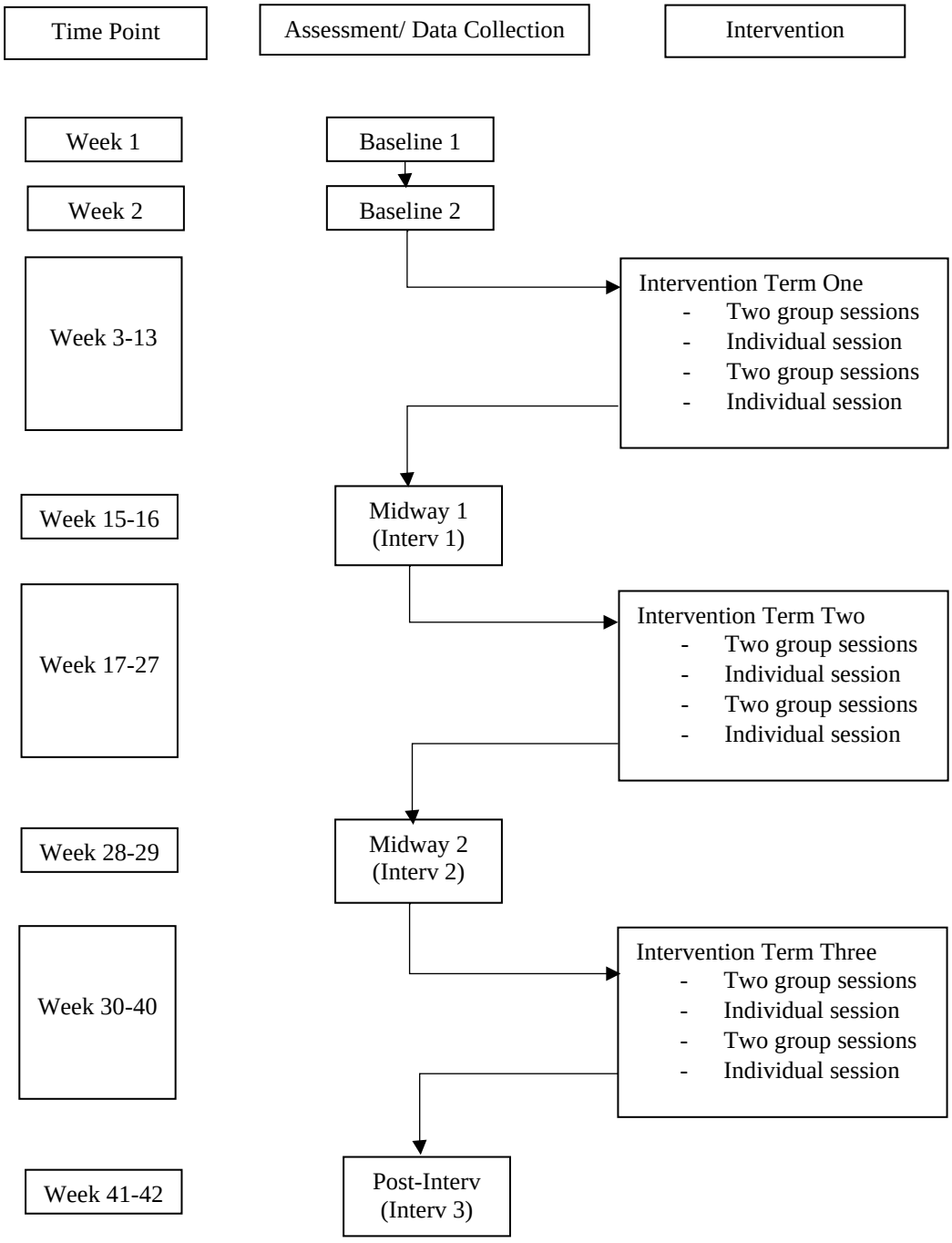

Figure 3.1. The general assessment and intervention process. Note certain participants may have withdrawn or joined the study at a later date throughout this process.

Table 3.5

Age of participants at each time point

Time	Wk 1	Wk 2	Wk 15/16	Wk 28/29	Wk 41-42
	B/L 1	B/L 2	Interv 1	Interv 2	Interv 3
Sophie	13m 15d	13m 19d	17m 7d	20m 17d	23m 3d
Jack	11m 28d	12m 5d	15m 24d	-	22m 2d
Conor	13m 15d	13m 23d	17m 4d	20m 15d	23m 5d
Daniel	11m 2d	11m 9d	15m 18d	18m 7d	21m 1d
Ellie	10m 25d	11m 2d	14m 22d	18m 3d	21m 3d

Time*		Wk 15-16	Wk 28/29	Wk 41-42
		B/L 1	Interv 2	Interv 3
Grace		17m 7d	20m 10d	23m 6d
Luke		12m 24d	15m 13d	18m 8d

Note. m = months; d = days; Wk = week; B/L = baseline; - = missing data for Jack at Intervention 2.

*= Participants Grace and Luke joined the study at a later date

3.6.1 Baseline assessments.

As previously stated, two baseline assessments were conducted prior to the intervention. The first baseline measures were taken two weeks before the PELD programme began. The researcher took these measures in the homes of the participants. This visit ranged from one to two hours depending on the engagement of each child (e.g. some children required breaks for feeding). At this visit the remaining demographic information was collected from the parent including a second language questionnaire if appropriate (See Appendix F). The researcher and the parent completed the Vocabulary Checklist 1- First 120 Words (Down Syndrome Education International, 2012). After the completion of the checklist, a numerical score for parental stress was obtained and recorded. Following this, two formal assessments were administered; the cognitive subtest of the Bayley Scales of Infant and Toddler Development III (Bayley, 2006) and the Preschool Language Scales Fifth Edition (PLS-5) (Zimmerman et al., 2011). The Bayley cognitive subtest was administered prior to the PLS-5 at all initial baseline sessions. The visit concluded

by observing and recording a seven-minute parent-child interactive play scenario using a set of pre-selected toys. The parent was instructed to play with their child as they typically would using the toys provided by the researcher. One week after the initial baseline was obtained, the researcher returned to the homes of the participants and recorded the parent-child interactive play observation again. This second baseline session lasted between 20-30 minutes depending on the engagement of child. All video recordings were then analysed using the Pivotal Behavior Rating Scale (Mahoney & Wheeden, 1998) and the parent interaction and language rating scale. Two baselines were collected for the five initial participants. The two participants who joined the study at a later date (Grace and Jack), completed their baseline assessments at Week 15/16, the time at which the other five participants completed their first midway assessment.

3.6.2 Midway assessments.

After the first term of the intervention was completed, the first of the midway assessments were collected at week 15/16. This assessment again took place in the homes of the participants and lasted 40-60 minutes. The Vocabulary Checklist 1- First 120 Words (Down Syndrome Education International, 2012) was updated and the parents were again asked to give a numerical value for parental stress. The seven-minute parent-child interactive play observation was then recorded. The same procedure was followed for the second midway assessment (Interv 2) that took place between week 28-29. After each midway, the video recordings were again analysed using the Pivotal Behavior Rating Scale (Mahoney & Wheeden, 1998) and the parent interaction and language rating scale.

3.6.3 Post-intervention assessment.

One to two weeks after the final intervention session (week 41-42), the researcher returned to the homes of the participants to complete the post-intervention assessment. The Vocabulary Checklist 1- First 120 Words (Down Syndrome Education International, 2012) was updated and the parents were again asked to give a numerical value for stress. The PLS-5 assessment (Zimmerman et al., 2011) was re-administered and another parent-child interactive play observation was recorded. The demographic information was also updated to establish if there were any

changes in the family circumstances/child's hearing/sight during the intervention period.

3.7 Analysis

3.7.1 Parent-child play interaction video analysis.

The videos were first rated using the Pivotal Behavior Rating Scale (Mahoney & Wheeden, 1998) and the parent interaction and language rating scale. The last five minutes of the video (minutes two to seven) were analysed in order to ensure the participants were engaged in the play from the onset of the analysis (Mahoney & Perales, 2005). This also ensured that the same five minutes were analysed across all measures. The researcher watched each video twice before scoring each rating scale.

Following this, the videos were transcribed using the Codes for the Human Analysis of Transcripts (CHAT) to generate detailed transcripts of the parent-child interactive play observation measuring the same five-minute observation (Venuti, de Falco, Esposito, Zaninelli & Bornstein, 2012). The researcher watched each video and transcribed verbatim each response from the child and the participant. Any unintelligible responses or words were coded using '*xxx*'. The child's communicative abilities were coded into three main categories; verbalisations, gesture and use of sign language. Verbalisations were categorised into vocalisations and babbling. Gestures were coded as deictic, conventional and iconic (Zampini & D'Odorico, 2009). Signs produced by the child were coded as imitated, prompted or spontaneous (Wright et al., 2013). See Table 3.6 for a detailed description of each category.

3.7.1.1 Parent language sample analysis.

Once the transcription was complete the researcher used the Computerised Language Analysis (CLAN) software to analyse the parent's language. Scores for total utterances, MLU utterances, MLU words, MLU morphemes, frequency types and frequency tokens were generated using the KIDEVAL programme from the CLAN software. Before running the KIDEVAL command, the researcher ran the MOR command which inserts a morphology and grammar tier into the transcript which is needed for the KIDEVAL command to run. Scores for each measure

obtained at baselines and the three intervention time points were plotted on line graphs for each participant as a means of demonstrating any change throughout the intervention.

3.7.1.2 Child analysis.

The FREQ command of the CLAN software was used to generate the frequency of each of the pre-lexical verbalisations, gestures and signs exhibited by the child during the free-play analysis. These scores were also plotted on line graphs for each participant as a means of demonstrating any change throughout the intervention.

Table 3.6

Description of child analysis coding

Category	Code	Definition
Pre-lexical Verbalisations		
Vocalisation	&=vocalises	All primitive sounds and single vowel sounds
Babbling	&=babbles	Vowel and consonant combination that consists of one or more syllables
Gesture		
Deictic	&=gesture:deictic	Gestures that single out a referent in the environment (e.g. pointing, showing)
Conventional	&=gesture:conventional	Gestures with a culturally defined meaning or form (e.g. waving)
Iconic	&=gesture:iconic	Gestures which refer to objects, people or events, reproducing the physical or function characteristics (e.g. actions for sleeping)
Signs		
Imitated	&=sign:imit	A child produces a sign or approximation within 8 seconds of the parent's model.
Prompted	&=sign:prom	A child produces a sign in response to a parents direct prompt or question
Spontaneous	&=sign:spon	A child produces a recognisable sign independent of a model occurring in the preceding 5 seconds

Note. Codes were entered using the CLAN software

3.8 Treatment Fidelity

The therapists adherence and delivery of the intervention was measured for treatment fidelity. It was reviewed on an ongoing basis throughout the programme so that feedback could be given with the aim of increasing fidelity in subsequent sessions. Thirty-five percent of the group sessions (6 /17) were recorded and these videos were coded for fidelity by two researchers. The therapists submitted a plan of the goals of the session to the researchers which were different to the goals that the outcome assessor was blind to during the intervention. The goals related to therapist implementation and were used to track and measure fidelity (Lieberman-Betz, 2015). Only the group sessions were included in the fidelity measures. The therapists did not record the individual sessions due to their sensitive nature of the ono-to-one sessions. Four of these videos were reviewed by two researchers and the remaining two videos were coded by one researcher due to the unavailability of one of the researchers. The videos had to be reviewed in a specific time frame as feedback was required in advance of the following treatment session. In addition, ethical requirements were such that the videos needed to be deleted in a timely manner.

The first session of each intervention theme was recorded and sent to the two researchers. For four of the videos, two researchers watched the videos together and individually rated them for adherence to the treatment protocol outlined in each session plan. Following this, inter-rater reliability was calculated and the researchers reached a consensus on the final agreement score (see Table 3.7). The remaining two videos were rated for adherence by only one researcher. Agreement of >93% was met for all four of the sessions that were coded for inter-rater reliability. Reliability for session four and five could not be calculated as only one person rated the videos. Scores for adherence for session one was low with only 59.6% noted. This may be due to the fact it was the first group session and that the clinicians may still have been adjusting to the delivery of the intervention and the importance of adhering to the session goals. In addition to this, a drop in adherence was again noted in session six (See tables 3.7). This may be related to the fact that a new clincian was involved in the implementation of this session as the one of the usual therapists was unavailable.

Table 3.7

Scores for adherence and inter-rater agreement

	Adherence %	Agreement %
Session 1	59.6	93.5
Session 2	83.7	95.3
Session 3	81.8	96.9
Session 4	83.7	-
Session 5	87.8	-
Session 6	70.3	96.2

Note. Session 4 and 5 were coded by one coder

3.9 Inter-rater Reliability

Inter-rater reliability was conducted on the video transcripts and the observational measures (the Pivotal Behavior Rating Scale and the parent interaction and language rating scale). The external rater was blind to the goals of the intervention and time of assessment as reliability was calculated post-intervention. Twenty percent of the transcripts (6 of 30 transcripts) were coded by the external rater. The external rater reviewed the six videos and differences were recorded. Agreement of >95% was obtained for all transcripts reviewed (see Table 3.8).

Table 3.8

Scores video transcripts inter-rater agreement

	Agreement %
Transcript 1	97.1
Transcript 2	98.5
Transcript 3	95.6
Transcript 4	98.5
Transcript 5	97
Transcript 6	99.1
Total agreement	97.7

Note. All six transcripts were selected randomly

The two observational rating scales were coded for reliability using Mahoney's (2009) procedure for establishing interrater agreement for the Maternal Behaviour Rating Scale (MBRS). The external rater was again blind to which videos were pre-intervention, midway assessments and post-intervention videos. Five videos were chosen at random and watched by the researcher and external rater. Both parties watched the videos together and scored them independently. After each video was played twice, their scores were discussed and a criteria for scoring was achieved. Each rater then watched and rated an additional five videos independently. Their reliability had to meet three criteria before additional videos were scored. The raters must a) have 100% agreement within +/- one point, b) have 80% exact agreement on the items, c) Cohen's Kappa of more than 0.61. As there was a lack of variability and spread in the participants scores, Cohen's Kappa was not generated for certain variables and did not always meet the criteria of 0.61 for others. Once this initial reliability was established, the raters each rated 20% of the remaining videos (4 of the 20 unrated videos) and their final scores for agreement were calculated. Agreement of above 85% was achieved for both measures. The breakdown for reliability is illustrated in Table 3.9.

Table 3.9

Final scores for inter-rater agreement

	CBRS	PRS
100% +/- one point	Achieved	Achieved
80% exact agreement	92.8%	86.1%
Cohen's Kappa (Range)	0.5-1.0*	0.556-1.0**

Note. CBRS = Child Behavior Rating Scale; PRS = parent interaction and language rating scale.

*Scores for one variable was not computed due to lack of variation in the children's scores.

**Only one variable did not meet the criteria of >0.61 for this rating scale

Chapter Four: Results

4.1 Introduction

The following chapter presents the results of the PELD intervention programme and the impact it had on the aforementioned child and parent outcomes. The results for each participant are presented in tables and figures under the three research questions. A descriptive analysis of the outcomes are presented in multiple line graphs for all participants under each variable to allow for a clear illustration of change over time.

4.2 Research Question 1: Does the PELD intervention programme encourage the development of language in infants with DS?

The first research question explored the impact the intervention had on the language development of the participating children. Language outcomes were captured using standardised testing (initial baseline; post intervention), parental report (initial baseline, after each intervention term, post intervention) and observational measures (collected at every baseline, after each intervention term, post intervention).

4.2.1 Standardised test scores.

The Preschool Language Scales Fifth Edition (PLS-5) (Zimmerman et al., 2011) was employed to assess change in each child's language growth. There were approximately 10 months between pre- and post- PLS-5 assessments for all participants excluding Grace and Luke who had approximately six months between pre- and post-assessments due to their later uptake of the intervention. Raw scores, scale scores, confidence intervals and scores for standard deviation (SD) are presented for each participant as a means of showing change (see Table 4.1). Raw scores were included as they do not consider the age of the child, which is important when using standardised tests on children with intellectual disabilities. Scale scores were reported as they control for maturation and these scores were then used to calculate each child's standard deviation from the mean (mean = 100; SD = 15). Scale scores also allowed for comparison of outcomes within the literature as previous studies have reported scale scores when measuring child language

outcomes in PCITs with children with DS (Wright et al., 2013). Table 4.1 presents a breakdown of scores per participant under the three PLS-5 subscales; auditory comprehension, expressive communication and total language score.

Table 4.1

Pre- and post-intervention PLS-5 scores for all seven participants

	Pre				Post			
	Raw Score	SS	95% confidence interval	SD	Raw Score	SS	95% confidence interval	SD
Auditory Comprehension								
Sophie	9	64	59-77	-2	17	73	68-81	-1
Jack	11	79	73-93	-1	14	61	57-70	-2
Conor	10	58	54-72	-2	16	69	64-78	-2
Daniel	10	74	68-93	-1	18	77	72-85	-1
Ellie	11	79	73-93	-1	22	92	86-99	WNL
Grace	13	73	67-85	-1	22	92	86-99	WNL
Luke	11	64	59-77	-2	15	66	61-75	-2
Expressive Communication								
Sophie	9	56	52-70	-2	20	81	75-90	-1
Jack	14	90	83-98	WNL	19	78	72-87	-1
Conor	14	73	67-85	-2	20	81	75-90	-1
Daniel	6	57	53-68	-2	22	88	82-96	WNL
Ellie	11	78	72-87	-1	22	88	82-96	WNL
Grace	18	89	82-99	WNL	22	88	82-96	WNL
Luke	11	63	58-76	-2	15	66	61-76	-2
Total Language Score								
Sophie	18	57	53-67	-2	37	76	70-85	-1
Jack	25	83	77-91	-1	33	68	63-77	-2
Conor	24	63	58-73	-2	36	74	69-83	-1
Daniel	16	63	58-73	-2	40	81	75-90	-1
Ellie	22	77	71-86	-1	44	89	83-97	WNL
Grace	31	80	74-89	-1	44	89	83-97	WNL
Luke	22	61	56-71	-2	30	64	59-74	-2

Note. SS = scale score; SD = standard deviation; WNL = within normal limits

4.2.1.1 Auditory comprehension subscale.

As illustrated in Table 4.1, there was a gain of between 3 and 11 points in raw scores for each participant from pre- to post-intervention. In terms of scale

scores (SS), there was an increase of between 2 and 19 points for all participants excluding Jack who was the only participant who showed a decrease in SS scores post-intervention. An increase of 10 points or more was noted in Conor, Ellie, and Grace's post-test SS scores. Two participants (Ellie and Grace) demonstrated age appropriate receptive language abilities according to the standardisation on the post-intervention PLS-5 assessment.

4.2.1.2 Expressive communication subscale.

Raw scores for expressive communication increased by a minimum of 4 points and a maximum of 16 points for the participants based on the pre- and post-assessment. Scale scores for the expressive communication subtest showed great variation among the participants. There was a gain of between 2 and 31 points for five of the participants with three of these participants increasing SS scores by 10 or more points. Two participants (Jack and Grace) decreased their SS scores from pre- to post-intervention although it is worthy to note that this decrease was by a single point for Grace's SS score. Three of the participants (Daniel, Ellie, Grace) demonstrated age appropriate expressive language abilities post-intervention according to standardisation on the PLS-5 assessment.

4.2.1.3 Total language score.

Total language scores displayed a minimum change of 8 points and a maximum change of 24 points noted in raw scores for the participants. Apart from Jack, there was a gain of between 3 and 19 points evident for total language scale scores. Four of these participants (Sophie, Conor, Daniel and Ellie) increased their SS scores by 10 or more points at post-intervention. Two participants (Ellie and Grace) illustrated age appropriate language abilities based on the PLS-5 standardised testing following the intervention.

4.2.2 Parental report.

Vocabulary growth was measured using the Vocabulary Checklist 1- First 120 Words (Down Syndrome Education International, 2012). This parental report recorded the first words learned by each child under the five headings described in the previous chapter. Multiple line graphs were plotted for each participant to show

change in vocabulary growth across all time points for each of the five items in Figure 4.1. Only the items that show change were plotted on the graph. That is, for graphs that only display some of the items, the participant scored zero across all time points for the items excluded. Scores for each participant are displayed in Figure 4.1.

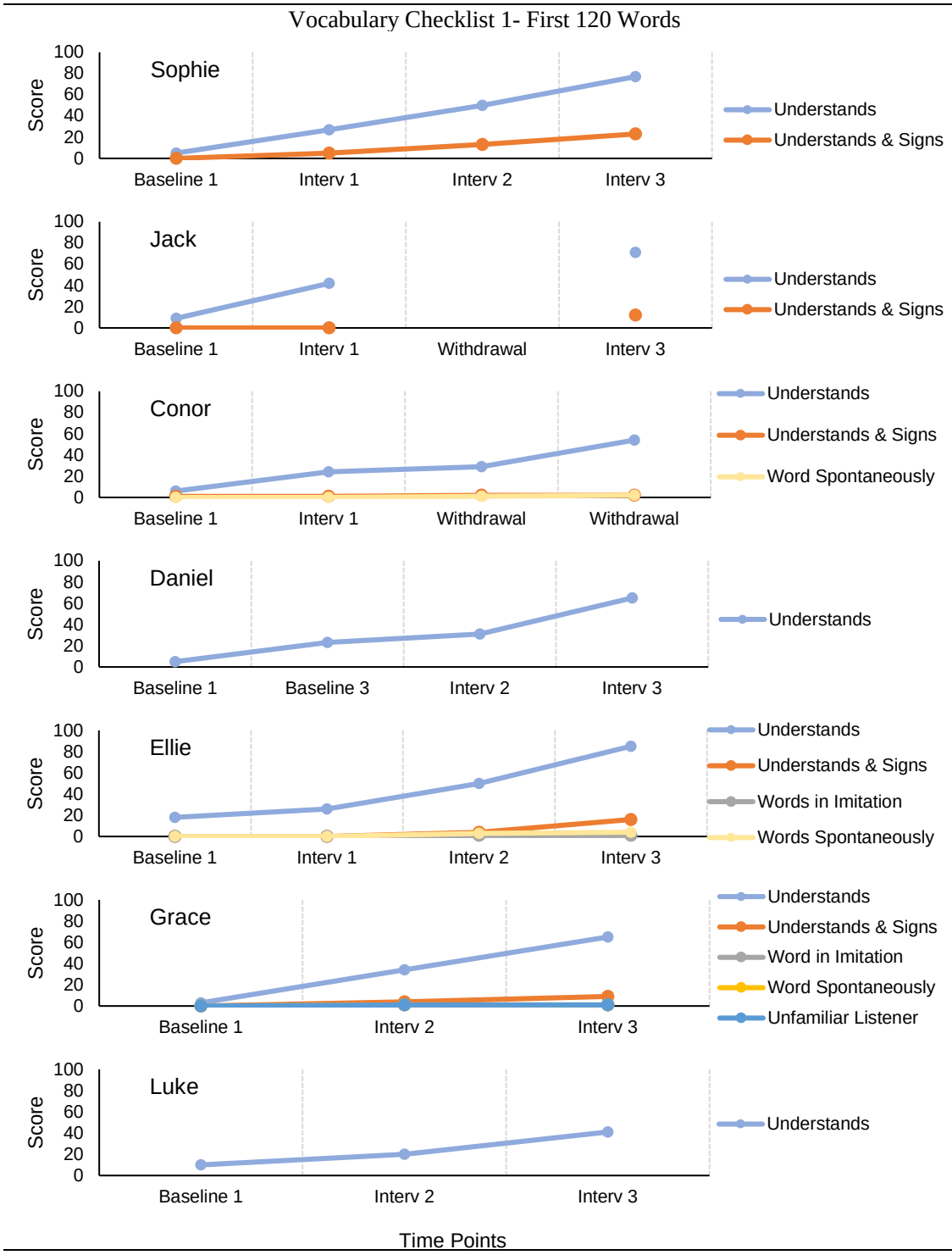

Figure 4.1. Vocabulary scores for all participants reported by parents using the Vocabulary Checklist 1- First 120 Words. For Grace, words in imitation, word spontaneously and unfamiliar listener scores were the same values so these three lines overlap. Jack is missing data for the third data collection time point. Grace and Luke started the intervention in at Intervention 2. Understands = total number of words the child understands; word spontaneously = number of words spontaneously produced by the child; unfamiliar listener = total number of words that can be understood by an unfamiliar listener; interv = intervention term; withdrawal = withdrawal from an intervention term.

4.2.2.1 Understands.

As illustrated in Figure 4.1, all participants demonstrated an increase in receptive vocabulary according to their parental report. The number of new words understood across the participants increased by a minimum of 31 words and a maximum of 72 words. Sophie demonstrated a steady and consistent increase for this variable across all time points as reflected by the checklist. She also demonstrated the largest increase in word understanding as she was reported to understand 77 words post-intervention compared to word knowledge of 5 words pre-intervention. Jack demonstrated a steep increase in his understanding of words following term one of the intervention. This was still showing an upward trajectory at the final term of intervention, however, it is impossible to comment on the rate at which he understood these words as there was missing data for the second term of intervention. Conor demonstrated an increase in his understanding of words after completing the first term of the intervention. After he withdrew from the intervention his understanding continued to increase, however, the rate varied across the remaining assessment points.

Daniel commenced the intervention during the second term where a steady but minimal increase in word knowledge was reflected by the checklist. It was reported that Daniel increased his word knowledge two-fold at the final term of the intervention. Ellie understood a total of 18 words at baseline which made her the participant with the highest number of words evident at this time point according to her parental report. Ellie showed a consistent increase in her understanding of words throughout the intervention and understood a total of 85 words at the end of the final intervention term. Grace's word knowledge consistently increased once she commenced the intervention at the second term of treatment. Grace's parent estimated she understood a total of three words pre-intervention and this increased to 67 words post-intervention. Finally, Luke displayed a slight increase in the number of words after his first term of intervention with this doubling in number once he completed his second term of intervention. The improvements noted in word knowledge across the participants were reflected in this checklist completed by each child's parent.

4.2.2.2 Understands and signs.

Conor was the only participant with a spontaneous sign, (sign for *bottle*), evident at the baseline measure. A total of five participants (Sophie, Jack, Conor, Ellie and Grace) all showed an increase in the number of signs used post-intervention. Increases in the number of signs ranged from a minimum of one new sign to a maximum of 23 new signs used by the participants. Sophie and Ellie demonstrated the greatest change with Sophie reported to use 23 new signs and Ellie using 16 new signs by the post-intervention assessment. This increase was in line with the increase noted in receptive vocabulary for these same two participants. Jack was reported to have 12 signs at post-intervention and Grace had a total of 9 signs. Conor increased his number of signs by one. The time of the largest increase of signs for all of the five participants was noted between intervention two and intervention three. Daniel and Luke had reportedly no signs by the end of the intervention. It was noted that nearly all of the signs that were reported by the parents were signs that were targeted throughout the intervention. Sophie was the only participant whose signed vocabulary consisted of signs targeted and not targeted in the intervention as reported by the parental checklist.

4.2.2.3 Says word in imitation.

Only two participants (Ellie and Grace) were reported to produce a word in imitation by the end of the intervention. The change for these participants was minimal with an increase of only one word per participant reported by their parents. This was acquired at the end of term two of the intervention for both participants.

4.2.2.4 Uses word spontaneously.

Three participants (Conor, Ellie and Grace) showed gains in spontaneous word use following the intervention with a minimum increase of one word and a maximum increase of four words produced by the children, as noted by parents. Ellie demonstrated the highest increase for this variable with an increase of four words noted. Grace demonstrated an increase of one word and Conor demonstrated an increase of two words. Conor's parent did not report any spontaneous word use during the intervention and these words emerged after he withdrew from the

programme. This increase in words was noted three and six months after his withdrawal from the intervention.

4.2.2.5 Words that can be understood by an unfamiliar listener.

This measures the number of words understood by a person outside of the child's immediate family. Grace increased the number of words understood by one and was the only participant to show change for this item. This was based on the parent's impression of their child's speech ability.

4.2.3 Observational measures.

A language sample analysis was conducted on five minutes of a free-play child and parent play activity. The child's communicative abilities were coded into three main categories; pre-lexical verbalisations, gesture and use of sign language. Pre-lexical verbalisations were subcategorised into vocalisations and babbling. Gestures were coded as deictic, conventional and iconic. Signs produced by the child were coded as imitated, prompted or spontaneous. Detailed descriptions of these variables are discussed in the previous chapter (See Chapter 3 pg. 61). None of the children used spontaneous signs throughout the intervention so this variable was not plotted on the graph. Figures 4.2-4.4 illustrate the change across each variable for each child. If a child showed no change for a particular variable, their results were not plotted. That is, for graphs that only display some of the items, the participant scored zero across all time points for the items excluded.

4.2.3.1 Pre-lexical verbalisations.

Figure 4.2 illustrates the two types of verbalisations; vocalisations and babbling, produced by the participants over time. Vocalisations were defined as any vegetative sounds, laughs/cries or single vowel sounds produced by the child (Olswang et al., 1987). Babbling was defined as any consonant and vowel combination that consisted of one or more syllables (Olswang et al., 1987).

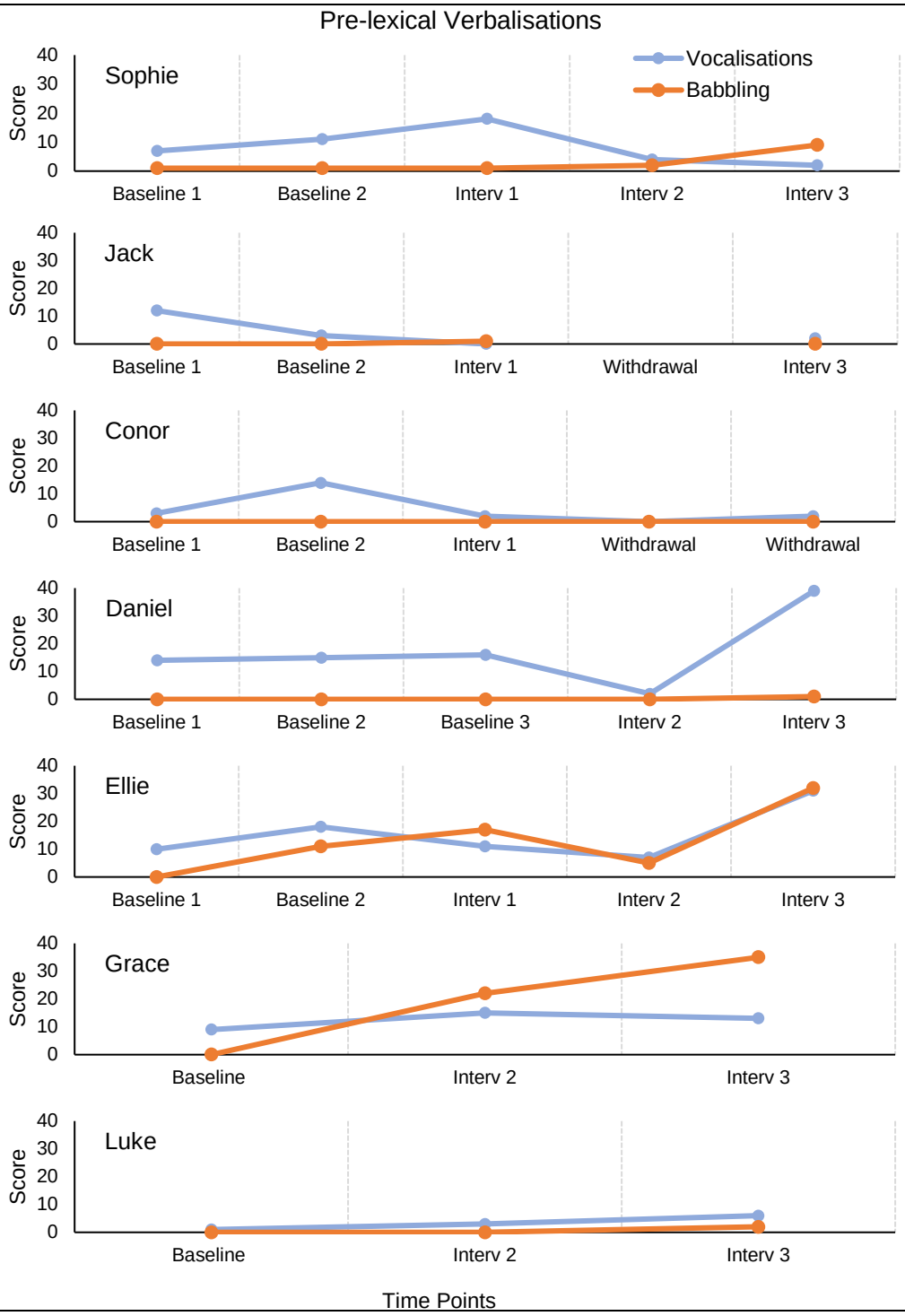

Figure 4.2. Pre-lexical verbalisation scores (i.e. the number of times a verbalisation was recorded during the 5-minute video recording) for each participant. Note there is missing data for Jack. Grace and Luke started the intervention in at Intervention 2. Interv = intervention; withdrawal = withdrawal from a term of intervention.

Considerable variation is evident in the above figure for verbalisations produced by the participants during the five-minute video samples that were obtained. Sophie's scores for vocalisations demonstrated a steady rise across the

baselines and peaked after intervention one. Her vocalisations then decreased and a notable increase in babbling was observed. Jack's vocalisations were relatively high at the initial baseline assessment and decreased across the remaining time points despite having commenced the intervention. It was noted that the child was teething during this time and was more restless than usual during these data collection points. In terms of babbling, no increase in babbling was noted for this participant. Conor demonstrated a spike in his number of vocalisations at the second baseline assessment but this was not maintained during the intervention. No increase in babbling was noted for this participant. Daniel on the other hand demonstrated increased variability for vocalisations. Across the baseline assessment, his scores were relatively high and consistent. A decrease was noted after intervention two but this is contrasted by a sharp increase at the end of the final intervention term. Daniel demonstrated the highest score for vocalisations elicited during the free-play video recording. Minimal to no change was noted in babbling for this participant.

Ellie showed the most variability for vocalisations and babbling across all participants. She was also the only participant who babbled during the baseline assessments. Vocalisations decreased after the first term of intervention and did not rise again until the final term of the intervention. Similarly, a decrease in babbling was noted after the second term of the intervention but this again rose sharply after the final term and surpassed this participant's scores for vocalisations. Grace's scores for vocalisations were relatively high at baseline and were maintained across the time points. Grace demonstrated a large increase in the amount of babbling as she scored zero at baseline compared to 35 at post-intervention. Luke was the only participant who showed minimal change for both variables across all time points based on the five-minute video sample analysis.

4.2.3.2 Gesture.

Figure 4.3 illustrates the three types of gestures; deictic, conventional and iconic, produced by the participants over the intervention period. Deictic gestures were defined as gestures such as pointing or showing in order to identify an object of interest. Conventional gestures were defined as actions that are understood across all contexts and have a universal meaning, for example waving. Iconic gestures were actions that convey the meaning of an object/person, for exampl action for sleeping.

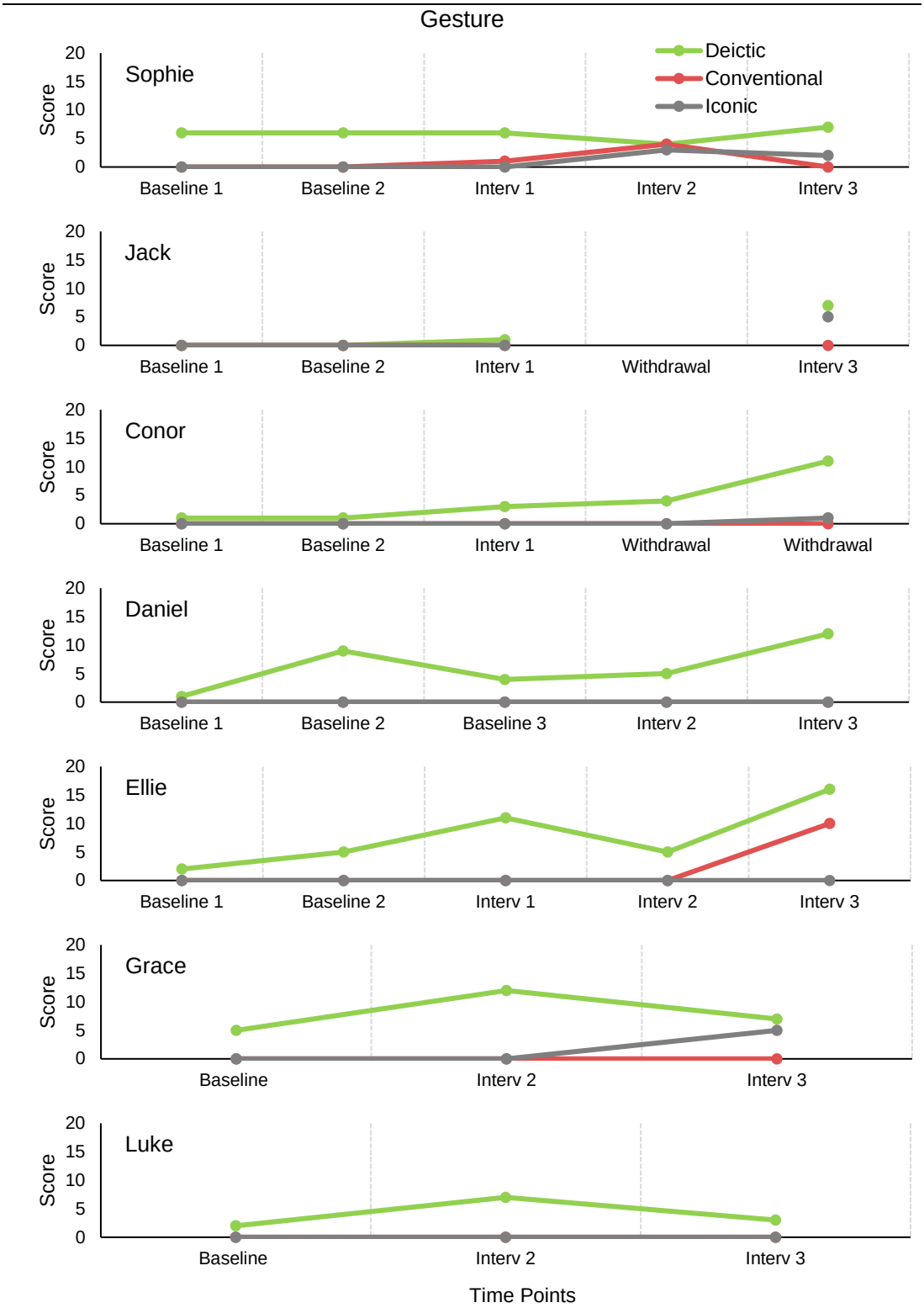

Figure 4.3. Scores for gestures (i.e. the number of times a gesture was recorded during the 5-minute video recording) for each participant. For Daniel and Luke, both conventional and iconic gestures sit on the same line as both received a score of 0 for both items. Note there is missing data for Jack. Interv = intervention; withdrawal = withdrawal from a term of intervention.

Considerable variability is also evident in terms of gesture use across the participants (see Figure 4.3). Firstly, Sophie demonstrated change across the three types of gestures. Sophie was the only participant who demonstrated a high and consistent use of deictic gestures across baseline measures. This was maintained throughout the intervention terms. A slight decrease in the number of deictic gestures was noted after intervention two, however this is contrasted by the increase in conventional and iconic gestures evident at this time point. Conventional and iconic gestures were evident at the final intervention assessment, however, they were less frequent than the deictic gestures elicited by this participant. Jack on the other hand demonstrated no gesture use across the baseline sessions. One deictic gesture was evident after the first intervention term and this rose to a total of seven by the final term. Jack also produced five iconic gestures at the final assessment with no change in conventional gestures noted.

Conor demonstrated the use of two deictic gestures across the baseline measures. An increase for this variable was noted after the first term of intervention which was maintained and increased after he withdrew from the therapy. Minimal to no change was noted in conventional and iconic gestures for Conor across the time points. Daniel demonstrated increased variability for deictic gestures before he began the intervention. A steady increase was noted for this variable once he commenced the intervention with a total of 12 deictic gestures observed by the final term. No change was observed for conventional and iconic gestures across the intervention. Again, Ellie's scores for deictic gestures varied across the time points with a total of 16 of these gestures evident at the final assessment. She was also the participant who demonstrated the most change for this item. Ellie's use of conventional gestures spiked from zero to 10 after the final intervention with no change in iconic gestures noted. Finally, Grace and Luke demonstrated an increase in the number of deictic gestures produced after their first term of intervention which decreased after the final term of the intervention. An increase in the number of iconic gestures were evident for Grace at the final assessment.

4.2.3.3 Signs.

Figure 4.4 illustrates the two types of signs that were evident among the participants. Imitated signs were defined as a sign/attempt of a sign that was

produced within eight seconds of the parent modelling same. Prompted signs were coded when a child responded with a sign following a question or statement posed by their parent. Spontaneous sign use was not plotted as none of the seven participants showed evidence of this skill.

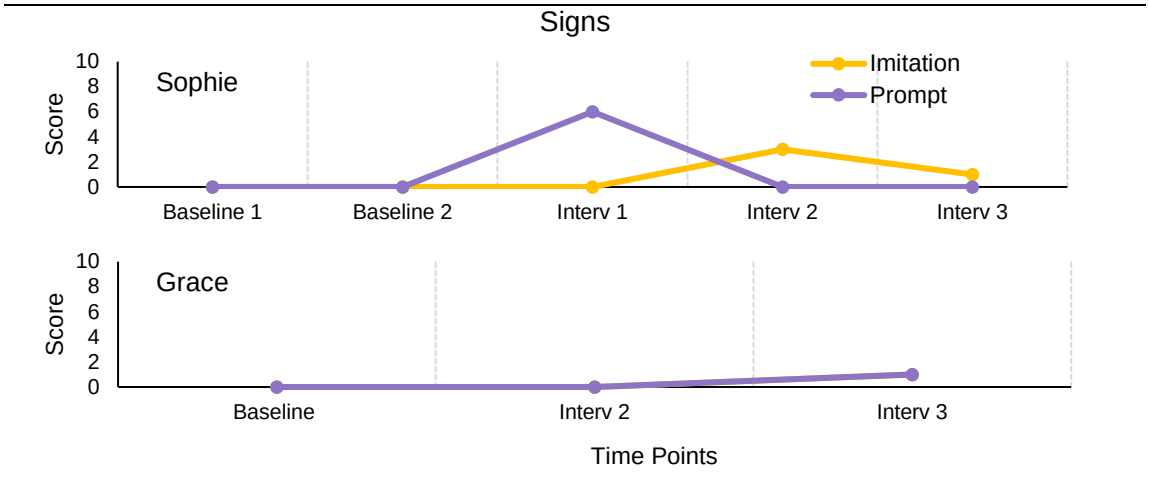

Figure 4.4. Scores for signs (i.e. the number of times a sign was recorded during the 5-minute video recording) for each participant. Imitated and prompted signs are plotted on the same line for Grace as she increased both items by a score of one after term three. Interv = intervention term.

Only two graphs were plotted for signs as only two participants (Sophie and Grace) demonstrated change over time (see Figure 4.4). The remaining five participants did not show evidence of any sign use in the videos across the time points and as a result no graphs were plotted for these participants. Sophie showed the greatest change in signs with a maximum of six prompted signs evident at intervention one and a maximum of three imitated signs evident at intervention two. Grace increased, both her prompted and imitated sign use, by one sign at the final intervention.

4.2.4 Summary

The Preschool Language Scales Fifth Edition (PLS-5) (Zimmerman et al., 2011) measured each child's receptive and expressive language skills. Although six of the seven children increased their SS scores for total language, considerable variability is evident within the participants which highlights the heterogeneous nature of the children in the current study.

Each child's receptive and expressive (signed and spoken) vocabulary was measured using the Vocabulary Checklist 1- First 120 Words (Down Syndrome

Education International, 2012) parental checklist. Based on this parental report, Grace was the only child who showed change across all 5 test items. Ellie showed change across 4 of the 5 items and Conor demonstrated change on 3 of the 5 items. Sophie and Jack showed change on 2 of the 5 items. Finally, Daniel and Luke showed gains on 1 of the 5 items. The words understood varied across the participants. The signs that were reported to be used by the children were mostly signs that were targeted in the PELD intervention programme.

In terms of pre-lexical verbalisations produced by the participants, four participants (Sophie, Jack, Ellie and Grace) were noted to increase their babbling by the end of the intervention. These four participants also showed a preference for vocalisations over babbling at baseline assessments. An interesting finding was noted for these participants as it was apparent that as the amount of babbling increased, the number of vocalisations decreased. Daniel was the only participant who demonstrated a sharp increase in vocalisations post-intervention with no change in babbling noted. Two participants (Conor and Luke) demonstrated minimal to no change for both vocalisations and babbling.

With regards to gestures, deictic gestures were the most common types of gestures to show change. Deictic gesture increased by a minimum of 1 and a maximum of 14 for the participants throughout the intervention. Interestingly, the two participants (Grace and Luke), who joined the programme later in the year, demonstrated an increase in deictic gesture after their first term of intervention and this decreased after their second term. However, the total number of deictic gestures for these participants did increase overall when compared to their baseline scores. For the five participants who had initially joined the study (Sophie, Jack, Conor, Daniel and Ellie), they increased their number of deictic gestures from baseline to post-intervention despite some fluctuations noted across the time points.

Conventional gestures showed the least amount of change following the intervention. Only two participants (Sophie and Ellie) demonstrated a change in conventional gestures after commencing the intervention. An increase of between four and 10 conventional gestures was evident for these participants. Interestingly, these were also the only two participants to complete all three terms of the intervention. A change in iconic gestures was noted for four of the participants (Sophie, Jack, Conor and Grace). These participants increased by a minimum of one and a maximum of five points following the intervention.

Finally, with regard to the use of sign, change was noted for prompted and imitated signs for two of the participants (Sophie and Grace) as per the video recording analysis.

4.3 Research Question 2: Does the PELD intervention programme affect the communicative interaction of infants with DS?

The second research question explored the change in the communicative interactions of the children across the different time points. A parent and child free-play activity was recorded and rated for the seven pivotal behaviours outlined in the Pivotal Behavior Rating Scale (Mahoney & Wheeden, 1998). The seven behaviours were then grouped into *attention* and *initiation*. These two variables are presented in Figure 4.5. The video recording was collected at the two initial baseline assessments (Baseline 1 and Baseline 2), after the first term of intervention (Interv 1), after the second term of intervention (Interv 2) and again after the third intervention term (Interv 3) for the first five the participants. Data for one of these initial five participants (Jack), was not collected after intervention two due to the personal circumstances of the family at the time. The final two participants (Grace and Luke), who joined the programme at a later date, had this measure collected at their initial baseline, after term two and after term three.

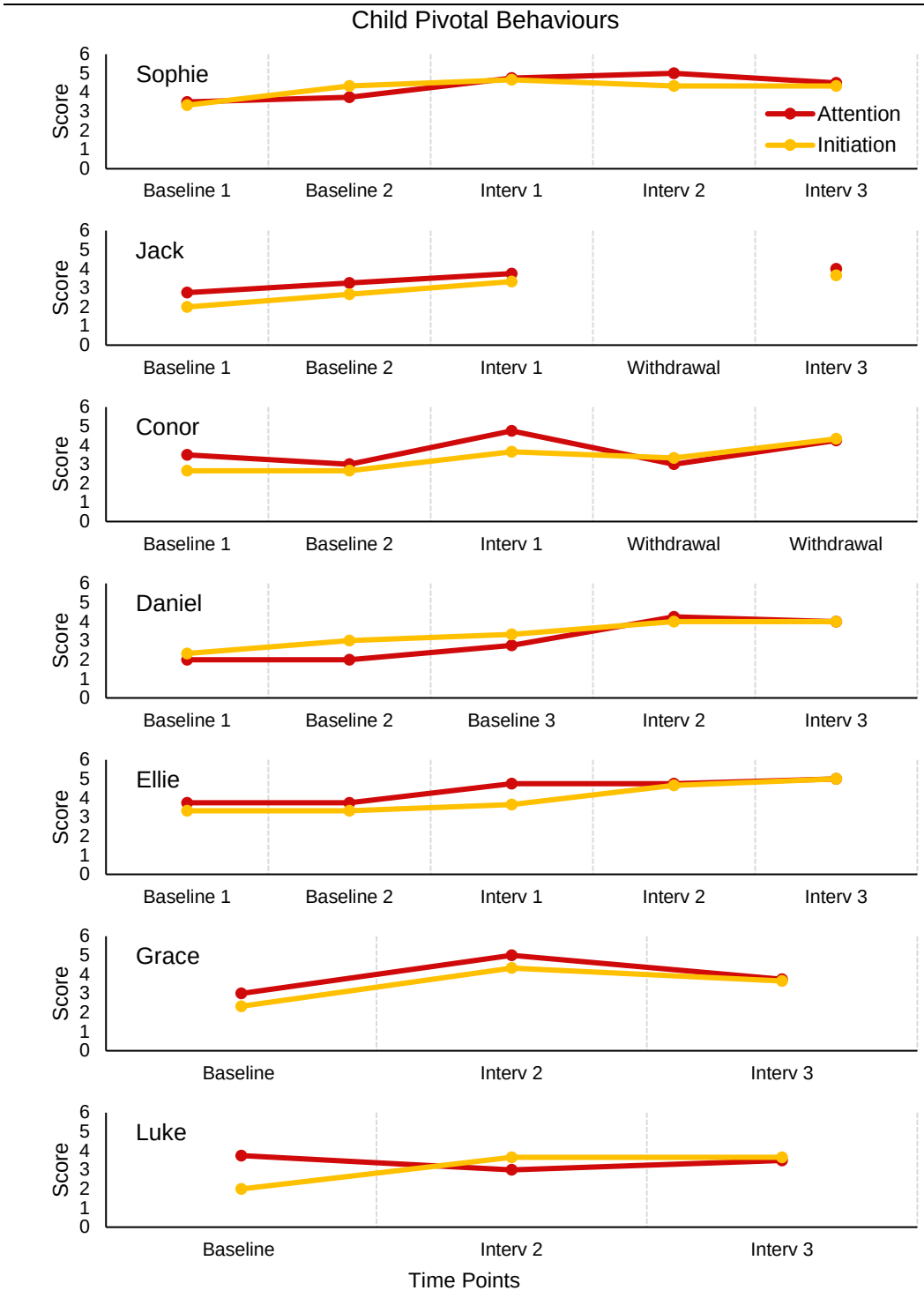

Figure 4.5. Attention and initiation scores for each participant. Note there is missing data for Jack. Grace and Luke started the intervention at Intervention 2. Interv = intervention; withdrawal = withdrawal from a term of intervention.

4.3.1 Attention.

Figure 4.5 presents scores for attention for each of the children across the time points. *Attention* described a child's overall ability to respond to joint attention

and combined scores for *attention to activity, persistence, involvement* and *cooperation.* Scores for these variables were summed and divided by the total number of variables. For the purpose of this study, this measure of attention will be discussed in terms of a child's ability to respond to joint attention. With the exception of Luke, all children demonstrated an increase in scores following the intervention. Sophie, Daniel and Ellie demonstrated an immediate increase following one term of the intervention and these scores remained high after additional terms of the PELD intervention were attended. A gradual increase was noted in Jack's score prior to the intervention and this continued to increase following the intervention. Conor's scores spiked after he completed one term of the intervention. A decrease was noted when this participant withdrew after term one. Grace showed a sharp increase for attention after one term of the intervention. A decrease was noted after the second term, however, her final score for attention was still greater than her baseline scores.

4.3.2 Initiation.

Figure 4.5 illustrates the *initiation* scores for each child across the time points. These scores were calculated by combining a child's score for *initiation adult, initiation activity* and *affect.* Scores for these variables were summed and divided by the three variables to produce an overall score for *initiation*. For the purpose of this study, the term initiation will be discussed in terms of a child's ability to initiate joint attention. All participants increased their scores for initiation when baseline scores are compared to end point scores. Despite all participants demonstrating an increase, this increase was not as substantial as the increase noted in attention for some of the participants. Three of the participants (Sophie, Jack and Daniel) scores showed an upward trajectory prior to the intervention and this continued to increase as the participants completed the intervention. Conor's scores for initiation increased by 1.75 after a term of the intervention. This participant withdrew from the intervention after this initial term and an increase in scores for initiation continued to increase despite therapy withdrawal. Ellie was the only participant who demonstrated an increase after a second term of intervention. Grace and Luke also increased their scores for initiation following a single term. Luke maintained these scores at the end point. A decrease in Grace's scores was noted

after the second term, however, her final score was still greater than her score at baseline.

4.3.3 Summary.

These findings suggest that the PELD intervention was effective in promoting the *attention* and *initiation* skills of the children. With the exception of Sophie and Daniel, all children demonstrated higher scores for attention over initiation at the baselines. At the final time point, five of the participants (Sophie, Conor, Daniel, Ellie and Grace) demonstrated similar scores for both attention and initiation. Jack's scores for attention were greater than his scores for initiation at the end point. Luke's scores for initiation were greater than his scores for attention at baseline.

4.4 Research Question 3: Does the PELD intervention programme change the way parents interact and communicate with their infants with DS?

The third and final research question explored the change in the way parents interact and communicate with their children with DS. This information was scored by analysing the participants' free-play video recordings. Observational measures were employed to rate the parent's interaction and communication skills. A language sample analysis was also conducted on the transcripts of the free-play parent-child activity.

4.4.1 Observational measures.

The five-minute video recording was reviewed and rated using a parent interaction and language rating scale. This rating scale was adapted from the Teacher Interaction and Language Rating Scale (Girolametto et al., 2000). Full description of the rating scale is outlined in the previous chapter (See Chp 3 pg. 56). The nine different items on the scale were rated for each participant across all time points and are presented in Figures 4.12-4.20.

4.4.1.1 Wait and listen.

Figure 4.12 presents each parent's change scores for this item across the five time points. *Wait and Listen* was scored based on the parent's ability to incorporate a time delay into interactions while also using a slow pace to allow the child to initiate.

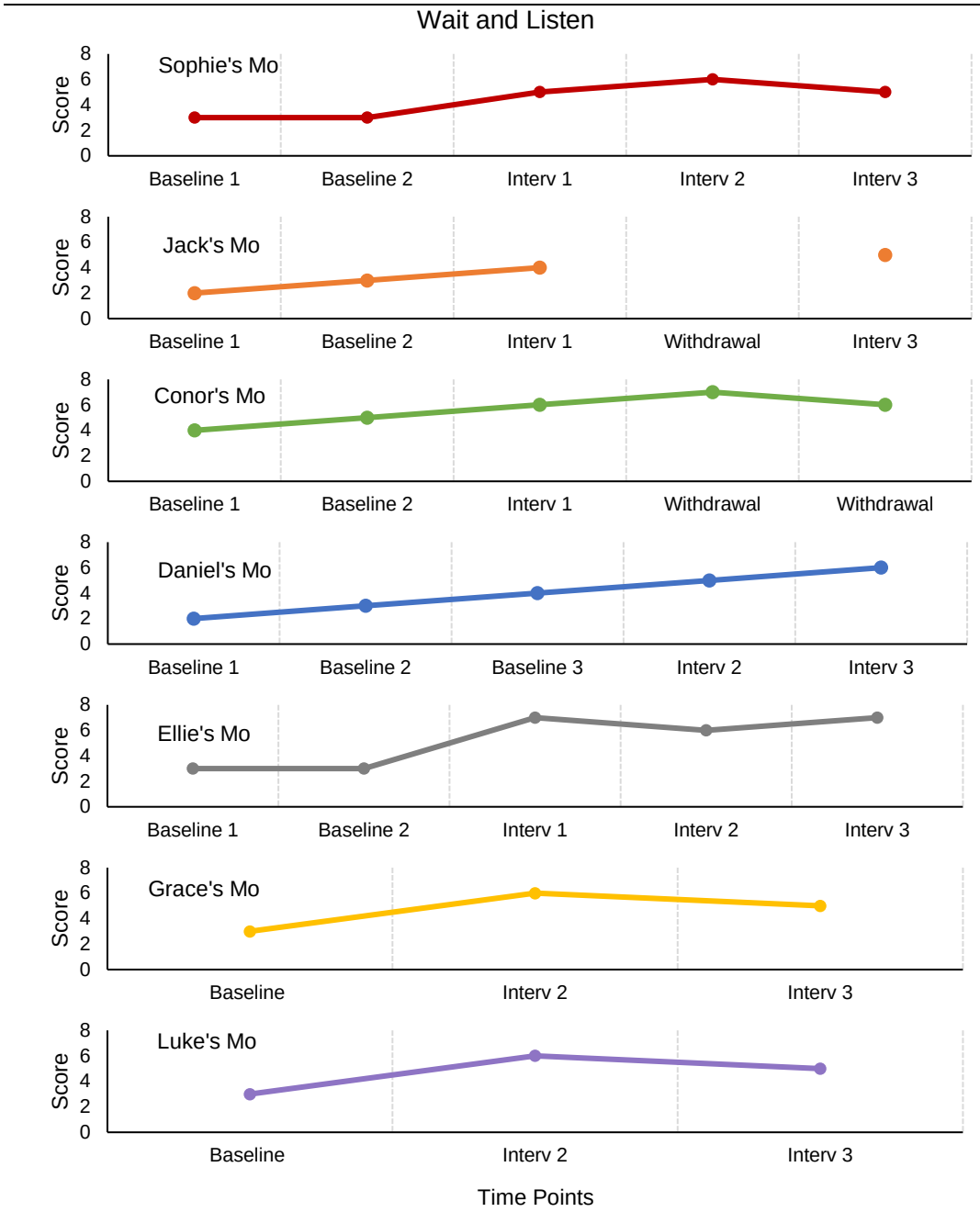

Figure 4.12. Scores for wait and listen for all parents of the participants across all time points. Interv = intervention term; withdrawal = withdrawal from a term of intervention; Mo = mother.

Both Sophie and Ellie's parents demonstrated a sharp increase in scores for this item after the first term of intervention and these scores were maintained for the duration of the intervention. Jack, Conor and Daniel's parents showed a steady

incline in scores during the baseline assessments with a continued increase noted after the participants completed one term of the intervention. Conor's parent demonstrated a slight decrease at the final assessment after they had not attended the intervention for two terms. Both Grace and Luke's parents increased their scores for this item once they started intervention with a slight decrease noted after the final term.

4.4.1.2 Following the child's lead.

Figure 4.13 presents scores for a parents ability to *follow their child's lead.* This measured and rated a parent's ability to respond verbally to their child's initiations or by being animated in response to the child's object of interest. Parents who achieved high scores avoided commands and vague responses to their child's initiations such as 'yeah', 'that's right', 'uh huh'.

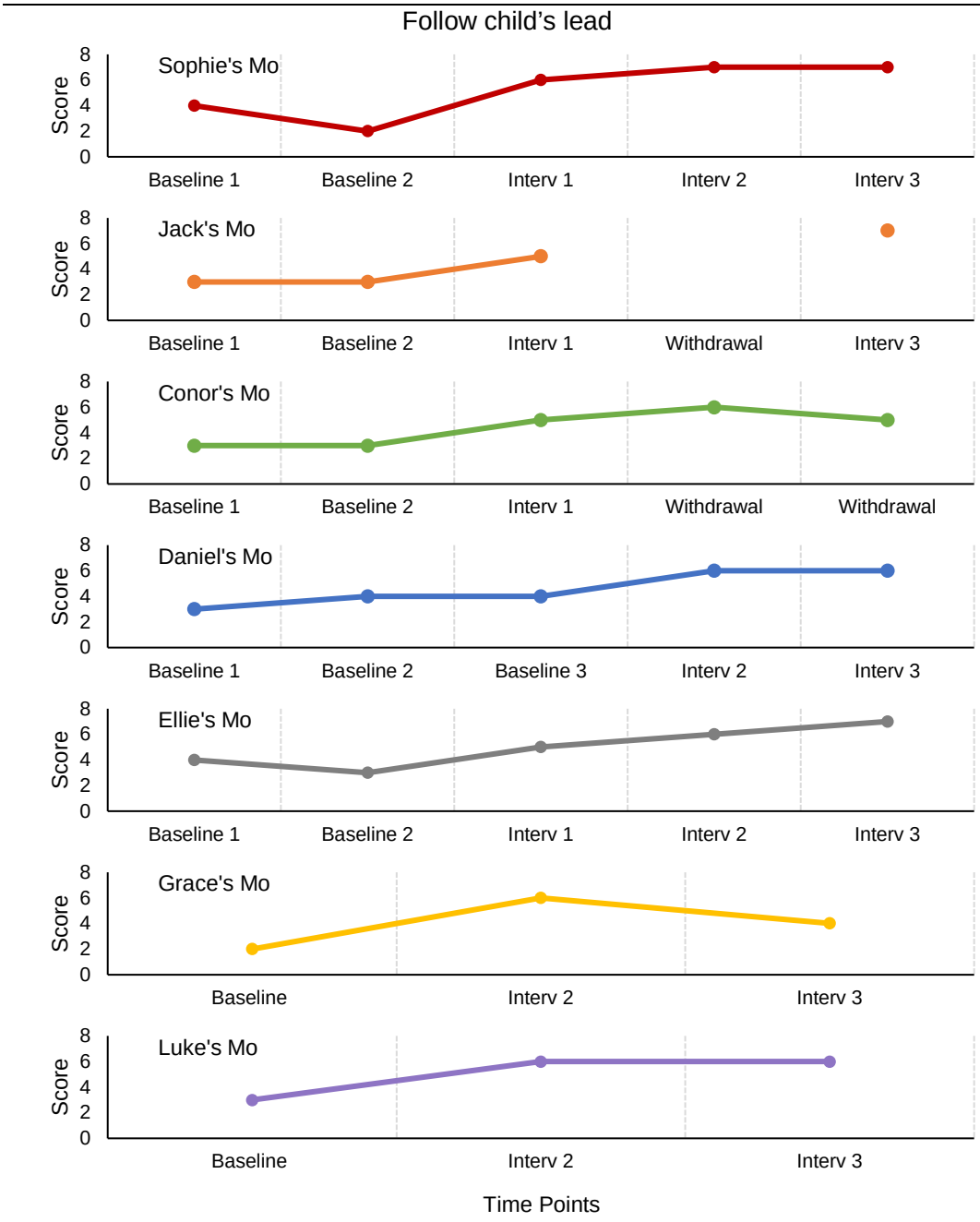

Figure 4.13. Scores for *follow child's lead* for all parents of the participants across all time points. Interv = intervention term; withdrawal = withdrawal from a term of intervention; Mo = mother.

As seen in Figure 4.13, all parents demonstrated an increase for this item when initial baseline scores and post-intervention scores were compared. All participants showed an increase of a minimum of 2 points and a maximum of 4 points once a family completed their first term of the intervention regardless of whether the family started the intervention at term one or term two. Sophie, Jack, Daniel, Ellie and Luke's parents maintained and/or increased their scores for this item after they attended the proceeding terms of the intervention. Interestingly,

Conor's parent increased her score at the fourth data collection point despite withdrawing from the therapy. A slight decrease was noted for this parent at post-intervention however these scores were still relatively high. Grace's parent also increased her scores by 4 points after the first intervention term. This decreased by 2 points post-intervention but again was still higher than the parent's initial baseline score.

4.4.1.3 Join in and play.

Figure 4.14 illustrates each parents scores for *join in and play.* This variable considers the number of times a parent actively joins in the child's play as a partner by building and expanding on the child's focus of interest and by playing without dominating the activity.

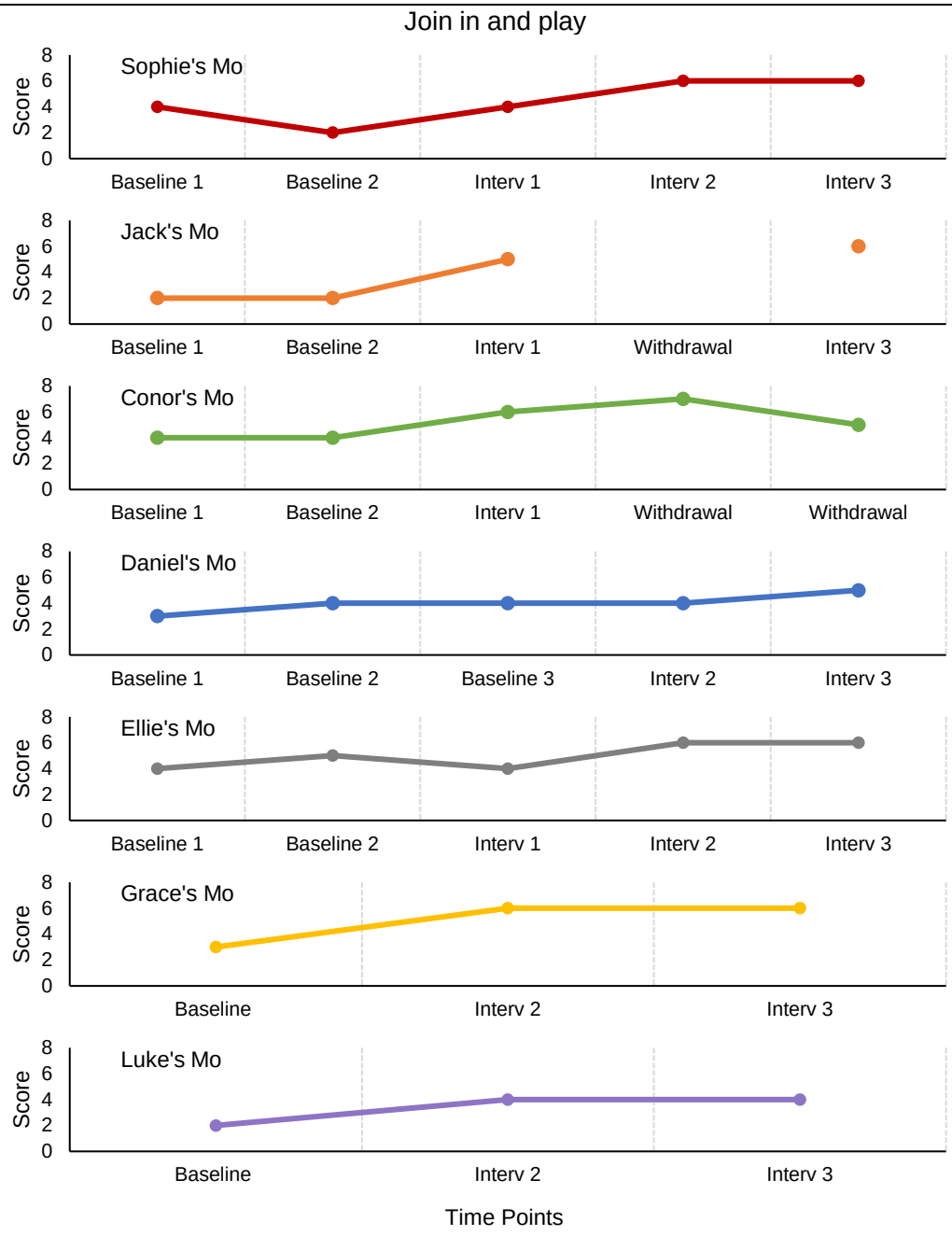

Figure 4.14. Scores for *join in and play* for all parents of the participants across all time points. Interv = intervention term; withdrawal = withdrawal from a term of intervention.

Visual inspection of Figure 4.14 shows the positive change for this item for all parents. All parents demonstrated an increase of between 1 and 4 points when first baseline and post-intervention scores were compared. Sophie, Jack, Conor, Grace and Luke's parents showed an immediate increase in scores following their first term of intervention. It was only after the second term of intervention that a change was noted for the parents of Daniel and Ellie on this measure. All

participants continued to maintain and/or increase these scores apart from Conor's parent, whose final score decreased at the post-intervention assessment.

4.4.1.4 Face to face interaction.

Scores for *face to face interaction* varied for each parent (see Figure 4.15). This measures the amount of times a parent adjusts their physical level by sitting on the floor facing the child or by leaning towards their child so that they are at the child's eye level.

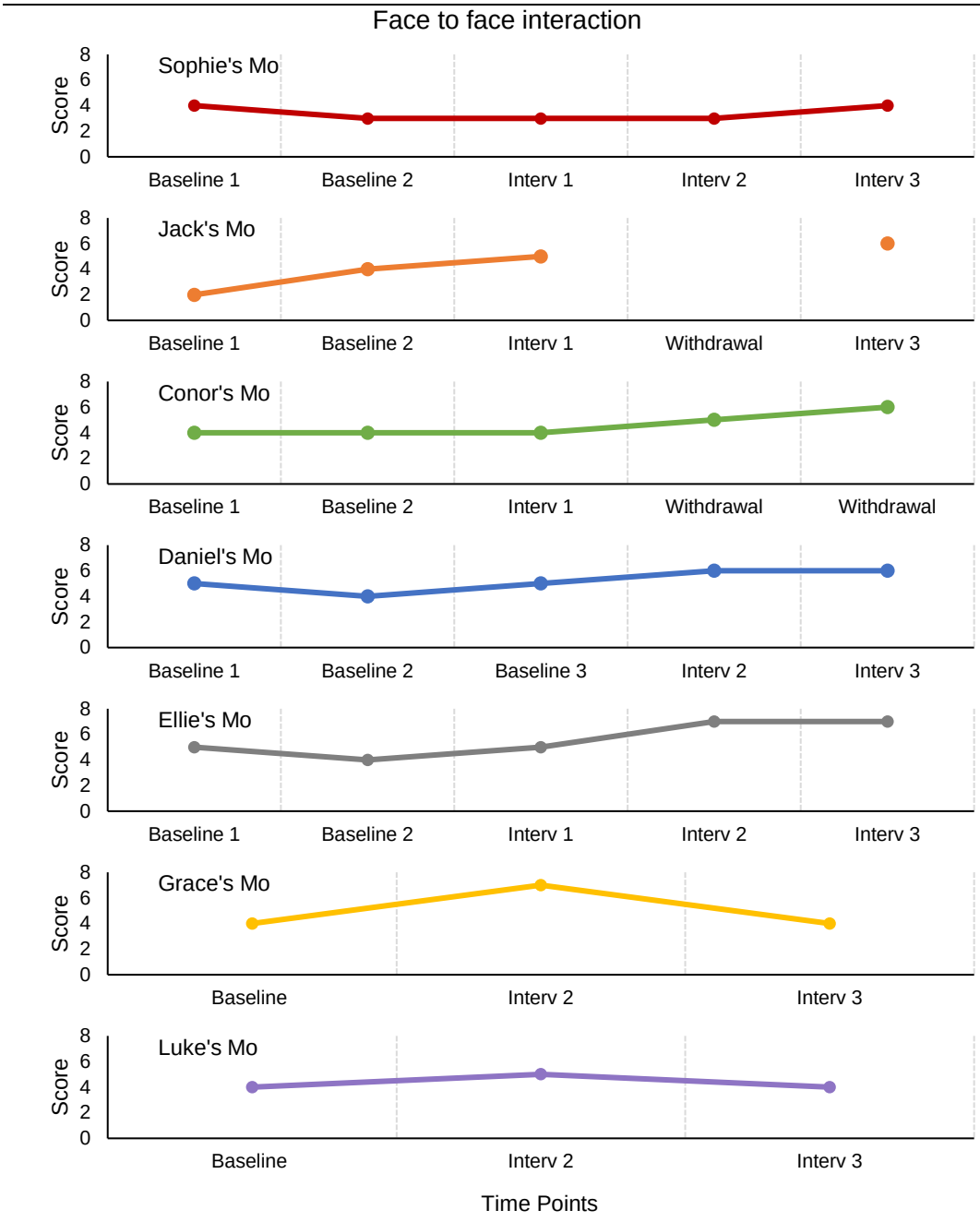

Figure 4.15. Scores for *face to face interaction* for all parents of the participants across all time points. Interv = intervention term; withdrawal = withdrawal from a term of intervention

Jack, Daniel and Ellie's parents improved this skill after the first term of intervention was completed. These improvements had been maintained and/or increased by the final intervention assessment. On the other hand, Sophie's parent did not show change until the final intervention term. It is important to note that this change was again minimal and matched the parents initial baseline score for this skill. No change was noted for Conor's parent's score after one term of intervention but an increase was noted after they withdrew from the intervention. Grace and Luke's parents showed an increase in scores after one term of intervention but regressed to their baseline scores after they completed their second term.

4.4.1.5 Encourage turn-taking.

Figure 4.16 presents parents scores' for *turn-taking* throughout the intervention. Parents encouraged turn-taking by balancing the number of child to adults terms, using animation when responding and linking comments/questions to invite the child to take a turn.

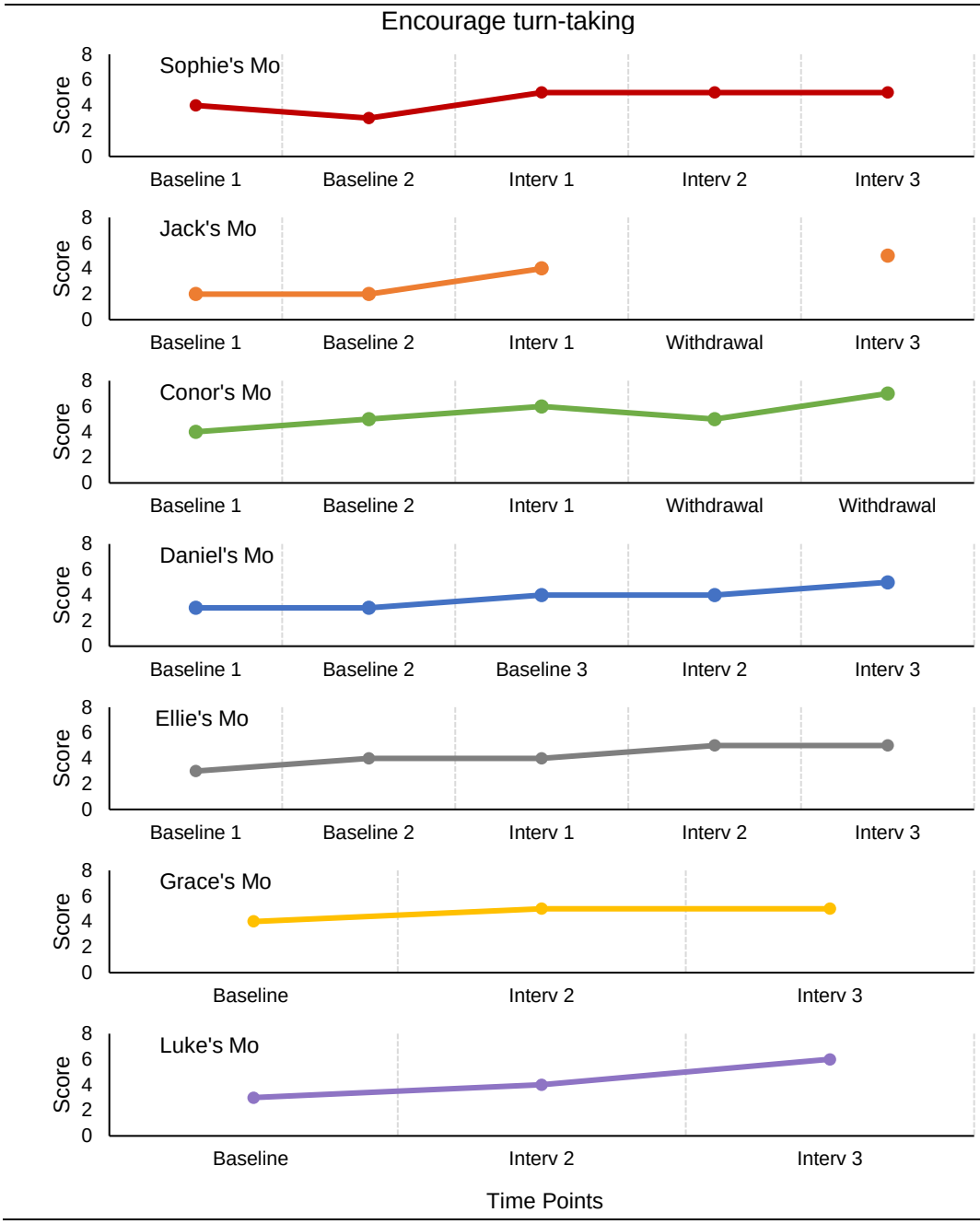

Figure 4.16. Scores for *turn-taking* for all parents of the participants across all time points. Interv = intervention term; withdrawal = withdrawal from a term of intervention; Mo = mother.

The above figure shows that scores for the parents' turn-taking encouragement improved across the time points for all participants. Positive change was evident after the first term of intervention for five of the participant's parents (Sophie, Jack, Conor, Grace and Luke). Whereas, Daniel's and Ellie's parents did not show change until after a second term of the intervention.

4.4.1.6 Gesture.

Figure 4.17 illustrates each parent's ability to use *gesture* to promote their communication and understanding. Parent gestures were scored every time a parent showed, gave, reached or pointed to convey an idea/meaning of a concept.

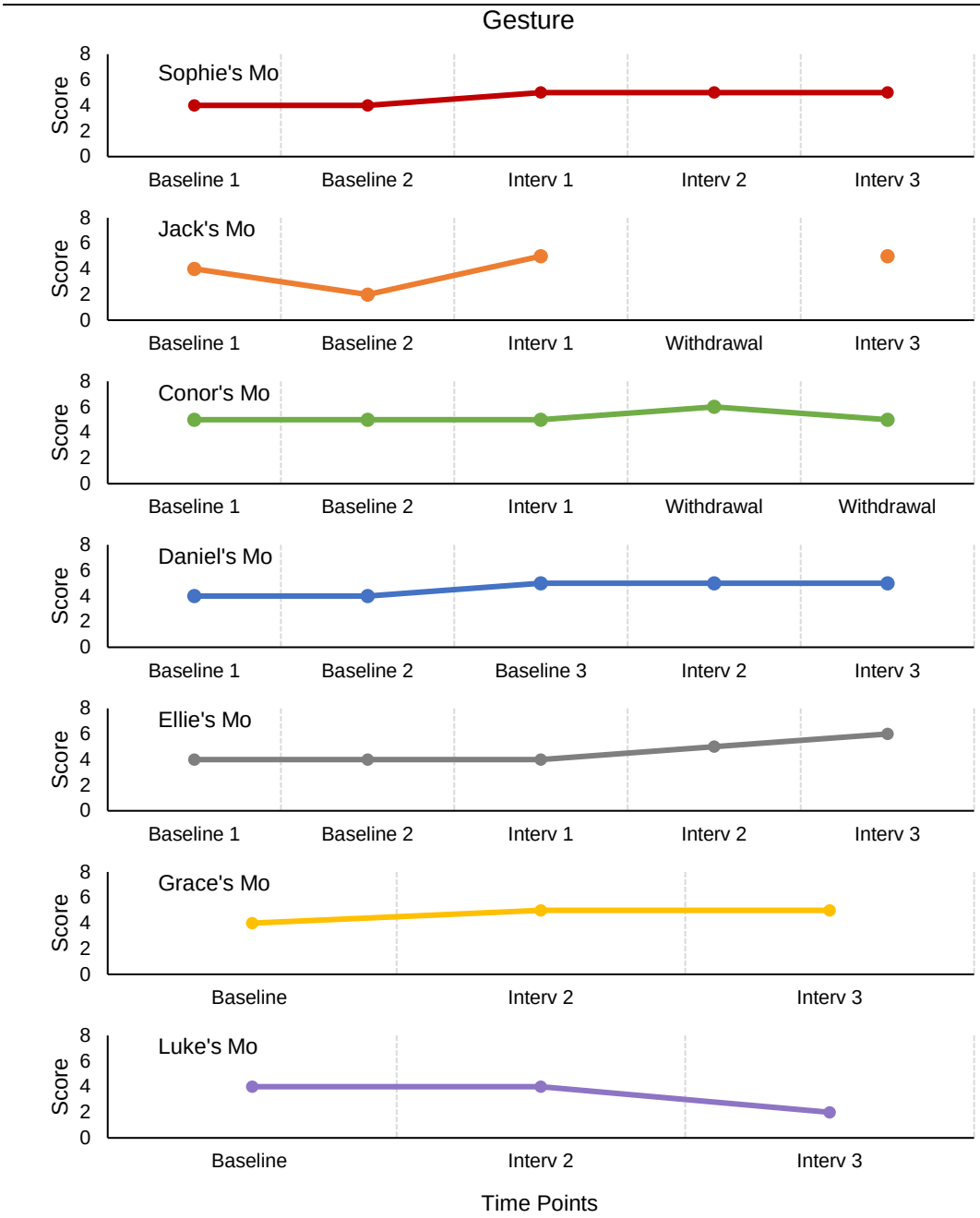

Figure 4.17. Scores *gesture* for all parents of the participants across all time points. Interv = intervention term; withdrawal = withdrawal from a term of intervention; Mo = mother.

As seen in Figure 4.17, parent's use of gesture varied with a range of scores presented on the graphs. The parents of Sophie, Jack and Grace all demonstrated an increase for this item after one term of the intervention and maintained this

improvement at post-intervention assessment. Ellie's parent improved after the second term of intervention and this score increased again by the final intervention assessment. An improvement in gesture use for Conor's parent was only evident after they withdrew from the therapy. Daniel's parent showed change prior to commencing the intervention and maintained this score as the intervention continued. Interestingly, Luke's parent maintained her baseline scores after one term and then regressed by two points (out of seven) after the second term of the intervention.

4.4.1.7 Lámh.

Figure 4.18 displays scores for each parent's use of Lámh signs. This variable measured the number of times a parents spoke and signed, signed alone or used hand over hand to promote communication with the child.

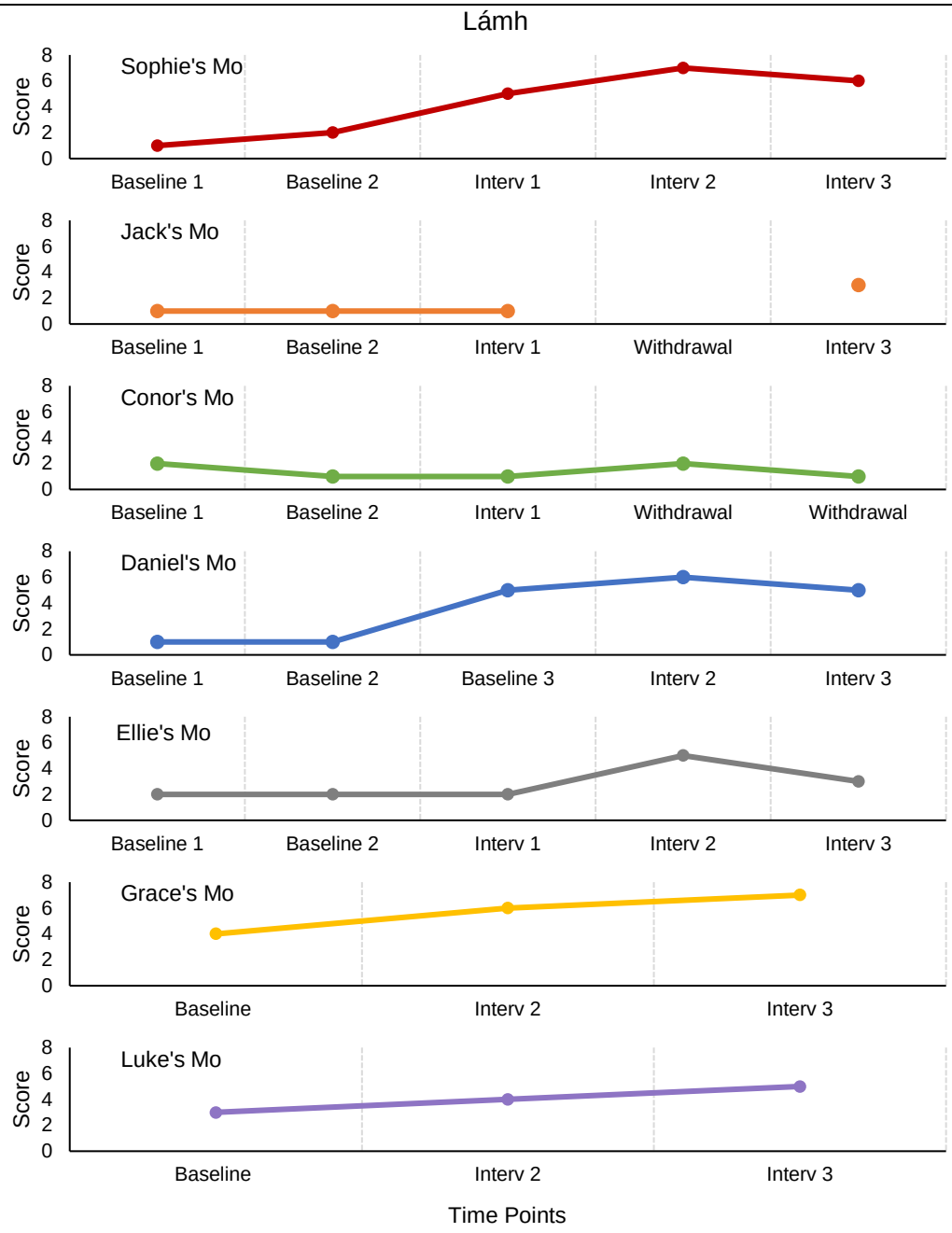

Figure 4.18. Scores *Lámh* for all parents of the participants across all time points. Interv = intervention term; withdrawal = withdrawal from a term of intervention; Mo = mother.

With the exception of Conor's parent, the use of Lámh increased for all parents by the time the final assessments were completed (see Figure 4.18). Over half of the parents (parents of Sophie, Daniel, Grace and Luke) showed an increase after one term of intervention. Daniel's parent demonstrated a great increase at baseline 3 before completing a term of the intervention. This continued to rise after commencing the intervention. The parents of Jack and Ellie increased their scores after a second term of intervention although a decline in scores was noted for Ellie's

parent by the time the final assessments were completed. Minimal change was noted for Conor's parent's use of Lámh with this parent's score remaining low throughout the time points.

4.4.1.8 Imitation.

Figure 4.19 presents parents' scores for *imitation*. This measured the number of times a parent mimicked the actions, gestures, sounds, facial expressions and/or words produced by their child.

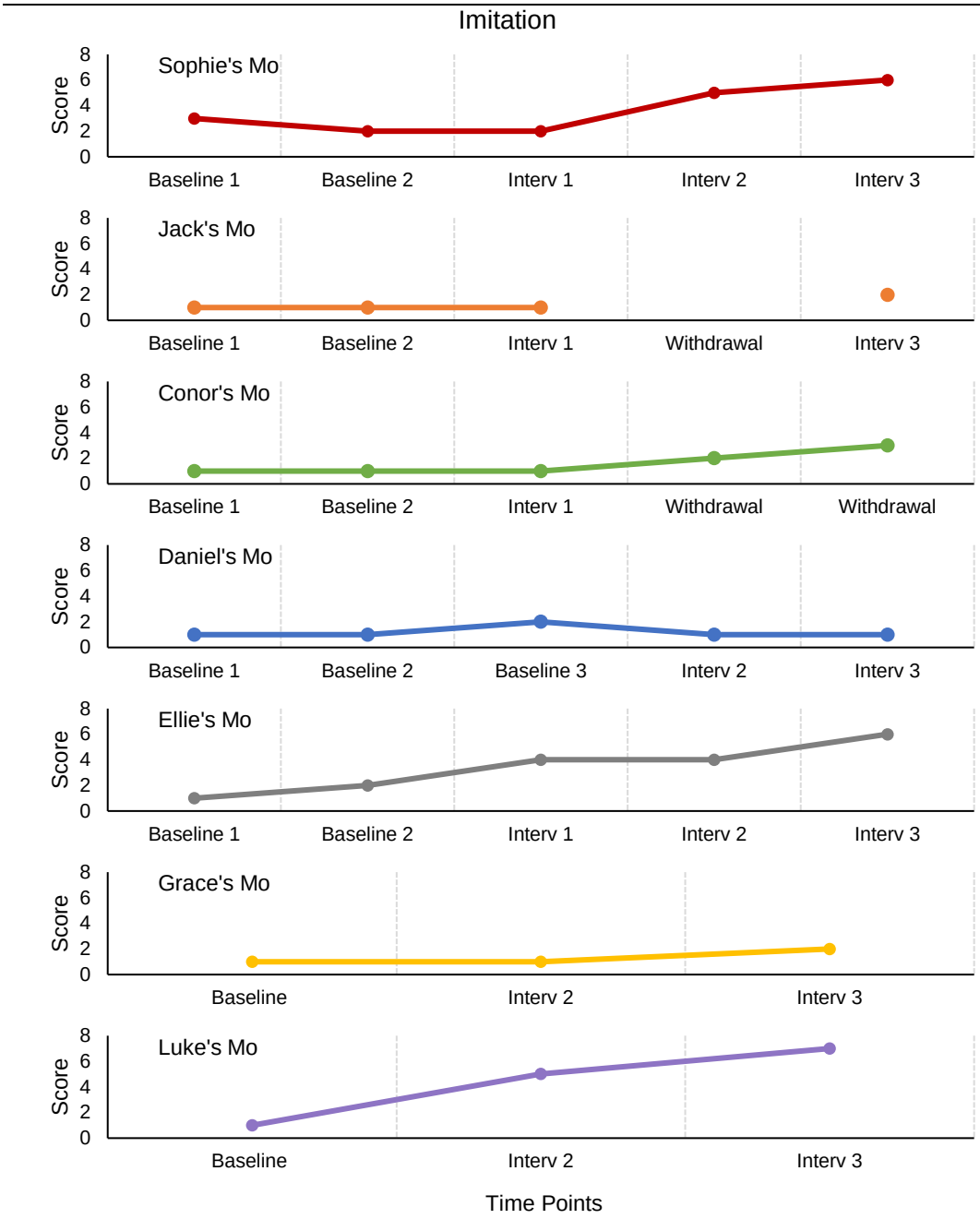

Figure 4.19. Scores for *imitation* for all parents of the participants across all time points. Interv = intervention term; withdrawal = withdrawal from a term of intervention; Mo = mother.

Visual inspection of Figure 4.19 shows a general improvement in the parent's ability to imitate throughout a free-play activity with their child. The parents of Sophie, Ellie and Luke demonstrated the greatest increase in scores for imitation at post-intervention with an increase in scores of three and six noted for these participants. Jack and Grace's parent's also demonstrated some improvement after the intervention although this increase was not as substantial as the other participants. Conor's parent improved after they withdrew from the intervention while Daniel's parent surprisingly regressed after they commenced their first term of intervention.

4.4.1.9 Variety of labels.

Figure 4.20 illustrates scores for parents' use of a *variety of labels.* This variable captures the variety of vocabulary used by the parents throughout the video. Parents who scored high for this variable used a range of word types (nouns, verbs, adjectives), repeated and emphasised key words and avoided the use of non-specific words such as 'it', 'that' and 'this'.

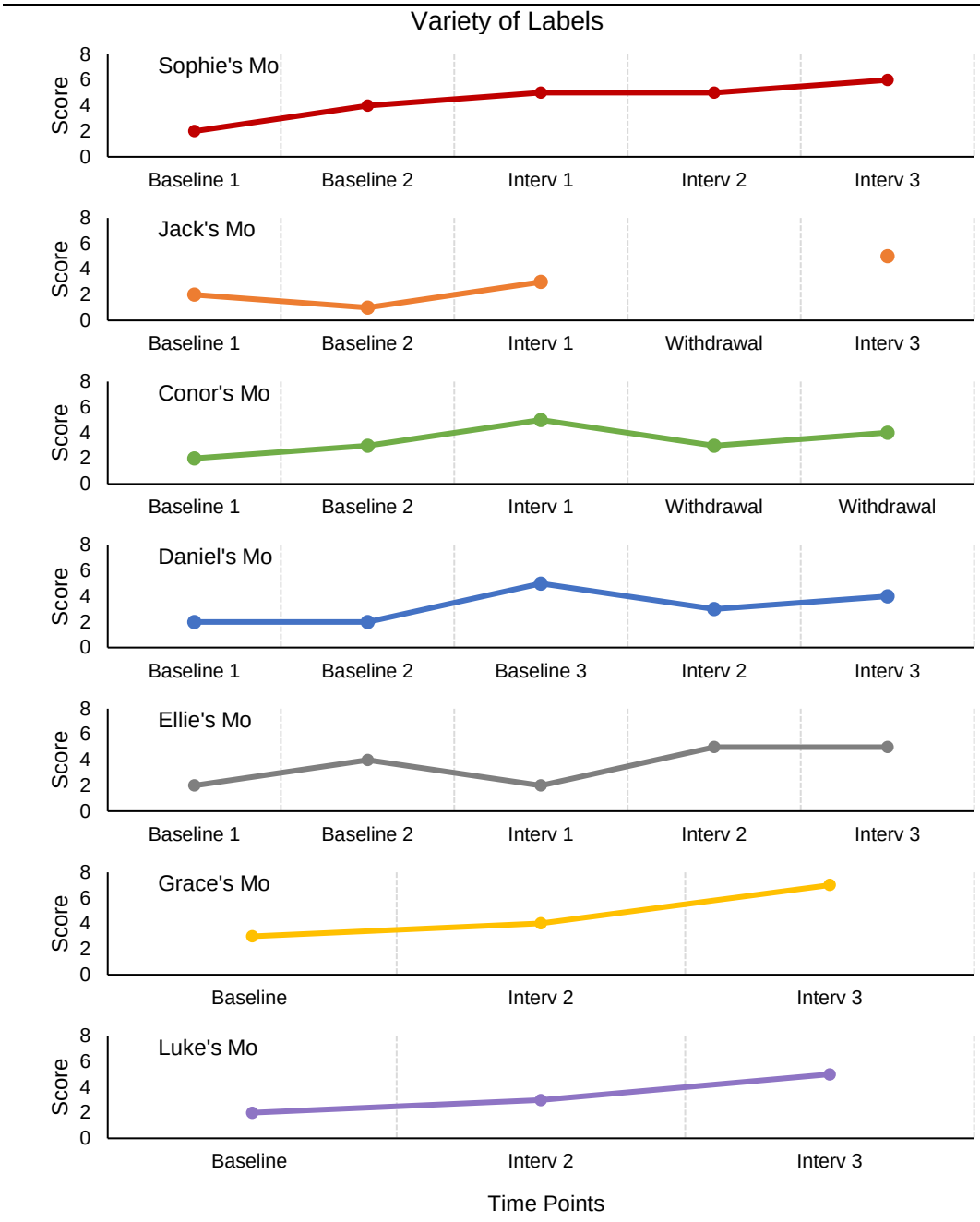

Figure 4.20. Scores for *variety of labels* for all parents of the participants across all time points. Interv = intervention term; withdrawal = withdrawal from a term of intervention; Mo = mother.

Visual inspection of the graph shows that for most parents (parents of Sophie, Jack, Ellie, Grace and Luke) the intervention promoted the use of a variety of labelling. Conor's parent increased scores for this item after one term of intervention but these scores were not maintained following their withdrawal from the programme. On the other hand, Daniel's parent's score peaked at baseline 3 and regressed after they started the intervention. Although these scores declined, they were still higher than their initial baseline scores.

4.4.1.10 Summary.

To summarise, the Figures 4.12-4.20 show that the PELD intervention programme was successful in promoting the use of some parent interaction and communication strategies although variation was evident across the variables for all parents. Three targeted areas from the parental outcome measures*; wait and listen, following the child's lead, join in and play* increased for all participants post-intervention. The remaining six items varied across the parents. Some parents demonstrated an increase for these variables while other parents showed no change.

4.4.2 Language sample analysis.

A language sample analysis was conducted on five minutes of a free-play activity between the child and parent. Scores for total utterances, MLU utterances, MLU words, MLU morphemes, frequency types and frequency tokens were generated using the KIDEVAL programme from the CLAN software (see Chp 3 pg. 60-61 for coding details). Values for type-token ratio (TTR) were generated, however, they were not reported as the single values for types and tokens provided more information for the parents language use than that of TTR since TTR is usually used with children and not adults. There were minimal differences between total utterances/MLU utterances and between MLU words/MLU morphemes. Therefore, only scores for total number of utterances and MLU morphemes were reported as they are more sensitive to language change. Figures 4.21-4.25 illustrate the findings for total utterances, MLU morphemes, frequency types and frequency tokens. Table 4.2 summarises the findings for all parents in the study and presents average baseline scores (pre-intervention scores) and post-intervention scores for the same four variables.

4.4.2.1 Total utterances.

Figure 4.21 presents the *total utterances* produced by parents during the five minute video recording throughout the intervention. Total utterances were defined as a unit of speech followed by a pause of one second, has a complete grammatical structure and/or ends with terminal intonation (Bernstein Ratner, Brundage & Fromm, 2016).

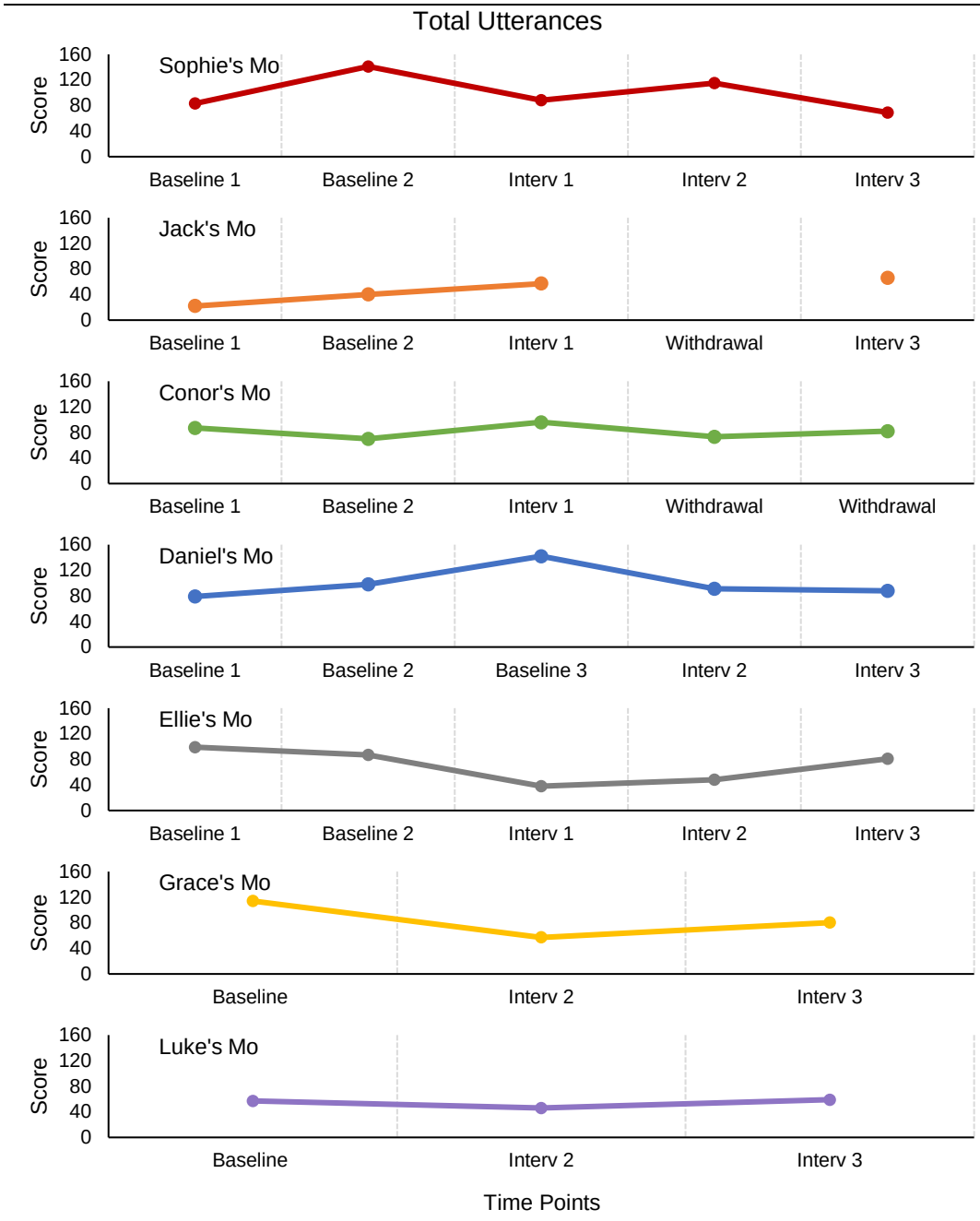

Figure 4.21. Scores for *total utterances* for all parents of the participants across all time points. Interv = intervention term; withdrawal = withdrawal from a term of intervention; Mo = mother.

Scores for total utterances showed substantial variation across the participants. Once scores at baseline were averaged, a huge variation of scores was noted with some parents producing an minimum of 31 utterances at baseline and others producing a maximum of 114 utterances at baseline. As seen in Figure 4.21, parents who used a high number of utterances at baseline decreased their number of utterances post-intervention. Conversely, parents who used minimal utterances at baseline (i.e. Jack's parent) increased in the number of utterances produced by the

final assessment. At the final data collection point, all parents produced between 59-88 utterances.

4.4.2.2 MLU morphemes.

Figure 4.22 presents scores for *MLU morphemes* produced by the parents. Scores for *MLU morphemes* were reported as opposed to MLU words as these scores were more sensitive to language change of the parents. *MLU morphemes* measures the number of morphemes per utterance used by the parents (Bernstein Ratner et al., 2016).

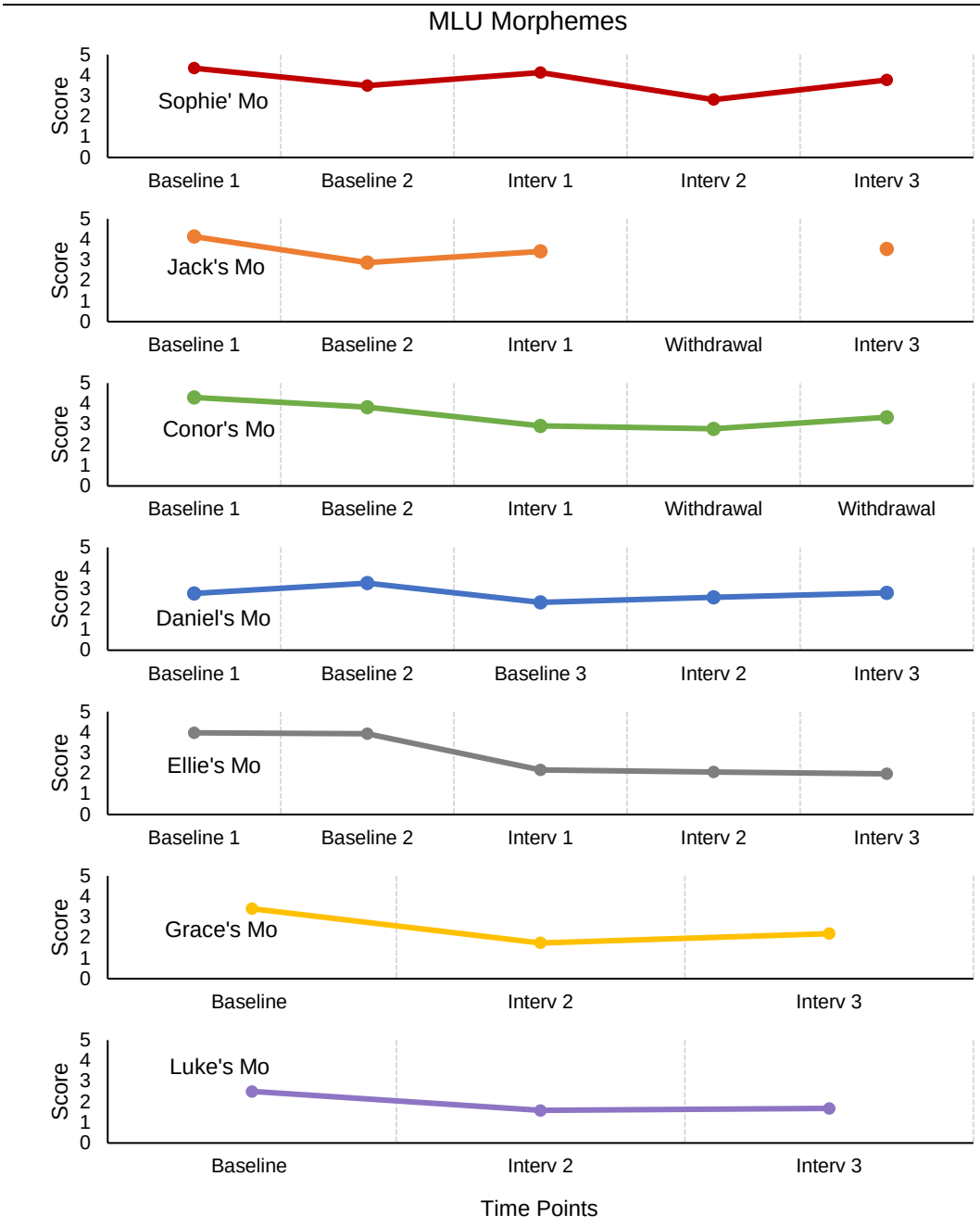

Figure 4.22. Scores for *MLU morphemes* for all parents of the participants across all time points. Interv = intervention term; withdrawal = withdrawal from a term of intervention; Mo = mother.

As seen in Figure 4.22, scores for *MLU morphemes* had generally decreased for all parents by the end of the intervention. Average baseline scores for MLUm were high and ranged from between 2.51-4.06. This decreased to a range of 1.68-3.77 by the end of the final intervention term. The parents of Sophie, Jack and Conor scored the highest for MLU morphemes post-intervention with scores of 3.34 and above noted for this measure. Sophie's parent's scores were consistently high throughout the intervention with a slight decrease noted after the second term of the

intervention. Jack and Conor's parents, who withdrew from certain therapy terms (Jack's parents did not attend intervention term two/Conor's parent did not attend intervention term two and three), also demonstrated a high score for MLU morphemes at post-intervention. The remaining four parents (parents of Daniel, Ellie, Grace and Luke) presented with low scores for MLU morphemes with a maximum of 2.80 noted for these parents.

4.4.2.3 Frequency types.

Figure 4.23 illustrates scores for *frequency types* used by the parents throughout the intervention. Frequency types are the number of different words used by the parent in the language sample analysis (Bernstein Ratner et al., 2016).

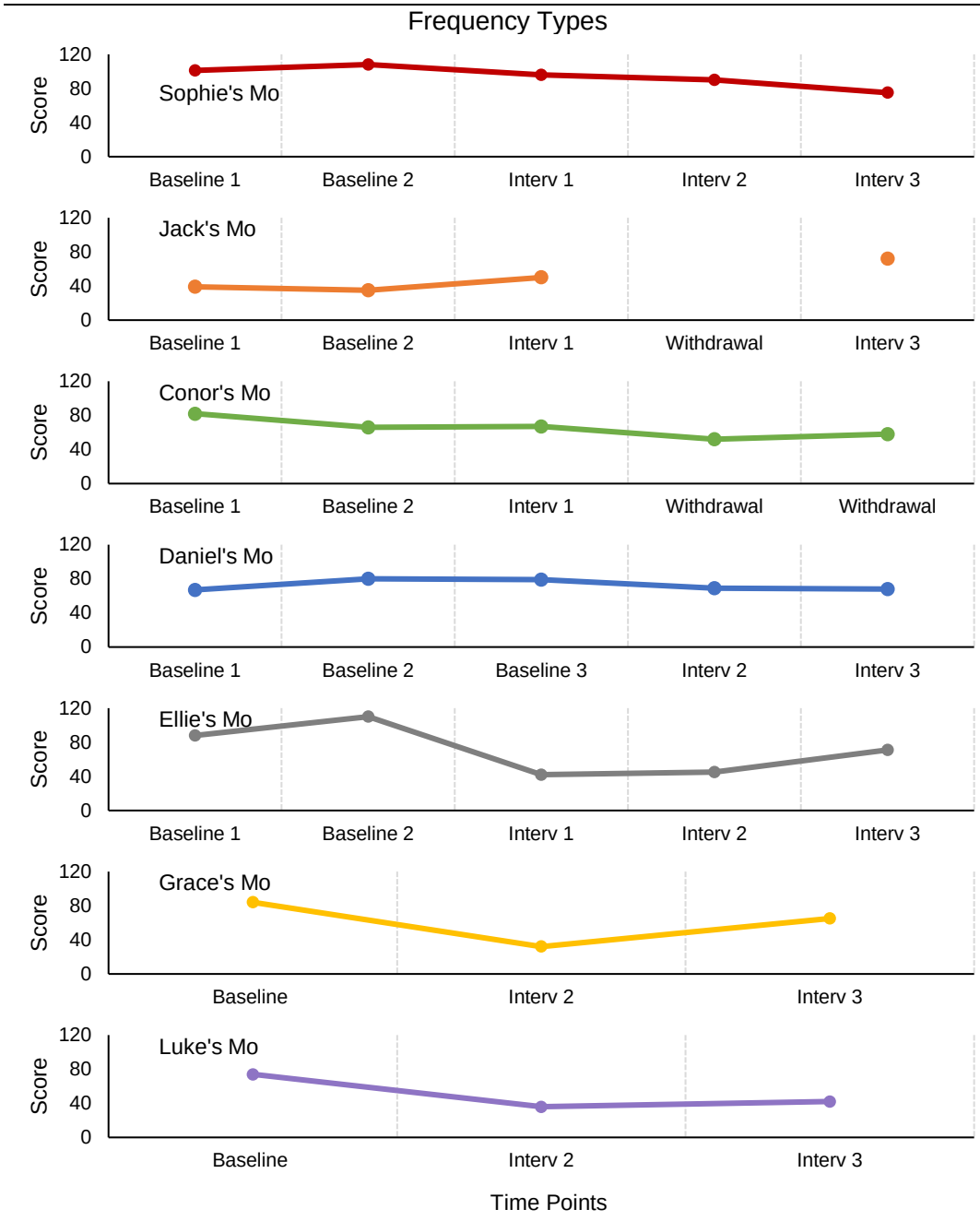

Figure 4.23. Scores for *frequency types* (total number of different types of words) for all parents of the participants across all time points. Interv = intervention term; withdrawal = withdrawal from a term of intervention; Mo = mother.

Average baseline scores for all parents ranged from 37 different words to 105 different words per sample (see Figure 4.23). This range decreased at post-intervention assessment with the number of different words used by the parents ranging from 42 words to 75 words. In general, the parents who had higher scores for frequency types at baseline (Sophie, Conor, Daniel, Ellie, Grace and Luke's parents) decreased their numbers for this following intervention. Contrastively,

Jack's parent who used a relatively low number of different words at baseline, increased this by the final intervention point.

4.4.2.4 Frequency tokens.

Figure 4.24 presents scores for frequency tokens used by the parents throughout the intervention. Frequency tokens are the total number of words used by the parent in the language sample analysis (Bernstein Ratner et al., 2016).

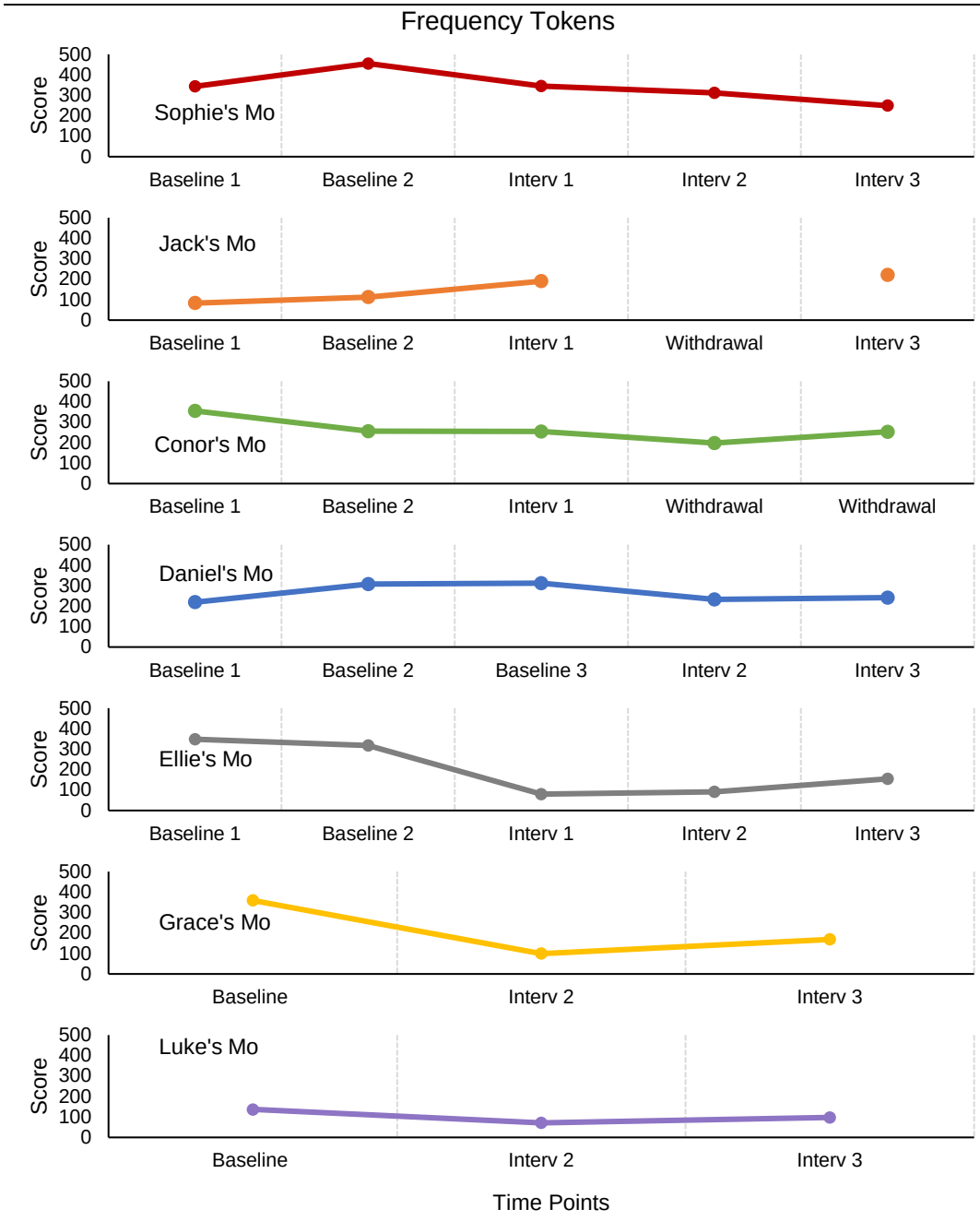

Figure 4.24. Scores for *frequency tokens* (total number of words) for all parents of the participants across all time points. Interv = intervention term; withdrawal = withdrawal from a term of intervention; Mo = mother.

In general, the number of words used by the parents decreased after the intervention with the exception of Jack's parent who increased after intervention (see Figure 4.24). It is important to note that this parent used notably fewer words than other parents at baseline assessment. The averaging of baseline scores demonstrated that the parent's number of words ranged from a minimum of 98 words to a maximum of 400 words which demonstrated a huge variation in the total words used

by the parents. At the post-intervention assessment, after each participant had received at least one term of intervention, this range decreased to a minimum of 97 words being used and a maximum of 253 words used at post-intervention.

4.4.2.5 Summary

Table 4.2. presents a summary of pre- and post-intervention scores for the parental language analysis.

Table 4.2
Summary of pre- and post- scores for language sample analysis

	Utt		MLUm		Types		Tokens	
	Pre	Post	Pre	Post	Pre	Post	Pre	Post
Sophie	112	69	3.91	3.77	105	75	400	249
Jack	31	66	3.50	3.53	37	72	98	221
Conor	79	82	4.06	3.34	74	58	306	253
Daniel	106	88	2.78	2.80	75	68	280	241
Ellie	93	81	3.94	1.98	99	71	333	155
Grace	114	80	3.40	2.19	84	65	359	169
Luke	57	59	2.51	1.68	74	42	137	97

Note. Pre-intervention scores were calculated by averaging the baseline scores for the participants. Utt = total utterances; MLUm = mean length of utterance in morphemes

To summarise, visual inspection of the table shows that four of the seven parents decreased their number of utterances post-intervention. It should also be noted that two of the three parents who increased their total utterances did so by a minimal amount (Luke and Conor's parents). With the exception of Jack's parent and Daniel's parent, all other parents decreased their scores for MLU morphemes. Finally, all parents, with the exception of Jack's parent, decreased their use of frequency types and frequency tokens when average baseline scores are compared to post-intervention scores.

4.4.3 Parental Stress Rating.

Figure 4.25 presents scores for stress reported by the parents. Throughout the intervention, the parents of the children were asked to give a numerical number for stress that they felt in that specific moment in time. A score of one meant the parents felt no stress parenting a child with DS and a score of ten meant the parents felt extremely stressed.

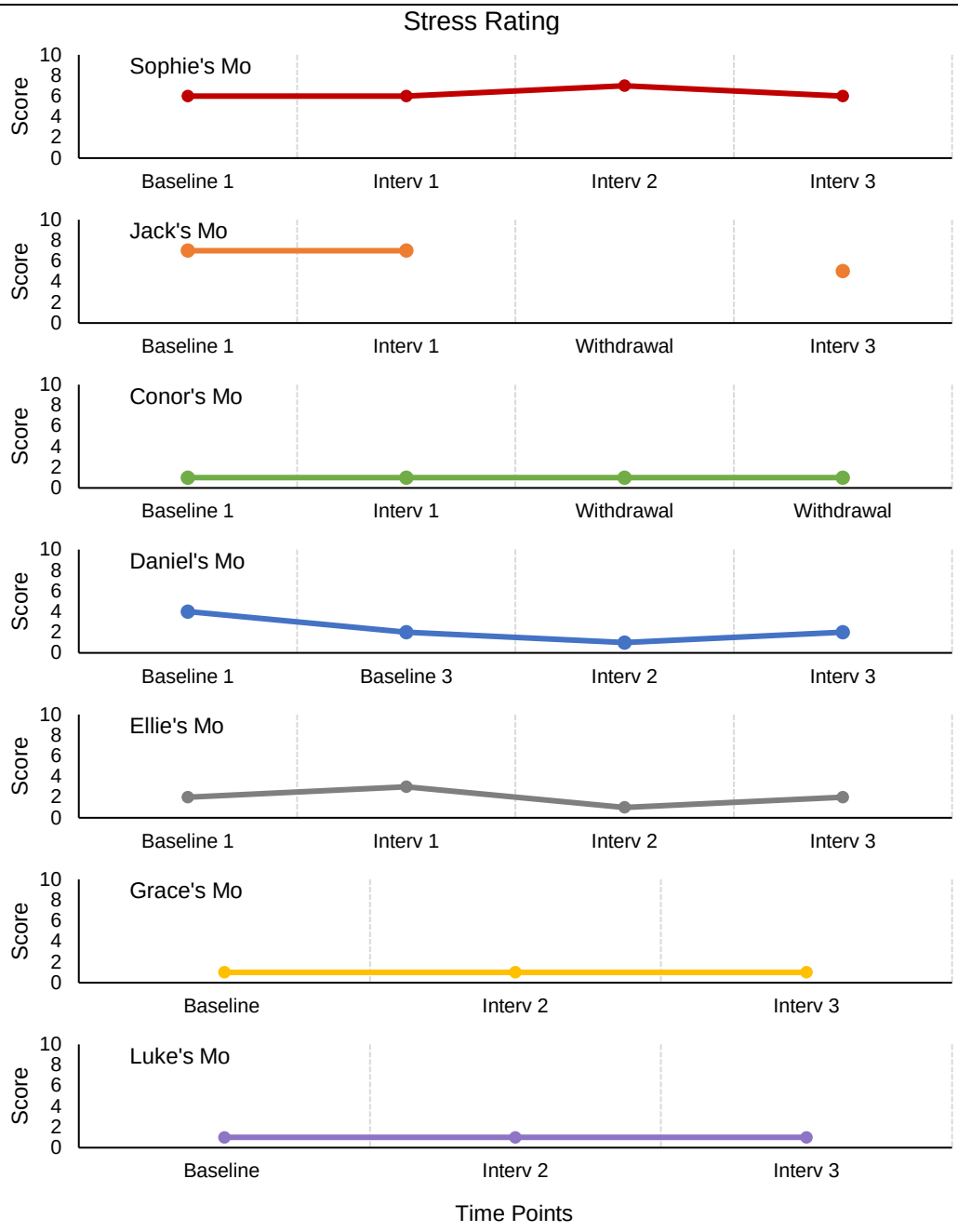

Figure 4.25. Numerical values for *stress* reported by the parents across s all time points. Interv = intervention term; withdrawal = withdrawal from a term of intervention; Mo = mother. Score of 1 = no stress; score of 10 = extremely stressed

Visual inspection of the graph shows that parental stress did not change throughout the intervention with all parents giving similar scores across the times points. Parents who gave higher scores for stress, reported that this was not directly related to their child with DS but was instead associated with the stress of trying to coordinate the numerous appointments that their child with DS is required to attend.

Chapter Five: Discussion

5.1 Introduction

The present study aimed to evaluate the effectiveness of a parent-child interaction therapy across a number of parent and child communication and interaction domains using a case series design. As this study adopted a single-case multiple-baseline design, each parent-child dyad acted as their own control. Following the analysis, there was some indication that this PCIT, known as the PELD programme, supported some aspects of child development and parental coaching although this was highly variable among the parent-child dyads with such gains also being noted on a variety of different outcome measures. Although there was considerable variation in the results for each dyad, patterns also emerged across the participants. The following chapter discusses the main findings from this study with respect to the three main research questions. Firstly, the results showed some indication that this PCIT promotes the development of non-verbal communication, particularly the use of gestures, sign, and receptive vocabulary in children with Down syndrome. Secondly, the findings suggest that the PELD programme may also encourage the development of communicative interactions of these children. Thirdly, there is some indication that the PELD intervention promoted change in parental behaviour in terms of communication and interaction skills with their children with DS. The results from this study also add to the current literature base with respect to the efficacy of a case series design to evaluate PCITs for DS given the heterogenous nature of the syndrome. For the purpose of this study, only variables that were targeted as the primary and secondary goals of the PELD intervention are discussed.

5.2 Early Language Development

The first question in this study explored whether the PELD intervention encouraged the language development of young children with DS aged 10-17 months. These results are discussed within the domains of receptive language and expressive communication.

5.2.1 Receptive language.

The first goal of this intervention was to promote the receptive language of the children in this study. As previously stated, receptive language abilities of children with DS in the early years are often in line with their mental age when compared to their expressive language abilities at the same age (Fidler, 2005; Miller 1999). Although receptive vocabulary is a relative strength for individuals with DS, expressive vocabulary development in the early years is often delayed and children with DS do not demonstrate a 'vocabulary explosion' that is evident in TD children (Abbeduto et al., 2007; Caselli et al., 1998). This study measured the children's receptive abilities using standardised testing and parental report.

5.2.1.1 Global measure of receptive communication.

Results from the standardised testing of the PLS-5 (Zimmerman et al., 2011) showed that with the exception of Jack, scale scores (SS) for receptive language increased for the remaining six participants. Three of these six children (Conor, Ellie and Grace) increased their SS scores by over 10 points immediately post-intervention. This increase in SS scores was much more substantial than the results presented by Wright and colleagues (2013) who explored the effectiveness of the EMT/JASPER Words and Signs intervention. Wright et al. (2013) also adopted the use of the PLS assessment in their research. Although two of their participants increased their SS scores by one standard deviation (SD) for receptive language they did not increase as notably as three of the participants in the current study who also increased by one SD, two of which demonstrated scores that were within normal limits as based on standardised testing. A possible explanation for this may be related to the fact that the children in the study by Wright et al. (2013) were older (age 23-29 months) than the participants in this study (age 10-17 months) at baseline. As previously mentioned, test items up to the age of two years on the PLS-5 assessment may be awarded if the parent/caregiver can provide an example of a time the child elicited a given behaviour. Considering the age of the children in the current study at baseline (age 10-17 months) and at post-intervention (age 18-23 months) this suggests that many of the items on the PLS-5 may have been awarded based on caregiver report. Therefore, the risk of parental bias cannot be ruled out considering all parents were aware that their child was receiving intervention. As the

children in the study by Wright et al. (2013) were older at baseline, they may not have achieved as high a score as parental report could not be accounted for on items past the 24-month mark, thus meaning the children in the study by Wright et al. (2013) had to elicit a given skill during the assessment session.

One unanticipated finding was noted in the post-intervention PLS-5 assessment scores of two of the participants. Ellie and Grace demonstrated receptive language scores that fell within normal limits at post-intervention, despite both participants presenting with mild receptive language difficulties pre-intervention. Although the results are preliminary in nature, these results are suggestive that the PELD intervention may promote the receptive abilities of young infants with DS to a point where they are similar to the early receptive abilities noted in TD children. Again, it is important to highlight that such items in the PLS-5 were based on parental report and results cannot be generalised to all receptive abilities of the children. Similarly, although the results are promising, the likelihood of such gains being maintained needs to be addressed. Often the receptive and expressive language difficulties noted in children with DS may not always be evident at 24 and 36 months (Roberts & Richmond, 2015) therefore follow-up visits would need to be conducted at these time points in order to see if the gains were maintained. Again, although the above findings were only noted in two of the participants, six of the seven children increased their scale scores post-intervention showing positive change for these children following the intervention. Huge variability was noted among the participants, thus highlighting the heterogeneity of the syndrome once more (Karmiloff-Smith et al., 2016).

5.2.1.2 Receptive vocabulary.

Receptive vocabulary was measured using an additional measure; The Vocabulary Checklist 1- First 120 Words (Down Syndrome Education International, 2012). As vocabulary comprehension is often intact in infants with DS (Abbeduto et al., 2007), this parental checklist offered an additional means of capturing change. Results from the checklist show that number of words understood by the children increased by a minimum of 31 words and a maximum of 72 words across the children. A rather interesting finding in relation to Sophie, who understood five words pre-intervention, increased to 77 words post-intervention as reported by her

parent. A potential explanation for this participant's lower score for word comprehension at baseline may be due to the fact that she is exposed to two languages in the home environment. Bilingualism and language acquisition have been widely researched with evidence showing that children who are learning two languages simultaneously often present with delayed receptive and expressive language abilities during the early years (Kohnert, 2010; Oller, Pearson & Cobo-Lewis, 2007). It can be hypothesised that Sophie's lower scores for vocabulary comprehension at baseline may be related to the fact that she was exposed to both English and an additional European language since birth. Although research within Down syndrome and bilingualism is limited, bilingualism in DS does not appear to impede on the development of children who present with language difficulties (Burgoyne, Duff, Nielsen, Ulicheva & Snowling, 2016; Cleave, Bird, Trudeau & Sutton, 2014). As the child's mother (who spoke English) was the parent taking part in the intervention, only the child's knowledge of English words were recorded. Vocabulary development in the second language was not recorded, therefore, it cannot be said for certain whether these gains were as a result of the intervention or as a result of maturation. This finding was also noted in relation to Grace's receptive vocabulary. Grace, who understood the least number of words at baseline (three words), reportedly understood 65 words post-intervention which showed she increased her word knowledge by 62. Although no measures for a second language were collected because at the time of the initial baseline, Grace's parent reported that Grace was only exposed to English, however as the intervention progressed it became apparent that the family had started to use more Irish with her.

On the other hand, children who were only exposed to one language throughout the intervention also demonstrated a wide variability of change. Results from this study suggest that although the PELD intervention promoted the receptive vocabulary of all children in the study, the children who understood more words at baseline demonstrated a greater increase throughout the duration of the intervention. For example, Ellie who understood the most words (18 words) at baseline, understood the highest number of words (85 words) post-intervention. This meant she increased her word knowledge by 67 words over the course of the intervention. This is in keeping with previous research in other populations (e.g. DLD) that children who have a better vocabulary and language base pre-intervention tend to respond better to treatment (Jackson, Leitao, Claessen & Boyes, 2019). Furthermore,

as children increased their receptive vocabulary at different rates, this highlights the individual differences and variability evident in this population and suggests that the PELD intervention is effective in promoting the receptive language abilities of all children regardless of their word knowledge pre-intervention.

It is important to note that both Sophie and Ellie understood the most words post-intervention and they were the only two participants who attended all three terms of the PELD programme. Therefore, this suggests that more intervention promotes the development of receptive vocabulary. This finding, although anecdotal, corroborates findings by previous authors who found that children with DS benefit from therapy that is implemented for a minimum of six months (Karaaslan & Mahoney, 2013; O'Toole et al., 2018). In addition to this, Yoder, Woynaroski, Fey, Warren and Gardner (2015) conducted a reanalysis of an RCT study by Yoder et al. (2014) who found that children with ID (with and without DS) who received a higher dosage of MCT (five one hour sessions a week over nine months) had better expressive vocabulary than the group who received a lower dose of the therapy once the children had functional play skills (one hour once a week over nine months). Yoder et al. (2015) conducted a re-analysis on the DS only group to explore why this increase in expressive language was noted. They found that a higher frequency of MCT intervention increased the receptive vocabulary and a child's canonical syllabic communication (e.g. production of a consonant-vowel combination imitating adult speech) which in turn increased their expressive vocabularies (Yoder et al., 2015). Although the results in the current study are indicative that more intervention is associated with greater outcomes for the receptive vocabulary of the children, there is risk of halo effect for these results given the fact that the parents were aware of the amount of intervention their children were receiving.

5.2.2 Expressive communication.

An additional goal of the PELD intervention was to encourage the development of expressive language of the children. As mentioned in the literature review, babbling is often delayed when compared to TD peers (Abbeduto et al., 2007) and first words do not emerge until the later age of between 18-36 months for children with DS (Roberts et al., 2007; Roizen, 2002) compared to TD children's first words appearing around their first birthday. Considering the child participants in

this study were between 18-23 months at the end of the intervention, change in the expressive communication was anticipated, therefore, making this aspect of language another primary goal of the PELD programme. Expressive language abilities of the children were captured using three different measures; standardised testing (PLS-5), parental report and an observational checklist.

5.2.2.1 Global measure of expressive language abilities.

The expressive communication subtest of the PLS-5 assessment offered a global means of measuring a child's level of expressive communication pre- and post-intervention. Mixed results were evident in this measure with five of the participants demonstrating an increase in SS scores of between 3 and 31 points while two of the participants (Jack and Grace) decreased in SS scores following the intervention. Of the five participants who increased their SS scores in this study, four children (Sophie, Conor, Daniel and Ellie) demonstrated change with an improvement in the SD scores also noted. The mixed results in terms of expressive abilities based on the PLS assessment were also noted in the study by Wright et al. (2013). Three of the four participants in this study increased their SS scores by between 2 and 10 points (Wright et al., 2013) thus showing fewer substantial gains than the participants in the current study. There are a number of possible explanations for this. First of all, the two participants in this study (Sophie and Daniel) who increased their SS scores by over 25 points following the intervention were the two participants who presented with the lowest SS scores pre-intervention. This meant they improved by one and two standard deviations with Daniel falling within normal limits for expressive communication abilities. Although positive, these findings should be viewed with caution as regression to the mean from extreme scores is a feature of standardised tests (Ebbels, 2017). In spite of this, five of the seven children demonstrated an overall increase in SS scores for this variable. A possible reason for the increase noted in the current study may be related to the protracted therapy duration. The children in the study by Wright et al. (2013) received 20 intervention sessions (approximately 20-30 minutes) twice weekly over 6 months (6-10 hours of intervention in total). Whereas the children in the current study received fortnightly intervention (45 minutes in duration) over 10 months (11.25 hours in total). This suggests that intervention programmes for more than six

months may result in better outcomes for the children (O'Toole et al., 2018) and that intervention implemented over a longer period of time may be more beneficial in developing the expressive language of children with DS.

5.2.2.2 Expressive vocabulary.

Expressive vocabulary was measured using an additional measure; The Vocabulary Checklist 1- First 120 Words (Down Syndrome Education International, 2012). According to this parental report, three of the seven participants (Conor, Ellie and Grace), made gains in the total number of spoken words following the intervention. These children gained between one and four words collectively. Wright et al. (2013) also measured the number of spoken words and all four of their participants used between three and nine new words following their intervention. The lower increase noted in this current study may be related to the fact the participants in the study by Wright et al. (2013) were between 23-29 months at the start of the intervention whereas the children in the current study were between 10-17 months at baseline. Again, the lack of words is not a cause for concern as Robert et al. (2007) and Roizen (2002) state that individuals with DS vary in their expression of first words which often do not emerge until 18-36 months. As the children in this study were between 18-23 months at the final data collection point, the use of verbal language and the emergence of words would only start to be expected around the time of the final term of the intervention. However, considering the age of the children at this time and given the fact that only three of the seven children showed a minor increase in spoken words, these findings are somewhat limited as such gains may be attributed to the natural maturation of the children.

5.2.2.3 Pre-lexical verbalisations.

As minimal to no change was expected in the number of words produced by the individuals given their young age, this study also measured the number of pre-lexical verbalisations used by the children during the free-play five-minute video. This study measured the pre-lexical verbalisations of the children using the first two levels of the three-stage model described by Olswang et al. (1987). Huge variability among the children was noted with some children showing increases in vocalisations alone, other children demonstrating increases in babbling, while other children's

scores for both babbling and vocalisations fluctuated throughout the intervention. The variability that was noted within pre-lexical verbalisations was again not unexpected given the heterogenous nature of DS (Karmiloff-Smith et al., 2016; Oliver & Buckley, 1994). This result was further corroborated by Parikh and Mastergeorge (2018) who found that of their 43 parent-child dyads (children with DS aged between 24-64 months) huge variability was noted in vocalisations of the children.

A rather interesting finding that was noted for some of the participants (Sophie, Ellie and Grace) was that as their scores for babbling increased their scores for vocalisations decreased. As this pattern of development is also noted in TD children (Morgan & Wren, 2018), this suggests that some children with DS follow a similar pattern of development albeit at a later stage (Kumin, 1996; Stoel-Gammon, 1997). An interesting finding to note is that the three children who demonstrated this pattern of development all attended two or more terms of the intervention consecutively. In addition to this, the parents of these three children all demonstrated high scores for *variety of labels* during the intervention which was a measure of parental responsiveness. Previous research has shown that all children require frequent exposure to words and sounds before they can produce the sounds and words for themselves (Stoel-Gammon, 2011). Therefore, the results show that there is some indication that as the parents were using more appropriate labelling, the children were exposed to more words thus potentially promoting their babbling development.

5.2.2.4 Gesture and Lámh.

Another goal of this study measured the use of gesture types and key word signing (Lámh) throughout the intervention. Using gesture as a means of communication is an area of relative strength for individuals with DS (Caselli et al., 1998; Franco & Wishart, 1995). Previous research has shown that gesture use is not only important for the development of language (Dimitrova et al., 2016), but has also been shown to predict the language abilities of children with DS at 24 and 36 months of age (Kaat-van den Os et al., 2015; Zampini & D'Odorico, 2011). For this reason, unaided augmentative and alternative communication (AAC), specifically the use of gestures and manual signs, has been incorporated into previous PCITs (Wright et al.,

2013), including the PELD intervention, as it provides young children with DS with a means of communication when they cannot communicate through verbal means.

Firstly, gestures were measured by analysing the five-minute parent-child interaction video. Gestures were defined using the descriptions reported by Zampini and D'Odorico (2009). The number of deictic gestures (e.g. pointing/reaching), conventional gestures (e.g. culturally recognised gestures such as waving) and finally iconic gestures (e.g. representational gestures such as actions for driving) were recorded. The PELD intervention aimed to build on the natural use of all of the above gestures. Deictic gestures appeared to increase regardless of the number of intervention terms attended and many participants showed evidence of deictic gestures prior to the study. This finding was not surprising considering the deictic gestures are often the first gestures acquired by children with DS (Ozcalisakan, Adamson, Dimitrova, Bailey & Schmuck, 2016).

On the other hand, conventional gestures were not employed by any of the participants pre-intervention. Following the programme, the only two participants (Sophie and Ellie) who demonstrated the use of conventional gestures as based on the observational measure, were also the only two participants who completed three full terms of the intervention. This is suggestive that more intervention may be associated with a greater increase in the use of conventional gestures. A potential explanation for this may be related to the fact that the first goal of every group intervention session orientated around greetings/waving, therefore, the children who attended all terms would have had increased exposure to these conventional gestures within the intervention setting. In addition to this, it is interesting to note that the increases noted in the current study are minimal when compared to the study by Chan and Iacono (2001) who reported conventional gestures were one of the most frequently used types of gestures in their study of children with DS (n = 3) aged between 17-19 months. Although both studies used a similar method (video recording parent-child play activity) of capturing gesture use, the study by Chan and Iacono (2001) recorded 15 minutes of a play activity whereas the current study only recorded seven minutes and analysed five minutes due to the younger age of the participants in the study (Mahoney & Perales, 2005). This is because Mahoney et al. (2007) found that children who are less than the 12-month developmental age can typically only sustain attention for five to seven minutes. Although the length of the video recording in the current study was guided by previous research, this

discrepancy may in part describe the varied results as perhaps the length of time in the current study was not long enough to capture all the gestures used by the children. Therefore future research should be cognisant of the fact that increased recording time may be necessary to capture such gestures.

Finally, the third type of gestures that were coded were iconic gestures. For the purpose of this study iconic gestures and Lámh signs were coded separately. However, it is important to note that there is some overlap between these two measures. As previously stated, Lámh is a key word signing system that is similar to Makaton vocabulary (Grove & Walker, 1990) used in the UK and other countries and signs in both sign languages are based on the real-life object/action they are representing (Glacken et al., 2019). For example, a Lámh sign for 'hug' would be similar to an iconic gesture of that type. This study coded such actions as Lámh signs and only the iconic gestures that were different to the Lámh sign of that action were coded as gestures. In this study four of the seven participants started to demonstrate the use of between one and five iconic gestures towards the end of the intervention. The lower number of iconic gestures in this study may be related to the fact that some of the iconic gestures may have been captured under the Lámh variable. In addition to this, iconic gestures often emerge when the children are older. A study by Stefanini et al. (2007) found that iconic gestures were the most prevalent in children with DS aged between 3;8-8;3 years of ages. This suggests that although the use of iconic gestures may start to emerge around 18-23 months, they are not the most commonly used gestures at this age (Stefani et al., 2007).

As previously mentioned the key word sign language, Lámh, was also incorporated into the PELD intervention and was both a child and parent goal of the intervention. The use of Lámh signs was captured using a video observation and parental report. Five of the seven child participants were reported to have increased their number of signs during the intervention based on the parental checklist. As expected, huge variability was noted among the participants during the intervention with some children demonstrating no increase in signs while other children increased their signs by 23. This study builds on current research that illustrates the huge variability in progress rates of young children with DS. A study by Kumin, Councill and Goodman (1998) noted similar variability when they explored the expressive vocabulary (both sign and speech) of 168 children aged 3 years. Results showed huge variability among the children with a range of 5 to 675 signed and/or spoken

words elicited by the children. Although the children in the current study are younger in age, the variability noted in the current study is in keeping with findings regarding the heterogenous nature of DS (Karmiloff-Smith et al., 2016; Kumin et al., 1998).

Although an increase in signs was noted for five of the seven children according to parental reports, such gains were not mirrored in all participants when this is compared to the data captured using the observational measure. Only two of these five participants (Sophie and Grace) demonstrated change on the observational measure, thus questioning the trustworthiness of the parental report. As all parents were aware that their children were availing of intervention, there is potential for the halo effect, therefore, the observational data may provide a more accurate representation of the outcomes. In addition to this, the observational measure also coded the different types of signs produced by the child. Lámh signs were coded as spontaneous, imitated and prompted (Wright et al., 2013). Both participants showed evidence of prompted and imitated signs with no children in the study demonstrating the use of spontaneous signs. As the observational video was an untrained activity, this falls in line with findings by Wright et al. (2013) who also noted an increase in signs in an untrained environment. They also found that the children used signs with untrained partners (Wright et al., 2013), however, the current study did not incorporate a measure to capture this.

In addition, although there is some indication that the intervention supported the use of signs for some of the children, this increase cannot be directly associated with the PELD intervention as all parents attended additional Lámh courses during the time of the programme. However, despite this, it is notable that the Lámh signs produced by the children post-intervention were all directly targeted in the PELD programme through the different songs and rhymes. Previous research found that teaching manual signs through familiar home routines/play activities is an empirical part of generalisation and maintenance of signs (Wright & Kaiser, 2017; Wright et al., 2013). Similarly, Glacken et al. (2019) found that parents reported they found it easier to maintain their use of Lámh in the home environment by using signs that were specific to their daily routines. Therefore, as the PELD intervention targeted manual signs through familiar home routines, it can be hypothesised that some of the improvements noted in the use of manual signs may be attributed to this intervention.

5.2.3 Summary of language development.

To summarise, although the effects of the intervention varied among the participants and outcome measures, the general findings suggest that the PELD intervention may be effective in promoting the early language skills of the children in this study. Children who attended three terms of the intervention demonstrated the most change in child measures. For these children, a specific increase was noted in the total number of Lámh signs, conventional gestures and receptive vocabulary. Although variability was noted among the remaining child participants, results suggest an overall positive increase for most of the children thus showing some indication that aspects of the PELD intervention may be effective in promoting the use of receptive and expressive language in young children with DS.

5.3 Communicative Interaction

With respect to the second research question that explored the interactional behaviours of the children, results suggest that the PELD intervention was also successful in promoting child pivotal behaviours. Communicative intentions, such as joint attention, are often primary goals of PCITs for preverbal children. In particular, joint attention has proven to play a key role in the social and language development of children with Down syndrome (Zampini et al., 2015). Given the age of the participants and the fact that they were all preverbal at the time of entry to the study, the PELD intervention recognised the importance of joint attention and made it a primary goal of the intervention. This study promoted the development of a child's ability to both respond and initiate joint attention. For the purpose of this study, a child's ability to respond to joint attention was described as a child's *attention* and a child's ability to initiate joint attention was described as *initiation* (Mundy et al., 2007). These variables were captured by video recording a parent-child interaction play activity and were then rated using the Pivotal Behavior Rating Scale (PBRS) (Mahoney & Wheeden, 1998).

5.3.1 Attention and initiation.

Firstly, attention was used to describe a child's response to joint attention initiated by their communication partner. The PBRS (Mahoney & Wheeden, 1998) rated a child's general attention, persistence, participation and their cooperation to

the activity. Children were assigned a score for each of these four aspects of interaction which were then combined to yield an overall score for *attention.* With the exception of Luke, all participants showed an increase in attention following the intervention. Secondly, the PELD programme also aimed to encourage the children in the study to initiate joint attention with a social partner. Zampini et al. (2015) found that a child's ability to initiate joint attention with a social partner could predict vocabulary development 6 months later in children who were aged 24 months at the time. To develop the initiation of joint attention was therefore another primary goal of the PELD intervention. Each observational video was reviewed and each child was awarded a score for their ability to initiate joint attention with an adult and within an activity. Affect was also measured. These three variables were then combined to yield an overall score for initiation.

Mixed results were evident across the participants with many children demonstrating an upward trend prior to the commencement of the intervention. This differs from the results by Karaaslan and Mahoney (2013) who also used the PBRS in their RCT of 15 children who were of preschool age. Children in the intervention group increased their attention by 54% and initiation by 57% compared to the control group who increased by 11% (attention) and 7% (initiation) (Karaaslan & Mahoney, 2013). Although scores for the children in the current study demonstrated a notable increase, this increase was not as substantial as the one noted by Karaaslan and Mahoney (2013). This discrepancy may be related to the different ages of the children noted between the studies.

An interesting point to note is that prior to the intervention, scores for initiation were generally lower than each child's score for attention. However, as the participants attended the intervention, all children demonstrated similar scores for both *attention* and *initiation* at the final assessment. A possible explanation for this increase in joint attention may be related to the parental training. Previous research has found that children with DS often have delayed auditory processing and in turn require more time to communicate. Research has found that parents of children with DS may not be aware of the amount of time needed for their child to process information and respond accordingly which therefore can result in clashes in communication (Berger & Cunningham, 1983; Jones, 1977; Lynch & Eilers, 1991). It can be hypothesised that these clashes in communication may be the reason for the

lower scores of initiation pre-intervention due to parents not waiting long enough to give their child an opportunity to respond.

The above hypothesis is supported when child scores for *initiation* and parent scores for *Wait and Listen* are visually compared. The term *Wait and Listen* measured a parent's ability to incorporate a time delay into interactions which was a primary parental goal of the current intervention as a means of encouraging initiation of the child (Weitzman, Girolametto & Drake, 2017). As soon as the child initiated in some way, be it through gestures or vocalisations, parents were encouraged to provide an appropriate response and expanded on their child's initiation. Results show that for each child, as their *initiation* scores increased, the parents scores for *Wait and Listen* also increased. This finding suggests that children are more likely to initiate an activity if their parents incorporate a time delay which has been documented in earlier research by Duker, Van Doeselaar and Verstraten, (1993). Although this is a preliminary result, it falls in line with results of a study by Meadan et al. (2014) who noted a similar pattern in their PCIT of children with DS aged 37-60 months whose parents engaged in a naturalistic and visual teaching strategies intervention. They noted in their study that when parents incorporated a time delay into an activity, the children increased their percentage of initiation when baseline and post-intervention scores were compared (Meadan et al., 2014).

In addition to this, another possible reason for the lower scores for initiation pre-intervention may be related to the fact that children with DS may take longer to disengage from their social partners face than TD peers (Chapman, 1997; D'Souza et al., 2015). This has implications for initiating joint attention as children must first engage in an object of interest in order to initiate joint attention (D'Souza et al., 2015; Wright et al., 2013). Although anecdotal, the findings of the current study suggest that the PELD intervention may also be effective in encouraging the children with DS to direct their attention to objects, however, further research and analysis is required in order for the true effects to be explored.

5.3.2 Joint attention and vocabulary.

In addition to this, this study also builds on findings by Zampini et al. (2015) and Mason-Apps et al. (2018) who suggest that the vocabulary development of children with DS is mediated by the quality of joint attention. The two participants

(Sophie and Ellie) who achieved the highest rating in the use of overall joint attention (both responding and initiating) following the intervention, were also the two participants with the highest reported scores for receptive vocabulary based on parental report. While this study did not set out to compare these two variables, these results, although anecdotal, add to the current literature base and support this hypothesis. Parents also play a role in the quality of joint attention achieved by the child as Harris et al. (1996) found that the duration and frequency a parent engaged in joint attention with their child is related to the receptive language development of the child. This study again supports these findings as the parents of Sophie and Ellie were the only parents to attend all three terms of the intervention.

5.3.3 Summary of communicative intentions.

All participants showed some evidence for *attention* and *initiation* skills during the pre-intervention period which suggests that these skills are apparent in young children with DS to a certain extent. This corroborates findings by Hahn et al. (2018) who found joint attention to be a relevant strength in comparison to other difficulties evident in this population In this study, scores for these variables increased, albeit by different quantities for each participant, following the intervention. This finding upholds recommendations by Hahn et al. (2018) that by capitalising these specific strengths of the individuals it can help promote the development of these communicative interactions and other areas of language.

One interesting point to note was that *attention* appeared to be more influenced by the intervention in comparison to scores for *initiation.* Most participants demonstrated an immediate change for attention after attending one or two terms of the PELD intervention. In addition to this, all children with DS achieved similar scores for attention and initiation post-intervention. Although natural maturation cannot be disregarded, the current findings suggests the PELD programme may be effective in promoting the joint attentional skills, particularly responding to joint attention, in young children with DS.

5.4 Parent Strategies

Another primary goal of the PELD intervention was to coach and train parents in communication and interaction strategies with the aim of fostering the

development of child communication skills. Mahoney et al. (1999) and Roberts and Kaiser (2011) highlight the importance of parent education and state that education of parents regarding their communication and interaction with their children should be a core aspect of all early interventions. Interaction strategies will first be discussed followed by the communication strategies adopted by the parents.

5.4.1 Parent interaction strategies.

The PELD intervention aimed to coach and train parents to become more responsive and less directive in order to facilitate richer parent-child interactions. The PELD intervention, like other PCITs, fostered the development of these skills by encouraging parents to follow their child's lead, incorporate time delays into interactions, encourage the use of turn-taking and to respond and expand on the child's object of interest. As previously stated these skills were measured using an adapted version of the *'Teacher Interaction and Language Rating Scale'* by Girolametto et al. (2000) using the parent-child observation video recording.

5.4.1.1 Follow the child's lead.

The strategy that was noted to increase the most for all parent's following the intervention was the parent's ability to *Follow their child's lead.* This strategy aimed to decrease parental directiveness by teaching parents to observe their child's behaviour and to follow their child's object of focus. It was apparent from several baseline videos that half of the parents led the activities during the five-minute free-play observation. They often introduced new toys despite their children showing interest in a previous or different toy. This directive play style is not uncommon in parents of children with DS and is often adopted by parents as they attempt to compensate for their child's delays in interaction or communication (Roach et al., 1998; Tannock, 1988). However, as research has found that this directive role can impede on the amount of communication and interaction of the child (Girolametto et al., 1994), the PELD programme aimed to reduce these behaviours by encouraging parents to follow their child's lead during play. Results suggest an immediate improvement for all parents after they completed just one of the PELD intervention terms. It is also apparent that these scores continued to increase or were maintained for most parents (5/7) if they attended more than one term of the PELD intervention

terms. These results fall in line with previous research by Karaaslan et al. (2011) and Karaaslan and Mahoney (2013) who noted parents of children with DS successfully decreased their directiveness following a Responsivity Teaching (RT) intervention. Similar to the current study, Karaaslan and Mahoney (2013) implemented RT over a substantial time period (6 months). Therefore, suggesting that interventions that are implemented over a protracted period of time may be effective in supporting change in the parenting style for parents of children with DS.

5.4.1.2 Join in and play.

A strategy that was found to go hand-in-hand with *following their child's lead* is a parent's ability to *join in and play*. While following a child's lead aimed to decrease parental directiveness, encouraging parents to join in and play encouraged parents to become more responsive to the child's object of interest. Maternal responsiveness describes positive and nurturing interactions where the parent responds appropriately to their child's initiations (Lorang, Sterling & Schroeder, 2018; Warren & Brady, 2007). All parents increased their scores for this variable with some parents (4/7) increasing their scores after one term of intervention and the remaining parents (3/7) increasing after a second term of intervention. These results show some indication that PELD intervention may be effective in promoting responsiveness in parents and this finding is in keeping with other PCITs within the DS population (Karaaslan & Mahoney, 2013, 2015; O'Toole et al., 2018; Roberts & Kaiser, 2011).

Moreover, as responsiveness has been shown to enhance a child's language learning environment and promote developmental skills in children with DS (Karaaslan & Mahoney, 2013), it can be suggested that all parents in the current study provided richer communication and language learning environments for their children. As parent scores for *join in and play* increased, a similar increase in the children's pivotal behaviours was noted. However, no correlational analysis could be conducted as the small sample size and study design restricted the statistical procedures that could be implemented. Although this observation is anecdotal, Karaaslan and Mahoney (2013) noted a similar observation and found a positive correlation between parental responsiveness and child engagement and joint attention. In addition to this, Lorang et al. (2018) explicitly explored parental

responsiveness to gestures and found that parents of children with DS demonstrated high levels of responsivity regardless of the age of the children (age 22-63 months) compared to parents of TD children who became less responsive as their child developed. Therefore, long term measures should be collected in order to see if the parents in the current study maintained and continued their use of such strategies as their young infants with DS progress into childhood.

5.4.1.3 Wait and listen.

Another parent strategy that appeared to increase following the PELD intervention was the parent's use of *Wait and Listen.* Parents were encouraged to introduce a time delay in order to encourage a response from their child. To do this, parents adopt the *OWLing* technique which stands for 'Observe, Wait and Listen' (Hanen Early Intervention Programme, 2011). All parents demonstrated an increase in this variable although some increase was noted for three of the seven parents during the pre-intervention period. This child-orientated behaviour is found to have positive affect on the child outcomes of children with a range of developmental disabilities, including but not specific to children with DS (Weitzman et al., 2017). By observing the child's behaviour, waiting expectantly for the child to initiate communication and by responding to the child's interest, this has been proven to promote the communication development of these children (Weitzman et al., 2017). However, children with DS may require a time delay more than children of other developmental disabilities due to auditory processing deficits noted within this population (Chapman & Hesketh, 2001; Jarrold & Baddeley, 1997). The introduction of an increased wait time between requests allows the child to respond (Duker et al., 1993), thus promoting their communication development and reducing the communication breakdown that can be present between children with DS and their parents (Berger & Cunningham, 1983; Lynch & Eilers, 1991). Therefore parents should not only be educated in the early lexical development of their child with DS (Buckley, 1993) but also trained in strategies to promote effective communication and language development (Roberts & Kaiser, 2011).

5.4.1.4 Interaction strategies with varied results.

Four interaction strategies (*face to face interaction*, *turn-taking*, *gesture* and *imitation*) demonstrated a varied response among the parents. Some parents increased their scores across all strategies following the intervention, some demonstrated no change, while others fluctuated during the intervention. A possible reason for the variability noted in participants responses may be related to the generalisability of these interaction strategies. All parent interaction strategies were captured using a video recording of a parent-child play activity that was recorded in the homes of the family. As this was an untrained activity (parents were asked to play with their child as they typically would), this meant that some parents incorporated songs or aspects of the PELD activities into this play while others did not. This suggests that the interaction strategies did not generalise to the home environment for some of the parents. This was surprising as previous research has demonstrated that parent interaction strategies tend to be more generalisable than parental language modeling strategies. This was noted by O'Toole et al. (2018) who conducted a re-analysis of the data in a study by Kaiser and Roberts (2013). It was found that parents of children with DS demonstrated the use of EMT strategies post-intervention, at the six-month follow up and again at the 12-month follow up (O'Toole et al., 2018). A possible reason for this discrepancy may be related to the fact that the children in the study by Kaiser and Roberts (2013) were of the preschool age whereas the children in this study were much younger (10-17 months at baseline). Therefore, the children in the Kaiser and Roberts (2013) study may have demonstrated more sophisticated communication skills and more interaction skills which may have given the parents more opportunities to demonstrate the use of the EMT strategies in untrained situations. Yoder and Warren (2002) and Vilaseca and Del Rio (2004) report that the amount a child communicates can have a different impact on parents' responsiveness. This is due to the fact that effective communication skills are acquired through bidirectional interactions between infants and parents (Berger & Cunningham, 1983). Slonims and McConachie (2006) explored how the communicative abilities of a child with DS can affect the communicative interactions of parents in their study which reported 23 parent-child dyads. Results showed that the interaction abilities of the infants with DS at eight and 20 weeks of age influenced the quality of the parent-child interaction (Slonims & McConachie, 2006). Therefore, considering the young age of the children in the

current study, the abilities of the children may have influenced the parents implementation of the targeted interaction strategies.

5.4.2 Parent's use of language.

The final goal of the PELD intervention involved encouraging parents to respond appropriately to their child's verbal and nonverbal communicative intentions. The PELD programme also encouraged parents to use responsive language that matches their child's level of communication and development (Girolametto et al., 1998). This was targeted through the following goals; promoting the use of sign language, using short sentences with developmentally appropriate vocabulary and finally emphasising key words and labels.

5.4.2.1 Lámh.

Despite the use of Lámh signs being specifically targeted in the PELD programme, incorporation of these signs into home routines was not evident for all parents. Three of the participants demonstrated an increase in the use of Lámh signs following a term of intervention whereas for other parents minimal to no change was evident. This is an unanticipated finding as one of the five main goals of the intervention targets the use of Lámh signs using daily routines. In addition to this, most parents also attended additional Lámh courses external to the PELD intervention therefore change was expected. Glacken et al. (2019) present a potential explanation for this finding when they qualitatively explored parents' experiences of using Lámh within the home. They found a number of factors were related to the attainment and maintenance of Lámh within the home some of which may describe the reasons for the mixed findings in the current study. Firstly, parents who have to encourage the use of an unaided AAC such as Lámh have to adopt the role of therapist while also fulfilling the maternal role (Singh, Hussein, Kamal & Hassan, 2017). This can be challenging for parents as they often find it difficult to find the balance between the two roles (Glacken et al., 2019; Singh et al., 2017). In addition to this, remembering the signs may also be a contributing factor as was indicated by peers of children with DS in a study by Bowles and Frizelle (2016). Furthermore, Wright et al. (2013) also noted varying degrees of signing among the four families in their study and noted that a child's use of sign was related to the parents use of sign

although the exact relationship between the two was not explored in detail. This point highlights the importance of encouraging the use of sign by parents given the positive outcomes it has for a child's expressive communication development.

On the other hand, perhaps the method used to measure this variable (observational measures of parent-child free-play activity video) was not effective in capturing change. As previously stated, the PELD intervention targeted Lámh signs using pre-specified themes and routines (e.g. dressing, mealtimes, outdoors). As the parents learn the signs in this context, the results suggest that they did not generalise to untrained activities such as the free-play activity for all parents. These results reflect those of Wright and Kaiser (2017) who reported in their study of four parent-child dyads that parents performed poorer on generalisation and maintenance probes, suggesting that parents require additional support when showing how EMT strategies could be used and adapted for the home environment. The varied results noted in the current study do not discredit this goal of the PELD intervention, however it does pose a question regarding how this goal should be measured in future research and how therapists can support parents in the use of Lámh in the home environment.

5.4.2.2 Encouraging developmentally appropriate language.

Another goal of the PELD intervention was to encourage parents to use developmentally appropriate language when communicating with their children with DS. During the pre-intervention phase, it was noted that some parents (Sophie and Ellie's parents) posed higher level questions to their children (such as *'Who's on the phone?'*) and often spoken in long sentences using numerous conjunctions (e.g. *'oh look at the dolly and she's running'*). Considering the age of the children in this study (age 10-17 months at baseline) and their developmental level, such language use may be considered too advanced for the children at this time point. This corroborates previous research that has shown parents of children with DS often increase their language input and use linguistically complex sentences as a means of promoting the language development of children with intellectual disability (Thiemann-Bourque et al., 2014). However, as such language adaptations have been proven to be ineffective with children with DS, current research has encouraged the use of developmentally appropriate language use.

The syntactic complexity of each parent's language was measured by calculating MLU in morphemes across all time points. Although adult language would usually be measured in MLU words, MLU morphemes was used in order to reflect subtle changes in the parent's language. Although a wide variation for average MLUm scores was noted at baseline, results showed a descrease in MLUm scores at post-intervention for that most parents, with four of the parents descreasing their MLUm by nearly a third. In addition to this, parental use of types (number of different words used) and tokens (total number of words) were also measured. Both types and tokens were seen to decrease following the intervention. The higher number of MLUs and greater number of different words used at baseline were not unexpected as work by Johnson-Glenberg and Chapman (2004) report that parents of children with DS use language that may be too syntactically complex for their children when compared to a group of parents of TD children. Girolametto et al. (1998) also measured MLU in morphemes and type-token ratio (TTR) in their study. The current study did not report TTR as the subtle differences between types and tokens used by the parents wanted to be captured. Findings in this study are contrary to the findings by Girolametto et al. (1998) who noted no difference in the MLUm or TTR of parents when the control group was compared to the intervention group. A possible reason for this difference may be related to the differences in the frequency as well as the overall duration of the PELD intervention. The parents in the study by Girolametto et al. (1998) received less frequent training (nine weekly 2.5 hour group sessions and four individual sessions) compared to the parents in the current study (12 fortnightly 45 minute session and six individual sessions). This suggests that increased therapy sessions over a longer period of time may be required before change is evident in parental use of MLUm and TTR.

When the overall decrease in MLUm, types and tokens is coupled with the fact that most parents improved their use of labeling, one can assume that parents were also repeating key words more often following the intervention as opposed to using too varied vocabulary without sufficient repetitions of each vocabulary item pre-intervention. Although there were fewer different words used in six of the seven participants post-intervention, it can be postulated that parents repeated these key words more frequently following the intervention. This shows that the PELD goal of '*using target key words and signs'* was effective in improving the quality of the parent's language input, as observed in a five-minute free-play routine within each

family's home. This is important as repetition of the stimulus is needed to address the short term verbal memory deficits noted in individuals with DS (Chapman, 1997; Decker et al., 2017). Conners, Rosenquist, Arnett, Moore and Hume (2008) and Mosse and Jarrold (2010, 2011) both reported that increased repetition of words can encourage vocabulary development within this population. Repetition of words has been studied both in TD children and children of different developmental disabilities and has also found to be linked with the improvements in comprehension and attention (Sokolov, 1993).

As previously stated, parents use of labeling also increased following the intervention according to the five-minute parent-child observation video. All parents demonstrated an increase for labelling with some showing change after one term and others demonstrating change after a second term. In theory, the free-play observation offered the perfect opportunity for labeling as the parent introduced a standard set of toys, some of which may be new and unfamiliar to the child. It is important to note that books were included in this set of pre-selected toys and for the parents who used a book during the free-play activity they demonstrated increased instances of labeling than those who did not. This may offer an additional explanation for the varied results noted across the parents' use of labelling.

5.4.2.3 Number of utterances.

The number of utterances used by the parents when interacting with their child also notably changed following the intervention. Prior to the intervention, average baseline scores for parents' utterances ranged from 31-114 utterances which indicates huge variability within parents who are communicating with children with Down syndrome of roughly the same age. An interesting finding was that following the intervention this range became smaller with all parents using between 59-88 utterances with their infant during a free-play activity (with 5/7 parents decreasing their total number of utterances and 2/7 increasing their total number of utterances post-intervention). A sub-goal of the PELD programme was to encourage the use of shorter sentences, developmentally appropriate language and to use slower rate of speech. These factors may have contributed to the positive improvement noted by the parents for total utterances used by the parents. For the parents who used a minimal 22 utterances at baseline (Jack's parent), they demonstrated an increase to

66 utterances at post-intervention. This was a positive effect for this parent as it showed that PELD encouraged this parent to communicate more with her child compared to when she first started the programme. Although this may be attributed to the fact that the parent became more comfortable with being recorded by the end of the intervention, it was noted that the quality as well as the quantity of language behaviours improved. During the pre-intervention period, results showed that this parent would repeat onomatopoeic words such as '*bang bang bang*' while the child was tapping the drum. This again is in keeping with results reported by Zampini et al. (2011). In their study of maternal language input with children aged 24 months with DS, Zampini et al. (2011) found that these mothers used more onomatopoeic words than parents of TD children and found that their overall language input was less complex than necessary (Yoder and Warren, 1998). A recent study by Ota, Davies-Jenkins and Skarabela (2018) explored the impact onomatopoeic words has on a child's vocabulary. In their study, the authors rated onomatopoeic words for iconicity and found that most onomatopoeic words referenced a sound effect (*e.g. 'the train goes choo choo')* as opposed to a noun (*e.g. 'look at the choo choo').* Following their analysis, results showed that iconicity was not related to vocabulary growth of the 47 children aged between 9 and 21 months (Ota et al., 2018). Following the PELD intervention, Jack's parent demonstrated richer language use and a higher quantity of labeling was noted at the final assessment, '*Drinking. Jack is drinking. Cup. Give Mommy the cup'*. Such linguistic mapping is necessary for a child's language development. Linguistic mapping consists of a parent responding to a child's verbalisations using simple complete grammatical sentences (for example, subject-verb-object) (Yoder & Warren, 1998). The use of such function words are important as children rely on function words to cue nouns and verbs (McDuffie & Yoder, 2010; van Kleeck et al., 2010). When parents use linguistic mapping, positive increases have been noted in the vocabulary development of TD children (Goldin-Meadow, Goodrich, Sauer & Iverson, 2007). This same theory can be applied to children with DS.

On the other hand, the parents of Sophie, Daniel and Grace were noted to use over 100 utterances during the pre-intervention period based on the observation video. As the PELD programme promoted a slower rate of speech, introduction of a time delay (interaction strategy) and the use of shorter sentences throughout the activities this meant the parents reduced the amount of input with their child.

Although 100+ phrases within a five-minute time period may seem excessive given the age of the participants, this is not surprising given the findings by Zampini et al. (2011). Following their research, they noted that parents of children with DS tended to use more utterances than that of the parents of TD peers, but produced a similar number of words when compared to mothers of children at the same linguistic level (Zampini et al., 2011). They hypothesised that this may be due to parents of children with DS attempting to compensate for their child's absence of verbal output and in turn attempt to fill pauses and stimulate language development (Zampini et al., 2011). However, in the study by Thiemann-Bourque et al. (2014), who used the LENA software analysis to measure the language environment of nine children (aged 9-54 months), found that there was no increase in the children's vocabulary for the parents who 'talk more' (Thiemann-Bourque et al., 2014). A possible reason for such outcomes may be related to the fact that parents who 'talk more' may not include a time delay, thus not giving the child an opportunity to initiate and respond which can have negative implications for the child's language and communication development (Weitzman et al., 2017). Therefore, the decrease in the number of utterances used by the parents in this study was not unexpected as all parents increased their use of a time delay post-intervention, thus showing that the interaction strategies and language modelling strategies intertwine and that parents should be upskilled in both aspects in order to optimise their child's language and communication development. The general decrease in utterances that was noted in the current study also corroborates the findings of Falkus et al. (2016), who also implemented parent-child interaction therapy (PCIT) with parents of children with DS aged 21-42 months of age. The authors found that when pre- and post-test videos were compared that the parents talked less and the children spoke more. However, as the children in our study were much younger at the time of the initial baseline, an increase in the child's spoken language was not anticipated. This highlights the need for follow up measures to be conducted in order to see if the children increase their verbal output as a result of the parents improved communication strategies.

5.4.2.4 Parental stress.

The results for parental stress reiterate findings found in the literature demonstrating mixed results for parental stress. In the current study, five of the seven

parents reported consistent low numbers for parental stress. This is not unusual as previous research has found this to be related to their children's amiable personalities and experience less maladaptive behaviours when compared to children with other developmental disabilities (Blacher et al., 2013; Hodapp, Ly, Fidler & Ricci, 2001). On the other hand, a study by Phillips et al. (2017) who compared the parenting styles and dimensions of parents of children with DS to parents of TD children found that not only did parents of children with DS get more stressed than TD parents, but that this stress also seemed to be related to the permissive parenting style noted in the parents in their study. It can be assumed as all parents reported similar scores for stress during baseline assessments and throughout the intervention, that completing the PELD intervention did not increase or decrease the level of stress experienced by the parents. However, further qualitative research is warranted to truly explore this anecdotal finding and future studies would benefit from exploring the parents perspective and experiences of completing this intervention programme.

5.4.3 Parent interaction and communication summary.

To summarise, the PELD intervention appears to be effective in promoting three main interaction strategies (*follow child's lead, join in and play* and *wait and listen*) while other interaction strategies demonstrated mixed results among the parents. In addition to this, the results suggest that parents also improved their language strategies with the majority of parents decreasing their number of utterances, MLUm, types and tokens post-intervention and increasing their repetition of key words and labelling.

5.5 Strengths and Limitations

This single-subject multiple-baseline study demonstrates the immense variability that is evident within children with DS (Karmiloff-Smith et al., 2016) and illustrates the individual differences that are evident at a very young age. This study design is also an appropriate way to measure change throughout an intervention programme as the collection of baseline measures ensured a level of control for each participant (Gast & Ledford, 2010). This design was deemed the most appropriate as the intervention is in its developmental phase and measures for capturing change in

the intervention needed to established before higher level research, such as randomised control trials, can be completed (Medical Research Council, 2000).

In addition to this, all measures were collected and rated by a researcher who was blind to the outcomes goals of the intervention. Similarly, a high percentage of the observational measures were double coded by an external coder who was blind to the goals of the intervention and blind to time, thus further decreasing the risk of observer bias. Furthermore, the current study adopted the use of a wide range of outcomes to capture change in both the parents and children. As previous research has implemented the use of parental report, observational measures and standardised assessment, very few studies have used all three methods for collecting data. Although the variety of measures used is a strength of this study, future research should incorporate an additional video recording analysis of a parent-child interaction during a daily routine in addition to the free-play activity that is commonly reported in the current study and previous studies by Girolametto et al. (1998), Karaaslan et al. (2011) and Wright et al. (2013). As the focus of this intervention is interaction and communication strategies within daily routines, an additional measure that captures this may provide a greater insight into the use of the parent interaction strategies in the home environment.

One limitation of the study relates to the lack of second language measures for the children in bilingual households. The true effectiveness of the PELD programme with bilingual children is unknown as no data was collected for the bilingual child's second language. These measures were not collected in the current study as the parent completing the study only spoke English with the child. Future research should measure change in all languages the child is exposed to in order to determine how the PELD intervention influences the language development of bilingual children.

Another limitation of the current study relates to the baseline assessments. Wright et al. (2013) collected three to five baseline assessments in order to establish a visual trend of the participants before the intervention commenced. However, due to constraints within the service and the commencement of the intervention within the community, only two baselines were collected for five of the participants and only one baseline was collected for the two final participants. In addition to this, the baselines were collected one week apart therefore it only captured change within the child over a very short period of time. According to Kazdin (2011), a minimum of

three baselines are needed in order for a pre-intervention pattern to be established so that any change noted in the children and parents can said to be caused by PELD intervention and not any external factors, for example the child's maturation. This therefore calls into question the internal validity of the current study.

Missing data was evident for one participant (Jack) during the second term of the intervention. It was unfortunate that the data could not be collected as this time as this was the only participant who attended the first term of the PELD intervention, withdrew from the study for term two and later recommenced the intervention at term three. As a result, the effect of therapy withdrawal followed by recommencement of therapy could not be explored due to this missing data.

A final limitation of the current study is the lack of control items. Ebbels (2017) recommend that within-participant designs that do not include a control group should aim to include control test items/targets in order to explore the true effectiveness of a study. Such control items should be of a similar nature and level to the test items and should be collected in line with the outcome measures. However, given the young age of the children in the current study, child outcomes were collected using naturalistic methods (free-play video recording), parental report and the PLS-5 assessment, therefore it was difficult to include control items that would correlate to such measures. Future research could adopt the use of control items for parent outcomes by potentially collecting data from themes or routines that were not targeted in the current intervention.

5.6 Future Research

As this is a preliminary study in terms of the effectiveness of the PELD intervention, there are many unanswered questions in terms of the relationship between the adult strategies taught in the PELD programme and the gains evident in the communication skills of the children. Future research should endeavour to explore what aspects of parent training have the greatest impact on child outcomes. This was beyond the scope of the current research given the design of the study, however, it would be beneficial to know what aspects of the PELD intervention parent training have the greatest impact on the child's communication and interaction abilities going forward. The use of statistical analysis and testing may also be beneficial in defining the true effect of the intervention. For example a

correlational analysis may be beneficial to explore which parents strategies corelate to changes in communication noted in the children. Similarly such an analysis could also be used to to compare child outcomes to total sessions attended and parental outcomes to total number of sessions attended in order to explore the true effect the duration and frequency of the PELD intervention had on child and parent outcomes.

In addition to this, it was noted that during some video recordings, the siblings of the children with DS often entered the room and distracted the parent which resulted in lower scores for certain variables (e.g. Daniels parent for imitation). This raises the question regarding the adaptability of the intervention strategies and how they can be integrated into family life. It may be beneficial for PCITs in the future to incorporate separate family sessions where parents are also coached on how to facilitate their child's communicative development within the busier home environment and also regarding how to incorporate the children's siblings into the activities. A possible way of addressing this in the current intervention could be the inclusion of a child's sibling into the individual sessions of the PELD intervention. This is an important consideration for the PELD intervention as all participants, with the exception of Jack, had one or more siblings. Trent-Stainbrook, Kaiser and Frey conducted a study in 2007 and trained the older siblings of three children with DS in two responsive interactions strategies (mirroring and verbal responses). Although the siblings were successful in learning and maintaining these two interaction strategies post-intervention and at the one-month follow-up, the strategies did not generalise to untrained activities (Trent-Stainbrook et al., 2007). Although the sample size was small, future interventions should consider including siblings into the intervention of children with DS as it may help to foster positive interactive relationships between siblings (Trent-Stainbrook et al., 2007; Trent, Kaiser & Wolery, 2005). This may create a richer language learning environment within the child's home as additional family members will be trained in communication training strategies.

As longer term measures were beyond the scope of this study, follow up measures should be collected in order to define the long-term benefits of the intervention. Research should examine the parents maintenance of strategies and also see if the communication and interaction gains, demonstrated in the children in the current study, facilitate the later communication and language development of the children. O'Toole et al. (2018) recommend that follow-up measures should be

collected 6 and 12 months after the intervention in order to determine its long-term effects.

In addition to this, qualitative research from the parent's perspective and experiences should be conducted in order to explore how they found the intervention and what aspects were most beneficial. As noted by Girolametto, Tannock and Siegel (1993), parental perspectives will be important for the further refinement of the programme in order to ensure it is feasible and acceptable for parents. It is important for this to be reviewed considering the mixed reports outlined by parents in the qualitative study by O'Loughlin et al. (2010) with regards to group interventions. Future research should also aim to measure parental implementation of the strategies outside the intervention sessions for adherence and dosage to further investigate the extent at which such strategies are used in the home environment (Wright & Kaiser, 2017).

Finally, as this study consisted of group and individual sessions, future research should explore the effects of whether more group or individual sessions are needed and which have the largest effect on the outcomes of the child (Lundahl, Risser & Lovejoy, 2006). Individual sessions provided an opportunity for the intervention to be specifically tailored and individualised to each families' circumstances (Kaiser & Roberts, 2013). While group intervention sessions provided an opportunity for the children to learn from each other, it also offered an opportunity for parents to share experiences (O'Loughlin et al., 2010). Therefore the amount and intensity of each style of intervention should be explored determine what ratio is most beneficial for young children with DS and their parents (Lundahl et al., 2006). In accordance with this, the individualisation of the programme could be increased with different individualised targets outlined for each parent-child dyad.

5.7 Clinical Implications

The PELD intervention has demonstrated some positive effects and outcomes for very young children with DS completing the first strand of the programme. As this is the first stage of the PELD intervention, the later strands should be evaluated in order to determine the true effectiveness of the PELD intervention. This study has established measures and means of measuring the effectiveness of this intervention.

Such measures can now be adopted as a means of capturing change within both the clinical setting and future studies.

There is some indication that specific aspects of PELD intervention may be effective in promoting the communication interaction of preverbal children with DS while also promoting their receptive and expressive language abilities. The PELD intervention suggests that such improvements can be achieved through fortnightly group and individual sessions across a period of 10 months. This is important as O'Toole et al. (2018) recommended that intervention should be implemented for longer than six months in order to see change in child outcomes. Similarly, as the two child participants who attended all three terms of the intervention demonstrated the most change, this adds to the current literature that intervention should be implemented over longer periods of time in order to maximise child gains and improvement. The positive parental outcomes also suggest that interventions, such as PELD programme, may be effective in teaching and coaching the parents of young children with DS and that longer interventions foster greater parental change.

Finally, the findings also suggest that the PELD intervention can be efficacious with families in a range of circumstances, although variety in the extent of changes was noted among the participant. The children in the study were from monolingual and bilingual households with a variety of parents who worked fulltime, part-time or who were on careers leave. The different home environments did not appear to have an impact on the effectiveness of the intervention which therefore suggests that the PELD intervention may be suitable for a range of different home environments. However, one point to note is that all families were middle class so the effect of the intervention in families of lower socioeconomic status is unknown.

5.8 Conclusions

This study contributes to the growing literature of parent-child interaction therapies and their effectiveness with young children with Down syndrome. This was the first study to measure an intervention programme that is specifically tailored to infants with DS and the findings suggest it can be effective with infants as young as 10 months of age. As discussed above, there is some indication that the PELD intervention can be effective in promoting the communication and interaction skills

of the children. In particular, improvements in receptive vocabulary, use of key word signs and gestures were noted in the majority of the children following the intervention. Similarly, the intervention encouraged the development of child pivotal behaviours, particularly a child's ability to respond to joint attention. This study also supports previous research regarding the effectiveness of PCITs that are implemented for more than six months. In the current study, children who attend all three terms of the intervention seemed to benefit the most from the PELD programme as they demonstrated the use of a wide range of gestures, understood the most words post-intervention and used the most Lámh signs as reported by their parents.

With regards to parent outcomes, the findings suggest that all parents were successful in adapting their parenting style with all parents demonstrating a notable increase in their abilities to follow their child's lead, expand on their child's play and incorporate a time delay into parent-child interactions. In terms of language strategies, parents also used developmentally appropriate language, increased the amount of effective labelling and repetition of key words post-intervention. Despite the huge variability of the participants in the current study, there is some indication that this intervention can be effective with young children with DS who present with a range of cognitive and language abilities and their parents. Results also showed that the children who completed all three terms of the intervention demonstrated greater gains in language and communicative intentions which suggests that intervention over a protracted period of time are more beneficial for children with DS and their parents.

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Appendix A

Summary of Parent-child Interaction Therapies

Summary of PCITs for young children with DS

	Girolametto et al. (1998)	Kaiser & Roberts (2013)	Wright & Kaiser (2017)	Wright et al. (2013)	Karaaslan & Mahoney (2013)	Fey et al. (2006) and Warren et al. (2008)	Yoder et al. (2014)	Meadan et al. (2014)	O'Loughlin et al. (2010)
PCIT intervention	Adopted aspects from Hanen Parent Programme using a focused stimulation approach	Enhanced Milieu Teaching (EMT)	Enhanced Milieu Teaching (EMT) with words and signs using a TMCR protocol	EMT/ JASPER Words and Signs	Responsivity Teaching (RT)	Responsivity education/ prelinguistic milieu teaching (RE/PMT)	Milieu Communic-ation Teaching (MCT)	Parent-Implemented Communication Strategies (PiCS) project	Little Owls
Aim of study	The effect training parents in responsive interaction strategies can impact their child's vocabulary development	To compare EMT that was administered by therapists alone to a group who received EMT by therapists and parents	To explore the effects of the TMCR model on parents use of EMT; To explore the effect of EMT words and signs on child language	To explore effect of EMT/ JASPER Words and Signs on the expressive language of children with DS	The effect of RT on parental responsiveness ; To explore the effect RT had on the pivotal behaviours of children with DS	The effect RE/PMT has on children's use of imperative, declarative and communicative acts; the effect RE/PMT has on parental language strategies	To explore the effect of dose frequency of the MCT intervention	To explore feasibility and effectiveness of PiCS of parent coaching strategies and social-pragmatic skills of the children	To explore the efficacy of Little Owls intervention from a parental perspective

Summary of PCITs for young children with DS (continued)

	Girolametto et al. (1998)	Kaiser & Roberts (2013)	Wright & Kaiser (2017)	Wright et al. (2013)	Karaaslan & Mahoney (2013)	Fey et al. (2006) and Warren et al. (2008)	Yoder et al. (2014)	Meadan et al. (2014)	O'Loughlin et al. (2010)
Duration and Frequency	Nine 2.5 hour sessions followed by four individual sessions. Overall duration not specified. Parents were trained in group setting	One 2 to 3 workshop, 24 twice weekly session, 30 minutes in duration and 12 home sessions, 20 minutes in duration	Twice weekly, duration was not reported	20 sessions (lasting 20-30 minutes) that were delivered twice weekly	Weekly 1.5 to 2 hour sessions over 6 months	RE = 8 one sessions over 6 months; PMT = 4 times per week, 20 minute sessions. Both groups received a low intensity booster prior to Warren et al. (2008) study	LDF group = one hour once a week for 9 months; HDF group = five hours once a weeks for 9 months	2 to 3 times a week for approximately 4 months	Not reported
Study design	RCT	RCT	Case series. Single-case multiple baseline	Case series. Single-case multiple baseline	RCT	RCT	RCT	Case series. Single-case multiple baseline	Qualitative. Semi-structured interviews
Participants	Children aged 29-46 months and their parents	Children aged 30-54 months and their parents	Children aged 28-33 months and their parents	Children aged 23-29 months and their parents	Children aged 2-6 years and their parents	Children aged 24-33 months and their parents	Children aged 18-27 months	Children aged 2-5 years and their parents	Parents of children with DS
Sample size	n = 12; 6 in intervention group; 6 in control group	n = 77; 8 children with DS in therapist only; 10 children with DS in parent & therapist group	n = 4	n = 4	n = 15; 7 in intervention group; 8 in the control group	n = 51; 26 had DS, 13 allocated to experiment group; 13 allocated to control group	n = 64; 16 children with DS in LDF group; 19 children with DS in HDF group	n = 5	n = 6

Summary of PCITs for young children with DS (continued)

	Girolametto et al. (1998)	Kaiser & Roberts (2013)	Wright & Kaiser (2017)	Wright et al. (2013)	Karaaslan & Mahoney (2013)	Fey et al. (2006) and Warren et al. (2008)	Yoder et al. (2014)	Meadan et al. (2014)	O'Loughlin et al. (2010)
Fidelity	Only parental attendance	Treatment fidelity and intervention adherence	Treatment fidelity and intervention adherence	Procedural fidelity	Treatment fidelity	Treatment fidelity	Fidelity of dose, dose frequency and duration	Treatment fidelity	N/A
Blinding	x	Rater of formal ax was not blind to experiment condition. Rater of observational measures was blind.	Coders were blind to time	Coders were not aware of experimental phase of child	Coders were blind to control and experiment group for all measures	Coders were not blind to groups at post-intervention analysis.	Coders were blind	Not reported	N/A
Measures and data collection	Pre-test and post-test video recording and child assessment of vocabulary	Pre-test; Post-test; 6 months follow up; 12 month follow up. Observational measures, standardised testing and parental report were used	Baseline assessment, intervention assessment, generalisation probes, maintenance assessments. Observational measures and transcription of a 10-minute video recording	Baseline assessments, intervention and home generalis-ation probes were collected. Observat-ional measures using video recording	Pre-test and Post-test assessment. Observational measures using video recording of parent-child play	Pre-test; Post-test; 6 months follow up; 12 month follow up.	Pre-test; 3 months of intervention; 6 months of intervention; post-test. Parental report and checklist	Pre-test and post-test. Other data collection points were not described. Norm-referenced standardised assessment and observational measures using a video recording of a parent-child play scenario	5 interviews over 3 months

Summary of PCITs for young children with DS (continued)

	Girolametto et al. (1998)	Kaiser & Roberts (2013)	Wright & Kaiser (2017)	Wright et al. (2013)	Karaaslan & Mahoney (2013)	Fey et al. (2006) and Warren et al. (2008)	Yoder et al. (2014)	Meadan et al. (2014)	O'Loughlin et al. (2010)
Results	No significant difference noted in children across the groups; Parents in experiment group used more focused stimulation techniques and target words	No difference in child language as based on formal testing; Parents in experiment group used more EMT strategies than control group at 12 month follow up across trained and untrained activities*	TMCR protocol is effective in training parents in EMT strategies. High level of fidelity can be achieved with the use of EMT strategies and TMCR protocol	EMT/ JASPER Words and Signs had a positive impact on the expressive language skills of the children although variability among the children was noted	Parents in the experiment group increased responsiveness and decreased directiveness; Increases in children in RT group increased their engagement.	Children in experiment group with DS showed positive effects however increase was not statistically significant. No difference between the control group and intervention group in relation to the way parents of children with DS responded. However, improvements were not noted at follow up	HDF noted a greater increase in their growth of vocabulary; children benefited more from MCT if they had functional play skills.	Parents require frequent coaching sessions to effectively implement communication strategies and one time training in insufficient	Parental perspective of potential barriers and strengths with PCITs in the Irish context

Notes. RCT = randomised control trial; x = not completed; TMCR = Teach-Model-Coach-Review, ax = assessment; LDF = low dose frequency; HDF = high dose frequency; N/A = not applicable

*Results from secondary analysis by O'Toole et al. (2018)

Appendix B

Clinical Research Ethics Committee Letters of Ethical Approval

COISTE EITICE UM THAIGHDE CLINICIÚIL
Clinical Research Ethics Committee of the Cork Teaching Hospitals

Tel: +353-21-4901901
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University College Cork
Lancaster Hall
6 Little Hanover Street
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Ireland

CREC Review Reference Number: ECM 4 (g) 04/09/18
& ECM 3 (eee) 13/08/19

Date: 13th August 2019

Dr Pauline Frizelle
Lecturer in Speech and Language Therapy
Brookfield Health Sciences Complex
University College Cork
College Road
Cork

Study Title: An effectiveness study of the [redacted] intervention ([redacted]) with children with Down syndrome.

Dear Dr Frizelle

The Chairman approved the following:

- Amendment Application Form signed 4th July 2019 (received 11th July 2019)
- Study population extension from 4 to 7 participants
- Revised Application Form.

Yours sincerely

Professor David Kerins
Chairman
Clinical Research Ethics Committee
of the Cork Teaching Hospitals

The Clinical Research Ethics Committee of the Cork Teaching Hospitals, UCC, is a recognised Ethics Committee under Regulation 7 of the European Communities (Clinical Trials on Medicinal Products for Human Use) Regulations 2004, and is authorised by the Department of Health and Children to carry out the ethical review of clinical trials of investigational medicinal products. The Committee is fully compliant with the Regulations as they relate to Ethics Committees and the conditions and principles of Good Clinical Practice.

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Coláiste na hOllscoile Corcaigh, Éire
University College Cork, Ireland

COISTE EITICE UM THAIGHDE CLINICIÚIL
Clinical Research Ethics Committee

Lancaster Hall,
6 Little Hanover Street,
Cork,
Ireland.

CREC Review Reference Number: ECM 4 (g) 04/09/18

Date: 31st August 2018

Dr Pauline Frizelle
Lecturer in Speech and Language Therapy
Brookfield Health Sciences Complex
University College Cork
College Road
Cork

Study Title: An effectiveness study of the [redacted] intervention ([redacted]) with children with Down syndrome.

Approval will be granted to carry out the above study at:

Down Syndrome Cork and parents homes

subject to receipt and approval of the following:

Revised application form	(a) Add Dr Ciara O'Toole as co-investigator, (b) form states that there will be no assent form but there is an assent form included for the study
Revised Information Leaflet	Make the following changes: (a) Change "The assessments will take place....." to "The evaluation assessments will take place....", (b) Change "However, participation is completely voluntary." to "However, participation in this evaluation of the [redacted] intervention is completely voluntary", (c) Change "...no bearing whatsoever on the speech and language therapy..." to "...no bearing whatsoever on the [redacted] programme or the speech and language therapy....", (d) Change "you are free to withdraw at any stage until the results....", Specify time. (e) Add details about visits, how many, how long, who will be present, researcher has been garda vetted ....
Revised Consent Form	Add "I understand that this study is an evaluation of the [redacted] programme and whether I decide to take part or not will not have any bearing on my involvement in the [redacted] Programme.

The following documents have been approved:

Document	Approved	Version	Date
Cover Letter	Yes		31st July 2018
Application Form	No, See Above		
CV for Chief Investigator	Yes		
Evidence of Insurance	Yes		
Study Protocol	None provided		
Data Collection Sheet	None		
Participant Information Leaflet	No, See above		
Consent Form	No, See above		
Study Questionnaire/Survey	Yes		
Interview Guide	No		
Garda Vetting Form	Yes		

UCC

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Clinical Research Ethics Committee

Lancaster Hall,
6 Little Hanover Street,
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University College Cork, Ireland

We note that the co-investigator(s) involved in this project will be:

Name	Occupation
Sarah Marie Cronin	MSc Student

Dr O'Toole will be added as a co-investigator on receipt of the revised application form.

Please note that permission is not yet granted to begin this study. When the document/documents required above have been submitted and approved by CREC, you will receive a full approval letter You cannot begin this study until you have received that letter. Original signed approval letters/conditional approval letters from CREC must be kept in your study master file for audit purposes.

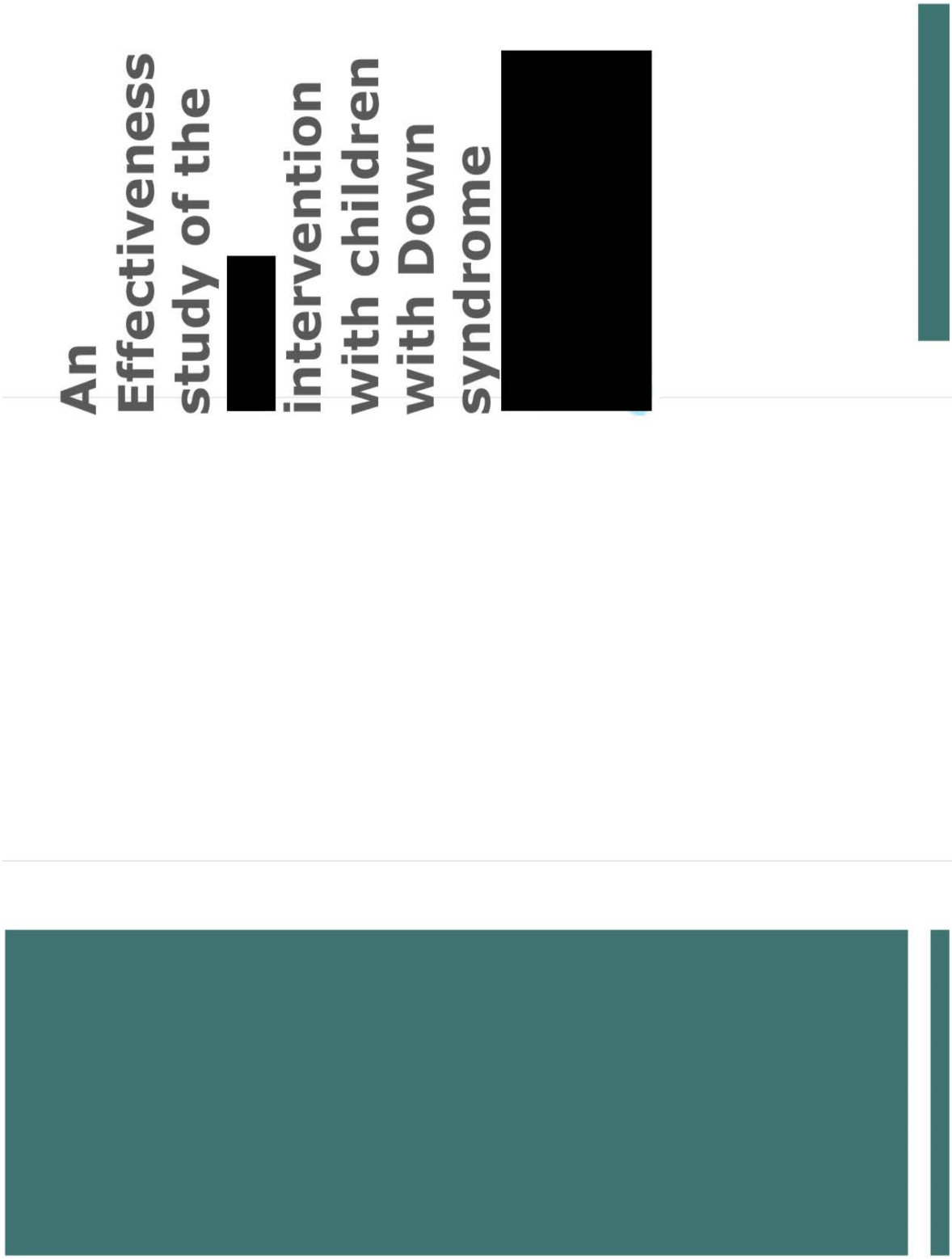
Yours sincerely

Professor Michael G Molloy
Chairman
Clinical Research Ethics Committee
of the Cork Teaching Hospitals

The Clinical Research Ethics Committee of the Cork Teaching Hospitals, UCC, is a recognised Ethics Committee under Regulation 7 of the European Communities (Clinical Trials on Medicinal Products for Human Use) Regulations 2004, and is authorised by the Department of Health and Children to carry out the ethical review of clinical trials of investigational medicinal products. The Committee is fully compliant with the Regulations as they relate to Ethics Committees and the conditions and principles of Good Clinical Practice

Appendix C

Information Leaflet



Purpose of the Study
The Speech and Language Therapists (SLTs) at the [redacted] would like to evaluate the effectiveness of their [redacted] programme and have asked for the help of researchers at University College Cork (UCC). For this study we are looking for at least four parents and their children to be involved in the evaluation.

What will the study involve? For parents who agree to take part, the study will involve an observation and assessment of communication skills targeted in [redacted] before, during and after the programme. The UCC researcher Sarah Cronin (who is Garda vetted) will carry out the observations and assessments. You as the parent will be present at all times. The therapy and assessments will be video-recorded to help with the analysis. The evaluation assessments will take place in your home and should take approximately one hour on the first, second and follow up visits and 20 minutes on the other two occasions. Sarah Cronin and her supervisors will analyze the results.

Why have you been asked to take part?
You have been asked to take part as you and your child are involved in the [redacted] programme. However, participation in this evaluation of the [redacted] intervention is completely voluntary. You are under no obligation to participate, and this will have no bearing whatsoever on the [redacted] programme or the speech and language therapy services you will receive from [redacted] If you agree to take part, you are free to withdraw at any stage until June 2019 at which time the results will have been made anonymous.

What information will be collected?
We will ask you some biographical information which will help us with our evaluation, and we will collect information on how you and your child are responding to the programme. The aim is to help us understand which aspects of [redacted] are useful for parents and children and which may need to be changed.

Will your participation be kept confidential?
We will preserve you and your child's anonymity by removing all identifying information and giving each parent and child a coded number which we will refer to throughout the study. Only the researcher Sarah Cronin (under the supervision of Dr Pauline Frizelle and Dr Ciara O'Toole) will have access to the information.

How will the information be kept?
All data will be stored in a locked filing cabinet on a password protected and encrypted laptop in the Department of Speech and Hearing Sciences Complex in UCC. On completion of the study, the video data will be kept for one-year and all other data, for a minimum of ten-years before being destroyed. If you agree, the videos can be used for educational purposes within the Department of Speech and Hearing Sciences in UCC.

What will happen to the results?
The results will be shared with the therapists at [redacted] and may be presented at conferences and in publications. At no stage will you or your child be identifiable.

What are the possible disadvantages of taking part?
We don't envisage any disadvantages, although, as the before and after assessments are not routinely part of [redacted], they will involve some extra time on your behalf.

Who has reviewed this study?
This study has been reviewed by the Clinical Research Ethics Committee of UCC.

Appendix D

Consent Form

[redacted]

An effectiveness study of the [redacted] intervention with children with Down syndrome

Parent Information & Consent Form

(***Please complete for the parent taking part in [redacted] Evaluation***)

Parent Name: _____ **Parent Age:** _____

Parent Education (please circle):

Primary Secondary Professional Training University Post-Graduate

Parent Occupation : _____ **Hours of Work/week:** _____

Ages of siblings (if any): Sibling 1 Aged: Sibling 2 Aged: Sibling 3 Aged:

Sibling 4 Aged:

Hours of Speech and Language Therapy received outside of [redacted] per month: _____**hrs**

Hours of other therapies per months: _____ **Name of other therapies:**

_____

Is your child attending a preschool programme? _____

Any other relevant details? _____

Consent:

1. I have read the information sheet about this study. **Yes / No**
2. I have received enough information about this study. **Yes / No**
3. I understand that I am free to withdraw from this study before, during or up to two-weeks after the intervention without giving a reason. **Yes / No**
4. I understand that this study is an evaluation of the [redacted] programme and whether I decide to take part or not will not have any bearing on my involvement in the [redacted] programme. **Yes / No**
5. I agree for me and my child to take part in this study. **Yes / No**
6. I give permission to video record the evaluation sessions. **Yes / No**
7. I give permission for the videos to be used for educational purposes within the Department of Speech and Hearing Sciences at UCC. **Yes/No**
8. I understand that anonymity will be ensured throughout the project. **Yes / No**
9. I give permission for my anonymised data to be kept and used for future research studies by researchers at UCC. **Yes / No**

Signature: _____

Date: _____

Name of Child: _____

Child's Date of Birth: _____

Email: _____

Appendix E

Pivotal Bahavior Rating Scale (Mahoney & Wheeden, 1998)

PIVOTAL BEHAVIOR RATING SCALE (Revised, 1998)

Gerald Mahoney

Note: The Pivotal Behavior Rating Scale has been reported in two published studies examining the influence of teachers' interactive style on the engagement of preschool-aged children with disabilities (Mahoney & Wheeden, 1998; 1999). Results from these studies indicated that the interactive behaviors measured by this scale are influenced by the way adults interact with children. These behaviors appear to be critical dimensions of children's active learning insofar as they are also related to children's level of developmental functioning. Factor analyses of these seven items indicate they measures two components of interactive behavior - Attention and Initiation. The following items have been arranged according to the scales they are associated with.

ATTENTION

1. **ATTENTION TO ACTIVITY: (Flittiness/Stay With-It-Ness)**.

This scale assesses the extent to which the child attends to activities. While the child may or may not be actively involved in the activity, the child rated as demonstrating high attention remains in the activity for an extended duration. The quality of the child's participation may be characterized as highly involved or uninvolved. In other words, the child may or may not appear to derive satisfaction from the activities. A child rated as low in attention may briefly participate in an activity and then physically remove herself or engage briefly in another activity. A child receiving a low rating in attention may frequently change or avoid activities, never seeming to attend to an activity for more than a few seconds at one time.

Rating of [1]: Very Low - The child never attends to a computer activity to more than a few seconds at a time. He or she may be completely inactive, avoidant of the activities, or may constantly change activities.

Rating of [2]: Low - The child can be described as generally inattentive for the activity. Although the child sometimes participates in the activity, she is more often inactive, avoidant of the activities, or engaged in changing activities.

Rating of [3]: Moderate - The child attends to the activities about as often as she does not. She has extended periods of time in which she participates in the activity as well as periods in which she is engaged in avoiding or changing activities.

Rating of [4]: High - The child "stays with" the activities during the majority of the session. She may have periods in which she is inattentive but these are short-lived and limited in number.

Rating of 5]: Very High - The child "stays with" the activities throughout the session. The child participates in the activities without periods of inattention.

2. **PERSISTENCE: (Practice/Problem Solving)**.

This scale measures the degree to which the child makes an effort to participate in activities. A child scoring high on persistence, makes several attempts at tasks when playing with the adult and continues to try solutions even though he may not successfully reach his or the adult's goal. Persistence also reflects the extent to which the child practices actions and vocalizations. A child receiving a high score may frequently perform the same action on same or different objects or practice vocalizations over and over again (imitate the adult or the computer's sounds/words). A child scoring low on the scale makes little effort to participate in the activities. He or she rarely practices behaviors or vocalizations and when encountering difficulty during an activity quickly gives up. Persistence is distinguished from compliance by being child-initiated as opposed to adult-initiated. In other words, if the child attempts to participate in an activity solely in response to the adult's request, this will be seen as compliance rather than persistence.

Rating of [1]: Very Low - The child never demonstrates repetition of a behavior. The child who is very low in persistence may never attempt a second try when having difficulty.

Rating of [2]: Low - The child infrequently demonstrates repetition of a behavior. She may occasionally make a second attempt when having difficulty but quickly gives up.

Rating of [3]: Moderate - The child has extended periods in which he or she seems to be practicing behaviors, but just as often has periods in which he does not practice. Similarly, there may be periods in which the child continues to try when having difficulty about as often as there are periods in which she quickly gives up.

Rating of [4]: High - Although the child has some periods in which she quickly gives up or during which repetition of behavior is rarely seen, in general, the child can be describe as high in persistence. She is often observed to practice behaviors or make second and third attempts when having difficulty.

Rating of [5]: Very High - The child frequently practices vocalizations or activities. He also may make repeated attempts at tasks when having difficulty. The child's persistence is a highlight of his behavior throughout the session.

3. **INVOLVEMENT: (DISTRACTIBILITY - Looking Around)**.

This scale reflects the intensity with which the child is involved in the activity. The child who is high in involvement is actively involved throughout the majority of the activity. This child appears to be highly motivated to engage in the activities regardless of whether they are adult or child initiated. He or she is intent on participating in the activities and seems to derive satisfaction from the activities. The child who is low in involvement is either passively involved during the activity, attempts to avoid participation, or is highly distractible during the activity. This child may "stay" with the activity but seems to derive little satisfaction from his or her involvement. This child may frequently look at the camera or leave the area.

Rating of [1]: Very Low - This child obviously does not derive satisfaction from his involvement in the activities. The child shows a great deal of neutral affect as well as some distress or avoidance of the activity. When the child does participate in the interaction, he seems to be "going through the motions" rather than actively participating. This child may be greatly distracted by other activities in the classroom.

Rating of [2]: Low - This child, for the most part, does not derive satisfaction from his participation in the activities. He may show largely

neutral affect and may appear passive during the interaction. His behavior may appear to be largely "rote" during the activities. Or this child may subtly or overtly demonstrate uninvolvement by being distracted during the majority of the session.

Rating of [3]: Moderate - The child derives some satisfaction from the activities. There are sustained periods in which he seems intent on what he is doing or uses gestures or vocalizations to express satisfaction with the activity. There are also extended periods in which the child seems to be "going through the motions" or is disinterested in the activities.

Rating of [4]: High - The child can be described as highly involved. During the majority of the session, the child appears to derive satisfaction from his participation in the activities.

Rating of [5]: Very High - The child is highly involved throughout the session. This child appears to be highly motivated to engage in the activities regardless of whether they are adult or child initiated. He or she derives a great deal of satisfaction from participating in the activities.

4. **COMPLIANCE/COOPERATION: (Does Child Avoid at All?)**

The degree to which the child attempts to comply with the requests or suggestions of the adult is measured using this scale. A child scoring high in compliance will make an effort to do what the adult asks or will respond quickly to the adult's subtle or overt suggestions. A child scoring low in compliance may refuse to cooperate with the adult. This child may actively avoid the activity by throwing materials or simply by ignoring the adults suggestions and engaging in other activities.

Rating of [1]: Very Low - The child may overtly demonstrate refusal to cooperate by throwing or pushing away materials, or may simply ignore the adult's suggestions.

Rating of [2]: Low - While the child may occasionally attempt to cooperate with the adult's suggestions, the child is not cooperative for the majority the interaction.

Rating of [3]: Moderate - The child attempts to cooperate with the adult's requests or suggestions about as often as he or she does not cooperate.

Rating of [4]: High - The child usually attempts to cooperate with the adult's requests or suggestions. He or she may occasionally refuse to cooperate but for the majority of the time attempts to follow the adults suggestions or requests.

Rating of [5]: Very High - The child consistently attempts to cooperate with the adult’s requests or suggestions. He or she responds quickly to both overt and subtle requests or suggestions.

INITIATION

1. **INITIATION: ACTIVITIES**.

This scale measures the extent to which the child initiates activities. A child who receives a high rating frequently attempts to initiate activities during the segment. (Examples of initiation: verbal initiation, start new game, change activities within game, ask for help. Child doesn't wait for adult guidance. Initiation is NOT trying the same thing over and over, ignoring opportunities to try something new). A child scoring low on this scale rarely attempts to initiate activities and may respond only to the adult's agenda rather than attempting to carry out her own agenda, or may appear uninterested in playing with the materials.

Rating of [1]: Very Low - The child almost never attempts to initiate activities. He or she may be extremely passive and inactive during the interaction or only engage in activities in response to the adult's requests.
Rating of [2]: Low - The child occasionally attempts to initiate activities. For the most part, however, the child either follows the adult's agenda or is very passive during the interaction.
Rating of [3]: Moderate - On several occasions the child attempts to initiate activities. There are also several periods in which the child is passive, uninvolved or responding only to the adult's agenda.
Rating of [4]: High - The child consistently attempts to initiate activities. Although the child initiates regularly, the child occasionally becomes uninvolved or passive during the interaction.
Rating of [5]: Very High - The child initiates activities throughout the session. He clearly has his own agenda and insists on following it.

2. **INITIATION: ADULT**.

The extent to which the child initiates interaction with the adult is measured using this scale. The child receiving a high rating, has frequent and lengthy bouts of eye-contact and other sharing behaviors such as vocalizations. This child tries to engage the adult by taking turns, or by using vocalizations, gestures and facial expressions. (Vocalizations may include: "come here", "your turn", "look at this"). A child scoring low in attention to adult may rarely have eye contact or attempt to share experiences or engage the adult by taking turns or through vocalizations or gestures.

Rating of [1]: Very Low - The child never attempts to share experiences with the adult. He or she never engages in periods of eye-contact or vocalization or attempts to engage the adult by showing or offering toys.

Rating of [2]: Low - The child occasionally attends to the adult by demonstrating eye-contact. For the most part, however, the child does not attempt to share experiences with the adult.
Rating of [3]: Moderate - The child attends to the adult approximately half of the time. He or she demonstrates periods of eye-contact or other sharing behaviors, but equally demonstrates periods of inattention. This child may also have extended periods of eye contact but more in response to the adult's behavior than in an attempt to engage the adult.
Rating of [4]: High - The child attends to the adult for the majority of the session. He or she is often observed to actively share experiences through eye-contact and vocalization and sometimes attempts to initiate activities with the adult.
Rating of [5]: Very High - The child has frequent and lengthy bouts of eye contact with the adult. He or she often vocalizes while looking at the adult or

attempts to share experiences by showing or offering toys or materials or otherwise initiating activities with the adult. The child is characterized by his frequent attempts to involve the adult.

3. **AFFECT.**

This scale reflects the child's general emotional state during the interaction. A child receiving a high score overtly demonstrates positive affect and enjoyment whether it be directed toward the adult or activity itself. This child may frequently smile, laugh or vocalize with the adult or during the activity. A child scoring low on this scale frequently demonstrates anger or distress during the interaction. He may cry, attempt to hit the adult, or throw materials and toys.

Rating of [1]: Very Low - The child demonstrates a great deal of distress during the interaction. He or she may cry, whine, or attempt to hit the adult or throw materials and toys.

Rating of [2]: Low - While the child does not demonstrate distress throughout the interaction, there are several sustained periods in which the child is distressed.

Rating of [3]: Moderate - The child, in general, displays low intensity enjoyment. Or this child can be generally characterized as sober or neutral in affect.

Rating of [4 High - For the most part, the child can be described as happy. He or she shows some neutral affect, but most often appears to be happy during the session.

Rating of [5]: Very High - The child often vocalizes, laughs or smiles when interacting with the toys or adult. He or she never demonstrates negative affect.

CHILD BEHAVIOR RATING SCALE (CBRS)
SCORING SHEET

CBRS ITEM	**Observation 1**	**Observation 2**	**Observation 3**	**Observation 4**
	Date _____	**Date** _____	**Date** _____	**Date** _____
ATTENTION				
1. Attention to Activity				
2. Persistence				

3. Involvement				
4. Compliance				
Scale Score (Att +Per + Inv + Com)/4				
INITIATION				
1. Initiation: Activity				

2. Initiation: Adult				
3. Affect				
Scale Score (Act + Adult + Aff)/3				
COMMENTS				

Appendix F

Parent Interaction and Language Rating Scale

Parent Interaction and Language Rating Scale

(Adapted and based on the Teacher Interaction and Language Rating Scale by Girolametto, Weitzman and Greenberg)

Instructions:

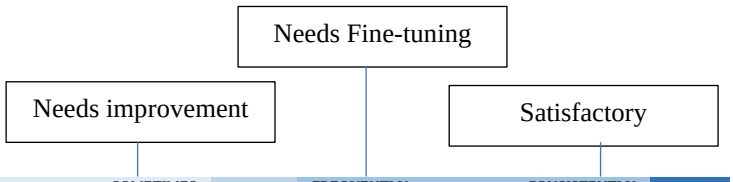
- Observe the videotaped interaction once all the way through without stopping. Make note of the frequency observations using the 'Comments' section. Use these comments to guide you in rating each of the 10 items on the rating scale.
- Observe the videotape a second time without stopping. Again, record your observations in the 'Comments' section. Then complete the rating scale of any of the 11 items you did not previously rate.
- On the 7-point scale, a rating of '**1**' indicates that the parent almost never uses the technique, whereas a rating of '**7**' indicates that the parent consistently uses the technique.
- Ratings of '**1-3'** indicate that the parent's use of the technique needs improvement and would be a therapy goal for future interactions.
- A rating of '**4**' indicates that fine tuning of the technique is required to achieve a rating of '5' or 'Frequently'. Therefore, this would be a therapy goal. If you think that the parent needs improvement on a particular technique, assign a rating of '3' or below. If the parent does not need improvement, assign a rating of '5' or above.
- A rating of '**5-7**' indicates that the parent's use of a technique achieves expectations. A rating of '5' or '6' is quite acceptable and should not be a goal for future therapy. However, improvement to a '7' is possible after the participation in the program if a parent is very motivated and makes outstanding changes across a number of techniques.
- Some items have 2 or more skills within their definition (e.g., Item 1, Wait and Listen includes both waiting and listening skills as skills to rate). If a parent uses some aspects of an item frequently (i.e., '5') and others only sometimes (i.e., '3'), you may assign and in between score (in this case, a '4').
- A rating of '**N/A**' should be rarely used. If there are very few examples of a technique or no examples of a technique you should use ratings from '1-3'. That is, the assumption is that opportunities for using the technique were missed. N/A should only be used in circumstances where:
    i. The technique is not appropriate to rate because of the activity (e.g., book reading is not conductive to joining in and playing), or
    ii. The child is beyond the age at which a particular technique is helpful (e.g., imitation for a preschool-aged child), or
    iii. The parent does not need to use the skill (e.g., the child is participating and interacting making certain items unnecessary).
- If rating a book-reading situation, do not rate the parent's reading of text since this does not constitute spontaneous communication.

- *Note:* if the child is using sign language, a picture communication system or some other AAC device, please interpret the words ‘gestures, sounds, words’ to include these forms of communication.

Parent Interaction and Language Rating Scale

Participant:

Date:



	ALMOST NEVER		SOMETIMES		FREQUENTLY		CONSISTENTLY	
1. Wait and Listen	1	2	3	4	5	6	7	N/A

Parent encourages the child to initiate verbally and/or nonverbally by:

- Waiting expectantly for initiations
- Using a slow pace which allows lots of time for child to initiate
- Listening to allow the child to complete their message

Comments:

	ALMOST NEVER		SOMETIMES		FREQUENTLY		CONSISTENTLY	
2. Follow the Child's lead	1	2	3	4	5	6	7	N/A

When the child initiates verbally or nonverbally, the parent follows their lead by:

- Responding verbally to their initiations
- Using animation
- Avoiding commands and vague acknowledgements (e.g. uh huh, yeah, that's right)

Comments:

	ALMOST NEVER		SOMETIMES		FREQUENTLY		CONSISTENTLY	
3. Join in and Play	1	2	3	4	5	6	7	N/A

Parent actively joins in the child's play as a partner by:

- Building on their focus of interest
- Playing without dominating

Comments:

	ALMOST NEVER		SOMETIMES		FREQUENTLY		CONSISTENTLY	
4. Be Face to Face	1	2	3	4	5	6	7	N/A

Parent adjusts their physical level by:

- Sitting on the floor or in child-sized chair
- Leaning forwards to facilitate face to face interaction
- If above child's level, bending to be close whenever possible

Comments:

	ALMOST NEVER		SOMETIMES		FREQUENTLY		CONSISTENTLY	
5. Encourage Turn-Taking	1	2	3	4	5	6	7	N/A

Parent encourages extended verbal turn-taking by:

- Linking comments and questions to invite the child to take turns
- Responding with animation
- Waiting expectantly for a response
- Balancing the number and length of adult to child turns
- Using sentence completion only with children at one-word stage

Comments:

	ALMOST NEVER		SOMETIMES		FREQUENTLY		CONSISTENTLY	
6. Gesture	1	2	3	4	5	6	7	N/A

Parent uses gestures to promote communication with the child by using:

- Pointing
- Giving
- Reaching
- Showing

Comments:

	ALMOST NEVER		SOMETIMES		FREQUENTLY		CONSISTENTLY	
7. Lámh	1	2	3	4	5	6	7	N/A

Parent employs the use of Lámh signs to promote communication with the child

- Speaks and signs
- Signs alone
- Uses hand over hand

Comments:

	ALMOST NEVER		SOMETIMES		FREQUENTLY		CONSISTENTLY	
8. Imitate	1	2	3	4	5	6	7	N/A

Parents imitates the actions, gestures, sounds or words made by the child

Comments:

	ALMOST NEVER		SOMETIMES		FREQUENTLY		CONSISTENTLY	
9. Use a Variety of Labels	1	2	3	4	5	6	7	N/A

Parent uses a variety of vocabulary (nouns, verbs, adjectives, adverbs) by:

- Emphasizing key words
- Repeating words
- Labeling objects, actions, attributes, events
- Avoiding non-specific words (e.g. it, this, that, there, thank you)

Comments:

Appendix G

Bilingual Language Questionnaire

Language Questionnaire

Name of child:
Gender:
DOB:
Chronological age:
Date:

Child's Language Exposure

What language(s) is your child exposed to and how often?

Language X:
Language Y:
Other Language:

	0 Never	**1 Rarely**	**2 Sometimes**	**3 Usually**	**4 Always**
Language X					
Language Y					
Other (specify)					

	Score/4
X	
Y	
Other	

2.) At what age did this exposure begin?

	Age in months
Language X	
Language Y	
Other (specify)	

Languages used with and by the child

What language(s) does the child's mother use with the target child?

Language Mother uses with CHILD						Language CHILD uses with Mother				
	0 Never	1 Rarely	2 Sometimes	3 Usually	4 Always	0 Never	1 Rarely	2 Sometimes	3 Usually	4 Always
Language X										
Language Y										
Other (specify)										

What language(s) does the child's father use with the target child?

Language Father uses with CHILD						Language CHILD uses with Father				
	0 Never	1 Rarely	2 Sometimes	3 Usually	4 Always	0 Never	1 Rarely	2 Sometimes	3 Usually	4 Always
Language X										
Language Y										
Other (specify)										

Does another adult regularly take care of your child (e.g. grandparent, babysitter, day care staff)? YES or NO
If YES, specify who this person is here _____ and complete the table below.

Language used by OTHER REGULAR CAREGIVER with CHILD						Language used by CHILD **with OTHER REGULAR CAREGIVER**				
	0 Never	1 Rarely	2 Sometimes	3 Usually	4 Always	0 Never	1 Rarely	2 Sometimes	3 Usually	4 Always
Language X										
Language Y										
Other (specify)										

Additional regular caregiver with child? _____

Language used by OTHER REGULAR CAREGIVER with CHILD						Language used by CHILD **with OTHER REGULAR CAREGIVER**				
	0 Never	1 Rarely	2 Sometimes	3 Usually	4 Always	0 Never	1 Rarely	2 Sometimes	3 Usually	4 Always
Language X										
Language Y										
Other (specify)										

What language(s) does the child's brother/sister use with the target child?

Language used by BROTHER/SISTER with CHILD						Language used by CHILD with BROTHER/SISTER				
	0 Never	1 Rarely	2 Sometimes	3 Usually	4 Always	0 Never	1 Rarely	2 Sometimes	3 Usually	4 Always
Language X										
Language Y										
Other										

Language used by BROTHER/SISTER 2 with CHILD						Language used by CHILD with BROTHER/SISTER 2				
	0 Never	1 Rarely	2 Sometimes	3 Usually	4 Always	0 Never	1 Rarely	2 Sometimes	3 Usually	4 Always
Language X										
Language Y										
Other										

Who does your typically child spend time with during the week?

	Weekday (waking hours per day)	**Weekend (waking hours per day)**
Mother		
Father		
Sibling		
Caregiver 1		
Caregiver 2		

What language activities is your child exposed to <u>each week</u> and in what language(s)?

	Language X			**Language Y**			**Other**		
Activities	0 Never or almost never	1 At least once a week	2 Every day	0 Never or almost never	1 At least once a week	2 Every day	0 Never or almost never	1 At least once a week	2 Every day
a. Sharing books									
b. Television									
c. Singing songs									
d. iPad/tablet									
f. Other									

<u>Language use among family members</u>

What language(s) are mainly spoken at home?

Language use AMONG FAMILY MEMBERS	√
Language X only	
Mostly Language X with some Language Y words	
Approximately equal proportions of Language X and Language Y	
Mostly Language Y with some Language X words	
Language Y only	
Language X, Language Y and additional language/s	