Evaluation of low-intensity therapy and parent training for young children w	vith autis	m
based on the early start Denver model		

by

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#### **ABSTRACT**

Autism spectrum disorder (ASD) is a pervasive developmental disorder that is characterised by deficits in social communication and restricted and repetitive behaviours, interests and activities. Recent developments in identification techniques mean that many children can be reliably diagnosed with ASD before the age of 2. Early identification creates the opportunity for early intervention. In fact, some research suggests that the earlier a child with ASD receives intervention, the greater the progress he or she is likely to make. Naturalistic developmental behavioural interventions are a relatively recent method of early intervention for children with ASD, which combine elements of previous intervention approaches (behavioural, naturalistic behavioural, and developmental/relationship-focused intervention). One such naturalistic developmental behavioural intervention is the early start Denver model (ESDM), which is designed for children with or at risk for ASD between the ages of 12 and 60 months (5 years). Research suggests that ESDM intervention may improve a range of child outcomes when delivered for at least 15 hours per week over at least 10 months. However, many families may not be able to access or afford such intensive intervention. Therefore, the two studies in this thesis evaluated the effectiveness of two ESDM delivery approaches that required relatively few hours of professional input per week. Specifically, low-intensity therapist delivered ESDM intervention, and ESDM parent training.

Study 1 used a multiple probe across participants design to evaluate the effectiveness of 3 hours per week of home-based ESDM therapy for 12 weeks for improving imitation, communication, and engagement for four young children with ASD. It also examined whether children showed increases in these outcomes with their mothers following the intervention. The results of this study suggest that, following the intervention, all four children increased their imitation skills and their engagement with the therapist. In addition three of the children had more functional utterances and one child increased his use of intentional vocalisations. These results were maintained four weeks after intervention and generalised to a lesser degree to each child's mother. This suggests that low-intensity therapist delivered ESDM intervention may improve outcomes for children with ASD.

The results of Studies 1 and 2 suggest that both low-intensity therapist delivered ESDM intervention and ESDM parent training may be promising intervention approaches for young children with ASD. This is particularly encouraging as both approaches involved relatively few hours of professional input per week. In theory, this could increase the number of families who are able to access such intervention. More research is needed to identify the most effective low-intensity ESDM intervention method, or combination of methods.

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#### **DECLARATION BY THE AUTHOR**

This thesis is comprised of my original work conducted for in fulfillment of a PhD under the supervision of Dr Larah van der Meer and Professor Jeff Sigafoos at Victoria University of Wellington. None of the research included within this thesis has been previously submitted for another degree or diploma. Research conducted by other authors has been carefully referenced in text.

The research included in this thesis was approved by the Victoria University of Wellington Human Ethics Committee under the project: Developmental, Relationship-Based Early Intervention for Children with Autism (Reference Number 22085, see Appendix J). Both my primary supervisor, Dr Larah van der Meer, and my secondary supervisor, Professor Jeff Sigafoos provided input into the design of Studies 1 and 2, assistance in the analysis of the data, and edits on all chapters. Dr Larah van der Meer also provided additional edits on the final manuscript. Aside from this input, the studies presented in this thesis were designed and conducted by me.

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#### CHAPTER 1

#### **AUTISM AND EARLY INTERVENTION**

#### **Historical Background**

In October 1938, a concerned mother and father brought their 5-year-old <sup>1</sup>son, Donald, to see Leo Kanner, a Ukrainian-born psychiatrist, for assessment (Kanner, 1943). Donald had an unusual ability to remember faces and names, could recite the alphabet before the age of two, and had perfect pitch. He also had frequent temper tantrums, loved to spin and throw objects, and was "happiest when left alone" (Kanner, 1943, p.217). Donald was the first of 11 children who would later be described by Kanner as having *infantile autism* (1943). In his 1943 paper, Kanner described these children as showing *extreme autism*, meaning that they ignored the external environment and, when possible, avoided social interaction. Each of these children also had *obsessive* behaviours, difficulties with communicating, anxieties and aversions, and an *obsessive insistence on the maintenance of sameness*. Kanner argued that these characteristics were symptomatic of a distinct disorder that was separate to *childhood schizophrenia* and *feeblemindedness*, which were common diagnoses at the time (1943).

Around the same time, Hans Asperger, an Austrian paediatrician, described a group of children who displayed a similar combination of behaviours and traits (1944). He called this condition *autistic psychopathy*. He also referred to these children as "little professors" due to their in-depth knowledge of certain specialist subjects (Silberman, 2015). They are now generally viewed as showing signs of higher cognitive and language functioning than the group described by Kanner (1943), but these children also had issues with the nuances of verbal and non-verbal communication, difficulties understanding the rules of social interaction, and they also showed repetitive behaviours and a restricted range of interests (Wing, 1981). It is often suggested that Kanner and Asperger's definitions of this disorder were developed independently, however, some researchers suggest that Kanner may have learned of the condition through collaboration with Asperger's former colleague (Silberman, 2015). Still, Asperger's work was comparatively less well known until it was translated into English by Lorna Wing in 1981.

Although Kanner and Asperger are the two researchers who are credited with formally identifying autism, this condition seems to have been described two decades earlier

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<sup>&</sup>lt;sup>1</sup> Ages for children under the age of five will be described in years or months, in line with the original source. Ages for children over five will always be described in years.

by a Russian psychiatrist named Grunia Sukhareva (as cited in Manouilenko & Bejerot, 2015). In 1926, she published a series of case reports describing six children with *schizoid psychopathology* (as cited in Manouilenko & Bejerot, 2015). Symptoms of this disorder included social isolation, *tic-like behaviours*, *pedantic rule-following*, and *talking in stereotypic ways* (Manouilenko & Bejerot, 2015). Manouilenko and Bejerot (2015) suggest that her work was not widely acknowledged at the time due to her gender, location and the fact that her work was not published in English.

There are even earlier descriptions of individuals who seem to have had symptoms resembling autism (Silberman, 2015). For example, in 1798, Jean Itard, a French physician, described the case of the *Wild Boy of Aveyron* who was found living by himself in the forest at the age of 11 or 12 (as cited in Wing, 1997). Itard described this boy as displaying limited eye contact, a lack of imitation skills, repetitive rocking back and forth, and an inability to form reciprocal social relationships. Several well-known historical figures may also have had autism. For example, Silberman (2015) suggested that Henry Cavendish, the British scientist who discovered hydrogen, had autism. Silberman describes Cavendish as an extremely shy individual who avoided social interaction whenever possible, wore unfashionable clothing, and went for a walk on the same route at the exact same time every day.

# **Diagnostic Criteria and Defining Characteristics**

Despite the possibility that researchers might have identified the condition now known as autism spectrum disorder as early as the 1920s (Maouilenko & Bejerot, 2015), a formal diagnostic category (i.e., infantile autism) was first included in the third edition of the Diagnostic and Statistical Manual in the 1980s (American Psychiatric Association, 1980). It was classed as a pervasive developmental disorder meaning that it was usually first diagnosed in infancy and persisted through the individual's lifetime (Matson & Horowitz, 2010). The diagnostic criteria for infantile autism were: (a) pervasive lack of responsiveness to other people; (b) gross deficits in language development; (c) peculiar speech patterns, if speech is present at all; (d) bizarre responses to the environment; and (e) an absence of delusions, hallucinations, loosening of associations, and incoherence as in schizophrenia. A child needed to meet all five of these criteria in order to receive a diagnosis of infantile autism.

Since 1980, the autism diagnostic criteria have undergone several iterations and name changes. For example, in the fourth edition of the DSM, infantile autism was renamed autistic disorder and was one of five pervasive developmental disorders that also included Rett's disorder, childhood disintegrative disorder, Asperger's disorder, and pervasive developmental disorder- not otherwise specified (PDD-NOS; American Psychiatric Association, 2000).

However, in the current edition (DSM-5), the pervasive developmental disorder category no longer exists (American Psychiatric Association, 2013) and autism is now classified as a neurodevelopmental disorder. All neurodevelopmental disorders are present early in development, and impair personal, social, academic, and/or occupational functioning. Further, the previous diagnoses of autistic disorder, Asperger's disorder and PDD-NOS have now been combined to form the diagnosis autism spectrum disorder (ASD). Thus, autism is now represented as a dimensional rather than categorical disorder. For example, it is assumed that those with high functioning autism (previously Asperger's disorder) differ only in severity from those with a greater degree of impairment rather than having a distinct disorder (Kamp-Becker et al., 2010; Kuriakose & Shalev, 2016).

In order for an individual to receive a diagnosis of ASD according to the DSM-5 criteria, he or she must present with impairments in two domains: social communication; and restricted, repetitive patterns of behaviours, interests, and activities. Each of these domains has several criteria within it. Table 1.1 delineates the diagnostic criteria for ASD in the DSM-5. The International Classification of Diseases and Related Health Problems (ICD-10) also provides diagnostic criteria for ASD (World Health Organisation, 1992). However these are similar to the DSM-5 criteria and are, therefore, not included in the table. According to these diagnostic criteria an individual must currently or by history meet all of the criteria in the domain A (social communication), and at least two of the criteria in domain B (restricted, repetitive patterns of behaviour, interests and activities) in order to receive a diagnosis of ASD. Individuals who meet the criteria for Domain A but not Domain B would receive a diagnosis of Social (Pragmatic) Communication Disorder rather than ASD (American Psychiatric Association, 2015). However, more research is needed to determine the diagnostic validity of the Social (Pragmatic) Communication Disorder diagnosis (Ozonoff, 2012). The individual also must not better meet the diagnostic criteria for intellectual disability or global developmental delay rather than ASD.

# Table 1.1

# **Autism Spectrum Disorder**

DSM-5 Diagnostic Criteria for Autism Spectrum Disorder

Diagnostic Criteria

- A. Persistent deficits in social communication and social interaction across multiple contexts, as manifested by the following, currently or by history:
  - 1. Deficits in social-emotional reciprocity, ranging, for example, from abnormal social approach and failure of normal back-and-forth conversation; to reduced sharing of interests, emotions, or affect; to failure to initiate or respond to social interactions.
  - 2. Deficits in nonverbal communicative behaviours used for social interaction, ranging, for example, from poorly integrated verbal and nonverbal communication; to abnormalities in eye contact and body language or deficits in understanding and use of gestures; to a total lack of facial expressions and nonverbal communication.
  - 3. Deficits in developing, maintaining, and understanding relationships, ranging, for example, from difficulties adjusting behaviour to suit various social contexts; to difficulties in sharing imaginative play or in making friends; to absence of interest in peers.
- B. Restricted, repetitive patterns of behaviour, interests, or activities, as manifested by at least two of the following, currently or by history:
  - 1. Stereotyped or repetitive motor movements, use of objects, or speech (e.g., simple motor stereotypies, lining up toys or flipping objects, echolalia, idiosyncratic phrases).
  - 2. Insistence on sameness, inflexible adherence to routines, or ritualized patterns or verbal nonverbal behavior (e.g., extreme distress at small changes, difficulties with transitions, rigid thinking patterns, greeting rituals, need to take same route or eat same food every day).
  - 3. Highly restricted, fixated interests that are abnormal in intensity or focus (e.g, strong attachment to or preoccupation with unusual objects, excessively circumscribed or perseverative interest).
  - 4. Hyper- or hyporeactivity to sensory input or unusual interests in sensory aspects of the environment (e.g., apparent indifference to pain/temperature, adverse

response to specific sounds or textures, excessive smelling or touching of objects, visual fascination with lights or movement).

- C. Symptoms must be present in the early developmental period (but may not become fully manifest until social demands exceed limited capacities, or may be masked by learned strategies in later life).
- D. Symptoms cause clinically significant impairment in social, occupational, or other important areas of current functioning.
- E. These disturbances are not better explained by intellectual disability (intellectual developmental disorder) or global developmental delay. Intellectual disability and autism spectrum disorder frequently co-occur; to make comorbid diagnoses of autism spectrum disorder and intellectual disability, social communication should be below that expected for general developmental level.

The DSM-5 also specifies the severity of ASD symptoms in terms of the level of support needed for each domain (American Psychiatric Association, 2015). For example, an individual with minimal impairment in the social communication domain would require support meaning that without supports in place he or she would have difficulty initiating interactions, trouble making friends, and/or may have decreased interest in social interaction overall. On the other hand, an individual who would require substantial support in this domain may have severe impairments in verbal and non-verbal communication skills such as limited to no spoken language and minimal response to social overtures from others. The same is true for the restricted and repetitive behaviours, interests, and activities domain. For example, an individual who requires support in this domain may have difficulty transitioning between activities and problems with planning and organisation, while an individual who requires substantial support may have extreme difficulty coping with change and may engage in a number of restricted/repetitive behaviours such as stereotyped movements, fixated interest, and avoidance of certain sensory inputs (e.g. the feeling of certain clothing or the texture of foods). The DSM-5 also specifies the presence or absence of additional impairments including intellectual impairment, language impairment, catatonia, and/or whether the diagnosis is associated with a known genetic or environmental factor such as Rett's syndrome.

ASD has a high level of co-morbidity with other disorders including obsessive compulsive disorder, attention-deficit/hyperactivity disorder, anxiety and mood disorders, epilepsy, and oppositional defiant conduct disorder (American Psychiatric Association, 2013;

Kim, Szatmari, Bryson, Streiner, & Wilson, 2000; Levisohn, 2007; Meier el., 2015; Reiersen & Todd, 2008; Simonoff et al., 2008). Intellectual disability (ID) is also considered to be a diagnosis commonly associated with ASD as it has been estimated that 40-70% of individual with ASD also have mild (IQ ~ 50-70) to severe ID (i.e., IQ < 50; La Malfa, Lassi, Bertelli, Salvini, & Placidi, 2004; Matson & Shoemaker, 2009). Further, many individuals with ASD develop challenging behaviours, such as aggression, self-injury and property destruction (Matson & Rivet, 2008).

Without effective early intervention, ASD has been associated with negative long-term outcomes including lower than expected academic success, limited friendships and peer relationships, and reduced participation in social/recreational activities in adolescence and adulthood (Estes, Rivera, Bryan, Cali, & Dawson, 2011; Orsmond, Krauss, & Seltzer, 2004). Further, many adults with ASD struggle to find employment, and although higher functioning individuals are often able to live independently, most continue to require support throughout their lives (Howlin, 2000).

#### **Prevalence**

It has been widely reported that the prevalence of ASD has increased substantially since the disorder was first described by Kanner in 1943 (Elsabbagh et al., 2012). For example, a 1966 evaluation of census data from 78,000 8- to 10-year-olds in the United Kingdom estimated the prevalence of ASD to be 4.5 per 10,000 individuals (Lotter, 1966). In contrast, a study conducted 43 years later, also in the United Kingdom which involved 3342 5- to 9-year olds, estimated the prevalence to be 157 per 10,000 individuals (Baron-Cohen et al., 2009). The newest estimates from the Centers for Disease Control (CDC) estimates that the prevalence of ASD in the United States is 1 in 68 individuals (Centers for Disease Control, 2014). These studies also found ASD was between 2.6 (Lotter, 1966) and 4.5 (Centers for Disease Control, 2014) times more common in boys than girls.

A recent literature review estimated the worldwide prevalence of ASD was 62 per 10,000 individuals and found that prevalence did not vary significantly between countries (Elsabbagh et al., 2012). However, the authors cautioned that this review contained prevalence estimates from a limited number of countries. Indeed, there appears to be no research into the prevalence of ASD in New Zealand (Ministries of Health and Education, 2008). Given the current New Zealand population of 4.5 million, and using the prevalence data from other studies, this suggests that the number of individuals with ASD in New Zealand could be between 28,000 (62 per 10,000 individuals) and 65,000 (1 in 68 individuals).

There are several possible explanations for the apparent increase in the estimated prevalence of ASD. First, it could be partially attributed to differences in study methodology including the methods of sampling, sources of data, age range of participants, and the method of diagnosis (Wing & Potter, 2002). Second, the diagnostic criteria for ASD has become broader over time, which could mean that a larger number of individuals now meet this criteria (King & Bearman, 2009). Third, professional and public awareness of ASD seems to have increased, which could mean there is increased screening and a greater chance of identifying a larger number of individuals (Wing & Potter, 2002). Fourth, research suggests that the increase in ASD diagnosis corresponds with a decrease in the prevalence of other disorders such as intellectual disability and learning disabilities (King & Bearman, 2009). This suggests that there might be some diagnostic substitution occurring, that is, the replacement of one diagnosis with another. Finally, the increase in ASD might reflect a true increase is the number of individuals with ASD. If this is the case, it remains unknown what might be responsible for this increase (Wing & Potter, 2002).

## **Aetiology**

ASD has been described as a "heterogeneous neurodevelopmental syndrome" for which there is no one unifying cause (Dawson, 2008; Geschwind & Levitt, 2007). In fact, some researchers suggest that ASD should be viewed as a combination of "syndromes" with a variety of underlying genetic and environmental causes (e.g. Geschwind & Levitt, 2007). There is evidence to suggest that ASD has a genetic basis. For example, in one study of identical and non-identical twins, Bailey et al. (1995) found that if one identical twin had ASD then there was a 92% chance that the other twin also had ASD. However, if a non-identical twin had ASD, then the chance that the other twin had ASD fell to 10%. There is also evidence that siblings (rather than twins) of children with ASD are more likely to receive a diagnosis of ASD than children who do not have a sibling with ASD, although estimates of this likelihood vary from a 3% to 18.7% (Bolton et al., 1994; Ozonoff et al., 2011). A number of genes have been implicated in increasing an individual's susceptibility to developing ASD (Dawson, 2008). These include genes related to the development of the cerebellum, serotonin reuptake, and functioning of the synapses (see Dawson, 2008, for a review).

There is also evidence that the brains of individuals with ASD develop differently from those without ASD, although none of these differences occur across all individuals (Geschwind & Levitt, 2007). For example, research suggests that some individuals with ASD have poor interconnectivity between brain regions, meaning that different regions of their brain are less able to "communicate" with one another (Murias, Webb, Greenson, & Dawson,

2007). This may make it more difficult for individuals with ASD to perform complex tasks. Similarly, several studies have found that many children with ASD have greater than average head growth in the first 2 years of life (Courchesne & Pierce, 2005). It is hypothesised that this is because unnecessary neurons are not "pruned" when they are no longer used, which leads to reduced neural organisation. However, it is not clear if differing brain development is a cause or a result of ASD.

Some children with ASD also seem to have reduced activity in areas of their brain related to social interaction and social motivation. Research by Dawson et al. (2002) found that children with ASD had reduced brain responses when viewing familiar faces and other social stimuli compared to children without ASD or developmental disabilities but that there was no difference between these two groups of children when viewing objects. Further, research suggests that, for some children with ASD, the areas of the brain related to the perception of social stimuli may not be well connected to areas related to reward (Dawson, Webb, & McPartland, 2005). Thus, for children with ASD, social stimuli may have a lower reward value than objects. This might help to explain why they are less motivated to attend to social stimuli. Due to this decreased motivation, children with ASD may then develop impairments in other more advanced social skills such as joint attention, imitation, and sharing emotions (Dawson, 2008).

Further, some authors suggest that many children with autism have dysfunctions within their mirror neuron system. This system consists of several brain structures and activates both when an individual carries out an intentional action or when the individual observes someone else carrying out an action (Williams, Whiten, Suddendorf, & Perrett, 2001). These dysfunctions may provide a plausible explanation for the low rates of imitation shown by many young children with ASD compared to typically developing young children (Rogers & Dawson, 2010). As young children generally acquire many new skills through imitating others, it is hypothesised that these deficits in imitation can lead to delays across all areas of development.

#### **Early Identification**

Differences between children with and without ASD may emerge very early in life (Dawson, 2008). Research suggests that many infants who will later receive a diagnosis of ASD show some behavioural markers within the first year of life (e.g. Barbaro & Dissanayake, 2010; Chawarska, Macari, & Shic, 2013; Iverson & Wozniaki, 2007; Jones & Klin, 2013; Maestro et al. 2002), and can often be "reliably diagnosed" before 2 years of age (Barbaro & Dissanayake, 2010; Chawarska, Klin, Paul, & Volkmar, 2007). This research is

typically based on the retrospective analysis of home videos of children who later received a diagnosis of ASD or the prospective analysis of infant siblings of children with ASD (highrisk siblings; Chawarska et al., 2013; Dawson, 2008). This is due to the increased likelihood that siblings of children with ASD will also have ASD (Ozonoff et al., 2001). For example, Chawarska et al. (2013) compared the eye gaze responses of 6-month-old high-risk siblings when viewing videos of social scenes to the responses of infants with no immediate relatives with a diagnosis of ASD (no-risk controls). Specifically, these scenes entailed an actor making a sandwich and using infant directed speech in an attempt to engage the child. The results of this study suggested that the infants who were later diagnosed with ASD spent less time looking at the actor and particularly looking at her face, than those who were not diagnosed with ASD. In another study, Iverson and Wozniaki (2007) compared videotapes of high-risk siblings who were aged between 5 and 14 months, to those of "no-risk" controls. They found that, compared to the "no-risk" controls, the high-risk siblings were more likely to have delayed development of motor skills and prelinguistic behaviours (e.g. reduplicated babbling such as "ba-ba" or "da-da"). Other potential behavioural markers in infants who will later be diagnosed with ASD include lack of eye contact (Jones & Klin, 2013; Osterling, Dawson, & Munson, 2002), failure to respond to name (Osterling et al., 2002), and decreased joint attention (Maestro et al., 2002). However, no one behavioural marker is present in all infants who will later be diagnosed with ASD, which reflects the complexity and variation that is inherent to this disorder (Geschwind & Levitt, 2007).

These early behavioural indicators have been incorporated into a number ASD screening tools which help to identify children as young as 16 months old who are "at-risk" for developing ASD (e.g. Barbaro, & Dissanayake, 2010; Baron-Cohen, Allen, & Gillberg, 1992; Robins, Fein, Barton, & Green, 2001, Rutter, Bailey, & Lord, 2003; Schopler, Van Bourgondien, Wellman, & Love, 2010). There are two levels of screening tools: level one screeners, which are designed to screen the entire population for ASD and can be quickly administered, and level two screeners which are typically used with children who may be "atrisk" for ASD, such as high-risk siblings (Kuriakose & Shalev, 2016). Level one screeners generally have high sensitivity, meaning that they accurately identify the majority of children who are at risk for ASD, but lower specificity, meaning that they do not always exclude children who are not at risk for ASD. Parents perceive that it is worse to "miss" a child who will later receive a diagnosis (low sensitivity) than to identify a child as "at-risk" when he or she will not receive a diagnosis (low specificity; Barton, Dumont-Mathieu, & Fein, 2012).

Examples of common level one screeners include the Checklist for Autism in Toddlers (CHAT; Baron-Cohen et al., 1992) which is designed for children 18 months and older, and the newer Modified Checklist for ASD in Toddlers (M-CHAT; Robins et al., 2001) which is designed for children between 16 and 30 months. Both the CHAT and the M-CHAT are parent questionnaires which can be administered in 5 to 10 min, although the CHAT also includes a clinical observation component. These questionnaires assess the presence of behaviours such as pointing, showing, eye contact, functional play, and pretend play. To the author's knowledge there are no empirically validated level two screeners for children under the age of two, however the Social Communication Questionnaire (SCQ; Rutter et al., 2003) and the Childhood Autism Rating Scale- second edition (CARS 2; Schopler et al., 2010), are both used with children who are 2 years or older. The CARS 2 is one of the most commonly used measures and has high sensitivity and specificity (Schopler et al., 2010). It is an 18-item clinician behavioural checklist which includes items related to verbal and non-verbal communication, imitation, anxiety, social relationships, and activity level.

In 2010, Barbaro and Dissanayake trained 184 maternal and child health nurses to monitor particular behavioural items related to ASD during each child's regular community health checks at 8, 12, 18 and 24 months. These behavioural items related to social attention and communication and varied depending on the child's age. For example, at the age of 8 months the nurses observed whether the children could play peek-a-boo, and at 24 months they observed whether they engaged in parallel play. In total 20,770 children participated in the study and, of these children, 216 were referred for further assessment. Of the 110 children who did undergo further evaluation, 89 received a diagnosis of ASD. They estimated that the sensitivity of the screening process was relatively high (69% to 83.8%) and that the specificity was very high (99.8% to 99.9%), which is different from the majority of other screening tools. Results of this study suggest that nurses are able to use screening tools with large populations of children, and that many children can be identified before the age of 2 using this procedure.

Once a screening tool has been used to identify a child as at-risk for ASD, diagnostic tools are used to confirm (or disconfirm) the results of the screener. Two of the most common such tools are the Autism Diagnostic Interview-Revised (ADI-R; Lord, Rutter, & Le Couteur, 1994) and the Autism Diagnostic Observation Schedule- Second Edition (ADOS-2; Lord, Rutter, et al., 2012). Both of these tools have modified versions with high sensitivity and specificity that can be administered with toddlers who are 12 months and older (Hus & Lord, 2014; Kim & Lord, 2012; Lord, Luyster, Gotham, & Guthrie, 2012; Kim, Thurm, Shumway,

& Lord, 2013). The toddler version of the ADI-R includes 35 items in addition to the 93 items from the standard interview and takes between 90 and 250 min to administer via a parent interview conducted by a trained examiner (Kim & Lord, 2012). It contains items related to: early development; communication; reciprocal interactions; restricted, repetitive behaviours and interests; and general behaviour. The toddler version of the ADOS-2 is a 30-60 min play-based observational assessment, in which trained examiners deliver a range of "presses" in an attempt to elicit social and communicative behaviours from the child (Lord, Luyster et al., 2012). The toddler modules of the ADI-R and the ADOS-2 summarise results in terms of *little-or-no*, *mild-to-moderate*, *or moderate-to-severe* concern because, due to the difficulty in providing diagnosis at such a young age, information from these diagnostic tools must be combined with clinical judgement before giving the child a formal diagnosis of ASD.

The autism observation scale for infants (AOSI; Bryson, McDermott, Rombough, Brian, & Zwaigenbaum, 2000) was developed for use with infant siblings of children with ASD who are aged between 6 and 18 months. It is a 20-min play-based observational assessment which evaluates the infant's social, affective, communication, visual, and motor responses to presses from a trained administrator. The AOSI is not currently recommended for clinical or diagnostic use due to low sensitivity but it has provided valuable information about the early behavioural markers of children who will later receive a diagnosis of ASD (Kuriakose & Shaley, 2016).

The existence of multiple accurate (high sensitivity and specificity) screening and diagnostic tools for young children with ASD has led several organisations to recommend routine autism screening for all children starting from the age of 18 months (Johnson & Myers, 2007; Ozonoff et al., 2011). Despite these recommendations, estimates of the average age of diagnosis in the United States range from 48 to 58 months (Centres for Disease Control, 2014; Shattuck et al., 2009) and around 49 months in Australia (Bent, Dissanayake, & Barbaro, 2015). Further, a retrospective study of 1300 parents of children with ASD in the United Kingdom found that, on average, parents first became aware of problems with their child's development around 18 months but that children did not receive a diagnosis until 4.5 years later (Howlin & Moore, 1997). Parents expressed frustration and a lack of satisfaction with the length of time between expressing concerns and receiving a formal diagnosis. There do not appear to be any estimates of the average age of diagnosis in New Zealand.

There are several factors which may affect the age of diagnosis (Kuriakose & Shalev, 2016). Research suggests that children are likely to be diagnosed later than average if: (a) they are female (Shattuck et al., 2009), (b) they have less severe autism symptoms (Mandell,

Novak, & Zubritsky, 2005), (c) they belong to an ethnic minority (Mandell, Listerud, Levy, & Pinto-Martin, 2002), (d) their family is near the poverty level (Mandell et al., 2005), or (e) they live rurally (Mandell et al., 2005). Further, professionals often report a lack of knowledge or trust in screening tools and a lack of training in ASD diagnosis (Morelli et al., 2014). These findings highlight the importance of further training of primary care providers and also increasing the accessibility of assessment and diagnosis for all individuals regardless of gender, symptom severity, ethnicity, family income or location.

# **Early Intervention**

The ability to identify children with or at risk for ASD when they are very young means that intervention can also begin earlier (Dawson & Bernier, 2013). In the words of Vismara, Colombi and Rogers (2009, p. 110): "the goal of diagnosis is treatment". Some research has suggested that the earlier a child with ASD receives intervention, the more progress he or she is likely to make (e.g. Bibby, Eikeseth, Martin, Mudford, & Reeves, 2002; Flanagan, Perry, & Freeman, 2012; Granpeesheh, Dixon, Tarbox, Kaplan, & Wilke, 2009; Harris & Handleman, 2000; Perry, Cummings, DunnGeier, Freeman, Hughes, & Managhan, 2011). However, other studies have found that children's age did not predict their response to intervention (Eikeseth, Smith, Jahr, & Eldevik, 2002; Klintwall, Eldevik, & Eikeseth, 2015; Magiati, Charman, & Howlin, 2007). It is not clear precisely why these studies found different results, although some authors attribute these disparities to factors such as varying (a) outcome measures, (b) settings, (c) symptom severity and presentation, and/or (d) research designs (e.g., Eikeseth et al., 2002; Fava & Strauss, 2014; Lang, Hancock, & Singh, 2016).

Most early intervention approaches are generally targeted at children with or at risk for ASD who are under the age of 5 or 6 (Perry et al., 2011). However, the youngest participants in most early intervention studies are around 2 years of age (Eikeseth, 2008). It is possible that intervention for infants with ASD who are under 2 years of age may be even more effective (Bradshaw, Steiner, Gengoux & Koegel, 2015). At least 10 studies have investigated the effectiveness of training parents to implement interventions for their infants with or at risk for ASD (see Bradshaw et al., 2015 for a review). The majority of these studies reported that, following intervention, most parents learned to implement the intervention techniques and most children's social communication and developmental skills improved. In one such study, seven parents were trained to implement modified early start Denver model procedures (ESDM; see Chapter 3) with their infants at risk for ASD (Rogers et al., 2014). Results of this study suggested that when these children were between 18 and 36 months old

they had fewer ASD symptoms and higher developmental scores than a comparison group who did not receive the intervention. Although the results of these studies are promising, much more high quality research is needed to determine whether very early intervention is more effective.

There are several theoretical reasons why early intervention may be particularly effective. First, there is evidence to suggest that the brain is more flexible, or "plastic" very early in life (Holland et al., 2014). The first few years of life can be described as a "critical period" for brain development, which is characterised by rapid brain growth, cortical specialisation, and the development of areas devoted to language and social learning (Courchesne & Pierce, 2005; Dawson, 2008; Holland et al., 2014). Environmental experiences have a large impact upon how the brain develops during this time (Fava & Strauss, 2014). Therefore, effective early intervention during this critical period may help to alter the brain while it is most malleable.

The social motivation hypothesis refers to research which suggests that areas of the brain related to "social reward" are less active in children with ASD (Dawson, 2008).

Therefore, early intervention that focusses on increasing children's attention to faces and developing positive social interactions may alter the brain and increase the reward value of social stimuli (Fava & Strauss, 2014; Rogers et al., 2014). This should increase the child's motivation to attend to social stimuli, which may allow him or her to learn more advanced social skills (Dawson, 2008). Research suggests that young children who received 2 years of intensive intervention which emphasised positive social interactions and fun sensory social games (ESDM; see Chapter 3), had similar activation in brain areas associated with attention to faces as typically developing children (Dawson, Jones et al., 2012). However, they did not show increased activation in brain areas associated with perceptual processing of faces or orienting towards the stimulus. The results of this study suggest that the intervention was effective in ameliorating some but not all of the brain processes associated with social interaction in young children with ASD.

It is hypothesised that, due to their deficits in social communication, and restricted and repetitive behaviours and interests, children with ASD are exposed to many fewer learning opportunities each day than typically developing children (Lang et al., 2016; Rogers & Dawson, 2010). This, in turn, causes them to fall further and further behind their peers. For example, if a child's main form of toy play is spinning the wheels of a toy car repetitively, it is unlikely that his peers will approach him to play and, therefore, he will not learn new play skills (Rogers & Dawson, 2010). Early interventions that target foundational skills such as

social motivation and attention, imitation, and functional toy play, may provide children with ASD with access to many more learning opportunities (Lang et al., 2016; Rogers & Dawson, 2010). The earlier children learn these foundational skills, the less "catching up" they will need to do with their peers. For example, if a child learns to roll a car back and forward with a peer, it is likely that his peers will be more motivated to play with him and he may learn new play skills such as crashing the car, making car sounds, and more advanced pretend play.

The effectiveness of early intervention might also be understood in terms of developmental trajectories, which are a representation of a child's changes in learning rate (Klintwall et al., 2015). Learning rate is a measure of a child's improvement in age equivalent scores for developmental outcomes over time (Klintwall et al., 2015). For example, if a child is speaking in two words utterances then he has an expressive language age equivalence of 2 years. If, after a year, he is consistently speaking in three to four word utterances then he would have an expressive language age equivalence of 3-years-old. As he has aged a year and his language age equivalence has also improved by a year, he has an expressive language learning rate of 1.0 (change in age equivalence/time). Children who have learning rates of less than 1.0, fall further and further behind their peers over time.

Effective intervention should improve children's learning rate (Klintwall et al., 2015). The earlier an intervention occurs, the more likely it is that these changes in learning rate will mean that that the child's developmental trajectory will "lift the child back into the zone of normal development" (Klintwall et al., 2015 p. 59). Figure 1.1 provides a theoretical example of the effect of very early intervention compared with the effect of intervention for an older child. Prior to intervention, Child A and B both have a learning rate of 0.5, which increases to 1.25 following intervention. However, Child A receives intervention at the age of one, whereas Child B receives intervention at the age of four. Within 2 years Child A has "caught up" with his peers, whereas Child B's developmental age is still considerably lower than his peers. This explains why very early intervention may theoretically help to "prevent" children at risk for ASD from ever receiving a diagnosis (Dawson, 2008).

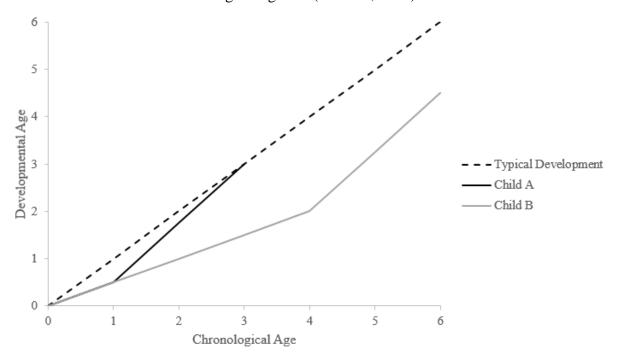


Figure 1.1. Hypothetical developmental trajectories for a child who receives intervention at the age of one compared with the age of four.

Although some research suggests that younger children with ASD tend to have better outcomes during intervention than older children, it is also important to consider the effectiveness of different types of early intervention approaches. This is because some types of early intervention may be more effective than others, or more suited to children with particular characteristics. Chapter 2 will describe the evidence-base for different types of early intervention and also moderators of treatment effectiveness.

# **CHAPTER 2**

# METHODS OF EARLY INTERVENTION FOR CHILDREN WITH ASD Overview

This chapter summarises four methods or approaches of providing early intervention to children with ASD and provides an overview of the evidence for the effectiveness of each method. These four methods are behavioural intervention, naturalistic behavioural intervention, developmental intervention, and naturalistic developmental behavioural intervention, which combines elements of the other three types. Each method has different underlying theories about the most effective approach for intervening on the core symptoms of ASD. Figure 2.1 illustrates how the methods influence one another and provides examples of the specific (i.e. "branded") models that use each method. This list of models is not exhaustive but does include the most commonly researched examples from each method.

# **Comprehensive Interventions**

Intervention methods for young children with ASD can be further separated into comprehensive interventions and focused interventions (Eikeseth, 2008; Odom, Boyd, Hall, & Hume, 2010; Wong et al., 2015). Comprehensive interventions, such as early intensive behavioural intervention (EIBI), target the core deficits that characterise ASD including impairments in language, cognition, play, and social skills (Odom et al., 2010; Rogers & Vismara, 2008). Other defining features of comprehensive interventions include: (a) descriptions of the intervention procedures in a manual or procedural guide with sufficient detail to allow replication, (b) the use of a clear conceptual framework, (c) intensive implementation (recommended 25+ hours per week), and (d) implementation for a long duration (recommended 10+ months; Odom et al., 2010). In contrast, focused interventions target one or more specific behavioural outcomes, and are typically less intensive and shorter in duration (Odom et al., 2010). Examples of focused treatments include the Picture Exchange Communication System (PECS; Frost & Bondy, 2002), Social Stories<sup>TM</sup> (Gray & Garand, 1993), and functional communication training (FCT; Carr & Durand, 1985). These programmes typically only target one area of impairment such as language or social skills (Odom et al., 2010). This chapter will predominantly focus on comprehensive interventions for young children with ASD.

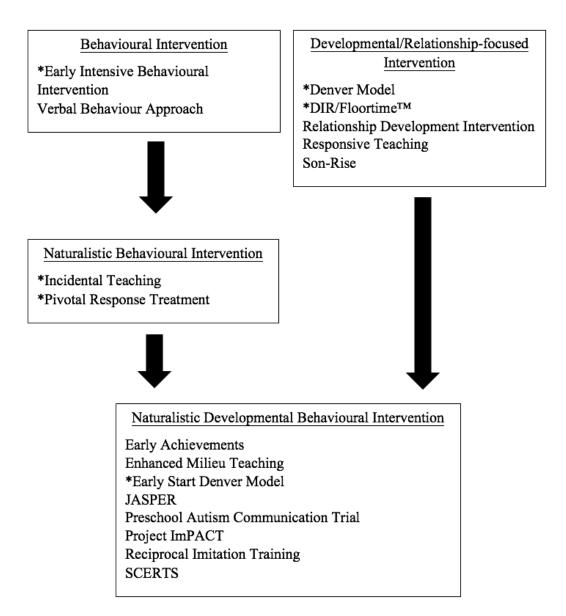


Figure 2.1. Overview of early intervention methods and specific programmes representative of each method. Note: \*Indicates a model of intervention which is described in detail within this thesis.

### **Evidence-based Practice in ASD**

When selecting a model of intervention for young children with ASD, one should consider whether or not there is high quality evidence for the effectiveness of this model. Evidence-based practice (EBP) refers to the process of integrating the best available research evidence with one's clinical expertise and stakeholders' perspectives to promote specific educational, health, or therapeutic outcomes in an individual (Frederickson, 2002). This term is also used to refer to specific procedures, programmes or interventions that have been shown to be consistently effective across several high quality empirical studies. Such procedures can be considered evidence-based practices. Thus EBP is both a process and a

reference to empirically-validated interventions (Reichow, Volkmar, & Cicchetti, 2008)

It is recommended that parents, caregivers, educators, and other professionals consider the research evidence for the effectiveness of a particular intervention before choosing to implement it with a child (National Autism Centre, 2015). This is because EBPs have been shown to result in objective improvements for the majority of children who have received the intervention and these finding have been replicated several times with different participants (Travers, Ayers, Simpson, & Crutchfield, 2016). Further, although models of intervention with a strong evidence base may not always be effective for a particular child or in every circumstance, they are perhaps less likely to be dangerous or cause harm to the child (Travers et al., 2016). On the other hand, there are some interventions which promise incredible progress or even a "cure" for ASD but for which: (a) there has been very little high-quality research conducted on their effectiveness, or (b) the high-quality research that has been conducted suggests that the intervention is not effective. These fad, pseudoscientific or *controversial* interventions often result in very few measurable gains in functioning, and may sometimes cause harm to the individual (Travers et al., 2016). Examples of seemingly popular interventions with a limited evidence-base include facilitated communication, the rapid prompting method, sensory integration therapy, the Gluten-Casein-free diet, and chelation therapy (Travers et al., 2016).

There are two key challenges when developing methods to evaluate the quality of evidence for different interventions for children with ASD (Reichow et al., 2008). The first is to develop a clear operational framework for evaluating both the quality of individual research studies and the number of high quality studies needed for a model to qualify as an EBP (Reichow et al., 2008). The second is to develop a framework which allows for the inclusion of single-case research as it is a very common type of experimental design that is widely used in the field of autism intervention. The National Autism Centre (2015) and Reichow et al. (2008) have both developed methods of determining EBP which address these two challenges.

The National Autism Centre (2015) criteria states that an "established EBP" has been shown to have beneficial intervention effects in at least two studies with a high quality group design or four studies with a high quality single-case design. "High quality" refers to studies that: (a) have a research design which includes random allocation to groups (group design) or several replications of the intervention effect (single-case), (b) include dependent variables that are measured using either standardised assessments or observational tools with high inter-observer agreement, (c) have replicable independent variables that are implemented

with at least 80% accuracy, (d) include participants who were diagnosed with ASD by a qualified ASD professional, and (e) include data on the generalisation and maintenance of intervention effects.

The Evaluative Method for Determining Evidence-Based Practices in ASD (Reichow, 2011; Reichow et al., 2008) states that a specific number of group and single-case studies with adequate or strong research rigor/quality are needed for an intervention to be considered an EBP (i.e., at least two group designs of strong research report strength conducted in different geographic locations or at least five single subject studies of strong research report strength, conducted by at least three different research teams, in at least three different locations, with a total sample size of at least 15 participants). Using this approach, each study is given a rating of "strong", "adequate" or "weak" depending on the number of primary and secondary quality indicators that it meets. Specifically, the quality rating is based on the quality/description of several primary and secondary quality indicators which vary depending on the study design (see Table 2.1).

Table 2.1

Primary and Secondary Quality Indicators from Reichow's Evaluative Method

Indicator	Group Design	Single Case
Primary	Participant characteristics	Participant characteristics
	Independent variable	Independent variable
	Comparison condition	Baseline condition
	Dependent variable	Dependent variable
	Link between research question and	Visual analysis
	analysis	Experimental control
	Use of statistical tests	
Secondary	Random assignment	Interobserver agreement
	Interobserver agreement	Kappa
	Blind raters	Blind raters
	Fidelity	Fidelity
	Attrition	Generalisation/maintenance
	Generalisation/maintenance	Social validity
	Effect size	
	Social validity	

The following sections will describe each method of early intervention and models which use this method, and will use the National Autism Centre guidelines (2015), Reichow's evaluative method (2011; Reichow et al., 2008), and any other relevant literature reviews, to determine the amount of evidence for the effectiveness of each model and method.

# **Behavioural Early Intervention**

Prior to the 1960s, it appears that some theorists and medical professionals believed that children with ASD were "untreatable" (Schreibman et al., 2015). This assumption began to change due to the increased use of interventions based on operant conditioning principles (Dawson & Bernier, 2013). Briefly, Skinner (1938) was among the first to systematically study what later became known as operant conditioning. His research and that of subsequent investigators (Sidman, 1953; 1962; Skinner, 1953) led to the formulation of a number of principles (e.g., reinforcement, punishment, extinction, stimulus control) of operant conditioning. In the initial operant conditioning research (Skinner, 1938) the basic paradigm was to place a hungry rat or pigeon inside an "operant conditioning chamber". In this chamber, the rat or pigeon could gain access to food by, for example, pressing a lever. By delivering food for closer and closer approximations to lever pressing or key pecking, Skinner (1953) discovered that the rats and pigeons would eventually learn to make these responses and appeared to have learned to do so because of the resulting access to the food. Thus, the delivery of food increased the likelihood that the rat would press the lever. Skinner (1953) named this effect "positive reinforcement", which refers to an increase in the probability that a behaviour will recur when it is followed by a specific type of consequence. When such a relation is observed the consequence can be defined as a reinforcer for that specific behaviour.

In other experiments (Sidman, 1953; 1962) a rat or pigeon in an operant conditioning chamber would receive intermittently delivered electric shocks. These shocks could be terminated by the rat or pigeon when a specific response occurred, such as pressing a lever or pecking a key. This relation was referred to as "negative reinforcement" and refers to a situation in which the removal of an aversive stimulus (electric shock) is contingent upon a specific response and consequently that response becomes highly likely to occur whenever the aversive stimulus is presented.

Through further research additional operant conditioning principles were formulated, including "punishment" (Alberto & Troutman, 2009; Catania, 2017; Skinner, 1953).

Punishment refers to a relation in which a response becomes less likely to occur when it is

followed by certain (punishing) consequences. "Extinction", in contrast, refers to a relation in which the probability of a previously reinforced response gradually decreases when that response is no longer followed by a (reinforcing) consequence. Another principle of operant condition is "antecedent control". This refers to control over responding by a stimulus (the discriminative stimulus [S<sup>D</sup>]) in which a response is more likely to occur when a specific stimulus is present compared to when that stimulus is absent. The stimulus is said to set the occasion for the behaviour to be reinforced.

Operant conditioning techniques were initially used with typically-developing children (e.g. Long, Hammack, May, & Campbell, 1958), however, in 1961 Ferster and DeMyer (1961) used a positive reinforcement procedure to teach new behaviours to children with ASD. In this study, an 8-year-old boy and a 9-year-old girl with ASD learned to press a key on a vending machine in order to gain access to lollies or a coin, which could be redeemed for other presumed reinforcers, such as access to music and a pinball machine.

Operant conditioning principles were subsequently used to teach more advanced skills to children with ASD. For example, in 1967 Risley and Wolf, taught four children with ASD between the ages of 7 and 12 to use functional speech instead of echolalia through a process of reinforcement, prompting, and prompt fading (see Discrete Trial Training for a description of these techniques). Specifically, the therapist gave each child a bite of a preferred food each time he or she imitated the name of an object (e.g. car), the therapist then faded the prompts until each child was able to name the object independently. The process of prompt fading involved the therapist first saying the whole word (e.g. "car"), then part of the word (e.g. "ca"), then mouthing the word without saying anything, and finally offering no prompt at all.

# **Discrete Trial Training**

The results of these initial behavioural studies suggested that children with ASD were able to learn functional skills when they were taught in highly structured environments using powerful extrinsic reinforcers. Lovaas (1987) suggested that the key to improving outcomes for children with ASD was to use the principles of operant conditioning to intervene intensively upon a wide range of behaviours as early as possible. He argued that the most important skills to teach included attending, imitating sounds and movements, matching, and complying with basic directions. Lovaas' comprehensive intervention approach for children with ASD is often incorrectly referred to as discrete trial training (DTT). However, DDT refers only to the manner in which skills are taught. That is, DTT involves teaching skills one at a time in "discrete trials". These are learning opportunities which have four components:

(a) a clear antecedent/ S<sup>D</sup>, (b) a single discrete response from the child, (c) a reinforcing

consequence for correct responses and some type of prompt or error correction for non-responding or incorrect responses, and (d) a brief inter-trial interval before presenting the an antecedent/ S<sup>D</sup> again to signal the start of the next trial (Lerman, Valentino, & LeBlanc, 2016). The first three components are also referred to as the "three term contingency" or ABC (antecedent, behaviour, consequence) format. A DTT teaching session includes a number of learning opportunities/trials [e.g., 10-20 opportunities] which are presented in rapid succession.

Prompts are defined as "antecedent stimuli that increase the probability of a correct response in the presence of the S<sup>D</sup>" (Lerman et al., 2016, p. 48). If a child does not respond to an S<sup>D</sup> or gives an incorrect response, then prompts are used to help him or her to perform the correct response. Examples include verbal prompts, modelling, physical prompts, gestural prompts, photographs, line drawings, and textual prompts (MacDuff, Krantz, & McClannahan, 2001). For example, if a therapist holds up a picture of a car and says *What's this?* (the S<sup>D</sup>) and the child does not respond or says something incorrect such as *Ball*, then the therapist may give a verbal prompt such as *Say car*.

In behavioural interventions, it is generally acknowledged that prompts need to be faded to ensure that the child learns to perform the behaviour in response to the S<sup>D</sup> rather than only in reaction to the prompt. There are several strategies for fading prompts including using a least-to-most prompting hierarchy, in which the therapist initially provides the least intrusive prompt (e.g. a verbal prompt) and gradually increases the amount of intrusiveness (e.g. a gestural prompt, then a partial physical prompt, then a full physical prompt) until the individual performs the correct response. Another prompt fading method is to use a most-to-least prompting hierarchy, in which the therapist initially delivers the most intrusive prompt necessary for the individual to successfully perform the skill. Research suggests that all of these prompt fading methods can be effective although there may be certain advantages and disadvantages to different methods of prompt fading for specific situations (MacDuff et al., 2001). For example, most-to-least prompting might be best suited for helping children with ASD quickly acquire new skills by minimising errors, whereas least-to-most prompting may prevent children from becoming overly dependent on the prompt.

In DTT, each correct response is followed by what will hopefully function as a reinforcing consequence such as providing praise and/or access to a preferred item (Lerman et al., 2016). Generally, the therapist will give the child an item that he or she has previously identified as reinforcing using a preference assessment (DeLeon & Iwata, 1996) or will offer the child a choice between potential reinforcers. Often prompted responses will receive a less

frequent, a smaller amount, or a less preferred/lower quality reinforcer than independent responses, or might not be reinforced at all. This differential reinforcement is intended to provide an incentive for the child to respond independently. Research suggests that such differential reinforcement does in fact lead to more rapid skill acquisition (Cividini-Motta & Ahearn, 2013).

Finally, the inter-trial interval is a pause between the end of one trial and the beginning of the next trial (Lerman et al., 2016). DTT is generally delivered at a rapid pace and, thus, the length of this pause is often short (e.g. 2-3s). Research suggests that children with ASD may acquire new skills faster when there is a short (0-8 s) rather than a long (10 s-several min) pause between trials (Majdalany, Wilder, Greif, Mathisen, & Saini, 2014). Short inter-trial intervals may be more effective because they help to maintain the child's attention and reduce the opportunity for the child to engage in off-task behaviour (Koegel, Dunlap, & Dyer, 1980).

DTT also incorporates regular measurement of the child's progress (Lerman et al., 2016). Often the therapist will record the outcome of each trial (i.e. whether the child made a correct or incorrect response) and will then plot these outcomes on a graph so as to be able to monitor the child's progress. If the data suggests that the child is not making progress then the programme is modified. However, collecting data on the outcome of each trial is labour-intensive so in some cases it might be more practical for the therapist to record outcomes for only some of the trials conducted in each session.

In DTT, new skills are also often taught in a highly-structured "distraction free" environment to ensure that the child is able to attend to the S<sup>D</sup> rather than extraneous cues within the environment (Lerman et al., 2016). In general, the child begins learning skills while seated at a small table facing the therapist, and items such as pictures, toys, and other interesting stimuli are removed from the environment. Once a child learns to respond correctly to the S<sup>D</sup> in this setting, the therapist might begin to target the skill in the natural environment in order to promote generalisation.

# **Early Intensive Behavioural Intervention**

In 1987, Lovaas published the results of a study that examined the effectiveness of an intensive behavioural intervention which made substantial use of a DTT approach. The intervention aimed to increase IQ and adaptive skills and reduce problem behaviour of young children with ASD. This study had a quasi-experimental multiple-group comparison design and included 38 children with a clinical diagnosis of ASD who were under 46 months at the start of the study. These children were assigned to the experimental group (n=19) which

received an average of 40+ hours of DTT per week for 2 or more years, or the control group (n=19), which received 10 hours or less of DTT per week for the same duration. Lovaas compared these results to another group of 21 children who received treatment-as-usual in a separate study. The results of this study suggest that, at the age of 6 or 7, the children in the experimental group had significantly better improvements in terms of cognitive abilities compared to both of the control groups and 47% of children experienced "recovery", defined as intelligence scores within the normal range and placement in a mainstream (rather than special education) classroom without additional support. A follow-up study also suggested that these results were maintained more than 4 years later (McEachin, Smith & Lovaas, 1993). This was the first study to suggest that 40 or more hours per week of early intervention could lead to drastic improvements for children with ASD (Schreibman et al., 2015). It helped researchers, practitioners, and parents to greatly increase their expectations for children with ASD and generated a large amount of research aiming to replicate these results.

Several other studies have replicated the Lovaas (1987) study and evaluated the effects of similar early intensive behavioural interventions (EIBI; generally 25+ hours per week of intervention). Indeed, there is now a considerable number of high quality studies on the effectiveness of DTT/the Lovaas approach and other similar EIBIs (Eikeseth, 2008; Rogers & Vismara, 2008). One such study which has been praised for its scientific rigor (e.g. Reichow, 2012) was conducted by Smith, Groen, and Wynn in 2000. In this study, children aged between 18 and 42 months with an independent clinical diagnosis of ASD or PDD-NOS were randomly assigned to an experimental group which received an average of 24.5 hours per week of DTT for 1 year (this was gradually reduced over the next 1 to 2 years) or a parent training group which received 5 hours per week of parent training on the principles of DTT for three to nine months. There were no differences between the groups prior to intervention, however, following intervention the DTT group had significantly higher scores on measures of intelligence, language, and academic achievement than the parent training group. The DTT group were also more likely to be placed in a mainstream classroom. However, there were no differences between the two groups in scores on measures of adaptive functioning or socioemotional functioning. Thus, the children in the DTT group had significantly better improvements for some, but not all, of the outcome measures.

At least 20 other studies have been published which compare the effectiveness of EIBI with eclectic treatment, treatment-as-usual, or less intensive intervention (<15 hours per week) (see Lerman et al., 2016 for a review). In general, these studies found that children who received EIBI had significantly better improvements on a range of outcomes including

intellectual functioning, language, adaptive behaviour, and social skills than the comparison groups. Further, these children showed greater reductions in autism symptom severity and were more likely to be placed in a mainstream classroom without additional support than those in the comparison groups.

There have been at least nine literature reviews published on the effectiveness of EIBI, most of which found that it is a "well established" or "established" EBP for young children with ASD (see Dawson & Bernier, 2013 and Lerman et al., 2016 for a full list). However, some reviews express concerns with the methodology used in some studies and/or state that some children do not respond as well to treatment as others. EIBI is also an "established evidence-based practice" according to the Reichow's evaluative method (2011; Reichow et al., 2008) and the National Autism Centre (2015). In fact, several reviews comparing EIBI with other early intervention approaches have found that EIBI is an effective, evidence-based approach for young children with ASD (e.g. Eikeseth, 2008; Rogers & Vismara, 2008).

## **Naturalistic Behavioural Early Intervention**

Although research suggests that EIBI is generally effective in improving a variety of outcomes for young children with ASD (e.g. Dawson & Bernier, 2013, Reichow, 2012), there are several potential limitations to this method of intervention. For example, due to the generally highly structured nature of the teaching environment, the gains made during treatment may not easily generalise to novel settings and people (Koegel, Ashbaugh, & Koegel, 2016; Smith, 2001). Further, some children receiving EIBI may not be motivated by the teaching activities and may engage in challenging behaviours in order to escape or avoid them (Koegel et al., 2016; Smith, 2001). Last, some children may become reliant on cues or prompts from the adult, and may, thus, be less likely to spontaneously perform target skills (Smith, 2001).

Naturalistic behavioural interventions were developed in an effort to increase child motivation, spontaneity, and generalisation of skills (Schreibman et al., 2015). Naturalistic behavioural interventions are based on behavioural learning theory, and employ many of the teaching techniques used in behavioural intervention such as (a) the use of the three-term contingency/ABC format; (b) the use of response prompting; (c) the selection of objective, measurable, socially-relevant target behaviours; and (d) the systematic recording of the occurrence of target behaviour(s) (Hart & Risley, 1968). One of the key ways in which these interventions differ from traditional ABA as applied by Lovaas (1987) is that skills are initially taught in the context of naturally occurring activities, such as play or daily routines,

rather than in contrived, structured environments (Hart & Risley, 1968; 1975). Correct child behaviours are also rewarded with natural reinforcers that are directly related to the target behaviour and children are given a greater amount of choice in activities and reinforcers (Koegel, Dyer, & Bell, 1987; Koegel & Williams, 1980).

### **Incidental Teaching**

The first model of naturalistic behavioural intervention was incidental teaching, which was developed by Hart and Risley in 1968. It involves teaching skills in the context of naturally occurring interactions (Hart & Risley, 1975). The key steps in this model are: (a) the child verbally or non-verbally indicates that he or she wants an item/activity, (b) the adult cues the target behaviour by asking a question or withholding access to the item/activity, (c) the adult prompts the correct behaviour if necessary, and (d) the adult provides the child with access to the item contingent on a correct or elaborated response (Hart & Risley, 1968; Pindiprolu, 2012).

Incidental teaching was first used to teach language skills to children from low-income families (Hart & Risley, 1968), but it has since been used to teach a variety of skills to children with ASD (Pindiprolu, 2012; Schreibman et al., 2015; Ingersoll, 2010b). In the first such study, McGee, Krantz, Mason, and McClannahan (1983) taught a 15-year-old boy and a 12-year-old boy with ASD to receptively identify food items in the context of a lunch preparation routine. Specifically, each young person was required to give his parent the item needed to complete the lunch routine and if he did not select an item or selected an incorrect item then the parent prompted the correct response before allowing him to access the item. The results of this study suggest that both young people learned to receptively identify the objects and that this learning generalised to a novel setting and activity. Incidental teaching procedures have also been used to teach children with ASD skills such as preposition use (McGee, Krantz, & McClannahan, 1985), reading (McGee, Krantz, & McClannahan, 1986), and reciprocal peer interactions (McGee, Almeida, Sulzer-Azaroff, & Feldman, 1992).

The Walden programme is a comprehensive early intervention approach for children with ASD which is based on incidental teaching procedures (McGee, Morrier, & Daly, 1999). It involves teaching social and language skills in the context naturally occurring routines both in children's homes and a specialised early childhood centre. The one study that has evaluated the effectiveness of the Walden programme found that, after at least 6 months of intervention, some children who did not speak upon entering the programme had begun to "verbalise meaningful words", and that 71% of children showed improvement in the time they spent in close proximity to other children (McGee et al., 1999). However, the quality of

this study has been rated as being weak based on the Reichow (2011; Reichow et al., 2008) evaluative method due to the one group pre- post-test design and the lack of inferential statistics. Thus, the certainty of evidence supporting the Walden model as an effective intervention for improving outcomes for young children with ASD must be viewed as inconclusive.

Aside from the one study investigating the effectiveness of the Walden programme (McGee et al., 1999), studies have primarily used incidental teaching procedures as part of focused treatments (i.e. treatments targeting one specific type of behaviour; Wong et al., 2015). The most thoroughly researched comprehensive naturalistic behavioural intervention for children with ASD is Pivotal Response Treatment® (PRT: Koegel & Egel, 1979), which is described in detail in the following section.

# **Pivotal Response Treatment**

PRT was developed in the late 1970s (Koegel & Egel, 1979). It is often delivered as a focused language intervention, but it is also a comprehensive intervention which targets a number of "pivotal areas" (Koegel et al., 2016). The developers of the model propose that targeting pivotal areas including initiations, self-management, response to multiple cues, and empathy will lead to extensive collateral gains in other areas, such as social skills, communication and a reduction of problem behaviour.

PRT is based on the theory of learned helplessness in children with ASD and other intellectual disabilities. Research in this area has found that humans and animals who are unable to exert control over the outcome of one situation are less likely to attempt to exert control in future situations (Hiroto, 1974; Hiroto & Seligman, 1975; Overmier & Seligman, 1967). For example, in a seminal study by Hiroto (1974), participants who had previously been exposed to an unescapable aversive loud noise were less likely to attempt to escape a loud noise in a new situation than those who had been exposed to an escapable loud noise, or no noise at all (Hiroto, 1974). For these participants attempts to escape did not lead to reinforcement, which reduced the likelihood that they would attempt to escape in a new situation.

The learned helplessness research has implications for individuals with ASD as they often present with delays across many areas of development (Kennedy & Courchesne, 2008). This may cause them to experience more failures than would be expected for a typically developing child (Koegel & Mentis, 1985). For example, a child's difficulties with social communication may cause him to have negative interactions with his peers. As the child's attempts to interact do not lead to reinforcement, this is likely to reduce his motivation to try

to engage with his peers in the future which may, thus, result in a reduction in his overall frequency of social interaction (Koegel et al., 2016).

PRT aims to increase the motivation of individuals with ASD to attempt and be successful in a range of developmentally and socially appropriate tasks (Koegel & Egel, 1979). It also aims to strengthen the individual's understanding of the relationship between making an attempt at a task and gaining reinforcement (Koegel & Egel, 1979). The five PRT procedures that have been developed to achieve these aims are: child choice, using direct and natural reinforcers, reinforcing attempts, task variation, and interspersing maintenance and acquisition tasks (Koegel et al., 2016). Researchers have investigated the effectiveness of each of these techniques, as well as the effectiveness of these techniques when combined in the comprehensive PRT package (Koegel et al., 2016).

Some of the PRT techniques, such as child choice, are also key components of incidental teaching (Hart & Risley, 1968). In PRT, child choice refers to importance of using materials and activities that the child likes or prefers during teaching (Koegel et al., 1987). To determine the items and activities which the child prefers, one can simply ask the child what he or she likes, provide the child with a choice between two activities (e.g. verbally, or by reaching/pointing), or observe the child during free play. Koegel et al.'s 1987 study supports the importance of allowing children to select reinforcers and to learn during preferred activities. In this study, ten children with ASD between the ages of 4 and 13 were found to show fewer social avoidance behaviours, such as gaze aversion, closed eyes, and moving away, during preferred activities, than non-preferred activities.

As with incidental teaching (Hart & Risley, 1968), another important PRT technique is the use of direct and natural reinforcers, that is, rewards which are directly related to the target behaviour as opposed to arbitrary rewards that are unrelated to the target behaviour (Koegel et al., 2016). For example, if a child says *Car* upon seeing a toy car, a direct and natural reinforcer for this behaviour would be to give the child the car. This should strengthen the child's understanding of the relationship between saying *Car* and gaining access to the toy car and, thus, should increase the likelihood that the child will say *Car* in response to seeing a toy car in the future. On the other hand, in traditional DTT, the child may be required to say *Car* in response to looking at a picture of a car and may be rewarded with a preferred snack such as a cookie. In this example, the child learns that when he says *Car* in response to a picture of a car he will receive a cookie. He may not necessarily generalise this learning to saying *Car* in response to seeing a toy car in his natural environment. Research by Koegel and Williams (1980) and Williams, Koegel, and Egel (1981) suggests that six

children with ASD between the ages of 4 and 7 learned target behaviours (language, imitation, and cognitive skills) faster when reinforcement was directly related to that behaviour rather than arbitrary.

The remaining three techniques are not components of incidental teaching and are based on learned helplessness theory (Hart & Risley, 1968). The technique of reinforcing attempts aims to decrease the number of failures experienced by children with ASD, which, in turn, should increase their motivation to participate in the task (Koegel & Mentis, 1985). This technique differs from shaping (Skinner, 1953) or reinforcement of successive approximations because all child attempts are consistently reinforced by the therapist, rather than the therapist limiting reinforcement to successive approximations of the desired behaviour (Koegel, O'Dell, & Dunlap, 1988). Koegel et al. (1988) used a reversal design to assess the effect of reinforcing attempts on the speech production and emotional affect of four non-verbal children with ASD who were aged between 3 and 12. They found that when the children were reinforced each time they made a speech sound, they were more likely to produce speech sounds, and were rated as more enthusiastic, happier and more interested than when they were only reinforced for sounds that increasingly resembled the target sound (motor shaping).

The fourth PRT technique is task variation. The authors suggest that using a variety of tasks may prevent the child with ASD from becoming "bored" and will therefore increase his or her motivation to continue participating in a therapy session. A study by Dunlap and Koegel (1980) found that a 5-year-old and a 7-year-old girl with ASD had more correct responses and appeared more enthusiastic, interested, and happy, when they were presented with a variety of tasks during a session, compared with doing the same task for the whole session.

The final PRT technique is interspersing maintenance and acquisition tasks. This is an extension of task variation and refers to including some tasks that the child can already do (mastered tasks) in all teaching sessions. The authors hypothesise that the use of maintenance tasks will ensure that the child will frequently experience success, which will prevent him or her from suffering learned helplessness (Koegel et al., 2016). Dunlap (1984) found that when five children with ASD between the ages of 5 and 10 were presented with a variety of maintenance and acquisition tasks during a session, they learned the acquisition skills faster and appeared more enthusiastic, interested, and happy than when they were presented with a variety of acquisition-only tasks or one constant task.

At least 18 studies have been conducted on the effectiveness of PRT techniques when

combined in an intervention package (Cadogan & McCrimmon, 2015; National Autism Centre, 2015). The majority of these studies utilised single-case designs and, thus, included only a few participants per study, but a least two randomised controlled trials (RCTs) have been conducted (Hardan et al., 2015; Mohammadzaheri, Koegel, Rezaee & Rafiee, 2014). In the study by Mohammadzaheri et al. (2015) the children were aged between 6 and 11 years and, therefore, this approach would not necessarily be considered to have been conducted on an early intervention population. The study by Hardan et al. (2015) examined the effectiveness of PRT parent training compared to a general parent psychoeducation programme for improving language in young children with ASD. Fifty-three children with ASD aged between 2 and 6 years and their parents were randomly assigned to one of the two groups. Each group received one training session per week for 12 weeks ( $8 \times 90$  min group parent-only sessions and  $4 \times 60$  min one-on-one sessions with the parent and child). Results of this study suggest that, following intervention, 21 of the 25 parents in the PRT group had learned to implement the PRT procedures with fidelity. Further, children in the PRT group had a greater total number of utterances and significantly higher scores on a measure of expressive and receptive language scores than children in the psychoeducation group.

PRT has been included in several literature reviews on effective early interventions for children with ASD (National Autism Centre, 2015; Rekap & Rekap, 2014; Rogers & Vismara, 2008). In the most recent such review, the National Autism Centre (2015) rated PRT as one of 14 established evidence-based practices for children with ASD. PRT would also be classified as an established EBP according to Reichow's evaluative method (2011; Reichow et al., 2008), because at least two studies with a strong group design conducted in different locations have found the intervention to be effective. However, most high quality studies (e.g. Hardan et al., 2015; Mohammadzaheri et al., 2014) on PRT have examined its effectiveness for improving language outcomes for children with ASD. It is not yet clear whether PRT is equally as effective for improving other outcomes such as social skills and behaviour or when used as a comprehensive intervention.

### **Developmental/Relationship-Focused Interventions**

Around the time that the first evidence was emerging for the effectiveness of EIBI (Lovaas, 1987), there was also a significant amount of research being conducted in the field of developmental science (Dawson & Bernier, 2013; Schreibman et al., 2015). Research into the typical and atypical development of skills such as communication, joint attention, imitation, and imaginary/symbolic play (e.g. Tager-Flausberg et al., 1993; Mundy, Sigman, Ungerer, and Sherman, 1987) contributed to the creation of a number of interventions which

are collectively referred to as developmental/relationship-focused interventions (Dawson & Bernier, 2013; Mahoney, Boyce, Fewell, Spiker, & Wheeden, 1998; Schreibman et al., 2015). These comprehensive interventions differ fundamentally from behavioural interventions in many ways, including the nature of the skills targeted, the order in which these skills are taught, and the use of "affectively rich" social interactions to encourage development. They also do not explicitly reference the techniques from behavioural learning theory such as the ABC format, prompting, and shaping but may employ some of these procedures to varying extents (Ingersoll, 2010b).

Developmental/relationship-focused interventions are heavily informed by a constructivist approach to development (Schreibman et al., 2015). Jean Piaget was one of the first constructivist theorists who posited, based on intensive and detailed observations of children, that there are four stages of cognitive development (sensorimotor, preoperational, concrete operational, and formal operational) and that children transition from one stage to the next based on their physical maturation and exposure to relevant experiences (Piaget, 1952). According to Piaget, learning occurs through assimilation, that is, processing new experiences using one's current level of understanding, or accommodation, that is, altering one's understanding based on new experiences. Thus, in order to maximise learning, one should ensure that instructional experiences are slightly more difficult than the child's current level of cognitive development (Feldman, 2014).

Lev Vygotsky, a Russian developmental psychologist, emphasised the role of social interaction in cognitive development (Vygotsky, 1978). He proposed that children's cognitive abilities increase when learning experiences occur in the "zone of proximal development". This is when a task is slightly too difficult for a child to do independently, but he or she is able to compete it with help from an adult or more advanced peer. This gave rise to the idea of "scaffolding" learning experiences, in which an older or more knowledgeable person initially assists the child with a task and then removes this assistance when the child can perform the task independently (Wood, Bruner, & Ross, 1976).

The constructivist approach has had a considerable influence on developmental/relationship-focused models of intervention for children with ASD and other disabilities (Schreibman et al., 2015). First, these models usually involve conducting an assessment of the child's developmental level and then selecting target skills which are slightly too difficult for the child to perform independently (Greenspan & Wieder, 1997; Schreibman et al., 2015). Further, the target skills are taught in the order that they would usually develop in a child without ASD, and new target skills are chosen when the child can consistently and

independently perform the current skill (Schreibman et al., 2015).

Research on the development of skills in children with ASD suggests that they often show similar, although delayed, patterns of development to those without ASD. Several studies suggest that children with ASD develop language in a similar way to those without ASD. For example, in an analysis of interactions between six children with ASD and their mothers, Tager-Flausberg et al. (1993) found that over a period of 12 to 26 months, these children showed similar increases in their mean length of utterance and improvements in grammatical structure to children with Down syndrome, and to children without developmental disabilities. Further, Toth, Munson, Meltzoff, and Dawson (2006) found that joint attention and immediate imitation strongly predicted language ability in a sample of 60 preschool-aged children with ASD. This is significant as imitation and joint attention are precursors to language development in children without ASD or developmental disabilities. Based on findings such as these, developmental approaches frequently focus on developing precursors to language learning such as babbling, joint attention, and use of gestures before targeting spoken language. This differs significantly from behavioural interventions, which traditionally advocated teaching language through the use of vocal imitation alone (Schreibman et al., 2015).

Developmental/relationship-focused interventions have a strong emphasis on building "affectively rich social relationships" between the child with ASD and the parent/therapist (Dawson & Bernier, 2013). The "social motivation hypothesis" suggests that the brains of children with ASD do not have the same reward response during social interaction as those of children without ASD (Dawson, Bernier, & Ring, 2012). Further, research suggests that children with ASD have difficulty sharing their emotions with others (Dawson, Hill, Spencer, Galpert, & Watson, 1990). Thus, these interventions use strategies to explicitly promote the sharing of positive affect and to increase the child's motivation to engage socially with a play partner (Rogers et al., 1986). This often includes adult use of positive affect and modelling of positive social interaction.

To the author's knowledge, there are no published systematic reviews on the effectiveness of developmental/relationship-focused interventions. Therefore, Table 7.1 and Table 7.2 in Appendix A summarise the participant characteristics, intervention characteristics, research rigor, and outcomes of the two most researched developmental/relationship-focused interventions for young children with ASD: the Denver model (previously the Playschool model; Rogers, Herbison, Lewis, Pantone & Reis, 1986) and the developmental, individual-difference, relationship-based, Floortime<sup>TM</sup> model

(DIR/Floortime<sup>TM</sup>; Greenspan & Wieder, 1997). These two models will be described in detail in the following sections. Other developmental/relationship-focused interventions for children with ASD include Responsive Teaching (RT; Mahoney & MacDonald, 2004), Relationship Development Intervention (Gutstein, 2001) and the Son-Rise programme (Kaufman & Kaufman, 1976). While three studies have been published on the effectiveness of RT (Karaaslan, Diken, & Mahoney, 2011; Mahoney & Perales, 2003, 2005), there appears to be only one weak quality empirical study on the effectiveness of both Relationship Development Intervention (Gutstein, Burgess, & Montfort, 2007) and the Son-Rise Program (Houghton, Schuchard, Lewis, & Thompson, 2013).

#### **The Denver Model**

The Denver model (also called the Playschool model) is a comprehensive developmental/relationship-focused group intervention that was created in the 1980s for children with ASD and/or other types of developmental disabilities (DD) between the ages of 2 and 6 years (Rogers et al., 1986; Rogers & DiLalla, 1991). Features of this model include the use of positive adult affect (e.g. smiling and laughing with the child) and fun "sensory social routines" (e.g., "peekaboo", "chase", and "hide and seek") to target a wide range of developmental skills. The Denver model is implemented in an early childhood setting and each child is allocated a "primary teacher" with whom they interact the most frequently (Rogers et al., 1986). Skills are targeted during play routines as the developers believe that "play is the primary vehicle for communicative, cognitive, and social/emotional development" in all children (Rogers et al., 1986, p.136). Other features of the Denver model include altering the classroom structure and routines to optimise the child's ability to attend to the therapist and to transition easily between activities (Rogers, Lewis, & Reis, 1987). Finally, challenging behaviours are dealt with through redirection and prompting positive behaviours (Rogers et al., 1986)

Two of the key skills targeted in this intervention are imitation and symbolic thought, which includes symbolic play, symbolic language and conceptual thought (Rogers et al., 1986). The developers hypothesise that these deficits prevent children with ASD from learning from their typically developing peers in their natural environment (Meltzoff & Moore, 1977; Rogers & Pennington, 1991). For example, a child who is unable to imitate a peer or adult during toy play is unlikely to learn to play with toys in a variety of functional ways. An article published on the Denver model in 2006 (Rogers et al.) specifies the use of a developmental curriculum to set individualised, developmentally appropriate goals for each child, although it is not clear whether this curriculum was also used when the model was

initially developed in the 1980s (Rogers et al., 1986; Rogers et al., 1987; Rogers & Lewis, 1989).

The model uses techniques from the IN-REAL outreach project to develop verbal and non-verbal communication (Weiss, 1981). These techniques include (a) the therapist narrating his or her own play, (b) the therapist narrating the child's play, (c) the therapist repeating and elaborating upon what the child has said, (d) the therapist mirroring the child's non-verbal communicative behaviours, and (e) providing intervention to the child in a naturalistic setting.

Five articles have been published on the Denver Model (Rogers et al., 1986; Rogers et al., 1987; Rogers et al., 2006; Rogers & DiLalla, 1991; Rogers & Lewis, 1989). However, the 2006 (Rogers et al.) article has been excluded from this section because the intervention appears to more closely resemble the more recent early start Denver model than the original Denver model. Each of the remaining studies examined the effectiveness of the Denver model of intervention when delivered by trained therapists to children with ASD who were younger than 6 years old in a modified classroom setting (Rogers et al., 1986; Rogers et al., 1987; Rogers & DiLalla, 1991; Rogers & Lewis, 1989). The ratio of children to teachers/teacher aides was 2:1 in all of the studies, but the intensity and duration of the intervention varied from 2 hours and 45 min of intervention per day, four times per week for 6 to 8 months (Rogers et al., 1986) to up to 4.5 hour of intervention per day, five times per week for 12 months (Rogers & Lewis, 1989) or for an average of 18 months (Rogers & DiLalla, 1991).

Results from these studies suggest that children who participated in the Denver model intervention showed improvements on a measure of adaptive behaviour following the intervention and there was no difference between the improvements shown by children with ASD and those of the children with other behavioural/emotional and developmental disorders (Rogers & DiLalla, 1991). Children in the Rogers et al. (1986) study and the Rogers and Lewis (1989) study also showed significant improvements on a measure of symbolic play and social communication and those in the Rogers and DiLalla (1991) study showed improvements on a variety of language measures. Half of the children in the Rogers and Lewis (1989) study showed a significant reduction in ASD symptoms. Further, Rogers et al. (1986) reported a significant increase in parent-child interactions involving child use of positive affect with the mother and social initiations to the mother.

Rogers et al. (1987), also evaluated the effectiveness of training 20 professionals and paraprofessionals to implement the Denver model. The teaching involved a 40-hour training

week followed by two training days which occurred 2 and 4 months into the intervention. The training procedures included didactic instruction, guided observation, feedback and teambuilding activities. On average, the participants found the intervention to be socially valid (understandable, useful, practical, applicable and effective). Video-analysis suggested that they were better able to implement the five key components of the Denver model after 3 months of intervention. These key components were staff members' ability to (a) use play as a main paradigm for learning and growth, (b) place emphasis on social-emotional development, (c) enrich the language environment using reactive techniques, (d) promote structure and routine of classroom, and (e) handle maladaptive behaviours (Playschool Observation Scale; Rogers et al., 1987).

According to the Reichow evaluative criteria (2011; Reichow et al., 2008) the Denver model is not an established or promising evidence-based practice because there have been no studies of strong or adequate rigor/quality published on this intervention (see Table 7.1, Appendix A; also see Table 2.1 for a full list of the criteria). The Reichow evaluative criteria were fully described on page 28, but, briefly, these criteria include seven primary quality indicators (e.g. participant characteristics, independent variable) and six to eight secondary quality indicators (e.g. random assignment, blind raters) which vary for single and group designs. A review of comprehensive psycho-educational interventions for young children with ASD also found that in two of the Denver model studies (Rogers et al., 2006; Rogers & DiLalla, 1991) "evidentiary support was so low that outcome data gave insignificant scientific meaning" (Eikeseth, 2008, p. 3). The National Autism Centre (2015) has not evaluated the effectiveness of the Denver model in isolation.

## DIR/Floortime<sup>TM</sup>

DIR/Floortime<sup>TM</sup> (Greenspan & Wieder, 1997) is another play-based developmental/relationship-focused model of early intervention for children with ASD. This model has three key components (Wieder & Greenspan, 2003). The first component, developmental capacities, refers to the six levels of "functional milestones" which are achieved early in life by typically-developing children. These milestones are (a) self-regulation and shared attention, (b) engagement and relating, (c) purposeful emotional interaction, (d) social problem solving, (e) creating ideas, and (f) thinking logically. For example, a child who is at the "engagement and relating" developmental milestone is learning to attend to a play partner, whereas a child at the "thinking logically" developmental milestone is learning to express ideas and opinions with words and through dramatic pretend play (Pajareya, & Nopmaneejumruslers, 2011). The second component, individual-

differences in information processing and motor planning, involves determining the child's sensorimotor preferences and deficits (Wieder & Greenspan, 2003). The final component, relationship-based, emphasises the type of interaction needed to foster the child's development and includes techniques such as observing the child's cues, following the child's lead, and "reading" the child's intentions. Parents and play partners are encouraged to implement the intervention during 15-20-min periods of "floortime" several times per day.

At least 13 studies have been published on the effectiveness of the DIR/floortime model, however, only four of these studies are included in Tables 7.1 and 7.2 in Appendix A due to the extremely low quality of the remaining studies (Liao et al., 2014; Pajareya, & Nopmaneejumruslers, 2011; Solomon, Necheles, Ferch, & Bruckman, 2007; Solomon, Van Egeren, Mahoney, Huber, & Zimmerman, 2014). Each of these studies focused on training parents to implement DIR/Floortime<sup>TM</sup> intervention with their children with ASD who were aged between 1.5 and 6 years. The intensity and duration of the training varied from a 3 hour workshop and 1.5 hours of home-based training with a follow-up session (Pajareya & Nopmaneejumruslers, 2011) to a full day workshop and 3 to 4 hours of home-based training per month for eight to 12 months (Solomon et al., 2007; Solomon et al., 2014).

The children in each study showed improvements in a variety of outcomes, for example, those in the Liao et al. (2014) and Solomon et al. (2007) studies showed significant improvements on a measure of "functional developmental progress" (Greenspan, DeGangi, & Wieder, 2001), while those in the Pajareya and Nopmaneejumruslers (2011) and Solomon et al., (2014) studies also had significantly better scores than the treatment-as-usual group on this measure. Further, children in two of the studies showed greater improvements in a measure of autism symptomology or diagnostic severity than the treatment-as-usual group. However, in the Solomon et al. (2014) study there was no significant difference between the DIR/Floortime<sup>TM</sup> group and the treatment-as-usual group for children's scores on measures of language or development.

Parent outcomes varied following the interventions. Liao et al., (2014) reported a significant reduction in parenting stress following intervention, while Solomon et al. (2014) did not find a significant difference in parenting stress between the DIR/Floortime intervention group and the treatment-as-usual group. In the first Solomon et al. (2007) study the authors reported that parenting interactional techniques did not change as a result of the intervention, whereas in the second study (Solomon et al., 2014) the authors reported that the DIR/Floortime group had significantly better interactional style than the treatment-as-usual

group following intervention. Pajaraya and Nopmaneejumreslers (2011) did not measure any parent outcomes.

Using Reichow's evaluative method (2011; Reichow et al., 2008) the DIR/Floortime<sup>TM</sup> model would be described as a promising evidence-based practice because at least two studies of adequate methodological strength found it to be effective (See Table 7.1, Appendix A). However, it cannot yet be considered to be an established evidence-based practice, which means it should be implemented with caution and closely monitored in case of harm/lack of progress (Reichow et al., 2011). DIR/Floortime<sup>TM</sup> is included in a chapter by Travers et al. (2016) on "fad, pseudoscientific, and controversial interventions" due to the limited scientific evidence of its effectiveness, despite the model having existed for over 20 years. The National Autism Centre (2015) has also labelled DIR/Floortime<sup>TM</sup> as an unestablished evidence-based practice meaning that there is extremely limited evidence for the effectiveness of this intervention and it may be possible that this intervention is harmful.

The lack of evidence for the effectiveness of this model is of particular concern considering that the website of the *Interdisciplinary Council of Development and Learning*, an organisation that was founded by the developers of DIR/Floortime<sup>TM</sup>, describes the model as having "the strongest research of any intervention to support its effectiveness in improving the core challenges of ASD" (Interdisciplinary Council of Development and Learning, n.d.). It could be relevant, however, that this organisation also funds the publication of articles supporting the effectiveness of DIR/Floortime<sup>TM</sup>.

# Naturalistic Developmental Behavioural Early Intervention

Naturalistic developmental behavioural interventions (NDBIs) are a relatively new form of early intervention for children with ASD (Schreibman et al., 2015). NDBIs use behavioural teaching procedures to target developmentally appropriate goals within the child's natural environment (Schreibman et al., 2015). The common features of NDBIs are drawn from each of the methods of intervention previously described in this chapter (behavioural, naturalistic behavioural, and developmental).

Similar to more traditional behavioural interventions, NDBIs involve teaching skills using ABA procedures. These teaching procedures include the use of the ABC format, modelling, and prompting for teaching skills, but each individual model varies in terms of the types of antecedents used to elicit behaviours and the method of prompting the behaviour (Schreibman et at al., 2015). Further, NDBIs emphasise the importance of selecting objective, measureable target behaviours and collecting data on the effect of the intervention on these behaviours. However, the methods of collecting data and selecting behaviours vary

depending on the type of NDBI.

Several of the components of NDBIs originate from naturalistic behavioural interventions. As with PRT and incidental teaching, all NDBIs teach skills in the context of play and/or daily routines, use natural reinforcers, and involve procedures to promote child choice (Hart & Risley, 1968; Koegel et al., 2016). Several NDBIs also incorporate other procedures from PRT such as task variation and reinforcing attempts (Schreibman et al., 2015).

Finally, the teaching targets used in these interventions reflect the growth of skills in typically developing children (Schreibman et al., 2015). Many of these interventions place particular emphasis on teaching skills which provide a foundation for further sociocommunicative development such as imitation, joint attention and prelinguistic language skills including gestures and intentional vocalisations (e.g. Rogers & Dawson, 2010). As with developmental interventions, NDBIs also use a relationship-focused approach to teaching. They value positive affect and aim to teach skills through "emotionally rich" social interactions.

NDBIs address some of the limitations or criticisms of other methods of intervention by combining intervention approaches. For example, the inclusion of behavioural learning principles is intended to increase the probability that the intervention will be effective because research suggests that behavioural interventions are often effective for improving a wide range of behaviours in young children with ASD (Lerman et al., 2016). Teaching skills in the context of play and daily routines may also be more likely to promote child generalisation of skills to the natural environment than traditional behavioural interventions such as DTT (Schreibman et al., 2015). Further, although there is limited evidence for the effectiveness of developmental/relationship focused interventions in isolation, an understanding of child development could guide in the determination of realistic treatment targets and effective intervention techniques, such as the use of developmentally appropriate verbal and non-verbal communication (Rogers & Dawson, 2010; Schreibman et al., 2015). Further, relationship-focussed strategies such as positive affect and increasing the reward value of social interaction may be particularly appropriate for young children with ASD as research suggests that young children without ASD are also very responsive to these strategies (Dawson, 2008; Kuhl, 2007).

Many of the more recent early intervention approaches for children with ASD fit under the NDBI umbrella. Examples of these interventions include reciprocal imitation training (e.g. Ingersoll, 2010a), enhanced milieu teaching (Hemmeter & Kaiser, 1994).

Project ImPACT (e.g. Ingersoll, Dvortcsak, Whalen, & Sikora, 2005; Ingersoll & Wainer, 2013), joint attention, symbolic play, engagement, and regulation (e.g. JASPER: Kasari, Gulsrud, Wong, Kwon, & Locke, 2010; Kasari, Kaiser, et al., 2014), social communication, emotional regulation, transactional support/the early social interaction project (e.g. Wetherby et al., 2014), early achievements (e.g. Landa, Holman, O'Neill, & Stuart, 2011), pre-school ASD communication trial (Green et al., 2010; Pickles et al., 2016), and the early start Denver model (Rogers & Dawson, 2010). There is empirical research to suggest that each of these interventions may be effective for improving some outcomes for young children with ASD, either from RCTs (Ingersoll et al., 2005; Kasari et al., 2010; Kasari, Lawton, et al., 2014; Wetherby et al., 2014; Landa et al., 2011) or multiple experiments with strong single-subject designs (Ingersoll et al., 2005; Ingersoll & Wainer, 2013).

# **Early Start Denver Model**

The ESDM appears to be an increasingly popular model of NDBI for children with or at risk for ASD aged between 12 and 60 months (Penner et al., 2015; Vivanti et al., 2014). It has been delivered by parents or therapists within a clinic, the child's home environment or in group settings such as early childhood education centres (Rogers & Dawson, 2010; Talbott, Estes, Zierhut, Dawson, & Rogers, 2016). The ESDM is based upon PRT and the Denver model. In line with PRT, the ESDM incorporates procedures to increase child motivation such a child choice, task variation, and using direct and natural reinforcers (Rogers & Dawson, 2010). It also teaches skills through the use of behavioural learning principles including the ABC format, response prompting and chaining. Skills are taught in the context of "joint activity routines" that occur naturally throughout the child's day rather than discrete trials. These routines include sensory-social play (e.g. songs, peekaboo, chase, bubbles, balloons), object play (e.g. puzzles, blocks, trains), greetings, tidying up, and daily living routines (e.g. meals, bath-time, bedtime, chores).

In addition, similar to the Denver model, the ESDM uses techniques that are intended to be useful for developing positive relationships with children and increasing the reward value of social interactions (Dawson, 2008; Rogers et al., 1986; Rogers & DiLalla, 1991). These techniques include: (a) the use of positive affect, (b) an emphasis on fun play with people, (c) copying the child's actions and noises, and (d) sharing control of the interaction. Like the Denver model, the ESDM also has a focus on teaching imitation, which the authors consider to be one of the key ways in which young children learn new skills (Meltzoff & Moore, 1977; Rogers & Dawson, 2010). The ESDM teaches imitation skills in the sequence in which they normally develop in children without ASD or developmental disabilities. First

a child is taught to imitate actions on objects and vocalisations, then to imitate gestures, and finally facial expressions (Rogers & Dawson, 2010).

The ESDM is a comprehensive intervention and uses a detailed developmental curriculum to target a range of skills in the order in which they would develop in children without disabilities between the ages of 12 to 48 months (Rogers & Dawson, 2010). These skills include receptive and expressive communication, joint attention, imitation, social skills, play skills, motor skills, behaviour, and daily living skills. In addition to imitation, the developmental domains that receive the most attention are non-verbal communication, verbal communication, social development, and pretend play (Talbott et al., 2016). The four levels of curriculum checklist correspond to skills which typically develop between 12 and 18 months, 18 and 24 months, 24 and 36 months, and 36 and 48 months.

The developers of the ESDM hypothesise that the delays experienced by children with ASD are due to receiving a limited number of learning opportunities compared to children without developmental disabilities (Rogers & Dawson, 2010). Therefore, they believe that intensive intervention is necessary to increase the number of learning opportunities that a child receives within any given day. It is for this reason that they aim to target skills within the context of each of the child's daily routines and activities. ESDM therapists also aim to provide learning opportunities every 10-20 seconds during therapy in order to maximise the amount of teaching that occurs in each session.

There are at least three published literature reviews on the effectiveness of the ESDM (Ryberg, 2015; Talbott et al., 2016; Waddington, van der Meer, & Sigafoos, 2016). In addition, several reviews of intervention programs for children with ASD have also included studies that implemented the ESDM (e.g. Bradshaw et al., 2015; Warren et al., 2011). The Waddington et al. (2016) was conducted to inform and guide the development of Studies 1 and 2 in this thesis and is included in full in Appendix B. Briefly, this review evaluated 15 studies published before June 2015 using Reichow's evaluative method (2011; Reichow et al., 2008) and concluded that "ESDM is a potentially promising intervention, but the limited number of high quality studies indicates the need for additional research to evaluate its effectiveness" (p. 93). Ryberg et al. (2015) only reviewed eight studies as they excluded studies with single case designs and case studies from the analysis. Like Waddington et al. (2016), they also concluded that the ESDM was promising and that the scope for future research was vast. Talbott et al. (2016) included 13 studies but did not systematically evaluate the quality of each study nor did they draw conclusions about the overall evidence for its effectiveness.

There may be several potential advantages of the ESDM over other NDBIs. First, it is the most thoroughly researched model, followed by JASPER (Kasari et al., 2010; Kasari, Lawton, et al., 2014). The ESDM is also one of the few comprehensive NDBIs for children with ASD (Odom et al., 2010; Schreibman et al., 2015). Most NDBIs typically target one or two areas of development. For example, JASPER focuses on joint attention and play (Kasari, Lawton, et al., 2014), and RIT focuses primarily on imitation (Ingersoll, 2010a). As ASD effects all areas of development, and particularly language, joint attention, social skills, and imitation, it would seem logical and efficient to target all of these skills in one intervention (Rogers & Dawson, 2010). Importantly, it is also one of the few models that has been evaluated when delivered by therapists and parents, both one-on-one and in groups, and at a variety of intensities.

### **Moderators of Treatment Effect**

Most studies have found that different types of early intervention are more effective for some children with or at risk for ASD than for others (Fava & Strauss, 2014). This has led many researchers to attempt to identify the characteristics of children who are more likely to respond better to certain interventions and those who are less likely to respond as well. As previously discussed, several studies have found that younger children may respond better to intervention than older children (e.g. Bibby et al., 2002; Dawson, Jones et al., 2012; Flanagan et al., 2012; Granpeesheh et al., 2009; Harris & Handleman, 2000; Perry et al., 2011; Remington et al., 2007; Vivanti et al., 2016) but several studies have also evaluated other child and family characteristics that may influence an individual's response to particular interventions.

Most studies examining these factors have focused on behavioural intervention. Specifically, several studies have found that children with higher pre-treatment scores on measures of intelligence (Bibby et al., 2002; Harris & Handleman, 2000; Klintwall et al., 2015; Lovaas, 1987; Magiati et al., 2007; Perry et al., 2011; Remington et al., 2007; Sallows & Graupner, 2005), language ability (Darrou et al., 2010; Flanagan et al., 2012; Magiati et al., 2007; Perry et al., 2011; Remington et al., 2007; Sallows & Graupner, 2005; Strauss et al., 2012) and adaptive behaviour (Remington et al., 2007; Sallows & Graupner, 2005) responded better to behavioural intervention than children with lower scores on these measures. However, other studies have found that children's scores on measures of intelligence (Eikeseth, Smith, Jahr, & Eldevik, 2007; Smith et al., 2000) language ability (Klintwall et al., 2015; Smith et al., 2000) and adaptive behaviour (Bibby et al., 2002; Eikeseth et al., 2007) did not influence their response to treatment. Similarly, some studies

have found the children with more severe ASD symptoms responded better to behavioural intervention (Sallows & Graupner, 2005), while others have found that those with less severe symptoms had better outcomes (Darrou et al., 2010; Smith et al., 2000) or that ASD severity did not significantly effect a child's response to treatment (Flanagan et al., 2012; Perry et al. 2011). In addition, one study found that children who scored higher on a measure of challenging behaviour showed greater improvements during behavioural intervention than children who did not score as highly (Remington et al., 2007), while another study found that those with less challenging behaviour had greater improvements (Strauss et al., 2012). Two studies reported greater increases during intervention for children who had higher pretreatment imitation abilities (Sallows & Graupner, 2005; Smith et al., 2000). Finally, Bibby et al. (2002) found that a child's gender had no impact upon his or her response to treatment.

Fewer studies have examined predictors of children's responses to naturalistic behavioural early intervention or NDBIs and, much like the research on behavioural intervention, most of these studies have found conflicting results. Specifically, Rogers, Estes et al. (2012) found that children with higher pre-treatment scores on an assessment of orienting to a non-social stimulus (i.e. a non-human noise) had better improvements on measures of cognition and autism symptom severity following parent delivered ESDM intervention than those with lower scores. They also found that children's pre-treatment imitation skills did not influence their response to the intervention. In contrast, Vivanti et al. (2013) found that children who had higher scores on a pre-treatment imitation task generally responded better to group ESDM intervention than those with lower scores. Those with higher functional object use and greater goal understanding also responded better to the intervention. Finally, Eapen, Črnčec, and Walter, (2016) found that children with lower pretreatment autism symptomology responded better to group ESDM intervention than those with lower scores. Scherer and Schriebman (2002) found that three children who had a behavioural profile indicative of being responsive to treatment (i.e. highly interested in toys, tolerant of adults in close proximity, low nonverbal self-stimulatory behaviour, and high verbal self-stimulatory behaviour) had improvements in communication and play following 6 months of therapist delivered PRT, whereas three children with the opposite behavioural profile did not improve (Sherer & Schreibman, 2005).

Several studies have also investigated the impact of family variables on a child's response to intervention. For example, Magiati et al. (2007) found that there was no link between a family's socio-economic status or education level and their child's progress during EIBI. Makrygianni and Reed (2010) reported that children whose parents received training in

the basic principles of applied behaviour analysis, made more improvements in adaptive behaviour following therapist delivered behavioural intervention than those whose parents did not receive training. They suggest that this may be because the parents who received training were better able to create a home environment that was consistent with the therapy environment.

As previously discussed, parental stress may also effect the child's treatment outcomes. Specifically, several studies have reported that parents whose children are receiving high intensity intervention report higher stress levels than those whose children are receiving low-intensity intervention (Osborne, McHugh, Saunders, & Reed, 2008; Strauss et al., 2012). Further, some studies have found that the children of parents who report a high level of stress may have reduced positive outcomes following treatment compared to children whose parents report a low level of stress (Osborne et al., 2008; Strauss et al., 2012). However, other studies did not find a significant relationship between parent stress and the child's response to treatment (Shine & Perry, 2010). Further, Estes et al. (2014) found that families who had experienced a greater number of negative life events in the 12 months preceding treatment, reported greater stress and a lower sense of parenting competence regardless of whether they were participating in the parent coaching intervention or treatment-as-usual condition.

Therefore, it is not yet clear which child or family characteristics are most likely to increase a child's chances of responding positively to intervention (Fava & Strauss, 2014). Further, no research to date has compared the effectiveness of two evidence-based treatments, therefore, it is not possible to determine whether certain child or family characteristics would mean that one type of intervention is likely to be more effective than another (Fava & Strauss, 2014). Thus, this is an important area for future research.

In summary, the four main methods of early intervention for children with ASD are behavioural intervention, naturalistic behavioural intervention, developmental/relationship focused intervention, and NDBI. Some of the specific models which use behavioural or naturalistic behavioural intervention methods, such as EIBI and PRT, have been shown to be consistently effective across several high quality empirical studies. However, research suggests that some children respond better to these interventions than others, although it is not currently clear which child or family characteristics would led an individual to be more or less responsive to a particular intervention. In contrast, specific models which use developmental/relationship-focused intervention methods, such as the Denver model and DIR/Floortime<sup>TM</sup> are not currently supported by any high quality empirical studies.

The ESDM appears to be a promising intervention approach which combines naturalistic behavioural intervention techniques with developmental/relationship-focussed strategies. One potential advantage of the ESDM is the versatility and flexibility of delivery approach which increases the range of circumstances and contexts in which the intervention can be delivered. Chapter 3 provides an in-depth analysis of each of these delivery approaches, which then serves as the main rationale for the two studies in this thesis.

#### **CHAPTER 3**

### **DELIVERY METHODS**

One potential strength or advantage of the ESDM over some other early intervention programmes is that it appears to lend itself to flexible delivery. Specifically, based on the literature, there are at least four distinct ways in which the ESDM can be delivered. These are: (a) intensive one-on-one intervention delivered by trained therapists and parents, (b) intensive group intervention delivered by trained therapists (c) low-intensity intervention delivered by trained therapists, and (d) parent training/coaching. This chapter outlines the potential strengths, weakness, and research evidence for each of these delivery approaches across all models of early intervention for children with ASD and the ESDM in particular. Reichow's evaluative criteria will also be applied to each of the specific ESDM delivery methods (Reichow, 2011; Reichow et al., 2008). The evidence and strengths of each delivery method will then provide the rationale for the two studies in this thesis, which will be presented in Chapter 4.

#### **Intensive One-on-one Intervention**

Research suggests that early intervention may be more effective when it is implemented for many hours per week (Klintwall et al., 2015). An intervention is generally said to be intensive when it is provided for 20 or more hours per week, as this is often stated as the minimum hourly provision for most EIBIs (Lerman et al., 2016). Most of the research into intensive early intervention has been related to behavioural interventions. For example, in his ground-breaking 1987 study, Lovaas found that young children with ASD who received 40 hours per week of early intensive behavioural intervention (EIBI) for 2 years showed significantly greater gains on measures of intelligence and were more likely to be placed in a mainstream classroom than children who only received 10 hours or less per week of the same intervention (see Chapter 2). Other studies have reported similar findings. For example, Reed, Osborne, and Corness (2007) found that 2.5- to 4-year-old children with ASD who received an average of 30 hours per week of behavioural intervention showed significantly better improvements on measures of intelligence and educational achievement after 10 months than children who only received an average of 12 hours of intervention per week. However, there were no differences between the two groups on a measure of adaptive behaviour. Further, Smith et al. (2000) found that children aged between 1.5- and 3.5-yearsold who received an average of 24.5 hours of therapist delivered behavioural intervention per week for 1 year had better scores on measures of intelligence, language, and academic achievement than those whose parents participated in a less-intensive parent training

programme which focussed on behavioural intervention techniques. Several meta-analyses also support the suggestion that intensive early intervention leads to better child outcomes compared to treatment-as-usual or eclectic treatment (e.g. Eikeseth, 2008; Peters-Scheffer, Didden, Mulders, & Korzilius, 2013).

Some researchers suggest that it is difficult to draw conclusions about the effect of treatment intensity on child outcomes because many studies do not provide specific data on the number of treatment hours each child received, particularly the children who were allocated to control groups (Stauss, Mancini, Fava, & the Specialisation in Cognitive Psychotherapy group, 2013). In addition, several studies have not found a link between treatment intensity and child outcomes (e.g. Fernell et al. 2011; Magiati et al., 2007; Sallows & Graupner, 2005). For example, Fernell et al. (2011) evaluated the effectiveness of behavioural intervention for 208 children with ASD aged between 20 and 54 months. They found that there was no significant difference in outcomes between children who received intensive intervention and those who did not. It is suggested that the conflicting results found in these studies could be due to additional factors other than treatment intensity, such as: (a) the quality of the treatment and the fidelity of implementation, (b) the setting in which the treatment was administered, (c) the degree of parent involvement, and (d) the degree to which the intervention was tailored to the child (Fava & Strauss, 2014).

Further, some government organisations, private insurers and service providers are critical of intensive intervention due to the associated expenses (Bouder, Spielman, & Mandell, 2009). Early intervention is estimated to cost between \$US40,000 and \$US80,000 per year (Chasson, Harris, & Neely, 2007). However, most studies have revealed that the costs associated with early intensive intervention are usually offset during the child's lifetime due to factors such as a decrease in the child's need for ongoing intervention, increased placement in mainstream classrooms, and increased chances of living independently (Chasson et al., 2007; Cidav et al., 2017; Penner et al., 2015). For example, Chasson et al. (2007) calculated that the use of EIBI would save the State of Texas \$208,500 per child due reduced special education costs and reduced years of receiving special education.

However, some researchers have suggested that there may be a point at which additional hours of treatment are no longer effective (Fava & Strauss, 2014; Matson & Smith, 2008; Reed et al., 2007). This may be due to child "burn-out", exhaustion or a loss of interest in the programme and the reinforcers used. Several researchers have directly examined the effect of each additional hour of treatment on child outcomes. For example, in a further analysis of data from 453 children who had previously participated in behavioural

interventions, Klintwall et al. (2015) found that for every additional hour of intervention a child's learning rated increase by 0.014, and that there was no point at which additional therapy had no added effect. Further, Granpeesheh et al. (2009) examined the number of behavioural objectives mastered by 245 children who had received behavioural intervention and found that for each additional hour of treatment, the child was likely to have mastered more behavioural objectives. Again, they found that there was no "point of diminishing returns".

In addition, the relation(s), if any, between the intensity of the intervention and its effectiveness may not be straightforward. For example, Granpeesheh et al. (2009) found that there was an interaction between the children's age when beginning treatment and their responses to the treatment. Specifically, for the children who received low-intensity intervention, those who were aged between 2 and 5 years mastered many more behavioural objectives than those who were aged between 5 and 7 years. However, for the children who received high intensity intervention, their age did not have an impact on the number of behavioural objectives that they mastered. This suggests that younger children may be more responsive to low-intensity intervention than older children.

Parental stress may also have an effect on the outcome of intensive treatment. Osborne et al. (2008) found that, in a sample of 65 children with ASD aged between 2.5 and 4 years, children who received more than 15.6 hours of intervention per week (50% of the sample), showed better outcomes on measures of intelligence, academic skills, and adaptive behaviour than those who received less than 15.6 hours of intervention per week. However, this relation only occurred if the parents did not report a high level of stress. Treatment intensity did not make a difference to outcomes for children whose parents did report a high level of stress. The authors suggested that parents should be encouraged to seek out extra counselling or stress management strategies before enrolling their children in an intensive early intervention programme.

## **Intensive One-on-one ESDM Intervention**

In 2010, Dawson et al. conducted the only RCT to date evaluating the effectiveness of intensive delivery of an NDBI. Specifically, they compared the effectiveness of intensive one-on-one therapist and parent<sup>2</sup> delivered ESDM to treatment-as-usual for 48 children with ASD aged between 18 and 30 months. Although they intended to provide the intervention

<sup>&</sup>lt;sup>2</sup> Parent training/coaching procedures are described in detail in the *Parent delivery of ESDM* section in this chapter

group with 20 hours per week of home-based, therapist delivered intervention, the children only received an average of 15.2 hours per week because of factors such as illness and holidays. However, these children also received a reported average of 16.3 hours of ESDM intervention per week from their parents, meaning that this study still meets the threshold for an intensive intervention (20+ hours per week). Results of this study suggested that children who received the intensive ESDM intervention had significantly greater improvements on measures of cognition and adaptive behaviour than the group who received treatment-asusual. A subset of the ESDM group also had higher brain activation in areas related to engagement and processing of faces (Dawson, Jones et al., 2012). Further, more children in the ESDM group than the control group also had a change of diagnosis from ASD to PDD-NOS during this time. However, there was no significant difference between groups on a measure of autism symptom severity or challenging behaviour. The outcomes for children in this study were comparable to those of children receiving EIBI, despite the children receiving fewer hours of therapist delivered intervention (Strauss et al., 2013).

A follow-up study was conducted 2 years after these children completed the intervention (Estes et al., 2015). It was found that the ESDM group maintained their improvement on measures of adaptive behaviour and cognition, despite receiving fewer hours of intervention than the treatment-as-usual group during the follow-up period. Further, at this time, the ESDM group had significantly greater improvements on a measure of autism symptom severity, which was not the case in the original 2010 study (Dawson et al.). The results of this study suggest that the early intensive ESDM resulted in prolonged positive outcomes for the intervention group.

Cidav et al. (2017) investigated the costs associated with providing this intensive ESDM intervention. They found that, although families of children in the ESDM group spent an average of \$US14,000 more than the control group on intervention services during the 2 years that they received the intervention, they then saved an average of \$19,000 each year following the intervention, due to a subsequent decrease in the need for intervention services. In other words, this intervention had "paid for itself" within less than 2 years because the children no longer needed to access as many intervention services including speech language therapy, ABA, occupational therapy, and social skills training. The results of this study are consistent with other research that has suggested that early intensive intervention reduces the cost of services in the long-term (Chasson et al., 2007; Penner et al., 2015).

Taken as a whole, the results of these studies suggest that intensive parent and therapist delivered ESDM intervention may lead to sustained improvements in children's

cognition, adaptive behaviour, and autism symptomology, and a reduction in the cost of intervention services over their lifetime. However, at this stage intensive one-on-one ESDM does not meet the criteria for a promising or established evidence-based intervention based on Reichow's criteria (Reichow, 2011; Reichow et al., 2008) because all of these results come from a single study, conducted by the developers of the model (Dawson et al., 2010). More research is needed to determine whether these results can be replicated with additional children in a different location.

## **Low-Intensity One-on-one Intervention**

While the findings about the effectiveness of one-on-one intensive early intervention are promising, there are several potential reasons why families might not be able to, or might not want to, enrol their children in such programmes (Grindle, Kovshoff, Hastings, & Remington, 2009; Johnson & Hastings, 2001; Yingling, Hock, Cohen, & McCaslin, 2017). Specifically, intensive intervention, if it is not publically funded, could be prohibitively expensive. In many areas there is also a limited number of professionals who are qualified to provide specialised early intervention services and, as such, families may have trouble recruiting and maintaining high-quality therapists. In addition, some parents have stated that participation in intensive intervention "overburdens" their child and it limits the time in which they are able to engage in other activities such as interaction with peers and siblings (Yingling et al., 2017).

In cases where EIBI is unavailable or unsuitable for a family, it is possible that low-intensity provision of similar intervention approaches (e.g. less than 20 hours per week) may still improve outcomes for children with ASD compared to receiving no treatment at all, or treatment-as-usual. Indeed, in their 2009 treatment intensity study Granpeesheh et al. found that children with ASD between 2- and 5-years-old who received the lowest number of hours of behavioural intervention (less than 50 h per month) still mastered more than 20 behavioural objectives on average each month. This is considerably less than the average of 60 objectives per month that were mastered by children of the same age who were receiving the highest number of hours of intervention (approximately 150 h per month), but it is possible that this is more objectives than the children would have mastered had they received treatment-as-usual.

Each of the EIBI studies which included a control group who received the same behavioural intervention, but at a lower intensity, found that the high-intensity group had better outcomes (Lovaas, 1987; Reed et al., 2007; Smith, Eikeseth, Klevstrand, & Lovaas, 1997). However, the Reed et al. (2007) and Smith et al. (1997) studies did not include an

additional control group who received treatment-as-usual. Thus, the low-intensity group may have had better outcomes than children who received regular treatment in the community. Although the Lovaas (1987) study found no difference between outcomes for children who received treatment-as-usual and those who received low-intensity intervention, the findings are limited in that the children in this additional control group may not have been comparable to those who received low-intensity intervention. Further, both the study by Lovaas (1987) and the study by Smith et al. (1997) merely stated that the control group received 10 hours or less of behavioural intervention per week. Therefore, it is possible that the children in this group actually received very few hours per week of intervention and they may have had larger improvements if they had received the full 10 hours each week.

There are few studies comparing the effectiveness of low-intensity behavioural intervention for young children with ASD with treatment-as-usual (Eldevik, Eikeseth, Jahr, & Smith, 2006; Peters-Scheffer, Didden, Mulders, & Korzilius, 2010; 2013). Each of these studies reported that children who participated in the low-intensity intervention showed more improvements on a variety of outcomes measures than the group who received treatment-asusual. However, they generally had more modest improvements than those reported in the majority of studies examining EIBI, particularly on measures of intelligence. For example, Eldevik et al. (2006) compared the effectiveness of 12 hours per week of behavioural intervention based on the Lovaas (1987) approach for children with ASD and intellectual disability under the age of 6 years to eclectic treatment for the same amount of time. They found that after 2 years of treatment, the behavioural intervention group showed more improvements on measures of intellectual functioning, language comprehension, expressive language and communication than the eclectic treatment group. However, there were no differences between groups on measures of non-verbal intelligence, behaviour, daily living, or socialisation. Further, in this study the gains in intellectual functioning were described as "small and of questionable clinical significance" (Eldevik et al., 2006, p. 211).

Several studies have also examined the effectiveness of low-intensity therapist provision of naturalistic behavioural intervention (Smith, Koegel, et al., 2010; Stock, Mirenda, & Smith, 2013) or NDBI (Colombi et al., 2016; Devescovi et al., 2016; Goods, Ishijima, Chang, & Kasari, 2013; Kaale, Smith, & Sponheim, 2012) for young children with ASD. For example Stock et al. (2013) compared the effectiveness of a low-intensity PRT programme with a higher intensity applied verbal behaviour programme (see Sundberg & Michael, 2001 for a description of the applied verbal behaviour programme). Specifically, the PRT programme (Nova Scotia EIBI) involved a maximum of 15 hours per week of one-on-

one intervention in home and preschool settings, while the verbal behaviour programme (Group Applied Behaviour Analysis) involved 15 to 25 hours per week of one-on-one and group-based intervention in a preschool setting. Results of this study suggest that, after 12 months, both groups showed statistically significant improvements on measures of cognition, receptive and expressive language, and problem behaviour, but there was no difference between the two groups. This suggests that the two groups were equally as effective, despite the Nova Scotia- EIBI group receiving fewer hours of therapist delivered intervention.

In each of the previously mentioned low-intensity studies, children received at least an average of 10 hours per week of intervention, however, in many countries this is a higher dosage than is publically funded by the government. Although there appears to be no data on the number of hours of intervention children receive in New Zealand, research suggests that young children with ASD in Italy typically receive between 2 and 6 hours of publically funded intervention per week (Colombi et al., 2016), children in Germany typically receive 2 to 5 hours per week (Freitag, Feineis-Matthews, Valerian, Teufel, & Wilker, 2012), and children in some parts of the United States receive between 2 and 3 hours per week (Vismara, Colombi, et al., 2009). Therefore, it is important to assess the effectiveness of therapy at an intensity that can be publically funded, and thus accessible to all families.

Two studies have used an RCT design to evaluate the effectiveness of very lowintensity therapist provision of a programme known as JASPER (Goods et al., 2013; Kaale et al., 2012). JASPER is an NDBI that focuses on teaching joint attention, symbolic play, engagement and regulation in the context of naturalistic play routines (Kasari et al., 2010; Kasari, Lawton, et al., 2014). This model incorporates several strategies that are characteristic of NDBIs, including following the child's lead, imitating the child, expanding on the child's utterances, and adjusting the environment to increase engagement. In 2013, Goods et al. evaluated the effectiveness of two 30-min sessions of JASPER per week for 12 weeks for minimally verbal young children with ASD who were not responsive to their 30 hour per week EIBI programme. Results of this study suggest that, at the end of intervention, the JASPER group had significantly higher scores on measures of spontaneous play, engagement, and initiation of requesting gestures than children who were in the EIBI programme but did not receive the additional JASPER intervention. There were no significant differences between the two groups on a measure of spontaneous joint attention. The findings of this study were particularly impressive given the nature of the population (non-responders) and the limited number of hours in addition to the EIBI programme. However, it is not clear how children who had not also received EIBI would have responded

to this intervention.

Overall, results of these studies suggest that low-intensity interventions can result in some improvements in outcomes for young children with ASD. However, in general, these improvements are not as large or as broad as the improvements shown by children with ASD in the majority of studies investigating EIBI (Dawson & Bernier, 2013; Lerman et al., 2016). This is a very important area of research as most publically funded interventions only consist of a few hours of individual therapy, if that, per week. Thus, it may be valuable for researchers to focus on maximising the effectiveness of low-intensity interventions for young children with ASD.

## **Low-Intensity One-on-one ESDM Intervention**

Two studies have examined the effectiveness of low-intensity therapist delivered ESDM intervention. Both were conducted in Italy (Colombi et al., 2016; Devescovi et al., 2016). In the first study, Colombi et al. (2016) compared the effectiveness of 6 hours per week of clinic-based ESDM therapy for 22 children with ASD aged between 18 and 48 months with treatment-as-usual for 70 children. After 6 months of treatment the ESDM therapy group had a significantly greater improvement on a measure of intelligence than the treatment-as-usual group, but there were no significant differences between groups on a measure of adaptive behaviour functioning. Devescovi et al. (2016) reported that children who received 3 hours per week of intervention for an average of 15 months showed significant increases on a measure of cognitive and language skills following intervention but no significant reduction in autism symptom severity.

The results of these two studies suggest that low-intensity therapist delivered ESDM intervention may improve some outcomes for children with ASD. However, it is not clear how these improvements compare to high-intensity ESDM intervention, as no studies have directly compared the effectiveness of varying intensities of one-on-one ESDM therapy. Further, due to the limited number of studies, and the absence of a control group in the Devescovi et al. (2016) study, low-intensity ESDM would not be considered to be a promising or an established evidence-based practice according to Reichow's evaluative criteria (Reichow, 2011; Reichow et al., 2008).

## **Intensive Group Intervention**

Group-based early intervention may be another more efficient alternative to intensive one-on-one intervention for young children with ASD (Vivanti et al., 2013). This is because it might allow professionals to deliver intervention to more than one child at a time which increases the number of children who can access services. Further, most group early

intervention is delivered in a preschool setting, which is considered to be more representative of a young child's natural environment than a clinic, at least according to the New Zealand Ministries of Health and Education (2008). Compared to one-on-one home-based intervention, preschool group intervention might also to reduce the burden on parents, as in this model they are not required to be home during the hours of the therapy (Vivanti et al., 2013). Further, group intervention programmes provide more opportunities to target social and play skills with a child's same age peers. When the group intervention programme includes typically developing peers, these peers can also serve as additional models of developmentally appropriate behaviour.

There are several studies on the effectiveness of group delivery of NDBIs, such as the ESDM (Eapen et al., 2016; Eapen, Črnčec, & Walter, 2013; Fulton, Eapen, Crnčec, Walter, & Rogers, 2014; Vivanti et al., 2014; Vivanti, Dissanayake, & Victorian ASELCC Team, 2016; Vivanti, Dissanayake, Zierhut, Rogers, & Victorian ASELCC Team, 2013;), Early Achievements (Landa et al., 2011; Landa & Kalb, 2012), and JASPER (Chang, Shire, Shih, Gelfand, & Kasari, 2016). For example, Chang et al. (2016) used an RCT to evaluate the effectiveness of training preschool teachers to implement JASPER procedures in their classroom. Twelve teachers were randomised to an intervention group who received 3 months of JASPER training or a waitlist control group. Results suggest that, 1 month after the training, the teachers in the JASPER training group had greater improvements in their use of the procedures than the control group. Children in the JASPER group also had greater improvements than the control group on measures of joint engagement, and joint attention language. Results were mixed for the remaining measures, in that children in the JASPER group had greater improvements in some aspects of the measures of initiations, joint attention gestures, mean length of utterances and play skills but not others. This data is somewhat limited in that it is not clear how many hours per week the teachers used these strategies. However, the authors suggested that children who received JASPER intervention had greater improvement overall.

### **Intensive Group ESDM Intervention**

Seven studies have examined the effectiveness of group ESDM delivered in an early childhood setting (Eapen et al., 2013; Eapen et al., 2016; Fulton et al., 2014; Vivanti et al., 2013; Vivanti et al., 2014; Vivanti et al., 2016; Vinen, Clark, Paynter, & Dissanayake, 2017), although of the children who participated in the Eapen et al. 2013 study, 10 also participated in the Fulton et al. 2014 study and 26 also participated in the Eapen et al. 2016 study. Further, all of the participants in the Vivanti et al. 2013 study also participated in the 2014 and 2016

studies, and 30 participated in the Vinen et al. (2017) study. Further, the main aim of the 2013 and 2016 studies by Vivanti et al. and the 2016 study by Eapen et al. was to identify predictors of treatment outcome, although each study also reported on treatment effectiveness. The children in each of these studies received intervention in a dedicated early childhood centre with other children with ASD and a teacher to child ratio between 1:3 and 1:4. The length of the intervention varied between 15 to 25 hours per week and lasted for between 10 and 12 months.

Each of these studies reported a range of positive child outcomes following intervention. Specifically, Fulton et al. (2014) found that, after 12 months of group ESDM intervention, the children's maladaptive behaviour had decreased and their cognitive skills had increased, but there was no significant improvement on a measure of cognitive ability or autism symptomology. Eapen et al. (2013) also found that, after 10 months of group ESDM intervention, the children showed improvements on a measure of cognitive skills, autism symptomology, and receptive and expressive language, but, as with Fulton et al. (2014) there was no significant improvement in cognitive ability. Both of these studies are limited by the absence of a control group, however in 2014 Vivanti et al. did include a group of children who received treatment-as-usual (Vivanti et al., 2014). In this study the authors compared the effectiveness of 15-25 hours per week of group ESDM intervention for 22 preschool-aged children with ASD to 30 children with ASD who were receiving treatment-as-usual within the community. After 12 months of intervention, the ESDM group had significantly greater increases on a measures of cognitive ability and receptive language than the treatment-asusual group but there were no significant differences between the groups in adaptive behaviour and autism symptom severity.

In 2017, Vinen et al. conducted a long-term follow-up study comparing outcomes for a subset of 30 children who had previously participated in the group ESDM intervention (i.e. some participants from the Vivanti et al., 2013; Vivanti et al., 2014; Vivanti, et al., 2016 studies) with outcomes for 28 children who had participated in eclectic community based intervention of the same intensity. Their results suggest that children between the ages of 6 and 9 who had participated in either of these early interventions had improvements on a measure of cognitive functioning compared to pre-intervention but also had an increases on a measure of autism symptom severity, which was due to increases in restrictive and repetitive behaviour following the intervention. There were no significant differences between the groups which indicates that the long-term outcomes for children in the ESDM group were not superior to those who received eclectic intervention.

Results of these studies suggest that intensive group ESDM intervention may improve some outcomes for children with ASD. However, due to the absence of a control group in all of the studies except Vivanti et al. (2014), this intervention could not be considered to be an effective, evidence-based practice (Reichow, 2008; Reichow et al., 2011). Rather, the data available currently suggest that intensive group ESDM may be a promising intervention approach. Further, no research has compared the effectiveness of one-on-one versus group ESDM intervention and, thus, it is not clear which of these two intervention approaches is the most effective, or if some children with ASD are more likely to respond better to one approach or the other.

## **Parent Delivery of Early Intervention**

Professionals have evaluated procedures for training/coaching<sup>3</sup> parents to implement various types of intervention techniques with their children. Many reasons have been offered for the potential value of training parents<sup>4</sup> to implement interventions with their children with or at risk for ASD (Oono, Honey, & McConachie, 2013; Ruppert, Machalicek, Hansen, Raulston, & Frantz, 2016). First, as parents are able to use intervention techniques during the child's daily routines and activities (e.g. meals, getting dressed, going to the playground etc.), they are perhaps then in a good position to provide their child with learning opportunities throughout the day (Ruppert et al., 2016). This might contribute to the recommended target of 25-40 hours per week of intensive early intervention to maximise child outcomes (Granpeesheh et al., 2009). Second, if parents can target skills across different natural environments, this may increase the likelihood of generalisation (Koegel et al., 2016). Third, it could be more cost effective and efficient to train parents to deliver the intervention rather than recruiting professionals for the same amount of hours (Dawson & Bernier, 2013). This is particularly important in countries and areas where there is limited funding or limited access to early intervention services.

There are also several potential disadvantages of having parents to serve as the main implementers of early intervention programmes. First, parents of children with ASD might experience more stress than parents of children without disability (Davis & Carter, 2008) and thus the expectation that these parents should implement many hours of intervention per

<sup>&</sup>lt;sup>3</sup> Some studies use the term parent coaching as a synonym for parent training, while others use specific "coaching procedures" see the Implications section in Study 2 for further information.

<sup>&</sup>lt;sup>4</sup> The term parent refers to parents or caregivers/legal guardians. This group will be referred to from herein as parents.

week with their children may further increase their stress. In fact, research by Strauss et al. (2012) suggested that parents who implemented a low-intensity intervention with their children reported significantly less parenting stress than those who implemented or facilitated high-intensity interventions. Research also suggests that intensive intervention may be less effective when parents are stressed (Osborne et al., 2008). Further, some parents experience more barriers to the implementation of early intervention strategies than others. For example, Bagner and Graziano (2013) found that the more barriers a parent faced including socioeconomic disadvantage, single parent households, minority status, and high stress, the more likely he or she was to withdraw from a parent training programme.

There are a variety of ways in which parents might become involved in their child's early intervention programme (Ruppert et al., 2016). These include: (a) implementing the intervention directly with their child, (b) providing input into the selection of intervention procedures and goals, (c) collecting data, and (d) helping with the generalisation and maintenance of skills. The research into the effectiveness of EIBI has mainly involved professional therapists as primary implementers of the intervention and parents often help with the generalisation and maintenance of skills (Strauss et al., 2013). In contrast, much of the research into the effectiveness of developmental interventions and NDBIs has involved training parents to be the primary implementers of the intervention (e.g. Kasari et al., 2010; Rogers, Estes, et al., 2012; Solomon et al., 2014). In parent training studies, mothers are usually the primary implementers of the intervention (Flippin & Crais, 2011). For example, Flippin and Crais (2011) recently reviewed 27 articles on parent-implemented early intervention for children with ASD and found that, of the 14 studies that specified the gender of the parent, only three included a father.

Parent training is usually delivered by a professional who has in-depth knowledge of the particular intervention and experience working with parents (Ruppert et al., 2016).

Training generally includes procedures such as: (a) lectures, (b) discussion, (c) modelling, (d) role-play, (e) coaching, (f) feedback, (g) reflection, and (h) praise (Ruppert et al., 2016).

These methods are based on the behavioural learning principles, in that the adult is provided with information (lectures and discussion) and the opportunity to practice the behaviour (role-play/modelling), then they are prompted to perform the correct behaviour (coaching/feedback) and receive reinforcement (praise). Research suggests that feedback is a particularly important component in adult learning and should include positive feedback for correct implementation, corrective feedback for incorrect implementation, and discussion of next steps (Hattie & Timperley, 2007).

Parents can receive training one-on-one or in a group either in their home, a clinic, or remotely via the internet. One-on-one parent training is the most researched modality and has been used to teach parents to implement a variety of interventions including DIR/Floortime<sup>TM</sup> (e.g. Solomon et al., 2014), the ESDM (e.g. Rogers et al., 2011), PRT (e.g. Hardan et al., 2015) and joint attention training (e.g. Kasari et al., 2010). There is limited research on group training for parents of young children with ASD. Group training is more cost effective than one-on-one parent training and it may help to create support networks between parents of children with ASD (Stahmer & Gist, 2001). For example, a study by Stahmer and Gist (2001) examined the effectiveness of a weekly parent support group in addition to a 12 week one-onone parent training programme based on the principles of PRT. The results of this study suggest that parents who participated in the weekly support group learned to use the PRT techniques better than those who did not. The authors suggest that this may either be because the discussion between the parents helped them to better understand the PRT techniques or the additional support reduced parents' stress which allowed them to better focus on the intervention. Internet-based parent training (also called telehealth delivery) is a low-cost training modality which allows individuals who live in remote areas, or areas without ASDspecific services to access intervention (Dudding, 2009). It allows parents to access intervention information and materials at any time and to be trained by an expert via videoconferencing.

At least nine literature reviews have been conducted on various aspects of parent-implemented or parent-mediated interventions for children with ASD or other developmental disabilities (Ruppert et al., 2016). Parent training research is described as using a "study within a study" approach, in that each study evaluates both (a) the effectiveness of the training for improving parent implementation of the intervention procedures, and (b) the effectiveness of the parent-implemented procedures for improving child outcomes such as communication, imitation, and challenging behaviour (Meadan, Ostrosky, Zaghlawan, & Yu, 2009). A systematic review by Oono et al. (2013) found that whilst parent-mediated early intervention was effective overall, it was more effective for improving parent implementation of the intervention rather than child outcomes. Specifically, this review included 17 RCTs with a total of 919 young children with ASD and found that there was strong evidence for changes in parents' fidelity (use of the intervention procedures) following intervention but that the children only showed small improvements in some outcomes such as language and autism symptom severity and did not show improvements in other outcomes such adaptive behaviour. In contrast, other literature reviews have found parent training to be effective for

increasing both parent and child outcomes, although the majority of these reviews question the overall quality of the research evidence (e.g. McConachie & Diggle, 2007; Meadan et al., 2009; National Autism Centre, 2016, Patterson, Smith, & Mirenda, 2012).

Although there is evidence to suggest that parent training/coaching can be effective for enabling parents to successfully implement early intervention programmes, it is not clear whether it is equally as effective as intensive early intervention delivered by trained professionals. In fact, some researchers state that "parent delivered intervention is not a substitute for evidence-based practices implemented by professionally trained, multidisciplinary teams" (Talbott et al., 2016, p.125). However, there are few studies comparing EIBI or other intensive intervention approaches with parent-mediated intervention (Smith et al., 2000). One such study found that 24.5 hours a week of DTT implemented by trained professionals led to better improvements in child outcomes than DTT implemented by trained parents (Smith et al., 2000). This may have been because parents did not implement the intervention as intensively. Further, several studies have also found that parent training did not result in significantly better child or parent outcomes than treatment-as-usual (Carter et al., 2011; Oosterling et al., 2010; Rogers et al., 2010).

# **Parent Delivery of ESDM Intervention**

Six studies (reported in seven publications) have examined the effectiveness of ESDM parent coaching (Estes et al., 2015; Rogers, Estes, et al., 2012; Vismara, Colombi, et al., 2009; Vismara, McCormick, Young, Nadhan, & Monlux, 2013; Vismara et al., 2016; Vismara & Rogers, 2008; Vismara, Young, & Rogers, 2012). Each of these interventions involved 1 to 1.5 hours of parent coaching per week for 12 weeks. This coaching was either delivered in person in a clinic setting (Vismara & Rogers, 2008; Vismara et al., 2009; Rogers, Estes, et al., 2012; Estes et al., 2015) or remotely via the internet (Vismara et al., 2012; Vismara et al., 2013; Vismara et al., 2016). Five studies reported that the majority of parents learned to implement the ESDM procedures with fidelity (according to the ESDM teaching fidelity rating scale) during the 12 weeks of coaching (Vismara et al., 2009; Vismara et al., 2012; Vismara, McCormick et al., 2013; Vismara et al., 2016; Vismara & Rogers, 2008), and, of these five, three reported an increase in imitation, functional verbal utterances, and attentiveness and social initiations for the majority of children during this time (Vismara et al., 2009; Vismara et al., 2012; Vismara & Rogers, 2008). Vismara, McCormick et al. (2013) also reported an increase in children's functional verbal utterances and expressive and receptive language during the 12 weeks of coaching but their joint attention initiations did not improve. Vismara et al. (2016) found that outcomes for children in the ESDM group did not

improve more than those of children in the control group.

The Rogers, Estes, et al. (2012) study did not find positive results either for the parents or their children. This was the largest study and involved 98 children with ASD between 14 and 24 months of age, who were randomly allocated to an intervention group who received the 12 week ESDM parent coaching intervention or a control group who received treatment-as-usual. The results of this study suggest that, following intervention, there were no significant differences between the groups in terms of the parents' use of the procedures or the children's cognitive or language abilities. In fact, the treatment-as-usual group had a significantly greater decrease in some aspects of autism symptomology (i.e. social affect on the ADOS; Lord et al., 2002) than the ESDM group. The authors suggest that the lack of difference was because the ESDM group received fewer hours per week of intervention than the treatment-as-usual group. The one positive finding in the Rogers et al. (2012) study was that parents in the ESDM group reported significantly stronger working alliances with the therapist/coach than those in the control group. Subsequent research by Estes et al. (2014) also suggested that parents in the ESDM intervention group reported less stress following the coaching than those in the treatment-as-usual group.

Due to these mixed results, ESDM parent coaching cannot yet be described as a promising, or evidence-based practice (Reichow, 2008; Reichow et al., 2011). The differences in results between these studies could be due to a number of factors including difference in: (a) the delivery setting, (b) the characteristics of the children and parents participating, (c) the use of proximal or distal outcome measures, and (d) the coaching procedures. More research is needed to determine the most effective methods and settings for delivering ESDM parent coaching.

Overall, there are advantages and disadvantages to each early intervention delivery approach and different methods may be better suited to different families and contexts. Based on the current research, none of the individual methods of ESDM delivery are established evidence-based practices (Reichow, 2008; Reichow et al., 2011). Thus, more research is need to determine the effectiveness of each of these delivery approaches. Chapter 4 will outline the rationale for the two studies in this thesis based on the advantages and disadvantages of the delivery approaches covered in this chapter, in addition to the previous ESDM research.

#### **CHAPTER 4**

### RATIONALE AND METHODOLOGY

Research suggests that intensive early intervention, and EIBI specifically, can be an effective approach for children with ASD. The research into the effectiveness of intensive one-on-one and group delivery of ESDM could also be seen as yielding promising results (Dawson et al., 2010; Vivanti et al., 2014). Indeed, in New Zealand, the country in which the thesis was conducted, the ASD Guidelines (a resource which provides evidence-based information and recommendations for professionals, individuals with ASD, and their families) recommend that "for children with ASD to make progress, they need to be engaged in developmentally appropriate activities or interactions for at least 15 to 25 hours a week" (p.89; Ministries of Health and Education, 2008). Nevertheless, in many countries, including New Zealand, intensive one-on-one or group early intervention is not funded by the government and there is a limited number of qualified professionals who can deliver these programmes (Colombi et al., 2016; Freitag et al., 2012; Ministry of Education, 2017; Vismara, Colombi et al., 2009). Thus, it is likely that many families might not be able to access or afford intensive early intervention programmes.

Given that most research suggests that early intervention for children with ASD may be particularly effective (Bibby et al., 2002; Flanagan et al, 2012; Granpeesheh et al., 2009; Harris & Handleman, 2000; Perry et al., 2011) it would seem potentially useful to attempt to identify effective methods of delivering early intervention that involve relatively fewer hours of professional input per week. The main focus of the empirical work reported in this thesis was to evaluate the effectiveness of two ESDM intervention delivery methods. Specifically, Study 1 evaluated the effects of a short duration, low-intensity therapist delivered intervention with four children, whereas Study 2 evaluated a short duration parent training programme involving five different children. These delivery methods could feasibly be provided to a large number of children at a limited cost to families or service providers.

# **Low-Intensity Therapist Delivered Intervention**

There is limited research on the effectiveness of low-intensity therapist delivered early intervention approaches. Thus, it is not currently clear whether models of early intervention that are evidence-based or promising when delivered at high intensities (e.g. EIBI; ESDM) are also more effective than treatment-as-usual when delivered at a low-intensity (Eldevik et al., 2006). There currently appear to be only two studies published on the effectiveness of low-intensity ESDM (Colombi et al., 2016; Devescovi et al., 2016). Both of these studies reported that children showed significant improvements on measures of

cognition and/or language following intervention. Study 1 in this thesis was designed to improve and extend upon these findings in the following ways:

- 1. Both the Devescovi et al. (2016) and the Colombi et al (2016) study were conducted in a clinic setting. Research suggests that individuals with ASD appear to have trouble generalising newly learned skills across environments (Koegel et al., 2016; Smith, 2001). Therefore, it may be better to teach children with ASD in the environment in which they will use their skills, such as in their home or kindergarten, rather than a clinical setting. Further, the New Zealand ASD guidelines (Ministries of Health and Education, 2008) also recommend the teaching of skills in the child's natural environment in order to maximise his or her participation and inclusion in society. The United Nations Convention on the rights of persons with disabilities states that intervention should be "undertaken in the least restrictive setting possible" (United Nations General Assembly, 2007). It might be argued that a clinic is a more restrictive setting than the home or school, for example. Therefore, Study 1 will evaluate the effectiveness of home-based low-intensity therapist delivered ESDM.
- 2. To the author's knowledge, none of the studies evaluating the effectiveness of therapist delivered early intervention for children with ASD have directly examined generalisation of child outcomes to their parent(s). This is of potential importance because if the child only uses his or her new skills with the therapist, this might not have any significant impact on the his or her overall functioning in daily life (Rogers & Dawson, 2010). Further, parents are arguably the most important individuals in a young child's life as they generally spend the most time with their child. Therefore, Study 1 will assess the generalisation of children's skills from the therapist to their mothers.
- 3. The majority of studies have evaluated the effectiveness of low-intensity therapist delivered intervention programmes which lasted for between 6 months (Colombi et al., 2016) to 2 years (Eldevik et al., 2006; Peters-Scheffer et al., 2013). Therefore, it is unclear whether shorter duration low-intensity intervention programmes are also effective. Study 1 will involve 12 weeks of intervention. A shorter duration of low-intensity direct therapy, if it were found to be effective, maintained over time, and generalised to parents, could open up the possibility of being able to obtain some benefit from a relatively brief intervention, which could reduce treatment costs and increase the number of children who could receive this type of intervention.

- 4. There are several limitations to the design of the Colombi et al. (2016) and Devescovi et al. (2016) studies. First, both of these studies used a quasi-experimental research designs, meaning that due to the lack of experimental control, any improvements shown by the ESDM group cannot necessarily be attributed to the intervention but could be due to other potential confounding variables (Johnson & Christensen, 2012). Specifically, the Devescovi et al. (2016) study had a one-group pre post-test design meaning that the reported improvements for the participants could have been due to the effects of history, maturation, testing, or instrumentation rather than the intervention. The Colombi et al. (2016) study used a non-equivalent comparison group design in that participants were allocated to the ESDM group on a "first come first served" basis rather than being randomised to groups. Therefore, improvements within the two groups could have been partially due to differential selection. Study 1 will use a multiple probe across participants design to address the these limitations (see Methodology section of this chapter)
- 5. Finally, Study 1 will examine different outcomes measures than those used in most previous research into comprehensive therapist delivered low-intensity intervention for children with ASD. The specific outcome variables and reasons for their selection will also be discussed in the methodology section of this chapter.

# **Parent Training**

One of the potential advantages of parent training is that it might be successfully completed in relatively few hours of professional input. Another potential advantage is that parents might be able to implement intervention techniques with their children for many hours a day in their natural environments (Dawson & Bernier, 2012; Granpeesheh et al., 2009). Research suggests that parent training is effective for improving parent use of intervention techniques and that increased parent use of these techniques often leads to improved outcomes for their children with ASD (e.g. McConachie & Diggle, 2007; Meadan et al., 2009; National Autism Centre, 2016, Patterson et al., 2011). The few studies that have examined the effectiveness of training parents to implement the ESDM have found conflicting results. Study 2 in this thesis aims to improve and extend upon the previous ESDM parent training research in the following ways:

1. All of the previous ESDM parent training/coaching studies took place in a clinic setting or remotely via the internet. While, research suggests that telehealth may be an effective intervention delivery approach for rural communities, or those with limited

access to services, there is no research to suggest that it should be used as a replacement for in-person delivery of intervention when it is possible for the therapist to do so (Boisvert, Lang, Andrianopoulos, & Boscardin, 2010; Vismara et al., 2016). Further, it is suggested that some families may benefit more from in-person intervention than from a telehealth approach (Vismara et al., 2016). Training parents in the home environment may also encourage generalisation of their own learning, as well as generalisation of their child's skills (Rogers & Vismara, 2015). Specifically, the home environment may involve distractions/challenges, which are not present in a clinic including the presence of other children, pets, and highly preferred electronic devices. Thus, training the parent in the home allows the parents to practice the skills in the real world environment. During home-based parent training, the therapist is also able to demonstrate the skills using the materials available at home and to observe the child as they behave in the home environment, rather than an unfamiliar clinic setting (Rogers & Vismara, 2015). Delivering the intervention in children's home might also help to eliminate the need for parents to travel to clinic settings, find childcare for siblings, or take additional time off work (Sweet & Applebaum, 2004). Therefore, Study 2 will examine home-based ESDM parent training/coaching as it is essential to investigate whether it is as effective as clinic or telehealth parent training.

- 2. To the author's knowledge no other parent training research has examined whether parent and child improvements generalise to a second family member such as the child's father. This could be particularly important because research suggests that fathers participate in parent training programmes less often than mothers, but that they might also play a large role in their child's development (Flippin & Crais, 2011). Therefore, Study 2 will examine whether or not a second close family member (father or grandfather), who did not participate in the parent training, will show an increase in his use of the ESDM procedures after the child's mother had completed the training, and, also, whether the child will demonstrate any increases in skills with this family member.
- 3. All studies investigating the effectiveness of the ESDM parent training/coaching have been implemented by a researcher who helped to develop the model. Thus, it is not clear whether this intervention is also effective when delivered by community-based therapists who have less intimate knowledge and experience with the model. Study 2 appears, at the present time, to be the first independent replication attempt of the ESDM parent training/coaching research. This will help to determine whether parent

- training is equally as effective when delivered by an independent researcher, (Waddington) who is also a certified ESDM therapist (Smith et al., 2007).
- 4. Although several studies have examined parent perceptions of ESDM parent training/coaching and parent training for young children with ASD more generally (e.g. Vismara et al., 2012; Vismara, McCormick et al., 2013), most of these studies only included brief acceptability questionnaires. In contrast, Ogilvie and McCrudden (2017) conducted a qualitative analysis of parent perceptions of therapist delivered ESDM therapy. One potential strength of Study 2 will be the inclusion of qualitative semi-structured open-ended interview about parent's experiences and perceptions of the intervention. This could provide detailed information about parents' perceptions of the ESDM intervention, training procedures, and potential modifications.

The two empirical studies reported in this thesis aimed to answer the following six research questions.

## **Research Questions**

# Study 1:

1. Does 3 hours per week of home-based ESDM therapy over 12 weeks produce an increase in imitation, functional verbal utterances, and engagement among four young children with ASD?

# Study 2:

- 2. Is 1 hour per week of home-based ESDM parent training over 12 weeks effective in enabling parents to implement ESDM intervention procedures with fidelity?
- 3. Does 1 hour per week of home-based ESDM parent training over 12 weeks produce an increase in imitation, functional verbal utterances, and engagement for five young children with ASD?
- 4. Do parents find the procedures used in ESDM parent intervention to be understandable, reasonable, efficient and effective?

The following question relates to Studies 1 and 2:

- 5. Do the skills learned in ESDM parent and therapist intervention generalise to a novel play partner (i.e. a parent or second family member)?
- 6. Are the skills learned in ESDM parent and therapist intervention maintained for 1 month following the intervention?

# **Hypotheses**

Most previous studies have suggested that low-intensity therapist delivered ESDM, and ESDM parent training/coaching has had an positive effect on at least some outcomes for children with ASD and/or their parents (Waddington et al., 2016). Therefore, it is hypothesised that most children in Study 1 will improve on at least some of the outcome measures during the 12 weeks of intervention. It is further hypothesised that, in Study 2, the majority of parents will learn to implement the ESDM techniques with at least 80% fidelity and that this will in turn lead to improvements in outcomes for their child with ASD (Vismara et al., 2016). It is also hypothesised that any improvements in child and parent skills will be maintained 1 month after the intervention, as this would be consistent with previous ESDM research (e.g. Vismara et al., 2012; Vismara, Colombi, et al., 2009). In line with previous social validity research (e.g. Vismara et al., 2012; Vismara, McCormick et al., 2013), it is anticipated that parents will find the parent implemented intervention to be understandable, reasonable, efficient and effective. Based on the principles of generalisation, it is hypothesised that, in Study 1, generalisation of the child's skills from the therapist to the parent is unlikely to occur because the child will not have been taught across multiple settings or people (Stokes & Baer, 1977). Further, in Study 2, generalisation to a second parent or family member will be moderated by the degree to which the parent who participates in the training learns to implement the procedures, and also the degree to which they explicitly share what they have learned with the second parent or family member (Stokes & Baer, 1977).

# Methodology

# **Experimental Design**

Single case designs are commonly used by researchers to gain an understanding of the effects of an intervention on an individual's behaviour (Baer, Wolf, & Risley, 1968). These designs are generally used when there are few participants in the study (Baer et al., 1968). Single case designs, which meet certain requirements, represent "true experimental designs" in that they allow the researcher to determine whether a functional relation exists between the independent and dependent variables (Kennedy, 2005). Such a relation suggests that changes in the independent variable (i.e. introduction of the ESDM intervention) have caused changes in the dependent variable(s) (i.e. the child and/or parent outcome measures). This is because they allow the researcher to control for the effect of confounding factors that may also influence behaviour such as maturation, history, instrumentation or testing effects (Johnson & Christensen, 2012).

Both of the studies in this thesis will use a multiple probe across participants design, which is a common single case design (Kennedy, 2005). In Study 1 the participants will be children with ASD and in Study 2 the participants will be children with ASD and their parent(s). Both studies will consist of four sequential phases: (a) baseline, (b) intervention, (c) generalisation, and (d) follow-up.

In the two studies, all participants will first complete a baseline phase, in which the dependent variable will be systematically measured prior to intervention (Kennedy, 2005). In this phase, 10-min play samples will be collected intermittently (e.g. not every week) in order to establish a pattern of child behaviour prior to the implementation of the intervention. This will provide a baseline against which to compare the effects of the intervention and thus has the potential to allow one to determine whether the intervention has in fact produced a meaningful change in the targeted behaviour compared to baseline levels. In this case, the baseline will be a "controlled" baseline meaning that conditions, such as the setting, materials available, and people present, will be held constant. Three baseline probes will be collected for the first participant. For the remaining participants the baseline probes will continue until the preceding participant has shown some improvement during intervention in one or more outcome measures. The only difference between the baseline and intervention phases will be the implementation/teaching of the ESDM procedures. If the target behaviours of all participants in the study improve following the introduction of the intervention, this would suggest that a "functional relation" has been demonstrated between the implementation of the intervention and an increase in target behaviours. That is, it would suggest that the intervention was responsible for the observed changes in the participants' behaviour.

## **Outcome Measures**

Fidelity of implementation. The primary outcome measure in Study 2 will be the percentage of parent's correct use of the ESDM techniques. Fidelity of implementation (also referred to as treatment fidelity or procedural integrity) is a measure of how well a parent or therapist adheres to the protocol for correct implementation of the procedures that should be used in a particular treatment model, as outlined either in the treatment manual or another standardised protocol (Cadogan & McCrimmon, 2015). It is recommended that parents and therapists receive sufficient training and continued monitoring to ensure that they implement the majority of intervention procedures correctly most of the time (Koegel et al., 2016; Matson, 2007). It is generally agreed that adherence of 80% or more to the treatment procedures represents an acceptable level of fidelity (Borelli et al., 2005). It is very important

to measure fidelity of implementation when evaluating intervention studies. First, if one does not measure fidelity it is not clear whether any changes in child outcome variables are due to the intervention as described in the manual/protocol or variations/deviations from the intended intervention (Bellg et al., 2004). If it is the second case, then the results of the treatment are unlikely to be replicable. Second, it is possible that incorrect implementation of the intervention procedures may be more harmful than providing no intervention at all (Koegel et al., 2016). Despite the importance of evaluating intervention fidelity, many studies do not include this measure. For example, Cadogan and McCrimmon (2015) found that just under a third of studies on the effectiveness of PRT did not include a measure of fidelity of implementation.

The ESDM employs a range of procedures to ensure high fidelity of implementation. In order to become a certified ESDM therapist one must work regularly with young children with ASD, have a graduate degree, and work in or have contact with an interdisciplinary team (Talbott et al., 2016). The steps in the ESDM therapist training process include: (a) attending an introductory training workshop to gain a theoretical understanding of the ESDM, (b) attending an advanced training workshop to gain practical experience using the ESDM and feedback from a certified trainer, and (c) submitting two to three videos using ESDM therapy with two different children. Therapists whose videos receive a rating of 80% fidelity or higher on the ESDM teaching fidelity rating scale then become certified ESDM therapists.

ESDM fidelity is measured on a 13-item fidelity rating scale, which scores each of the ESDM teaching components on a 5-point Likert scale, where a score of 1 represents "no competent teaching", and a score of 5 represents "extremely competent teaching" (Rogers & Dawson, 2010). These 13 items are: (a) management of child attention, (b) quality of the ABC teaching episode, (c) quality of instructional techniques, (d) modulation of child affect and arousal, (e) management of unwanted behaviour, (f) dyadic engagement, (g) child motivation, (h) adult affect, (i) adult sensitivity and responsivity, (j) communicative opportunities and functions, (k) appropriateness of adult's language, (l) elaboration of activities, and (m) transitions between activities.

Of the nine studies that involved therapist delivery of intervention, only four included an on-going measure of fidelity of implementation (Colombi et al., 2016; Dawson et al., 2010; Vismara, Young, et al., 2009; 2013). Of the remaining studies, Eapen et al. (2013) stated that therapists were formally trained in ESDM but not whether they were certified or implemented the procedures with ongoing fidelity. Fulton et al. (2014) and Vivanti et al. (2013; 2014) both stated that at least some of the therapists implementing the study were

certified, but they too did not include an on-going measure of fidelity. Devescovi et al. (2016) did not measure fidelity and stated that therapists were only certified in the model after the completion of the study. Study 1 will include an ongoing measure of therapist fidelity.

All of the ESDM parent coaching studies have measured fidelity using this 13-item rating scale (Rogers, Estes, et al., 2012; Vismara, Colombi, et al., 2009; Vismara, McCormick, et al., 2013; Vismara et al., 2016; Vismara & Rogers, 2008; Vismara et al., 2012). However, only half of these included a measure of the fidelity with which the coach implemented the training procedures (Rogers, Estes, et al., 2012; Vismara, McCormick, et al., 2013; Vismara et al., 2016). Therefore, it is not clear whether the coaches were consistent in their use of the procedures outlined in the ESDM coaching manual (Rogers & Vismara, 2015) or the treatment protocol (e.g. Vismara & Rogers, 2008). Study 2 will improve upon this limitation by including a measure of the coach's fidelity, as well as the parent's fidelity.

Imitation. The percentage of intervals containing imitation was selected as one of the main child outcome measures in both studies because the developers of the ESDM hypothesise that children with ASD generally have deficits in imitation which prevent them from learning from those around them (Rogers et al., 1986; Rogers & Vismara, 2010). This is supported by research which suggests that some, but not all, children with ASD have problems related to mirror neuron systems, which inhibit their ability to imitate others (Williams et al., 2001). Therefore, directly teaching children with ASD to imitate others should improve their ability to learn from peers and adults, and thus, reduce their developmental delays across all areas. Hence, due to the emphasis of the model on teaching imitation skills, it was important to assess whether this increased during intervention. Also, due to having such limited duration of intervention, teaching each child the skills to learn from others would seem particularly beneficial.

Few studies have evaluated the effect of early intervention on the imitation skills of young children with ASD. Three ESDM parent coaching studies have included the frequency of spontaneous imitation as an outcomes measure (Vismara et al., 2012; Vismara, Colombi et al., 2009; Vismara & Rogers, 2008). Spontaneous imitation was defined as unprompted imitation of the parent's actions (with or without objects), vocalisations, or words. Further, several focussed imitation interventions for young children with ASD have reported on the percentage of intervals containing object and gestural imitation (Ingersoll & Gergans, 2007) or the child's rate of spontaneous imitation per minute (Wainer & Ingersoll, 2015). Even fewer studies have investigated the effectiveness of therapist delivered intervention for improving the imitation skills of young children with ASD, and those that do mostly involve

interventions which focussed exclusively on imitation skills (Ingersoll et al., 2010a; Ingersoll, Lewis, & Kroman, 2007; Ingersoll & Screibman, 2006). Indeed, none of the ESDM direct therapy studies included imitation as a main outcome measure. Therefore, due to the hypothesised role of imitation in the observed deficits of children with ASD, and the limited research on the effectiveness of early intervention for improving imitation skills, it was considered to be an important outcome measure in both studies.

**Functional utterances.** The percentage of intervals containing functional utterances was selected as one of the main outcomes measures in both studies for several reasons. First, measures of expressive and/or receptive language are included in the majority of studies evaluating the effectiveness of early intervention approaches for children with ASD, regardless of the delivery method or study design (Lerman et al., 2016; Meadan et al., 2009; Waddington et al., 2016). Therefore, inclusion of this outcome measure would facilitate comparison of the effectiveness of the current studies with previous research. Second, deficits in social communication are one of the defining features of ASD (American Psychiatric Association, 2013). Speech onset is generally delayed in children with autism and studies suggest that between one quarter and half of all children with ASD develop "fluent speech", while approximately a third could be considered nonverbal (using very limited consistent words; Anderson et al., 2007; Pickett, Pullara, O'Grady, & Gordon, 2009; Wodka, Mathy, & Kalb, 2013). Further, parents report communication development to be amongst their top intervention priorities for their children with ASD (Pituch et al., 2011; Rodger, Braithwaite, & Keen, 2004; Whitaker, 2007). Thus, communication would seem to be a particularly important and socially valid target for early intervention.

There are many different methods of measuring children's communication skills using single case design methodology. Previous ESDM parent training studies have measured the frequency of spontaneous utterances, which are defined as unprompted utterances which contain a phonetically correct word or word approximation and are "relevant to the interaction" (Vismara et al., 2012; Vismara, Colombi, et al., 2009; Vismara & Rogers, 2008) or "directed to the parent" (Vismara et al., 2016; Vismara, McCormick, et al., 2013). Several studies have also evaluated the effectiveness of other early intervention approaches for improving children's frequency or percentage of intervals containing utterances (e.g. Hancock & Kaiser, 2002; Kasari, Lawton, et al., 2014; Kaiser & Roberts, 2013; Koegel et al., 1988). A disadvantage of this method of measurement is that it only provides information about the quantity of utterances, rather than the complexity or the variety. Therefore, some early intervention studies for children with ASD have included measures of each child's

mean length of utterance (MLU) in words (Kaiser, Hancock, & Nietfeld, 2000; Kasari, Kaiser, et al., 2014) or the number of different utterances during each session (Kaiser et al., 2000; Kasari, Kaiser et al., 2014; Seung, Ashwell, Elder, & Valcante, 2006). Studies 1 and 2 will include a measure of functional utterances and engaged functional utterances in order to examine whether the total number of utterances improves during the intervention. It will also include an additional evaluation of each child's MLU and variety of utterances in order to examine whether the complexity and diversity of utterances also improves during this time.

Intentional vocalisations. NDBIs generally advocate for teaching skills, including communication skills, in the sequence in which they typically develop in children without developmental disabilities (Schreibman et al., 2015). In the ESDM children are taught prerequisites to spoken language such as non-verbal gestures, eye contact, and intentional vocalisations prior to being taught spoken language (Rogers & Dawson, 2010). Considering the age of the children participating in the ESDM studies, it would appear logical to assume that many of them would not yet have developed these prerequisites for spoken language. Therefore, it would seem important to measure these children's progress on pre-linguistic communication as well as spoken language. Specifically, Studies 1 and 2 will evaluate the percentage of intervals containing intentional vocalisations for the children for whom this is deemed to be a developmental appropriate intervention target. Intentional vocalisations are purposeful vocal sounds that do not contain a phonetically correct word or word approximation, and they are considered to be a precursor to language development in children without ASD or developmental disabilities (Vihman, Macken, Miller, Simmons, & Miller, 1985).

Interestingly, none of the ESDM studies with a single case design have included a measure of pre-linguistic communication, such as intentional vocalisations, despite the model's emphasis on this area of development (Rogers & Dawson, 2010). Indeed, very few studies evaluating the effectiveness of early intervention for children with ASD have included a measures of pre-linguistic communication. Further, the studies that have included this measure are generally focussed interventions which exclusively target pre-linguistic communication such as pre-linguistic milieu teaching (Franco, Davis, & Davis, 2013; Warren, Yoder, Gazdag, Kim, & Jones, 1993). The focus on the effectiveness of interventions for improving spoken communication could mean that children's progress on other non-spoken forms of communication may not be identified. Therefore, Studies 1 and 2 will include a measure of the percentage of intervals containing intentional vocalisations, in

addition to the percentage of intervals containing functional utterances, only for the children for whom this measure is deemed to be developmentally appropriate.

**Engagement.** The percentage of intervals containing engagement was selected as the final child outcome measure in both of these studies. This is because another defining characteristic of ASD is a reduced interest in, or ability to respond appropriately to, social interactions (American Psychiatric Association, 2013). Further, as mentioned in Chapter 1, the social motivation hypothesis suggests that some individuals with ASD may have reduced activation in areas of the brain related to reward when viewing faces, compared to individuals without ASD (Dawson et al., 2005). For these reasons, the ESDM, and other NDBIs place particular emphasis on increasing these children's motivation for, and interest in, social engagement with adults and peers (Schreibman et al., 2015). Thus, it would seem particularly important to evaluate whether the intervention procedures in Studies 1 and 2 produce an increase in this measure.

Several previous ESDM studies have included a measure of child attentiveness and social initiations (Vismara et al., 2012; Vismara, Colombi, et al., 2009; Vismara & Rogers, 2008). Specifically, all of these studies used the child behaviour rating scale (CBRS), which includes behaviours such as attending to the adult, cooperating with instructions, initiating play ideas, and sharing enjoyment and enthusiasm (Mahoney & Wheeden, 1998). However, this measure is rather limited as behaviours are measured retrospectively on a 5-point likert scale. Thus, this could increase the subjectivity of the measure and also prevent detection of small changes in behaviour. Several other NDBIs have included a measure of the "quantity" of social engagement, including the percentage of joint engagement (e.g. Kasari et al., 2010; Kasari et al. 2014), child responsiveness to bids for joint attention (Aldred, Green, & Adams, 2004), eye contact (Vernon, Koegel, Dauterman, & Stolen, 2012), and directed positive affect (Vernon et al., 2012). Studies 1 and 2 will include a broad measure of the percentage of intervals containing engagement which will correspond with the first fidelity item on the ESDM teacher fidelity rating scale (i.e. the therapist or parent's ability to attract attention to their face, voice, and actions). Examples of child behaviours that could indicate engagement include eye contact, shared smiles, turn taking, imitation, giving, sharing, and showing.

# **Data Analysis**

All of the primary outcomes/dependent variables for each study (imitation, functional utterances/intentional vocalisations, and engagement, as well as fidelity of implementation for Study 2 only) will be graphed for each child and/or parent and each phase of the study. These graphs will be visually analysed for observable changes in behaviour due to

intervention. This will involve visual inspection to determine the level, trend, and slope of the data paths (Kennedy, 2005). Visual analysis is commonly used to infer a functional relationship between the independent and dependent variables in single-case research. Descriptive statistics will be provided to strengthen and support the results of the visual analysis. Additional outcome measures (MLU, frequency, and variety of utterances, and fidelity of implementation in Study 1) will be presented in a table with the mean for each child, outcome and phase of the study.

The non-overlap of all pairs (NAP) statistic will be calculated to determine the effect size of improvements from baseline to intervention and baseline to follow-up for each primary outcome measure (Parker & Vannest, 2009). NAP is calculated by comparing each data point in baseline with each data point in intervention or follow-up and noting whether the intervention or follow-up point overlaps (overlapping pair), does not overlap (non-overlapping pair) or ties (tied pair) with the baseline data point. The number of non-overlapping pairs is then divided by the total number of pairs to give an outcome between 0 and 1.0. A score of 0 indicates that all data points in intervention or follow-up overlap with all data points in baseline, whereas a score of 1.0 indicates that none of the data points in intervention or follow-up overlap with any of the data points in baseline. According to Parker and Vannest (2009), NAP scores between 0 and .65 indicate "weak effects", scores between .66–.92 indicate "medium effects", and scores between .92–1.0 indicate "strong effects".

# **Ethical Considerations**

The study was assessed and approved by the Victoria University of Wellington Human Ethics Committee (Reference Number 22085). In Studies 1 and 2 informed consent will be obtained from parents for their own participation and their child's participation (see Appendix C). In Study 2 consent will also be obtained from a second family member for participation in the generalisation phases of the study (see Appendix C). Further, each study will also comply with the ethical standards of the American Psychological Association (American Psychological Association, 2016) and the New Zealand Psychological Society (Evans, Rucklidge, & O'Driscoll, 2007).

Special consideration will be given to the vulnerable nature of participating children and care will be taken to consult carefully with families and to keep them fully informed throughout the research process. For example, the therapist will ensure that she updates the parents about the child's progress after every session and will address any parent questions or concerns as, and when, they arise.

The therapist will meet with parents who are interested in participating in the proposed research, will explain the information sheet and consent form, and will inform parents that they can withdraw from the study at any time without negative consequences. Due to the age of the children participating in this study, and potential delays in receptive and expressive language and cognition, it will not be possible for them to give informed consent to participate (American Psychological Association, 2016). However, their assent to take part in the study will be determined by their apparent willingness to participate in the play/routines-based activities with their parent or the therapist, such as willingly accompanying the adult to the activities and engaging in these activities. If any child shows prolonged behaviour indicating that they would like the session to be terminated, for example, crying, screaming, hitting and other challenging behaviour, and leaving the area, then the session will be terminated for that day. If the child shows this type of behaviour across multiple sessions in a row, the therapist and parent(s) will discuss whether or not the child should continue to participate in the intervention.

The following two chapters will provide an in-depth description of the methods used to answer the six specific research questions detailed earlier in this chapter, as well as the results of each study and a discussion of these results. Specifically, Chapter 5 will describe the method and results of Study 1: Low-Intensity Therapist Delivered ESDM Intervention and Chapter 6 will describe the method and results of Study 2: Parent Training based on the ESDM.

#### **CHAPTER 5**

# STUDY 1: Low-Intensity Therapist Delivered ESDM Intervention Method

# **Participants**

Four children and their parents were recruited for this study. The parents of two of the children were referred to the primary therapist (Waddington) by the District Health Board. The other two parents made contact with the primary therapist after attending an ESDM introductory workshop delivered by Elizabeth Aylward, a certified ESDM trainer. That workshop was conducted at Victoria University of Wellington. During the course of the workshop, the primary therapist explained that she was planning to undertake the present research. She offered these parents the opportunity to participate in the study because their children met the inclusion criteria.

Inclusion criteria were: (a) the child was under the age of 5 years (60 months) at the start of the study; (b) the child had a clinical diagnosis of ASD or met criteria for an ASD diagnosis based on an ADOS assessment (Lord, Rutter, et al., 2012); (c) the child did not have another serious or specific medical, genetic, neurological or sensory condition (e.g., Down syndrome, fragile X) and (d) the child was not receiving intensive early intervention of any type at any time during the study. Table 5.1 gives a summary of each child's age, gender, and results of diagnostic and adaptive behaviour assessment.

Table 5.1

Child Demographic Characteristics and Vineland-II and Social Communication

Questionnaire (SCQ) Results.

	Charlie	Alan	Chris	Jeevan
Age at start of study	2:3	4:3	3:2	3:8
(Years: Months)				
Gender	Male	Male	Male	Male
Vineland II				
Communication	Low	Low	Mod. low	Low
(Adaptive level)				
Receptive	0:1	0:6	1:11	1:6
Expressive	0:3	1:2	1:11	1:8
Written	N/A	2:5	6:9	3:1
Daily Living	Low	Low	Adequate	Low
(Adaptive level)				
Personal	0:10	2:6	0:10	1:4
Domestic	0:7	1:6	4:6	0:7
Community	0:7	2:5	6:10	2:3
Socialisation	Low	Low	Low	Low
(Adaptive Level)				
Interpersonal	0:3	1:1	1:4	0:8
Play	0:7	1:4	0:9	0:7
Coping	1:6	1:6	1:1	0:7
Motor skills	Adequate	Mod. low	Low	Mod. low
(Adaptive Level)				
Gross	2:0	2:5	1:6	3:2
Fine	2:2	2:10	1:8	2:2
Maladaptive	Elevated	Average	Elevated	Clin. sig.
behaviour index				
Internalising	Clin. sig.	Average	Clin. sig.	Clin. sig.
Externalising	Average	Average	Average	Average
SCQ	Risk of ASD	Risk of ASD	Risk of ASD	Risk of ASD

Note: Clin. sig. = Clinically significant, Mod. low= Moderately low

**Charlie.** Charlie (pseudonym) was 2 years and 3 months old at the start of the study and was the sixth of seven siblings. He lived with his mother, father, and all of his siblings. He scored as being at risk for ASD on the lifetime SCQ (Rutter et al., 2003; see Materials for a description of this measure) and had been diagnosed with ASD by a multidisciplinary team at his local District Health Board at the age of 2 years and 2 months. The Child Development team includes an ADOS-2 (Lord, Rutter, et al., 2012), administered by trained professionals, as part of the diagnostic process. His scores on the Vineland-II (Sparrow et al., 2005; see Materials for a description of this measure) indicated that his total score for communication, including both receptive and expressive communication, was low (i.e. two or more standard deviations below the mean for children of his age). His written communication score was not calculated due to his age. His overall adaptive level for the daily living skills and socialisation subdomains was also low, whereas his motor skills were rated adequate, which means that his overall score for that domain was within one standard deviation of the mean for children of his age. His internalising score on the maladaptive behaviour index was clinically significant and his externalising score was average for his age. Charlie's mother reported that, prior to the intervention, Charlie's only spoken word was go in response to someone saying Ready... . set. She also reported that in the past he had learned other words that he no longer seemed to use. Charlie's mother stated that he was an "affectionate boy" who enjoyed cuddles, was quite active, and loved playing outside. Charlie attended play centre every morning, from 9:00 to 11:00 am and had one weekly session of early intervention involving services from a speech language therapist, an early intervention teacher, a music therapist and a physiotherapist. Charlie's parents had not received any ASD-related training or coaching but Charlie's mother had read the ESDM parent manual (Rogers, Vismara, et al., 2012).

Alan. Alan was 4 years and 3 months old at the start of the study. He lived with his mother, father and younger sister. Alan scored as being at risk for ASD on the lifetime SCQ (Rutter et al., 2003) and was diagnosed with ASD by the local Child Development team at the age of 2 years and 11 months. His scores on the Vineland-II (Sparrow et al., 2005) indicated that his overall adaptive level for communication were low, and that his written age equivalency was higher than his receptive and expressive age equivalency. His adaptive level on the daily living skills and socialisation subdomains was also low, whereas his adaptive level for the motor skills subdomain was moderately low, which meant that his overall score for this domain was between one and two standard deviations lower than the mean for children his age. His levels of internalising behaviour and externalising behaviour on the maladaptive behaviour index were average for his age. Alan's mother reported that his most

consistent word was *night-night*, but he also sometimes said *more* and *no*. She also reported that he had learned to request preferred items using the Picture Exchange Communication System (PECS; Frost & Bondy, 2002), but did not consistently use this communication mode. Alan's mother also stated that he had previously been able to say more words and that he would become frustrated due to being unable to communicate. She also said that he often put non-edible objects in his mouth. He was reported to enjoy sensory play, outdoor play (especially the trampoline), and treats such as chocolate and ice blocks. Alan went to kindergarten 4 days a week and attended the same early intervention centre as Charlie. Six months prior to this study, Alan was receiving 10 hours a week of one-on-one ABA therapy but his parents chose to discontinue this service. Alan's parents had previously participated in an ASD parent education course.

**Chris.** Chris was 3 years and 2 months old at the start of the study. He lived with his mother and older sister. He scored as being at risk for ASD on the lifetime SCQ (Rutter et al., 2003) and was diagnosed with ASD by the local Child Development team at the age of 2 years and 11 months. He had a moderately low overall adaptive level on the communication domain of the Vineland-II (Sparrow et al., 2005), but had a very high age equivalency (6:9) for written communication. Specifically, Chris was assessed as being able to print more than 20 words from memory and read up to a second grade level. His adaptive level for daily living skills was average, and his adaptive level for socialisation and motor skills was low. Chris's level of internalising behaviour on the maladaptive behaviour index was clinically significant and his externalising behaviour was average for his age. Prior to intervention, Chris's mother reported that he knew well over 100 words and often communicated using short phrases, for example [the letter] A goes here and I play. His mother stated that he enjoyed reading and spelling and read bedtime stories to his older sister. He was very interested in transportation and also liked social interaction, including playing chasing games with his sister. Chris attended in home childcare three times per week and did not attend kindergarten as his mother reported that he found it too overwhelming. He was also participating in a food intervention and hydrotherapy. Chris' mother had not received any ASD-related training or coaching

**Jeevan.** Jeevan was 3 years and 8 months old at the start of the study. He did not have any siblings. He lived with his mother, father, uncle, aunt, and two cousins. The family primarily spoke Oriya and Hindi at home, but Jeevan was reported to speak English most of the time. Jeevan scored as being at risk for ASD on the lifetime SCQ (Rutter et al., 2003) and was diagnosed with ASD by his local Child Development team at the age of 3 years and 1

month. His scores on the Vineland-II (Sparrow et al., 2005) indicated that his overall adaptive level for communication was low, and that his age equivalency for written language was higher than his receptive and expressive language. His adaptive level for the daily living skills, and socialisation subdomains was also low, whereas his motor skills were moderately low. Jeevan's level of internalising behaviour on the maladaptive behaviour index was clinically significant and his externalising behaviour was average for his age. Jeevan's mother and aunt stated that he had a wide vocabulary, however, he did not often use this language to communicate. He was reported to have vocal stereotypy and frequently sang nursery rhymes to himself. He was also reported to like holding three items at a time such as spoons, straws, and alphabet letters. His mother stated that he really liked cuddles and tickles. Jeevan attended kindergarten every day from 1:00 pm to 5.00 pm and began to attend the same early intervention centre as Charlie and Alan on the 10<sup>th</sup> week of intervention. Jeevan's parents had not received an ASD-related training or coaching

## Setting

The main room used for therapy for each child was decided upon following a discussion with the parents. All of Charlie's sessions were conducted in the downstairs room of his house. This room included a bed, a child sized table and chairs, and a chest of drawers on which the box of toys was placed. Baseline, generalisation, and follow-up sessions for Alan were conducted in the family's living room, which included a couch, a table, a childsized table and chairs, an inflatable "peanut chair", and a television. During generalisation and follow-up there was also a small trampoline in the middle of the room. Intervention sessions were conducted both in the living room and outside. The outside area was well fenced and included a trampoline, a swing, several child-sized bikes, a table and chairs, and an area for water play. All baseline, intervention, and generalisation sessions for Chris were conducted in the living area which included a couch, a television, a cardboard pretend shop, and a bookshelf with toys and books. The house was open plan, thus, the kitchen and dining areas were accessible from the living room but were not used during therapy. The final follow-up session for Chris was conducted in a relative's house because the family were relocating to a different city. This room contained a couch, a desk with a computer, and a chair. All sessions for Jeevan were conducted in the living room of his home, which had two couches, a television, and a dining table.

#### **Materials**

During therapy sessions, the children were able to play with any toy that was already in the room. For example, Charlie had access to a magnet toy and blocks; Alan had access to blocks, trains and cars; and Chris had access to a wide variety of toys including cars, books, blocks, a ten pin bowling set, and finger puppets. Jeevan did not choose any of his own toys during the sessions.

In addition, the therapist brought a large transparent plastic box containing a number of specific "therapy toys" for each session. The box contained an assortment of the toys described in Appendix D. Additional toys included shaving foam, simple cause and effect games such as "Don't Spill the Beans" and a marble run. During baseline, the therapist ensured that each child's favourite toys (as reported by their parents) were available each session. For Charlie, these were bubbles, balloons, blocks, Lego® Duplo, books, and vehicles. For Alan this included cars, playdoh, and items for water play. Chris's favourite toys were reported to be cars, and Jeevan's were alphabet letters. Sessions were filmed using an iPhone® 5s on a small tripod.

The Vineland-II and SCQ were administered via an interview with the parents prior to the start of baseline. The Vineland-II examines adaptive functioning across four developmental domains: communication, daily living skills, self-care and motor skills and several subdomains, for example, receptive, expressive and written language in the communication domain (Sparrow et al., 2005). The Vineland-II takes between 25-min and 1-hr to complete. The SCQ is a brief parent questionnaire designed to screen for ASD (Rutter et al., 2003). It contains 40 dichotomous (yes-or-no) questions related to autism symptoms and scores of over 15 on this measure indicate that the child may be "at risk" for ASD.

# **Dependent Variables**

Data on each dependent variable were collected from the 10-min videos undertaken during each baseline, generalisation, and follow-up session, and on two of every three intervention sessions. Each 10-min of video was divided into 60, 10-s intervals (see Data Collection Studies 1 and 2, Appendix E). Four child-related dependent variables were defined and recorded as being either present or absent for each 10-s interval. These were: (a) imitation, (b) functional utterance (c) meaningful/intentional vocalisation, and (d) engagement with the therapist. Functional utterances included recording the frequency and variety of utterances and the mean length of utterance (MLU). However, variety was not recorded for Chris because he had a wide variety of utterances in the baseline phase. The extent to which parents implemented the ESDM procedures correctly (i.e., with fidelity) during generalisation probes was also recorded.

**Imitation:** Imitating was defined as performing an action with or without an object, or producing a vocalisation, within 10-s of an adult model and without prompting from an

adult, such as the adult saying *Do this* or physically helping the child to perform the action. Partial-interval recording (Kennedy, 2005) was used to record whether or not any instances of imitation had occurred during each 10-s interval. The percentage of intervals containing imitation (out of 60 possible) was calculated for each 10-min play sample.

Functional utterance: A functional utterance was defined as any utterance by the child that: (a) occurred without adult prompting or modelling of the utterance within 10-s of its occurrence, (b) was contextually related to the interaction or task, for example, not unrelated speech, not repetitions of the child's own speech, and not repetitions of adult's prior speech, and (c) contained a phonetically correct approximation of the correct word or word combination (e.g. not saying horse when labelling a cow). In addition, an engaged utterance was an utterance directed to the therapist, or directly related to the therapist's communication, for example *Look at the car*. Unengaged utterances were those which were functionally related to the task, but not directed at the therapist, for example, a child facing away from the parent and spontaneously labelling a toy. Partial-interval recording (Kennedy, 2005) was used to record whether or not any instances of utterances and engaged utterances had occurred during each 10-s interval. The percentage of intervals (out of 60 possible) containing utterances and engaged utterances was calculated for each 10-min play sample. In addition, event recording (Kennedy, 2005) was used to document the frequency of functional utterances in the same 10-min play sample.

**Mean Length of Utterance (MLU):** MLU was defined as the mean number of morphemes (meaningful units of language) per functional utterance in each 10-min play sample. For example, the sentence *Give me apple*, contains three morphemes. Repetition of the same word, for example *Go go* was coded as one morpheme rather than two. Rote learned utterances such as counting and reciting the alphabet were counted as having an MLU of 1. Songs were excluded from this analysis to avoid artificially inflating the MLU.

Variety of utterance: Variety of utterance was a measure of the number of different functional utterances and combinations of functional utterances spoken by each child during each 10-min play session. Different conjugations of verbs (e.g. *go* and *going*) and different combinations of words (e.g. *you*, *go*, and *you go*) were each counted as a unique and different utterance. Repetitions of the same word (e.g. *go go* instead of *go*) and different word approximations of the same word, (e.g. *ba*, and *buba*, for bubbles) were counted as the same utterance. This measure was not calculated for Chris because his variety of utterances was very high during baseline.

Meaningful/intentional vocalisations: A meaningful/intentional vocalisation was

defined as any utterance by the child that: (a) occurred without adult prompting or modelling of the vocalisation, (b) was related to the interaction, for example, not unrelated speech, stereotypy or echolalia, (c) did not contain a phonetically correct approximation of the word or word combination, and (d) did not consist of any whining, screaming, crying, or laughing. In addition, the child needed to be at least partially oriented towards the adult and the vocalisation needed to occur within 10-s of an adult action or utterance and/or the child appeared to be making a request, rejecting response, or comment as indicated by other behaviours such as reaching for or moving towards an object, pushing away an object, or pointing at the object. This measure was only recorded for Alan because it was appropriate for his language level but was not for the other three children. Specifically, Alan had ESDM expressive language goals related to increasing intentional vocalisations and non-verbal communication, but the remaining three children had expressive language goals related to increasing functional utterances. Partial-interval recording (Kennedy, 2005) was used to record whether or not any instances of meaningful/intentional vocalisations had occurred during each 10-s interval. The number of intervals (out of 60 possible) containing meaningful/intentional vocalisations was calculated for each 10-min play sample.

**Engagement with therapist**: Engagement with therapist was defined as any clear indication that the child was attending to the therapist's face, voice, and actions, as well as any instances of the child showing social initiation. More specifically, this dependent measure was recorded when the child was observed to be: (a) orientated towards the therapist, that is facing the therapist; (b) smiling and/or laughing in response to the therapist's action; (c) looking in the direction that the therapist was pointing/indicating; (d) giving, sharing, or showing objects to the therapist; (e) imitating the therapist's actions; (f) taking turns with the therapist; (g) following directions given by the therapist; (h) communicating with the therapist through words, vocalisations, and/or gestures; and/or (i) continuing or elaborating on the therapist's play actions. Engagement did not include repetitive play with objects, taking materials without looking at the therapist, facing away from the therapist, running away from the therapist, engaging in challenging behaviour (e.g. hitting, biting, or screaming), ignoring questions or instructions, and/or repetitively asking for activities or objects that are not available (e.g. making more than three of the same requests in less than 60 s). A measure of partial engagement was obtained by using a partial-interval recording method (Kennedy, 2005). Specifically, the presence or absence of engagement was recorded for each 10-s interval of each 10-min session. A measure of whole engagement was also recorded using a whole-interval recording method (Kennedy, 2005). Specifically, in whole

engagement, the child was required to show behaviours indicating engagement for the entire 10-s of each interval.

Parent fidelity of implementation. This is a measure of parents' correct use of the ESDM techniques. During generalisation probes, the parents' fidelity of implementation was determined using an 18-item questionnaire based on the ESDM fidelity checklist (Rogers & Dawson, 2012; see Appendix F). The questionnaire had 13 fidelity categories: management of attention, ABC format, instructional techniques, affect and arousal, management of unwanted behaviours, dyadic engagement, motivation, positive affect, sensitivity and responsivity, multiple and varied communication, appropriateness of adult language, joint activity structure and elaboration, and transition between activities. Each of these categories contained 1 to 3 items and resulted in a maximum score of 12 points. Thus, if a category contained 3 items, the maximum score for each item was 4 points, whereas if a category only had one item, the maximum score for that item was 12 points. Examples of items included "adult attracts attention to face and auditory cues" and "teaching opportunities occurred more than once every 30 s". Each item was scored on a five point Likert-scale, where a score of "never" resulted in zero points, a score of "occasionally" resulted in one quarter of the possible points for the item (e.g. 1 if there were 4 possible points or 3 if there were 12 possible points), a score of "sometimes" resulted in half of the possible points for that item, a score of "usually" resulted in three quarters of the possible points for the item, and a score of "consistently" resulted in all the possible points for that item. The item "multiple and varied" communication" was scored differently: a score of "none" resulted in 0 points, a score of "one" resulted in 3 points, a score of "two" resulted in 6 points, a score of "three" resulted in 9 points, and a score of "four +" resulted in the full 12 points. The questionnaire resulted in an overall percentage of fidelity based on the sum of the parent's score divided by the total scores for all the items (136-156 points depending on whether all phases of an activity were observed in the video)  $\times$  100%.

# **Experimental Design**

The effects of the intervention were evaluated using a multiple probe across participants design (Kennedy, 2005). Each child/parent dyad participated in the following sequence of phases: baseline, intervention, generalisation, and follow-up. This design was considered an effective analytic tool for determining if the therapist's use of the ESDM procedures was responsible for increases in the four child-related dependent variables.

#### **Procedures**

**Baseline.** One baseline session occurred per week for each child. Session times varied, but occurred primarily in the morning for Charlie, Chris, and Jeevan, and in the afternoon for Alan. At the start of each session, the therapist started the video recorder and then placed the box of toys in front of the child. The therapist waited 10-s and if the child did not chose a toy, then she placed a selection of toys from the box in front of the child. If the child did not select a toy within 10-s then the therapist selected a toy and probed an imitation skill or vocal utterance before handing the toy to the child. The session continued for 10-min from the time the child selected a toy. During the filming, the therapist interspersed 10 naturalistic imitation probes (e.g. banging on a drum and looking at the child expectantly) and 10 naturalistic probes for functional verbal utterances (e.g. holding up a car and saying What's this) during each the session. These were conducted when an opportunity arose, for example, if the child were playing with a car, the therapist would also grab a car and say *Vroom vroom*, giving the child the opportunity to imitate. No other response prompting was used at any time during baseline. The therapist responded in an appropriate and friendly manner to all social initiations from the child. For example, if a child held up a fire truck and looked at the therapist, she might say Wow, what a cool fire truck. Parents and siblings were instructed not to interact with the child during the 10-min period.

**Baseline generalisation probe.** Each child's mother participated in the generalisation probe that took place during the baseline phase for Alan, Chris, and Jeevan. This probe occurred during the 2nd week of intervention for Charlie due to difficulties with scheduling the probe during baseline. For the probe, the mother was instructed to "play with your child as you normally would". Once the child was settled into play with his mother, the therapist began filming the 10-min session. The therapist did not make any comment or give any feedback about the child's play or the mother's interaction with the child during or after the filming.

Intervention. Intervention was based on the principles of the ESDM, as detailed in the intervention manual (Rogers & Dawson, 2010). During the first intervention session, the primary therapist (Waddington) played in a naturalistic way with the child for approximately 1 hr. A second therapist then noted on the ESDM curriculum checklist (Rogers & Dawson, 2010) whether or not the child displayed a range of developmental skills and would instruct the therapist on further skills to probe during the play. The second therapist was a graduate student who had completed the advanced ESDM workshop. Based on this first session and in consultation with the parents, two to three goals were selected for each child for each of the nine developmental domains (i.e., receptive communication, expressive communication,

social skills, imitation, cognition, play, fine motor, gross motor, and behaviour). Chris was the only child who was at a developmental level to have goals in the joint attention domain. Table 5.2 shows the items from the curriculum checklist that were used for each child's goals. An example of a goal for Charlie was "When an adult presents Charlie with two preferred objects, one in each hand, he will make eye contact and point with his index finger in order to choose an item, on four out of five opportunities across three consecutive sessions and two different adults and settings".

During the remaining sessions, the goals were embedded into the therapist's play with the child and were taught by applying the following behaviour analytic teaching principles: (a) the delivery of learning opportunities at least every 30s, (b) delivery of clear antecedents, (c) the use of reinforcement, and (d) the use of instructional strategies such as prompting, shaping, chaining, and fading. At the beginning of each session, the therapist would greet the child and then allow him to verbally or non-verbally (e.g. leading the adult to the activity or object, pointing) choose the first activity, either by presenting a limited choice of two options or allowing the child to choose any toy or activity. Teaching occurred through the use of two types of routines: sensory social routines, in which the therapist played with the child without an object (e.g. songs, chase, tickles), or using special sensory objects that the child did not operate (e.g. balloons, bubbles, shaving foam); and joint activity routines, in which the therapist and the child played together with an object (e.g. blocks, books, balls). Throughout these routines, the therapist maintained a positive affect, employed strategies to maximise the child's motivation (e.g., offering choices, following the child's lead, and being sensitive and responsive to all attempts at communication), and made conscious attempts to attract and maintain the child's attention to her face, voice, and actions. When the therapist determined that there were no more learning opportunities during the chosen activity, the child seemed bored, or the child initiated finishing an activity, the therapist would instruct the child to tidy up and, once the child had done so, he could select a new activity. Sessions were terminated after 1-hr or when the therapist determined from the child's behaviour that he or she no longer wanted to continue. For example, during the first couple of weeks of intervention, Alan would leave the therapy area after about 40-min of intervention, and would try and turn on the television.

Data were collected in the first 2 of 3 weekly sessions with the exception of the first week as the first session was used for the curriculum assessment. On rare occasions data were collected during the third session if there was a problem with filming or the child was out of camera for a significant proportion of one of the other 10-min play samples. The therapist

usually began filming at the start of the session however, data was collected for 10 min from the start of the activity that began closest to 20-min of settled play. This may have been slightly before or after 20-min depending on the length of the activities and structure of the session.

During intervention Alan showed challenging behaviour associated with requesting access to cartoons at the start of many sessions. It was found that conducting the beginning of sessions outside caused a reduction in this challenging behaviour. In line with ESDM procedures and maximising child motivation, sessions were conducted for Alan in varying locations around the house including outside, in his bedroom, and in the therapy room used in baseline. For this reason, an additional phase involving probes in the therapy room was conducted, to ensure consistency between baseline and intervention. During this phase, Alan was required to begin the session in the therapy room before being allowed to go outside and filming took place in the first 10-min of the session, rather than after 20-min

**Generalisation.** This phase took place one week after the final intervention session for each child. Procedures were identical to the baseline generalisation probe, except that three 10-min videos were taken during a single week.

**Follow-up.** Follow-up took place four weeks after the last intervention session for each child. These sessions were conducted by the therapist using the same procedures as intervention. These sessions lasted 10 min and the entire session was filmed.

Table 5.2

Curriculum Checklist Items Used to Inform Goals for Each Child.

Domain	Charlie	Alan	Chris	Jeevan
Receptive	1-12 (Level-Item):	1-5: Follow a proximal point to	3-6: Follows two or more	1-5: Follow a proximal point
Communication	Responds by stopping actions	place objects in containers,	instructions given in	to place objects in containers,
	momentarily in response to	puzzle pieces, etc.	situational routines	puzzle pieces, etc.
	inhibitory words (e.g., "no",	1-13: Gives object as verbally	3-8: Differentiates early size	1-13: Gives object as verbally
	"stop").	requested when paired with	concepts – big/little.	requested when paired with
	1-13: Gives object as verbally	adult's outstretched hand.	3-12: Understands pronoun	adult's outstretched hand.
	requested when paired with		referents "mine" and	1-14: Performs a one step,
	adult's outstretched hand.		"yours".	routine instruction involving
	1-14: Performs a one step, routine			body actions paired with
	instruction involving body actions			verbal/gesture cue
	paired with verbal/gesture cue.			
Expressive	1-3: "Asks" for help by handing	1-3: "Asks" for help by	1-8: Points to indicate a	1-8: Points to indicate a choice
Communication	object to adult.	handing object to adult.	choice between two objects.	between two objects.
	1-8: Points to indicate a choice	1-8: Points to indicate a choice	2-10: Shakes head and says	2-2: Produces 6-10 single
	between two objects.	between two objects.	"no" to refuse.	words or approximations
	2-2: Produces 6-10 single words		2-12: Asks (approximates)	within the context of familiar
	or approximations within the		"What's that?" when	routines, sensory-social
				routines, songs.

	context of familiar routines,		encountering something	2-10: Shakes head and says
	sensory-social routines, songs.		unfamiliar.	"no" to refuse.
Joint Attention	N/A	N/A	2-5: Spontaneously "shows"	N/A
			objects.	
			2-7: Spontaneously points to	
			interesting objects.	
Social Skills	1-2: Uses motor prompt to initiate	1-7: Has a repertoire of $5-10$	2-2: Verbally requests or	1-2: Uses motor prompt to
	or continue a sensory social	sensory social games.	physically initiates familiar	initiate or continue a sensory
	routine.	1-9: Responds to greeting by	social games.	social routine.
	1-7: Has a repertoire of $5-10$	gesture or vocalisation.	2-4: Uses gesture or words	1-9: Responds to greeting by
	sensory social games.		to attain adult's attention.	gesture or vocalisation.
Play	1-6: Plays independently with	1-6: Plays independently with	2-3: Carries out single	1-6: Plays independently with
	toys requiring several different	toys requiring several different	action with a prop on a doll	toys requiring several different
	motor actions	motor actions	or animal.	motor actions
	1-8: Completes play task and puts	1-8: Completes play task and	2-4: Combines functionally	1-8: Completes play task and
	away.	puts away.	related actions on a play	puts away.
			theme.	
Imitation	1-1: Imitates 8-10 one step actions	1-1: Imitates 8-10 one step	1-4: Imitates six oral-facial	1-1: Imitates 8-10 one step
	on objects.	actions on objects.	movements.	actions on objects.
		1-2: Imitates 10 visible motor	2-8: Imitates two movement	1-2: Imitates 10 visible motor

	1-2: Imitates 10 visible motor actions inside song/game routines.	actions inside song/game routines.	sequences in song/game routines.	actions inside song/game routines.
Cognition	1-1: Matches/sorts identical	1-1: Matches/sorts identical	2-7: Matches/sorts in two	1-1: Matches/sorts identical
	objects.	objects.	dimensions.	objects.
	1-2: Matches/sorts identical	1-2: Matches/sorts identical	3-6: Plays games involving	1-2: Matches/sorts identical
	pictures.	pictures.	memory for hidden objects.	pictures.
Fine Motor	2-3: Copies three or more simple	1-7: Uses a pincer grasp and a	1-6: Takes apart pop beads,	1-8: Stacks three big blocks in
	block designs.	three-finger grasp as	Duplos.	a tower (or stacking cups).
	2-5: Imitates five or more simple	appropriate to toy.	1-8: Stacks three big blocks	1-9: Makes marks, lines,
	actions on play dough (roll, poke,	1-9: Makes marks, lines,	in a tower (or stacking	scribbles, and dots with
	pat, squeeze).	scribbles, and dots with	cups).	markers/crayons.
		markers/crayons.	1-9: Makes marks, lines,	
			scribbles, and dots with	
			markers/crayons.	
Gross Motor	1-1: Kicks big ball.	1-1: Kicks big ball.	2-2: Jumps off step and over	1-1: Kicks big ball.
	1-8: Rolls ball back and forth with	1-8: Rolls ball back and forth	obstacles.	1-7: Throws ball and beanbags
	another person.	with another person.	2-6: Kicks ball into target.	any direction.

# **Interobserver Agreement**

Interobserver agreement (IOA) was assessed by having an independent observer collect data on the four child-related dependent variables during 29 to 100% of the 10-min videos for the baseline, intervention, generalisation, and follow-up sessions. The primary therapist (Waddington) taught the independent observer, a Masters of Educational Psychology student, how to use the data collection sheets and ensured that she understood the operational definitions of each dependent variable. The observer then practiced coding a video of each of the children and discussed any issues that arose with the primary therapist, who directed her to the relevant sections of the operational definitions. These practice videos were not included in the overall IOA.

IOA percentages were calculated using interval agreement (Kennedy, 2005). An agreement occurred if both the primary therapist and the independent observer recorded an occurrence of the behaviour or if both did not record an occurrence of the behaviour during that interval. A disagreement occurred if one observer recorded an occurrence of the behaviour and one did not. The overall percentage of agreement for each session was calculated using the formula: Agreements/(Agreements + Disagreements) × 100%. Table 5.3 indicates that mean IOA was between 80 and 98% for all children and outcomes, except for agreement on Chris' whole engagement which was a mean of 78%.

Table 5.3

Means (and Ranges) for IOA Percentages Across Children and Dependent Variables.

	Charlie	Alan	Chris	Jeevan
Imitation	89%	90%	91%	93%
	(77 - 97%)	(78 - 98%)	(83 - 98%)	(73 - 100%)
Total Utterances	88%	98%	90%	91%
	(73 - 93%)	(93 – 100%)	(85 - 97%)	(77 - 98%)
Engaged Utterances	88%	98%	87%	90%
	(73 - 93%)	(93 – 100%)	(77 - 97%)	(75 - 100%)
Intentional Vocalisations	N/A	84%	N/A	N/A
		(77 - 98%)		
Partial Engagement	90%	95%	92%	92%
	(73 - 97%)	(85 – 100%)	(72 - 100%)	(83 - 100%)

Whole Engagement 84% 82% 78% 88% (60 - 98%) (75 - 100%) (37 - 98%) (68 - 100%)

# **Procedural Integrity**

This is a measure of how well the therapist followed the procedures in each phase of the study. During the baseline and generalisation phases, procedural integrity (PI) was assessed using a checklist that was completed by the same independent observer who conducted the IOA check (see Appendix G). The checklist described each step of the procedures during that phase, for example "the session lasted ten minutes from the child taking a toy", and "therapist responded appropriately to any child attempts to initiate play or interaction". The percentage of PI was calculated using the formula: steps correct/total steps  $\times$  100%. Mean PI during these phases was 100% for Charlie, Alan and Jeevan, and 98% for Chris (range = 92 – 100%).

Only one therapist (Waddington) conducted all phases of the study. She had several years of experience working with children with ASD and became a certified ESDM therapist in the  $13^{th}$  week of the study, which meant that she had completed the introductory and advanced ESDM workshops, and had submitted videos of two 30-min ESDM sessions which were judged by a certified ESDM trainer to meet the criteria of over 80% fidelity on the ESDM teaching fidelity rating system (Rogers & Dawson, 2010). Further, ongoing ESDM fidelity/PI during intervention and follow-up was evaluated by the independent observer using the same measure of fidelity of implementation as was used with parents during the generalisation phase (see **Dependent Variables**; Appendix F). Mean PI during these phases was 93% for Chris (range = 87 - 100%), 93% for Jeevan (range = 87 - 98%), 92% for Alan (range = 87 - 96%) and Charlie (range = 89 - 94%).

### **Results**

# **Imitation**

Figure 5.1 shows the percentage of intervals containing independent (unprompted) instances of imitation for each child across sessions and during the generalisation probes.

# Imitation.

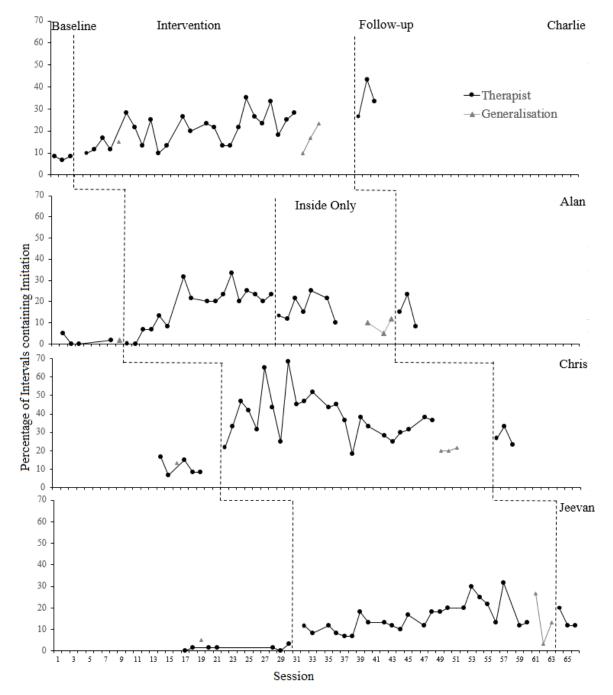


Figure 5.1. Percentage of 10-s intervals containing at least one independent (unprompted) instance of imitation per 10-min play sample for each child across phases.

Figure 5.1 indicates that during the baseline phase Charlie had a mean of 7.8% of intervals containing imitation per 10-min play sample which increased to a mean of 20.5% of

intervals during the intervention phase (NAP=1.0). Visual analysis of the graph indicates a variable but increasing trend from the first to last intervention session. The percentage of intervals containing imitation increased again during follow-up to a mean of 30.4% of intervals (NAP=1.0). With Charlie's mother, 15% of intervals contained imitation during the generalisation probe at the start of intervention compared to a mean of 16.7% of intervals during the post-intervention generalisation phase.

During the baseline phase, Alan had a mean of 1.7% of intervals containing imitation per 10-min play sample which increased to a mean of 17.3% during intervention (NAP= 0.94). Visual analysis of the graph indicates that the percentage of intervals containing imitation increased steadily during intervention and was maintained during the Inside Only procedural modification. This increase was also maintained during the follow-up phase, with a mean of 15.6% of intervals containing imitation (NAP=1.0). With Alan's mother, the percentage of intervals containing imitation increased from 1.7% during the baseline generalisation probe to a mean of 8.9% during the post-intervention generalisation phase.

During the baseline phase, the mean percentage of intervals containing imitation for Chris was 11%, which increased to 38.5% in intervention (NAP=1.0). Visual analysis of the graph indicates that imitation increased for the first nine recorded sessions and then decreased but still remained higher than baseline (NAP=1.0). The mean percentage of intervals containing imitation decreased during follow-up to 27.7% but still remained higher than baseline levels (NAP=1.0). The percentage of intervals containing imitation with Chris' mother increased from 13.3% during the baseline generalisation probe to a mean of 20.5% in the post-intervention generalisation phase.

During the baseline phase the mean percentage of intervals containing imitation for Jeevan was 1.4% which increased to a mean of 15.5% during intervention (NAP=1.0). Visual analysis of the graph indicates a slight increasing trend during the intervention phase. The increase in imitation was maintained during follow-up, with a mean of 15.5% of intervals containing imitation (NAP=1.0). The percentage of intervals containing imitation with Jeevan's mother increased from 5% during the baseline generalisation probe to a mean of 14.4% during the post-intervention generalisation phase.

## **Functional Utterances/Intentional Vocalisations**

Figure 5.2 shows the percentage of 10-s intervals containing intentional vocalisations, functional utterances, and engaged functional utterances for each child across sessions and during the generalisation probes.

#### Follow-up Charlie Baseline Intervention Utterance- Therapist Utterance- Mother -Engaged- Therapist Engaged-Mother Percentage of Intervals containing Vocalisations, Functional Inside only Alan Utterances, and Engaged Functional Utterances --- Vocalisation- Therapist -∆-Vocalisation- Mother Chris

**Functional Utterances.** 

1 3

Figure 5.2. Percentage of 10-s intervals containing vocalisations, functional utterances, and engaged functional utterances per 10-min play sample for each child across phases.

5 7 9 11 13 15 17 19 21 23 25 27 29 31 33 35 37 39 41 43 45 47 49 51 53 55 57 59 61 63 65 67

Session

Figure 5.2 indicates that during baseline, Charlie had a mean of 2.8% of intervals containing functional utterances and 1.1% of intervals included engaged utterances. Thus, 35% of all utterances in this phase were engaged. During intervention this increased to a mean of 30.1% of intervals containing functional utterances (NAP = 0.98), and a mean of 24.9% of intervals included engaged utterances (NAP = 1.0). In this phase 83% of all utterances were engaged. Visual analysis of the graph suggests an increasing trend for

functional utterances until the  $6^{th}$  recorded session which remained relatively stable until the  $18^{th}$  recorded session, when it increased again. There was also an increasing trend for engaged functional utterances until the  $6^{th}$  recorded intervention session, which stayed relatively stable for the remainder of intervention. The mean percentage of intervals containing functional utterances increased again during the follow-up phase with a mean of 46.1% (NAP = 1.0) for functional utterances and 31.1% for engaged utterances (NAP = 1.0). Sixty-six percent of utterances during this phase were engaged. With Charlie's mother, the percentage of intervals containing functional utterances increased from 10% in the generalisation probe, to a mean of 46.1% during the post-intervention generalisation phase. The percentage of intervals containing engaged utterances with his mother also increased from 6.7% to 31.1%. Table 5.4 shows the mean MLU, variety of utterances and frequency of utterances for all of the children across each phase of the study. This table indicates that Charlie's MLU was highest during the follow-up probe and lowest during the  $1^{st}$  month of intervention. His frequency and variety of utterances were both highest during the follow-up probe and lowest during the follow-up probe and lowest during baseline.

During the baseline phase of the study Alan had a mean of 4.2% of 10-s intervals containing meaningful/intentional vocalisations per 10-min play sample, this increased to a mean of 29% of intervals (NAP = 0.96) during the intervention phase. Visual analysis of the graph indicates a steady increase in meaningful/intentional vocalisations during intervention which was maintained during the inside only procedural modification. The mean percentage of intervals containing meaningful/intentional vocalisations increased again during follow-up to 52.2% (NAP = 1.0). The percentage of intervals containing intentional vocalisations with Alan's mother also increased from 8.3% during the baseline generalisation probe to a mean of 37.7% during the post-intervention generalisation phase.

All of Alan's functional utterances were engaged across all phases of the study. During baseline the mean percentage of functional utterances for Alan was 0.4%, which increased slightly in intervention to 1.6% (NAP = 0.59). Visual analysis of the graph indicates that most functional utterances occurred during the inside only phase of intervention. This slight increase was maintained during follow-up with a mean of 2.8% of intervals containing functional utterances (NAP = 0.96). The percentage of intervals containing functional utterances with Alan's mother increased from none in the baseline generalisation probe to a mean of 4.4% during the post-intervention generalisation phase. Table 5.4 indicates that Alan's MLU remained stable at 1.0 during baseline, intervention and follow-up. His variety and frequency of utterances both increased slightly from 0.3 in

baseline to 1.0 and 1.6 respectively.

During the baseline phase for Chris there was a mean of 36.3% of intervals containing functional utterances and a mean of 17.7% of intervals containing engaged utterances. 45% of all functional utterances during this phase were engaged utterances. This increased to a mean of 69% of intervals containing functional utterances during intervention (NAP = 0.95), and a mean of 60.3% of intervals containing engaged utterances (NAP = 0.99). Eighty-four percent of all functional utterances during this phase were engaged utterances. Visual analysis of the graph indicates an increasing trend for functional utterances, including engaged functional utterances, during baseline, which continues in intervention. The percentage of intervals containing functional utterances increased again during follow-up to a mean of 80.6% (NAP = 1.0), and a mean of 71.7% for engaged utterances (NAP = 1.0). Thus, during this phase engaged utterances comprised 84% of all functional utterances. The percentage of intervals containing functional utterances with Chris' mother increased from 28.3% during baseline to a mean of 73.9% after the intervention. The percentage of intervals containing engaged utterances also increased from 20% to 66.1%. Table 5.4 indicates Chris' MLU and frequency of utterances. It indicates that Chris' MLU increased steadily from 2.4 in baseline to 3.5 at follow-up. His frequency of utterances also increased steadily from 26.4 in baseline to 72.3 in follow-up. The variety of utterances was not calculated for Chris because he had a wide variety of utterances in baseline.

During the baseline phase for Jeevan there was a mean 19.8% of intervals containing functional utterances and a mean of 3.1% of intervals containing engaged utterances. Thus, 13% of utterances were engaged during this phase. This increased to a mean of 69% of intervals for functional utterances (NAP = 0.93) and 56.5% of intervals for engaged utterances (NAP = 1.0). During this phase 90% of utterances were engaged. Visual analysis of the graph suggests a sharp increase in the percentage of functional utterances, including engaged utterances, from the  $4^{th}$  to  $5^{th}$  intervention session, which was variable but remained high for the remainder of the intervention phase. During follow-up the percentage of intervals containing imitation decreased to a mean of 60% (NAP = 1.0). The percentage of intervals containing engaged utterances also decreased to 50% (NAP = 1.0). During this phase 82% of functional utterances were engaged. The percentage of intervals containing functional utterances with Jeevan's mother increased from 11.7% during the baseline phase to a mean of 55.6% following intervention. The percentage of intervals containing engaged utterances also increased from 6.7% to 38.9% following intervention. Table 5.4 indicates that Jeevan's MLU increased slightly during the intervention but that this increase was not maintained at follow-

up. His variety of utterances increased steadily during intervention and remained at a similar level at follow up. His frequency of utterances increased from baseline to the  $2^{nd}$  month of intervention before decreasing in the final month of intervention and follow-up.

Table 5.4

Mean MLU, Variety and Frequency of Utterances (1 d.p.) for All Children Across Baseline,
Intervention, and Follow-up Phases.

	MLU	Variety	Frequency
Charlie			
Baseline	1.2	1.3	2.0
Month 1 intervention	1.1	3.8	12.3
Month 2 intervention	1.2	9.1	24.4
Month 3 intervention	1.3	14	24.9
Follow-up	1.6	21	33.0
Alan			
Baseline	1.0	0.3	0.3
Month 1 intervention	N/A	N/A	0
Month 2 intervention	1.0	1.0	1.25
Month 3 intervention	1.0	1.4	1.9
Follow-up	1.0	1.0	1.6
Chris			
Baseline	2.4	N/A	26.4
Month 1 intervention	2.7	N/A	44.3
Month 2 intervention	2.9	N/A	53.9
Month 3 intervention	3.2	N/A	60.6
Follow-up	3.5	N/A	72.3
Jeevan			
Baseline	1.1	11.4	14.3
Month 1 intervention	1.3	15.0	36.5
Month 2 intervention	1.5	27.6	70.3
Month 3 intervention	1.6	32.9	52.9
Follow-up	1.1	32.3	48.3

Note. The mean for MLU was calculated only from the sessions in which the child had one or more functional utterances.

# **Engagement**

Figure 5.3 shows the percentage of 10-s intervals containing partial or whole engagement for each child across sessions and during the generalisation probes.

# Engagement.

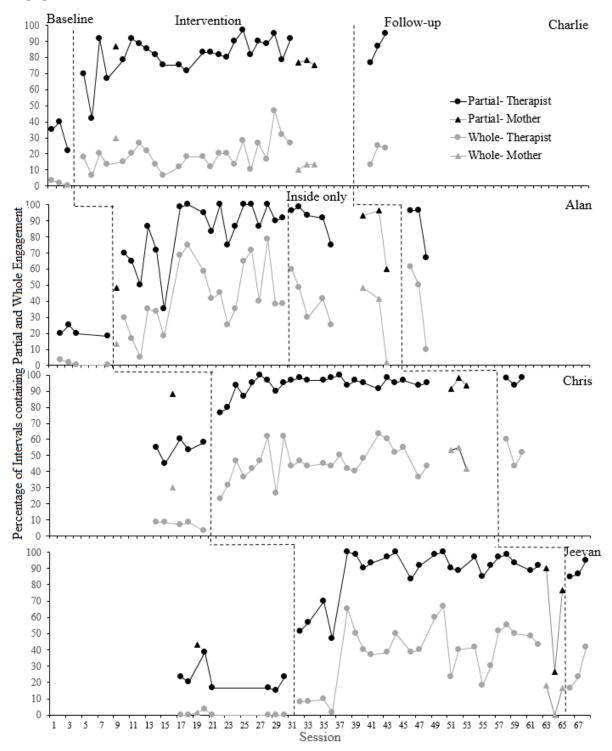


Figure 5.3. Percentage of 10-s intervals containing partial engagement and whole engagement per 10-min play sample for each child across phases.

Figure 5.3 indicates that, during baseline Charlie had a mean of 32.2% of intervals containing partial engagement and 1.7% of intervals containing whole engagement which increased to a mean of 81.5% of intervals containing partial engagement (NAP = 1.0) and 19.2% of intervals containing whole engagement (NAP = 1.0) during intervention. Visual analysis of the graph indicates that partial engagement increased during the first six recorded intervention sessions before decreasing slightly until the 12<sup>th</sup> intervention session and then increasing again. The increase in intervals containing partial and whole engagement was maintained at follow-up with a mean of 86.1% of intervals (NAP=1.0) and 19.2% of intervals (NAP=1.0) respectively. Charlie's partial engagement with his mother decreased from 86.7% of intervals in the generalisation probe at the start of intervention to a mean of 76.7% of intervals in the post-intervention generalisation probe at the start of intervention to a mean of 12.2% of intervals in the post-intervention generalisation probe at the start of intervention to a mean of 12.2% of intervals in the post-intervention generalisation probe.

During the baseline phase, Alan had a mean of 20.8% of intervals containing partial engagement and 1.7% of intervals containing whole engagement which increased to a mean of 85% of intervals (NAP = 1.0) containing partial engagement and 42.6% of intervals containing whole engagement (NAP = 1.0) during the intervention phase. Visual analysis of the graph indicates a variable but increasing trend from the first to last intervention session for both partial and whole engagement, which was maintained during the inside only procedural modification. The increase in the percentage of intervals containing partial and whole engagement was maintained during follow-up with a mean of 86.1% of intervals containing partial engagement (NAP=1.0) and 20.6% of intervals containing whole engagement (NAP=1.0). The percentage of intervals during the baseline generalisation probe to a mean of 83.3% of intervals during the post-intervention generalisation phase (NAP = 1.0). The percentage of intervals containing whole engagement also increased from 13.3% in the baseline generalisation probe to a mean of 40.6% in the post-intervention generalisation phase (NAP = 1.0).

During baseline Chris had a mean of 54.3% of intervals containing partial engagement and a mean of 7% of intervals containing whole engagement. During intervention this increased to a mean of 93.9% of intervals containing partial engagement (NAP = 1.0) and 37.6% of intervals containing whole engagement (NAP = 1.0). Visual

analysis of the graph suggests that Chris's partial engagement increased during the first six recorded sessions and then stayed relatively stable for the remainder of the intervention phase. The increase in percentage of intervals containing both partial and whole engagement was maintained during follow-up with a mean of 96.6% of intervals (NAP = 1.0) and 51.6% of intervals (NAP = 1.0) respectively. The percentage of intervals containing partial engagement with Chris' mother increased from 88.3% in the baseline generalisation probe to a mean of 94.4% during the post-intervention generalisation phase. The percentage of intervals containing whole engagement also increased from 30% in the baseline generalisation probe to a mean of 50% during the post-intervention generalisation phase.

For Jeevan, the mean percentage of intervals containing partial engagement during baseline was 21.9% and the percentage of intervals containing whole engagement was 0.5%. During intervention this increased to a mean of 87.4% intervals containing partial engagement (NAP = 1.0) and 31.8% of intervals containing whole engagement (NAP = 0.99). Visual analysis of the graph indicates a sharp increase in partial and whole engagement from the 4<sup>th</sup> to the 5<sup>th</sup> recorded session, which was variable, but remained relatively high for the remainder of intervention. The increase in intervals containing partial and whole engagement was maintained during follow-up with a mean of 88.9% of intervals (NAP = 1.0) and 27.2% of intervals (NAP = 1.0) respectively. The percentage of intervals containing partial engagement with Jeevan's mother increased from 43.3% during the baseline generalisation probe to a mean of 64.4% during the post-intervention generalisation phase. The percentage of intervals containing whole engagement also increased from 1.7% during the baseline generalisation phase.

# **Parent Fidelity of Implementation**

Table 5.5 shows each parent's percentage of fidelity of implementation in the generalisation probe before the intervention (or during the 2<sup>nd</sup> week of intervention for Charlie) and the mean of the three generalisation probes following intervention. Mean fidelity increased for Alan, Chris, and Jeevan's mother following intervention and remained relatively stable for Charlie's mother.

Table 5.5

Mean Parent Fidelity of Implementation of ESDM Techniques Prior to, and After,
Intervention for All Children.

	Charlie	Alan	Chris	Jeevan
Baseline	81.1%	53.8%	58.0%	33.7%
After Intervention	82.7%	66.9%	73.6%	55.0%
Increase	1.6%	13.1%	15.6%	21.3%

#### **Discussion**

One aim of this study was to evaluate the effectiveness of 3 hours per week of therapist delivered ESDM therapy for 12 weeks for improving imitation, functional utterances, and engagement for four young children with ASD. A second aim was to assess if any treatment gains would maintain over time and if parents could implement the procedures and maintain child treatment gains during generalisation probes. The results of this study suggest that, following the 12 weeks of therapy, each child increased his engagement with the therapist and imitated the therapist more. Three of the four children increased their use of functional utterances, while the fourth child increased his use of intentional vocalisations. These improvements were maintained four weeks after the intervention had finished. The children also showed some increases in each of these skills with their mothers, although in some cases these increases were not as large as with the therapist. A study conducted on parent perceptions of this intervention also suggested that parents found the intervention to be highly acceptable (Ogilvie & McCrudden, 2017). Overall, these results suggest that the present application of ESDM, which was a relatively short-duration and low-intensity therapy appears to have produced some positive outcomes for the four participating children.

# **Engagement**

All of the children were more engaged with the therapist during the intervention compared to baseline and maintained this improvement at follow-up. It does not appear that any previous research evaluating therapist delivered ESDM has included a measure of engagement. However, two of the ESDM parent training studies did measure the child's attentiveness to the therapist and also found that this generally increased during the intervention and was maintained at follow-up (Vismara & Rogers, 2008; Vismara, Colombi, et al., 2009). Further, several studies have also found that child engagement increased during

therapist delivery of a targeted joint attention intervention to young children with ASD (Chang et al. 2016, Goods et al., 2013; Kaale et al., 2012). Thus, similar to previous research, the results of this study suggested that short-term, low-intensity ESDM intervention did lead to an increase in child engagement that was maintained after the treatment ended.

Each child became immediately more engaged with the therapist from the first intervention session. This increase was most likely due to many of the ESDM techniques which are hypothesised to increase engagement (Rogers & Dawson, 2010). This includes the use of relationship-focused strategies, such as positive affect and fun sensory social games, and strategies to increase child motivation such as child choice, encouraging attempts, and mixing maintenance and acquisition tasks. As these techniques were all implemented at the same time, it is not clear whether: (a) one of these techniques was the most effective for increasing engagement, (b) the combination of techniques was the most effective, or (c) different children responded differently to each of the techniques. Thus, more research is needed on the effect of each of the ESDM techniques, and different combinations of these techniques, on child engagement.

Although each child immediately became more engaged with the therapist during intervention, their engagement continued to increase until the 2<sup>nd</sup> or 3<sup>rd</sup> week of intervention. This may have been partly because the therapist was not familiar to them prior to the study. Therefore, the increasing engagement could merely represent increasing familiarity with the therapist. Research suggests that children with ASD, much like their typically developing peers, may be generally more engaged with people they know well compared to unfamiliar people (Dissanayake & Crossley, 1996). However, the improvements in engagement are unlikely to be solely due to familiarity because, for each child except for Charlie, their average level of engagement with the therapist in intervention was higher than their engagement with their mother during baseline. On the other hand, this increase could have been due to increasing therapist familiarity with each child and his toy and activity preferences. Specifically, during the first few intervention sessions the therapist may not yet have identified the child's most preferred toys or sensory social games, or actions within those games. An additional reason for this increase could be that the therapist was directly targeting and reinforcing behaviours related to engagement (Dawson & Rogers, 2010). For example, ESDM therapists should only deliver cues and reinforcement when the child is attending to them. To illustrate, the therapist may only give the child access to a preferred item when the child makes eye contact, which should encourage the child to independently make eye contact when requesting items in the future.

Jeevan had a sharp increase in partial and whole engagement between the 2<sup>nd</sup> and 3<sup>rd</sup> week of intervention. This sharp, rather than steady, increase suggests that Jeevan was perhaps capable of attending to adults prior to the intervention, but was not doing so with his mother or the therapist (Johnson & Christensen, 2012). This increase was most likely due to the therapist identifying highly motivating activities for Jeevan, as research suggests that children with ASD may show fewer social avoidance behaviours during highly preferred activities (Koegel et al., 1987). Specifically, although Jeevan's parents had stated that he was highly motivated by songs, the therapist discovered in the 3<sup>rd</sup> week of intervention that Jeevan particularly enjoyed being picked up and swung around during a variety of songs. The therapist was then subsequently able to identify many more activities that Jeevan enjoyed. Further, as with the other children, his sustained increase in engagement may have also been due to the therapist continually targeting and reinforcing behaviours related to engagement (Rogers & Dawson, 2010).

During intervention, many of Alan's play samples took place outside. This was because he was highly motivated by the play equipment that was outside such as a trampoline and a swing set. However, as all baseline sessions had taken place in his living room, it was not possible to determine whether his improvements in engagement and the other outcome variables were due to the ESDM therapy or factors such as his increased motivation to play outdoors (Baer et al., 1968). For this reason, the final seven play samples were conducted in the living room at the beginning of the hour long session. Alan's engagement remained high during this phase, which suggests that his improvements were not solely due to the potentially motivating outdoor environment.

All four children were consistently more engaged with the therapist 1 month after intervention compared to baseline, however, Alan's engagement decreased dramatically on the final follow up session with the therapist. In fact, his engagement during this session was lower than all but one of the intervention sessions. Alan also engaged in a lot of challenging behaviour during this session, in an attempt to gain access to a preferred food item which his parent had told him was not available. Thus, it is possible that Alan was less engaged because (a) he was engaging in challenging behaviour, (b) he was not motivated for the activities compared to the food item, (c) his challenging behaviour affected the therapist's ability to deliver the ESDM procedures, and/or (d) some other unexplained reason.

Results of this study suggest that, during intervention and follow-up, all four children were partially engaged (i.e. engaged for part of each 10-s interval) with the therapist for the majority of most 10-min play samples. However, each child was only fully engaged (i.e.

engaged for the whole 10-s interval) with the therapist for an average of 20% (Charlie) to 50% of intervals in each play sample. This suggests that, while the children attended to the therapist intermittently for the entire play sample, they demonstrated sustained attention for a much more limited time. While it seems unrealistic to expect any child to show behaviours indicating engagement for the entire 10-min play sample, it is not clear whether or not the level of engagement shown by these children is comparable to that of children without ASD or developmental disabilities. Future research should compare the engagement levels of children receiving ESDM therapy, with those of typically developing children.

These results also suggest that whole engagement may be a more sensitive measure of children's attention to the therapist's face, voice, and actions than partial engagement. This is because there was evidence of a ceiling effect for partial engagement for each of the participants (Johnson & Christensen, 2012). Specifically, after the first few weeks of intervention there were several play samples in which each child was partially engaged with the therapist during 90-100% of intervals. Further, Charlie and Chris had high partial engagement with their parents during baseline. This indicated that there was very limited room for improvement on this measure. On the other hand, children's whole engagement with the therapist was generally very low in baseline, but did not reach 100% during any of the intervention or follow-up sessions.

Results suggest that Charlie was generally less engaged with the therapist during intervention than the other three children. This was likely due in part to his activity preferences. Specifically, Jeevan and Alan were highly motivated by sensory social games and songs, whereas Charlie was most motivated by play with toys such as cars, play dough, and animal figures. It is likely that children will be highly motivated to attend to the therapist during highly preferred sensory social games because he or she is the sole source of interest. In contrast, when a child is playing with highly preferred objects, the therapist must in some cases compete with the object for the child's attention. Specifically, Charlie, like many children with ASD, had a tendency to play with toys in a restricted way, and did not always want to involve the therapist in his games (American Psychiatric Association, 2013).

Although Chris was also generally more motivated by objects than sensory social games, he was also highly engaged with the therapist during baseline, and was eager to include the therapist in his games. Therefore, although Charlie was partially engaged with the therapist throughout the play sample, there were many instances in which he was attending to objects rather than the therapist compared to the other three children.

#### **Imitation**

The percentage of intervals containing imitation increased for all four of the children during the 12 weeks of intervention. None of the previous studies evaluating the effectiveness of therapist delivered ESDM intervention have included imitation as an outcome measure and there is limited research on teaching imitation to young children with autism in general. However, the results of this current study are consistent with the few previous studies evaluating the effectiveness of other early intervention approaches for teaching imitation. Specifically, these studies found that children's spontaneous and/or elicited, gestural and/or object imitation skills improved following intervention (Ingersoll et al. 2007; Ingersoll et al., 2010a; Ingersoll & Schreibman, 2006). Results of this study further suggest that the increase in imitation was also maintained for each of the children at follow-up, which is also consistent with previous research (Ingersoll et al., 2007; Ingersoll & Schreibman, 2006). This is a particularly important finding as all of the aforementioned studies focussed exclusively on teaching imitation. This suggests that the imitation skills of young children with ASD may improve during low-intensity comprehensive intervention, as well as focused imitation intervention.

For each of the children there was very little overlap between the percentage of intervals containing imitation in the baseline and intervention phases. This suggests that the therapist delivered ESDM intervention had an immediate positive effect on imitation (Johnson & Christensen, 2012). This may have been partially due to corresponding increases in child engagement during the intervention, in that an individual needs to be attending to the therapist in order to imitate him or her. Thus, the more a child is attending to the therapist, the greater the number of opportunities for imitation. However, for Charlie, Jeevan, and Alan (prior to the 'inside only' procedural modification), although the percentage of intervals containing imitation varied from session to session, there was a generally increasing trend from the beginning to the end of the intervention phase. This suggests that the improvements in imitation were not solely due to increased engagement, which remained relatively stable after the first couple of weeks of intervention, and may have been due to a true increase in their ability to imitate the therapist over time.

Chris showed the greatest improvement in the percentage of intervals containing imitation during the intervention. This may have been because he had the highest percentage of intervals containing imitation in baseline, which indicated that he was already able to imitate the therapist in some circumstances, and that he may have been "developmentally ready" to learn further imitation skills (Ingersoll, 2010a). Research also suggests certain child

characteristics may lead them to respond better or worse to imitation intervention. For example, Ingersoll (2010a) found that spontaneous play acts predicted a child's response to imitation intervention. As Chris was highly motivated by toys, he may have been more interested in the actions involving toys and more likely to imitate them. Also, as Chris had the greatest pre-intervention expressive abilities on the Vineland-II, he may have been more likely to imitate the therapist's spoken language than the other participants (Sparrow et al., 2005).

During intervention, Charlie, Alan and Jeevan imitated the therapist during a maximum of 40% of intervals. This is comparable to the findings of other studies aimed at teaching imitation to young children with ASD (Ingersoll & Schreibman, 2006; Ingersoll et al., 2007). However, during the 6<sup>th</sup> and 8<sup>th</sup> week of the intervention there were sessions in which Chris imitated the therapist during 60% to 70% of intervals. It is possible that this is indicative of excessive or stereotyped imitation, which is also a symptom of ASD (American Psychiatric Association, 2013). Another potential explanation for this effect is that, at this stage of the intervention, Chris may have overgeneralised his learning of this skill (Alberto & Troutman, 2009). His subsequent reduction in the percentage of intervals containing imitation, to a level similar to that found in previous research (Ingersoll & Schreibman, 2006; Ingersoll et al., 2007), suggests he may then have determined when it was appropriate or not appropriate to imitate the therapist. In future, researchers should evaluate the percentage of intervals containing imitation for typically developing young children of varying ages to enable clinicians to better determine the amount of imitation that is developmentally appropriate.

## **Functional Utterances**

The results of this study suggest that the percentage of intervals containing functional utterances improved for all four children during the intervention but, for Alan, this improvement was minimal. Further, Charlie, Chris and Jeevan's MLU, variety of utterances, and/or frequency of utterances also increased during the intervention. Several previous studies have also found that therapist delivered ESDM intervention led to improvements in children's expressive language (e.g. Dawson et al., 2010; Devescovi et al., 2016; Vivanti et al., 2014) but the differences in the language measures used in these studies prevent direct comparison with the current study. However, several researchers have used comparable measures to evaluate the effectiveness of targeted naturalistic language interventions for young children with ASD and have also reported similar improvements (e.g. Hancock & Kaiser, 2002; Kasari, Kaiser, et al., 2014; Kaiser & Roberts, 2013; Koegel et al., 1988). The

findings of this study again suggest that comprehensive low-intensity ESDM intervention was successful in improving functional utterances in a similar manner to targeted language interventions.

The results of this study also suggest that, for each of the children, the improvements in functional utterances were maintained 1 month after the intervention finished. Several of the aforementioned studies on naturalistic language interventions also found that children's language improvements were maintained for 3 (Kasari et al., 2014), 6 (Kaiser & Hancock, 2002) or 12 months (Kaiser & Roberts, 2013) from the end of treatment. As the follow-up duration was shorter than that reported in previous studies, it is not clear whether these improvements in functional utterances would be maintained in the long-term.

For all of the children except Alan, there was very little overlap between the percentage of intervals containing functional, and engaged functional, utterances in baseline and intervention. For Charlie and Jeevan, this suggests that the intervention had an immediate positive effect upon their use of functional, and engaged functional, utterances. As with imitation, the increase in engaged utterances could be partially due to an increase in their engagement with the therapist at the beginning of intervention. However, Jeevan's mean number of intervals containing functional utterances in intervention was 3x higher than his mean in baseline, and Charlie's was 10x higher than baseline, which represents a considerable increase in overall language use. In contrast, it is not possible to attribute Chris' improvements in intervals containing functional utterances to the ESDM intervention. This is because his baseline data suggests that his utterances were increasing prior to the start of intervention (Johnson & Christensen, 2012). Therefore, the further increases in Chris' utterances during intervention could have simply been due to maturation, or another extraneous variable, rather than the intervention itself.

Visual analysis of the graph would suggest that, for Chris, Jeevan, and Charlie, functional utterances stopped improving after the 1<sup>st</sup> month of intervention. However, for Chris and Jeevan, this may have been due to ceiling effects for this variable (Johnson & Christensen, 2012). Specifically, there were some sessions during intervention in which Chris and Jeevan produced functional utterances during almost 100% of intervals. This suggests that frequency, rather than percentage of intervals containing utterances was a more appropriate measure for these two children. Indeed, Chris' frequency of utterances continued to increase during each month of intervention and at follow-up and Jeevan's frequency of utterances was also higher in the 2<sup>nd</sup> and 3<sup>rd</sup> month of intervention than the 1<sup>st</sup>. Further, although there were no ceiling effects for Charlie's utterances, his MLU and variety of

utterances continued to increase during intervention and follow-up. This suggests that although he was not speaking more often, his language was becoming more varied and sophisticated as the intervention continued.

It is also possible that it was not developmentally appropriate to measure the quantity of utterances for Chris or Jeevan, as they both had a high percentage of intervals containing functional utterances prior to intervention. Indeed, both of these children had intervention goals related to increasing the quality and functionality of their spoken language rather than the quantity. For example, Chris' expressive language goals related to saying "no" and asking "what's that?". Further, although Jeevan and Chris had a similar percentage of intervals containing functional, and engaged functional, utterances during intervention, further analysis of these utterances revealed that Chris' communication was more sophisticated than Jeevan's. Specifically, Jeevan's mean MLU was 1.0 during baseline and increased to 1.6 in the final month of intervention, while Chris's mean MLU was 2.4 and increased to 3.2. Further, Jeevan had a limited variety of utterances during baseline, which increased during each month of intervention, while Chris had a wide variety of utterances during baseline. Both of these factors suggest that it may be preferable for researchers to examine children's progress towards specific developmental language goals rather than solely measuring the quantity of language.

In baseline, Jeevan seldom directed utterances towards the therapist, but in some of the sessions he did produce a considerable amount of non-socially directed functional utterances. Specifically, during this phase he would often label objects or sing songs about his actions with toys, without making eye contact with the therapist or orienting towards the therapist (e.g. singing *Old MacDonald had a Farm* while moving the corresponding toy). This suggests that prior to intervention, Jeevan was able to use language, but was not using it to communicate socially, which is common for many children with ASD (American Psychiatric Association, 2015). Jeevan's utterances and engaged utterances increased sharply during the 3<sup>rd</sup> week of intervention, which coincided with his increased engagement with the therapy. This suggests that, during intervention, Jeevan learned to use his pre-existing language skills in a more socially-oriented way. In other words, the intervention may have helped to motivate Jeevan to use language in the context of social interaction.

Charlie and Alan had the fewest functional utterances during baseline and, while Charlie's functional utterances increased greatly during the intervention, Alan's did not. There were several of Charlie's individual characteristics that may have helped him to respond better to the intervention than Alan. First, Charlie was 2-years-old at the start of the

study whereas Alan was 4-years-old. Some studies suggests that younger children may have better outcomes following early intervention than older children (e.g. Bibby et al., 2002; Granpeesheh et al., 2009; Harris & Handleman, 2000), and, specifically, that younger children who received direct ESDM intervention showed greater improvements on a measure of language skills than older children but did not differ on any other measure (Vivanti et al., 2016). Therefore, Charlie may have had greater improvements in functional utterances because he was younger than Alan. Research also suggests that children with greater language skills also respond better to early intervention (e.g. Kasari et al., 2008). Alan only produced one functional utterance during the baseline phase, whereas Charlie produced two utterances during each session. Although this is perhaps not much of a difference, it may suggest that Charlie was more "developmentally ready" than Alan to acquire additional spoken language. This finding highlights the need for further research on which individual characteristics lead a child to respond better to a particular intervention.

#### **Intentional Vocalisations**

In baseline, Alan only used one word approximation and very few intentional vocalisations (purposeful vocal sounds that do not contain a word or word approximation) with his mother or the therapist. As the ESDM approach advocates teaching skills in the sequence in which they would normally develop in children without ASD or developmental disabilities, it was determined that the therapist should focus on increasing Alan's intentional vocalisations (rather than functional utterances) during the intervention (Rogers & Dawson, 2010). Results from this study suggest that Alan's vocalisations increased during intervention, and were maintained 1 month later. Although none of the previous ESDM research has included intentional vocalisations as an outcome variable, this finding is consistent with several studies evaluating the effectiveness of prelinguistic mileau teaching (PMT) for young children with ASD and/or other developmental disabilities (see Peters-Scheffer, Huskens, Didden, & van der Meer, 2016, for a review). As the name implies, PMT focusses on developing children's intentional prelinguistic communication and the majority of children who participated in therapist delivered PMT intervention improved their use of prelinguistic communicative acts such as intentional vocalisations, eye contact, and gestures (Franco et al., 2013; Warren et al., 1993).

Alan's use of intentional vocalisations increased steadily in intervention and remained relatively stable during the 'inside only' procedural modification, before increasing again during the first two follow-up sessions 1 month later. This suggests that Alan was slowly learning to use intentional vocalisations in the context of his interactions with the therapist

and that he maintained his use of vocalisation after the removal of the intervention. This is a particularly promising finding considering the limited intensity of the intervention and the fact that Alan was the oldest child in the study, with the most significant language delay.

Results suggest that Alan was beginning to use slightly more functional utterances towards the end of intervention, although this remained relatively low, at an average of just under two utterances per session. It is possible that the large increase in his use of intentional vocalisations contributed to this slight increase in functional utterances. Intentional vocalisations precede language development in typically developing children, and research suggests that language develops similarly in children with ASD (Tager-Flausberg et al., 1993; Vihman et al., 1985). Therefore, it is possible that, for Alan, the use of intentional vocalisations was a necessary pre-requisite for further spoken language development. It is also possible that by reinforcing intentional vocalisations the therapist taught Alan that he needed to "use his voice" in order to gain access to preferred items and activities, which could have resulted in this small improvement in functional utterances (Koegel et al., 1988). Unfortunately, due to the short duration of the intervention, and limited follow-up period, it remains unclear whether Alan's use of functional utterances would have continued to improve in a meaningful way.

#### Generalisation

Alan, Chris, and Jeevan had a greater percentage of intervals containing imitation, functional utterances (intentional vocalisations for Alan), and engagement with their mothers following intervention compared to baseline. Although Charlie had a greater percentage of intervals containing functional utterances with his mother following intervention, his percentage of intervals containing imitation remained the same and his percentage of intervals containing engagement decreased. Further, the improvements that each child showed with their mothers were not always as great as their improvements during intervention with the therapist. To the author's knowledge, no other studies have examined the generalisation of child gains during direct therapy to a parent or family member. However, these results are consistent with Ingersoll et al. (2007) who found that five young children with ASD generalised their improvements in imitation skills to a novel professional therapist.

Each child's percentage of intervals containing imitation at the end of intervention was higher with the therapist than with their mothers. In contrast, Charlie, Alan, and Chris' percentage of intervals containing functional utterances (intentional vocalisations for Alan) at the end of intervention was similar with the therapist and their mothers. This may be because

their parents were more motivated to create opportunities for expressive communication than imitation. This is supported by research which suggests that communication development is frequently mentioned by parents as a top intervention priority whereas imitation is not (Pituch et al., 2011; Rodger et al., 2004; Whitaker, 2007). Therefore, parents may not have been creating sufficient opportunities for the child to imitate them compared to the therapist. Specifically, the therapist may have intentionally created many opportunities for imitation, as it is one of the key skills targeted in ESDM intervention (Rogers & Dawson, 2010). However, more research is needed into the number of opportunities that parents of young children with ASD create for their children to perform different types of developmental skills.

All of the parents except Charlie's mother increased their correct use of the ESDM techniques by at least 10% following the intervention. As these parents did not participate in any ESDM parent training, it is possible that these increases were due to observational learning, in which the parents observed the ESDM sessions, and then imitated some of the techniques used by the therapist (Bandura, Ross & Ross, 1963; Mazur, 2006). This is supported by the fact that Alan, Chris and Jeevan's mothers were able to observe many of the ESDM sessions which generally took place in a central room in the home whereas Charlie's mother did not observe any of the sessions because they took place in a different floor of the house. However, as Charlie's mother's fidelity was already very high during baseline this indicates that she was already using many of the ESDM techniques prior to the intervention and may have had limited room to improve. Further, it should be noted that, although the remaining parents did improve in their use of the techniques following intervention, they did not reach the level of 80% correct use of the ESDM procedures which is generally considered to be the lowest level of acceptable implementation. This suggests that parents may benefit from direct training in the ESDM techniques (e.g. coaching, practice, feedback etc.) in additional to simply observing the therapist (Rogers & Vismara, 2015).

It is possible that Alan, Jeevan, and Chris' mothers' increasing use of the ESDM techniques contributed to their children's improvements in some areas following the intervention. For example, each of these parents improved in their use of some of the ESDM techniques related to child attention and motivation such as: management of attention, sensitivity and responsivity, use of positive affect, motivation, and management of affect and arousal. Therefore, it is possible that increased use of some or all of these techniques caused their children to be more engaged, and to use more engaged utterances, with their mothers compared to baseline. This is supported by the fact that Charlie's mother had the highest fidelity at the start of the intervention and Charlie also had high levels of engagement with

her at this time. It is not clear whether the children would also have shown similar levels of engagement or use of engaged utterances with their mothers if each mother's fidelity had remained at the same level as baseline. More research is needed to examine the effect of the different ESDM fidelity components on child outcomes.

It is also possible that some of the child improvements in imitation, functional utterances/vocalisations, and engagement, with their parents following intervention represented true generalisation of their learning with the therapist (Stokes & Baer, 1977). Specifically, as the parents were not generally implementing the ESDM techniques with a high degree of fidelity, it is possible that the children were simply using new skills that were effective with the therapist with a novel person, regardless of this person's use of ESDM techniques. In future, research could evaluate the generalisation of child learning to an individual with no knowledge of ESDM techniques.

## **Social Validity**

This study did not include an evaluation of social validity because an in-depth evaluation of parent perceptions of this intervention was conducted by Ogilvie and McCrudden (2017). In the Ogilvie and McCrudden (2017) study, at least one parent of the four children who participated in Study 1 completed a Treatment Acceptability Rating Scale-Revised (TARF-R; Reimers, Wacker, Cooper, & de Raad, 1992; see Chapter 6 for a description) and took part in a qualitative semi-structured interview about each their perceptions of the aims, procedures and outcomes of the therapist delivered ESDM intervention. Results of the TARF-R suggest that all four parents found the intervention to be acceptable and, specifically, that: (a) the intervention was reasonable in terms of the procedures used, (b) the intervention was effective for improving child outcomes, (c) they would be willing to continue the intervention, (d) the intervention resulted in minimal side effects. Three of the four parents also reported that the intervention was minimally disruptive. Three key themes that were thought to contribute to the high parent-rated acceptability of the intervention emerged from the qualitative analysis. These were: (a) the importance of good rapport between the child and the therapist, (b) the alignment between the ESDM therapy and the parents' parenting style, and (c) the social significance of the child's improvements in behaviour. Taken as a whole, these results suggest that the parents of the children in this study perceived the low-intensity therapist delivered ESDM to be socially valid and acceptable.

These results are comparable to the findings of Colombi et al. (2016) who also included a measure of the feasibility of low-intensity therapist delivered ESDM intervention

and concluded that the programme was acceptable, due to high child retention rates, and was in high demand, as 200 families applied for the programme. However, the Ogilvie and McCrudden (2017) study appears to be the only one which has provided in-depth qualitative analysis of parent perceptions of the low-intensity therapist delivered ESDM intervention. This suggests that more research is needed in this area as parent perceptions of social validity affect their likelihood of continuing the intervention and recommending the intervention to others (Foster & Mash, 1999).

## **Implications**

Results of this study suggest that home-based ESDM therapy was effective in improving outcomes for four children with ASD. This is an important finding as it appears that the two previous studies evaluating low-intensity therapist delivered ESDM intervention took place in a clinic setting (Devescovi et al., 2016; Colombi et al., 2016). There are several potential advantages to providing treatment at home rather than in a clinic. First, it is possible that home is a more convenient setting for parents than a clinic, as the parents do not need to travel to the location, find childcare for siblings or take additional time off work (Sweet & Applebaum, 2004). It could also be argued that home is a more natural setting to receive intervention than a clinic, and both the New Zealand ASD guidelines (Ministries of Health and Education, 2008) and the United Nations Convention on the rights of persons with disabilities (United Nations General Assembly, 2007) advocate for teaching children in their natural environments. Further, it is possible that teaching the children at home facilitated their generalisation of skills to their parents, with whom they interact frequently in the home environment (Stokes & Baer, 1977). These advantages, and the fact that this study found generally positive results suggests that, when possible, therapists should suggest to parents the possibility of providing at least some therapy sessions in the child's home.

The therapist delivered intervention in this study only lasted for 12 weeks, whereas in previous research the low-intensity therapist delivered intervention lasted for between 6 months (Colombi et al., 2016) and 2 years (Eldevik et al., 2006; Peters-Scheffer et al., 2013). Therefore, the fact that each of the children showed improvements during the intervention, and that these improvements were maintained 1 month later, suggests that short duration low-intensity direct therapy may also be effective in improving outcomes for young children with ASD. This is an important finding, as a shorter duration of low-intensity therapy could reduce treatment costs and increase the number of children who are able to access the intervention.

The finding that imitation and engagement increased for all four children during the intervention is particularly important in light of the short duration of the intervention.

Specifically, one is not able to target all possible development goals within a 12 week period, however, increasing a child's imitation and engagement skills arguably improves his or her ability to learn from others in their natural environment (Rogers et al., 1986; Rogers & Vismara, 2010). This should in theory increase the possibility that the child will continue to learn new skills from others after the intervention has finished. Therefore, interventions with a limited time and duration should perhaps place the most emphasis equipping children with the skills to learn from adults and peers, such as imitation and social interaction skills. More research is needed into the skills needed to promote generalisation of outcomes following therapist delivered intervention for young children with ASD.

The results of this study suggest that the children's improvements during intervention with the therapist did generalise somewhat to their mothers. However, in some cases, the improvements with their mothers were not as great as the improvements with the therapist. This suggests that parents may need to directly target skills such as imitation, and engagement, in addition to the therapist, in order to facilitate generalisation. Further, although three mothers increased their ability to implement the ESDM procedures, they were not implementing these procedures with an acceptable level of fidelity. This suggests that observational learning alone may not have been sufficient for teaching parents to implement ESDM procedures with their children with ASD, and thus should perhaps be complemented by other strategies such as didactic teaching, practice, and feedback. These findings present a compelling argument for training parents in addition to direct therapy.

#### Limitations

Chapter 7: General Discussion, will include general limitations that apply to both Studies 1 and 2. However, this study is limited in several ways that Study 2 was not. First, although the therapist had completed the advanced ESDM workshop, she did not become a certified ESDM therapist until the 3<sup>rd</sup> month of the study. At this stage she was already delivering intervention to Charlie and Alan but not to Chris and Jeevan. Therefore, it is possible that Chris and Jeevan received a higher quality of intervention than Charlie and Alan, as the therapist had had more practice with the intervention and was a certified ESDM therapist for the entire time that she worked with them. Further, it is possible that child outcomes may have been better if the intervention had been implemented by a more experienced ESDM therapist.

During the initial weeks of intervention Alan engaged in behaviours that indicated that he wanted to finish the session early such as attempting to turn on the television or leaving the therapy area. When these behaviours occurred, the therapist terminated the

sessions in order to maintain a positive relationship with Alan and to prevent challenging behaviour. For this reason, the therapist also spent longer "becoming a play partner" with Alan than with the other children in order to increase his motivation to participate in the session for the entire hour (Rogers & Dawson, 2010). This included strategies such as narrating his play, imitating his actions, and providing help but did not include direct teaching. Therefore, Alan did not receive the full "dose" of intervention during the first 2 weeks. It is possible that his outcomes would have been different if he had received the full hour of therapy right from the start of intervention.

Study 1 is also limited in that the number of learning opportunities was fixed during baseline but, due to the naturalistic nature of the ESDM therapy, it was not possible to control for the number of learning opportunities during the intervention or follow-up sessions. Therefore, there may have been fewer opportunities for the children to imitate the therapist, or to use functional utterances in baseline than in intervention or follow-up. Although the therapist provided a probe for an imitation skill or a functional utterance every 30s during baseline it is possible that these opportunities occurred more frequently during intervention and follow-up, in the context of the ESDM therapy. Therefore, the increase in these outcomes during these phases could be partially due to the child having more opportunities to perform the skill.

Finally, mean IOA on whole engagement for Chris was 78%, which is slightly below the 80% benchmark, generally considered to be the minimum level of acceptability (Kennedy, 2005). Therefore, the data for Chris' whole engagement may be less accurate than the data for the other dependent variables for Chris, and whole engagement for the other participants. It is possible that this IOA on whole engagement was lowest for Chris because he had the most sophisticated play behaviour and used the most complex functional utterances. This may have made it more difficult to determine whether Chris' behaviour was directed towards the therapist for the entire 10-s interval.

In summary, the results of this study suggest that home-based, low-intensity therapist delivery of ESDM may be effective in improving imitation, functional utterances/intentional vocalisations and engagement for some young children with ASD. These improvements also appear to have been maintained 1 month after the treatment had finished. Further, the results suggest that children may generalise the skills learned with the therapist to their mothers to some degree. Chapter 6: Parent training based on the ESDM will include the methods, results, and a discussion of Study 2 of this thesis.

#### **CHAPTER 6**

# STUDY 2: Parent Training based on the ESDM Method

# **Participants**

Five children and their parents were recruited for this study. The parents of two of the children were referred by a local organisation that provides home-based autism information sessions to families. Another two parents were referred by a local area District Health Board. The final parent made direct contact with the trainer (Waddington) after finding her details on a website listing certified ESDM therapists. Inclusion criteria for participating in this study were: (a) the child was under the age of 5 years (60 months) at the start of the study; (b) the child had a clinical diagnosis of ASD or met criteria for an ASD diagnosis based on an ADOS-2 assessment (Lord, Rutter, et al., 2012); (c) the child did not have another serious or specific medical, genetic, neurological or sensory condition (e.g., Down syndrome, fragile X) and (d) the child was not receiving intensive early intervention of any type at any time during the study. Parents also had to have provided consent to allow videotaping of their interactions with the child during parent training sessions and a second family member had to provide consent for the generalisation probes. All of the participating parents had been involved in no more than one other autism specific parent training/support programme, but they were not participating in any other parent training programmes during the present study. Table 6.1 provides a summary of each parent's demographic characteristics and Table 6.2 provides a summary of each child's demographic characteristics and the results of diagnostic and adaptive behaviour assessments.

Table 6.1

Parent Demographic Characteristics

	Dean	Rick	Sean	Idris	Alex
Primary participant	Mother	Mother	Mother	Mother	Mother
Generalisation- family	Father	Father	Grandfather	Father	Father
member					
Marital status	Married	Long-term	Married	Married	Married
		relationship			
Employment	None	Part-time	None	Full-time	None
Education	High School	Bachelor's	High School	Master's	High School
Ethnicity	Cambodian	NZ European	NZ European	Indian	Māori

Table 6.2

Child Demographic Characteristics and Vineland-II and Social Communication

Questionnaire (SCQ) Results.

	Dean	Rick	Sean	Idris	Alex
Age at start of study	3:0	4:11	4:0	1:11	2:11
(Years:Months)					
Gender	Male	Male	Male	Male	Male
Vineland II					
Communication (Adaptive level)	Mod. low	Mod. low	Low	Low	Low
Receptive	1:3	2:2	1:3	<0:1	0:11
Expressive	2:0	2:5	1:1	0:8	0:9
Written	N/A	5:10	1:10	N/A	N/A
Daily Living	Mod. low	Mod. low	Low	Adequate	Mod. Low
(Adaptive level)					
Personal	1:1	2:9	1:8	1:11	1:1
Domestic	1:10	3:5	0:10	1:2	1:2
Community	1:6	3:6	2:6	2:5	1:6
Socialisation (Adaptive Level)	Low	Low	Low	Mod. low	Mod. Low
Interpersonal	0:5	0:7	0:9	0:7	0:8
Play	0:9	2:1	1:2	0:3	1:1
Coping	1:6	1:1	1:1	0:10	2:3
Motor skills (Adaptive Level)	Mod low	Adequate	Low	Adequate	Mod. Low
Gross	2:2	3:2	2:7	1:9	1:10
Fine	1:11	4:3	1:10	2:10	1:10
Maladaptive behaviour index	N/A	Clin. sig.	Elevated	N/A	N/A
Internalising	N/A	Clin. sig.	Clin. sig.	N/A	N/A
Externalising	N/A	Elevated	Elevated	N/A	N/A
SCQ	Risk of ASD				

Note: Clin. sig. = Clinically significant, Mod. = Moderately

**Dean.** Dean was 3-years-old at the start of the study and lived with his mother, father, two older sisters, and his grandmother. His parents spoke Khmer and English at home. Dean understood and spoke some Khmer, but he mainly communicated in English. His mother stayed at home to look after him and had previously completed an ASD education course but had not completed any other parent training. His father, who took part in the generalisation probe, had completed a 1 day parent training course focusing on play with children with ASD. Dean scored as being at-risk for ASD on the SCQ (Rutter et al., 2003) and was diagnosed with ASD by multidisciplinary team at his local District Health Board at the age of 2 years and 2 months. The Child Development team administers an ADOS-2 as part of the diagnosis process (Lord, Rutter, et al., 2012). His scores on the Vineland-II (Sparrow et al., 2005) indicated that his overall adaptive level for communication was moderately low. His adaptive level for the daily living skills and motor skills domains was also moderately low and his adaptive level for the socialisation domain was low. He did not receive a score on the maladaptive behaviour index or for written communication because he was under 3 years at the time of the assessment. Dean's mother reported that, prior to intervention he primarily communicated using single words or two word phrases related to preferred items and activities, for example yellow truck and horse. Her main concern was that he did not always seem to understand what she was saying. She stated that his preferred activities included playing repetitively with transportation toys and running around with his sister. At the start of the study Dean went to a local kindergarten twice a week and attended a playgroup with his mother for 1 hr, 3 times a week. He also received monthly visits from a speech-language therapist.

**Rick**. Rick was 4 years and 11 months old at the start of the study and lived with his mother and father. He did not have any siblings. His mother worked 3 days a week and looked after Rick on the other 2 days. She had previously completed an ASD education course and a 1 day introductory workshop about playing with children with ASD. Rick's father had not participated in any parent training. Rick scored as being at risk for ASD on the SCQ (Rutter et al., 2003) and was diagnosed with ASD at the age of 3 years, 5 months by the local Child Development team. His scores on the Vineland-II (Sparrow et al., 2005) indicated that his overall adaptive level for communication was moderately low and that his age equivalency for written language was higher than his age equivalency for receptive and expressive language. Specifically, he could identify all letters of the alphabet, read at least 10 words aloud, and was able to print at least three words from an example. His adaptive level

for daily living skills was also moderately low, his adaptive level for socialisation was low, and his adaptive level for motor skills was adequate. His internalising score on the maladaptive behaviour index was clinically significant and his externalising score was elevated. Rick's mother reported that, prior to intervention, Rick communicated using scripted phrases and by completing sentences said by an adult. She stated that his interests included trains, tunnels, physical play such as running and jumping, and being outside. She also reported that he was a caring boy who enjoyed cuddles and trying to make her laugh. Rick went to day care from 8.00 am to 3.00 pm three times a week. For the 1st month of the study, Rick was participating in a food therapy intervention and visited an occupational therapist once per month, but this was terminated when he turned 5 years of age.

**Sean**. Sean was 4-years-old at the start of the study and lived with his mother, father, and older brother. His paternal grandparents also frequently visited Sean and took him on outings. His paternal grandfather took part in the generalisation probe as his father worked long hours. Sean's mother did not work and cared for him for 2 days a week when he was not at kindergarten. She had previously completed an ASD education course. Sean's father and grandfather had not received any formal education or training about ASD but Sean's mother reported that she shared useful information with family members. Sean scored as being at risk for ASD on the SCQ (Rutter et al., 2003) and was diagnosed with ASD at the age of 3 years and 3 months by a private paediatrician based on the DSM-5 ASD diagnostic criteria (American Psychological Association, 2015). His scores on the Vineland-II (Sparrow et al., 2005) indicate that his overall adaptive level was low for all domains (communication, daily living, socialisation, and motor skills). His internalising score on the maladaptive behaviour index was clinically significant and his externalising score was elevated. Sean's mother reported that he communicated using generalised words such as okay, yeah, yes, and go, in response to adult phrases such as ready, steady. His parents had taught him to point and he frequently used pointing to communicate. His mother stated that Sean loved physical "rough and tumble" play and particularly enjoyed playing with outdoor equipment such as scooters, slides, and trampolines. She reported that he was well-behaved but would sometimes have challenging behaviour related to certain noises and crowded spaces. He went to kindergarten from 8:30 am to 2:30 pm three times a week, was participating in a weekly food therapy intervention and had previously received services from a speech language therapist.

**Idris**. Idris was 1 year and 11 months old at the start of the study and lived with his mother and father, although his father frequently travelled to a different city for work. His mother and father both worked full-time and, at the start of the study, Idris was cared for by

his maternal grandmother. When his maternal grandmother returned to India, Idris' mother worked from home in order to look after him. The family spoke Tamil at home and the few words that Idris had said were also in Tamil. Idris' mother and father had not received any autism specific education but had six meetings with an occupational therapist regarding strategies to support Idris' development. Idris scored as being at risk for ASD on the SCQ (Rutter et al., 2003) and, at the start of the study, he was on the waiting list for an ADOS assessment from the local Child Development team. He was diagnosed with ASD and global developmental delay (GDD) by this team in the 10<sup>th</sup> week of intervention. His scores on the Vineland-II (Sparrow et al., 2005) indicated that his overall adaptive level for communication was low. His adaptive levels for daily living skills and motor skills were adequate and his adaptive level for the socialisation domain was moderately low. He did not receive a score for written communication or maladaptive behaviour due to his age. At the start of the study, Idris' mother reported that he frequently babbled but his only spoken word was Mum in Tamil which he did not say very often. He would sometimes vocalise intentionally but he did not point to communicate. She reported that his favourite toys included a shape sorter and stacking boxes and he also enjoyed tickles and listening to her sing. She stated that he frequently walked and ran around the room in a repetitive fashion and often put non-edible objects in his mouth. In the 9<sup>th</sup> week of intervention, Idris began to attend kindergarten from 1:15 pm to 3.15 pm 3 times a week. Throughout the duration of the study he was not receiving any additional support.

Alex. Alex was 2 years and 11 months old at the start of the study and lived with his mother, father, older sister, and younger brother. His mother stayed at home to look after him and his siblings. She had previously completed a 12 week group training programme aimed at improving communication for children with ASD. Alex's father had not received any parent training. Alex scored at risk of ASD on the SCQ (Rutter et al., 2003) and was diagnosed with ASD at the age of 2 years and 1 month by the local Child Development team. His scores on the Vineland-II (Sparrow et al., 2005) indicated that his overall adaptive level for communication was low. His adaptive levels for daily living skills, socialisation, and motor skills were all moderately low. He did not receive a score for written communication or maladaptive behaviour due to his age. At the start of the study his mother reported that he did not have any spoken language but he was occasionally observed to vocalise to request desired items and activities. His mother stated that his preferred play items were cause-and-effect toys, balls, and cars and that he enjoyed messy play including shaving cream, paint, and sand. She also reported that he enjoyed playing "people games" like tickling and singing songs.

Alex attended kindergarten from 8:30 am to 3:00 pm three times a week and was visited monthly by a speech language therapist.

## **Setting**

All of the parent training sessions took place in each child's home. The trainer (Waddington) delivered the parent training via PowerPoint® presentations (see section on Intervention/Parent training) in Sean's family's dining room and in the living room with the four remaining families. The location of the play activities with the trainer and the parent varied depending on the skill being taught and the activity. For Dean, all play activities began in his living room, which contained several boxes of toys, two couches, a television, and a child sized table and chairs. During some play activities he would leave the living room and enter his parent's bedroom. Rick's play activities took place in three main areas: his bedroom which had shelves of toys and a bed shaped like a race car; the living room which had two couches, a bean bag, a mirror, and boxes of toys; and the backyard, where he could access water play, the sandpit, toy construction vehicles, and the trampoline. The majority of Sean's play activities took place in the living room, which contained two couches, a television, and his toy cars; or outside where he had access to a trampoline, a slide, his scooter, and an exercise ball. During some play activities Sean also entered his parents' bedroom to play. Idris' family moved house between the 5<sup>th</sup> and 6<sup>th</sup> intervention session. Prior to moving, all play activities took place in his living room, which contained two couches, a small plastic slide, a television, and his toys. After moving, play activities took place in the new living room, which had two couches, a television, a slide and an exercise ball, and in Idris' "play room" which contained his toys. For Alex, play activities took place in the living room which contained two couches, a television, and draws of toys.

## **Materials**

During play activities the participants had access to any available toy and materials in the home. Commonly used play items for each child were as follows: Dean- wooden number puzzles, play dough, toy emergency vehicles, toy cars and a ramp, a plastic tea set, and a tunnel; Rick- a train set, a metal construction toy, large plastic construction vehicles, the trampoline, water play with buckets and pipes, a tent, and books; Sean- a marble tower, toy cars and a plastic garage, bubbles, balloons, an exercise ball, a pillow, and the trampoline; Idris- nesting cups, a bead maze, cause-and-effect toys, an exercise ball, and bubbles; Alex-books, toy cars, balls, crackers, and a Swiss ball. The trainer occasionally brought additional toys to demonstrate particular skills (e.g. a Swiss ball, a toy monkey with accessories, and shaving foam) but these were not used during the initial 10-min of the parent training session.

During the curriculum assessment, the trainer brought a large transparent plastic box containing an assortment of developmentally appropriate toys (see Appendix D).

Sessions were filmed by the trainer using an iPhone® 5s and the PowerPoint® presentation was delivered on the trainer's MacBook air. The parents were given paper copies of the child's weekly goals and reminders at the end of each session. The *Vineland-II* and SCQ were administered via an interview with the parents prior to the start of baseline (See Chapter 5 for a description of these assessments).

## **Dependent Variables**

Data on each dependent variable were collected from the 10-min videos undertaken within each baseline, intervention (Sessions 3 to 12), generalisation, and follow-up session. Each 10-min video was divided into 60, 10-s intervals (see Appendix E). As in Study 1 (see Chapter 5), four dependent variables related to child behaviours were defined and recorded as being either present or absent for each 10-s interval. These four dependent variables were (a) imitation, (b) functional utterances (including mean length of utterance [MLU], frequency, and variety of utterances for Dean, Rick, and Sean), (c) meaningful/intentional vocalisations (Idris and Alex only), and (d) engagement with the parent. Meaningful/intentional vocalisations were only recorded for Idris and Alex because they both had ESDM expressive language goals related to increasing intentional vocalisations and non-verbal communication, but the remaining three children had expressive language goals related to increasing functional utterances. As in Study 1 there was also one parent-related dependent variable and that was the extent to which parents implemented the ESDM procedures with fidelity which was scored using an 18-item checklist (see Appendix F).

# **Experimental Design**

The effects of the intervention were evaluated using a multiple probe across participants design (Kennedy, 2005). Each child/parent dyad participated in the following sequence of phases: baseline, intervention/parent training phase, and follow-up. Generalisation probes were also conducted during baseline and follow-up. This design was considered an effective analytic tool for determining if the parent training programme was effective in enabling parents to implement the ESDM procedures with fidelity and if parent use of the ESDM procedures was responsible for increases in the four child dependent variables.

#### **Procedures**

**Baseline.** Baseline involved weekly sessions in the homes of each of the participants. Sessions occurred in the morning for Sean and Alex, in the afternoon for Dean and Rick, and

predominantly in the afternoon for Idris. Alex's younger brother was present during most sessions, whereas the siblings of all other children were not usually present. At the start of each session, the trainer instructed the child's mother to *Play with your child as you normally would*. Once the child was settled into play with his mother, the trainer began filming for 10-min The trainer did not give any comment or feedback about the play or the parent's interaction with the child during or after the filming.

**Baseline generalisation probe.** The procedures for the baseline generalisation probe were identical to those used in baseline, except the child's father or grandfather (Sean) played with the child. The generalisation probe for Sean and his grandfather occurred during the 3<sup>rd</sup> week of intervention due to difficulties with scheduling.

**Parent training/Intervention.** This phase lasted for 12 weeks, with one 1-hr long parent training session per week. During the first parent training session, the trainer (Waddington) conducted the curriculum assessment in order to determine developmentally appropriate goals for each child. No data was collected in this session. The curriculum assessment involved the trainer playing in a naturalistic way with the child for approximately 1 hr. A second ESDM therapist (PhD students who had completed the advanced workshop) then noted on the ESDM curriculum checklist (Rogers & Dawson, 2010) whether or not the child displayed a range of developmental skills and instructed the trainer to probe additional skills during the play. Based on this first session and in consultation with the parents, one to three goals were selected for each child for each of the nine developmental domains (i.e., receptive communication, expressive communication, social skills, imitation, cognition, play, fine motor, gross motor, and behaviour). Dean and Rick were the only children who were at a developmental level to have goals in the joint attention domain. Table 6.3 shows the items from the curriculum checklist that were used to inform each child's goals. An example of a goal for Dean was "When an adult offers Dean a toy or food item and says "Do you want [item]", Dean will nod his head and/or say yes most of the time."

After the 1<sup>st</sup> week, each subsequent 1-hr session was based upon a chapter from the ESDM parent manual *An Early Start for your Child with Autism* (Rogers, Dawson, et al., 2012). Each of these chapters focuses on a different skill for interacting with children with ASD or a different area of child development. The 11 chapters of the book that formed the basis for the parent training were: Capturing your child's attention, having fun with sensory social routines, building back-and-forth interactions, how children learn, sharing interests with others, it's playtime, let's pretend, moving into speech, and putting it all together. The corresponding 11 intervention sessions had the following structure (see Figure 6.1): (a) the

parent demonstrated the techniques discussed in the previous week with their child for 10 min, this data served as the 10-min play sample for each parent training session (did not take place in Week 2); (b) the trainer gave the parent feedback including at least two positive comments and one area for improvement; (c) if necessary the trainer modelled the skill with the child; (d) the parent and trainer discussed the previous week's progress in terms of both child goals and adult use of ESDM techniques (this sometimes occurred prior to the parent demonstration of skills); (e) the trainer delivered the PowerPoint® presentation which provided a brief overview of the current week's chapter and also related the chapter to the child's specific goals (the wording of the PowerPoint® presentation was simplified for Dean's mother because she was not fluent in English); (f) the trainer demonstrated the new technique(s) with the child for 5 to 10-min (did not take place in Week 12); (g) the parent practiced the technique(s) from the new chapter with the child for 5 to 10-min (did not take place in Week 12); (h) the trainer gave the parent feedback including at least two positive comments and one area for improvement (did not take place in week 12); and (i) the parent and the trainer discussed the child's developmental goals for the upcoming week and the adult techniques that could help the child to achieve these goals, and the trainer answered any additional questions (did not take place in Week 12). Sessions for Rick and his mother were split across 2 days after the 2<sup>nd</sup> week of intervention because it was determined that his mother was not able to focus on the PowerPoint® presentation and monitor Rick at the same time. In the 1st session of the week Rick and his mother demonstrated the previous week's skills and received feedback from the trainer (steps f to h). Data was collected in this session. In the 2<sup>nd</sup> session of the week Rick's mother and the trainer discussed the previous week's progress, went through the PowerPoint® presentation, and discussed the goals for the upcoming week (steps d - e and i). The  $8^{th}$  intervention session for Idris and his mother was also split over 2 days because he was napping when the trainer arrived.

**Follow-up generalisation probe:** This phase was identical to the baseline generalisation probe and occurred approximately 1 week after the final parent training session.

**Follow-up probe:** This occurred 4 weeks after the final parent training session and was identical to baseline. Following this probe the trainer offered to meet with the parent(s) to discuss new developmental goals for the parent to continue teaching their child. Three parents agreed to this, however, Idris and Alex's parents wanted to continue targeting the goals from intervention.

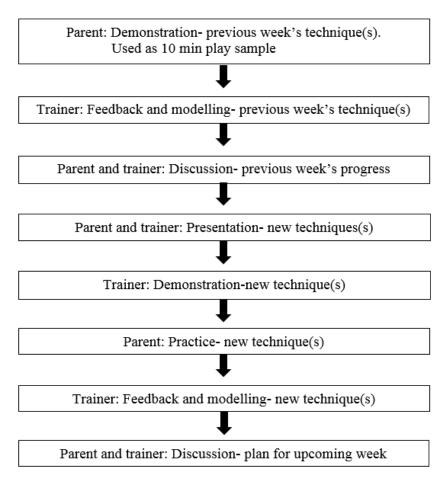


Figure 6.1. Structure of the 11 parent training sessions.

Table 6.3

Curriculum Checklist Items Used to Inform Goals for Each Child.

	y .	3			
Domain	Dean	Rick	Sean	Idris	Alex
Receptive	2-2 (Level-Item):	3-6: Follows two or more	1-5: Follow a proximal	1-5: Follow a proximal	1-5: Follow a proximal
Communication	Follows 8–10 one-step	instructions given in	point to place objects in	point to place objects in	point to place objects in
	verbal instructions	situational routines.	containers, puzzle pieces,	container.	container.
	involving body actions	3-12: Understands	1-13: Gives object as	1-14: Performs a one	1-15: Performs a one
	and actions on objects.	pronoun referents "mine"	verbally requested when	step, routine instruction	step, routine instruction
	2-4: Responds to verbal	and "yours".	paired with adult's	involving body actions	involving body actions
	instruction to		outstretched hand.	paired with	with no gesture
	give/point/show for 8-10			verbal/gesture cues,	
	specific objects in natural			puzzle pieces.	
	play, dressing, eating				
	routines				
Expressive	1-8: Points to indicate a	3-5: Comments and	1-8: Points to indicate a	1-2: Vocalises with	1-6: Points proximally to
Communication	choice between two	requests using early	choice between two	intent.	request desired object.
	objects.	possessive forms (mine,	objects.	1-6: Points proximally to	1-4: Takes turns
	2-11: Nods head "yes"	yours).	2-2: Produces 6-10 single	request desired object.	vocalizing with
	and says "yes"	3-6: Gestures or	words or approximations		communication partner.
	to affirm.	vocalises "I don't know"	within the context of		
		in context.	familiar routines.		

			sensory-social routines,		
			songs.		
Joint Attention	2-4: Responds to "Show	2-3: Gives or takes	N/A	N/A	N/A
	me" by extending object	object from other person,			
	to adult.	coordinated with eye			
		contact.			
Social Skills	1-5: Has a repertoire of 5	2-5: Responds to social	2-5: Responds to social	1-5: Has a repertoire of 5	1-9: Responds to
	– 10 sensory social	greeting with "Hi" or	greeting with "Hi" or	– 10 sensory social	greeting by gesture or
	games.	"Bye-bye", and waves	"Bye-bye", and waves	games.	vocalisation.
	2-2: Verbally requests or	imitatively.	imitatively.	1-9: Responds to	2-2: Verbally requests or
	physically initiates	2-7: Consistently		greeting by gesture or	physically initiates
	familiar social games.	coordinates eye contact		vocalisation.	familiar social games.
		with vocalisation and/or			
		gesture to direct			
		communication.			
Play	2-3: Carries out single	1-8: Completes play task	1-6: Plays independently	1-6: Plays independently	1-2: Plays independently
	action with a prop on a	and puts away.	with toys requiring	with toys requiring	and appropriately with
	doll or animal.	2-3: Carries out single	several different motor	several different motor	10 one-step toys.
		action with a prop on a	actions.	actions.	1-8: Completes play task
		doll or animal.	1-8: Completes play task	1-8: Completes play task	and puts away.
			and puts away.	and puts away.	

Imitation	1-2: Imitates 10 visible	1-1: Imitates 8-10 one	1-1: Imitates 8-10 one	1-1: Imitates 8-10 one	1-1: Imitates 8-10 one
	motor actions inside	step actions on objects.	step actions on objects.	step actions on objects.	step actions on objects.
	song/game routines				
Cognition	1-3: Matches/sorts	4-3: Gives "one",	1-1: Matches/sorts	1-3: Matches/sorts	1-1: Matches/sorts
	pictures to objects.	"some", "a lot", "a	identical objects.	pictures to objects.	identical objects.
		little", "all of them",			
		"more" and "most.			
Fine Motor	2-3: Copies three or	2-3: Copies three or	1-1: Places one to two	1:8: Stacks three big	1-1: Places one to two
	more simple block	more simple block	shapes in a shape sorter.	blocks in a tower (or	shapes in a shape sorter
	designs.	designs.	1-3: Completes three-	stacking cups).	1-3: Completes three
			piece wooden handle		piece wooden puzzle.
			puzzle.		
Gross Motor	2-6: Kicks ball into	3-8: Hops on one foot.	2-6: Kicks ball into	1-1: Kicks big ball.	1-6 Walks around objects
	target.		target.		on floor rather than
					stepping on them.

## **Social Validity**

One week after the final intervention session, each parent participated in a face-to-face semi-structured open-ended interview (Laverty, 2003; Rapley, 2001) about their experiences and perceptions of the intervention. This type of interview enables researchers to understand the world from the subject's point of view (Brinkmann & Kvale, 2015). This interview took place in their home and was conducted by an independent person (an intern educational psychologist at Victoria University). See Appendix H for the questions used in the interview. The analysis of these interviews was informed by thematic analysis (Braun & Clarke, 2006) and Van Manen's (1990) work. The steps of this process included: (a) transcribing the data, (b) a naive first reading and an informed second reading of the data, (c) coding interesting features of the data, (d) identifying potential themes, (e) checking the themes against the data, (f) naming the themes, and finally (g) reporting the findings. The analysis was deductive, as it was informed by existing theories of social validity (e.g. Reimers et al., 1992), and descriptive, as it involved presenting the data without providing additional interpretation (Johnson & Christensen, 2012).

At this time, parents also completed the Treatment Acceptability Rating Form — Revised questionnaire (TARF-R; Reimers et al., 1992). The TARF-R is a 20-item questionnaire which provides a measure of the social validity of the parent training programme based on the parents' perceptions of the program's: (a) reasonableness, (b) their willingness to participate, (c) side effects, (d), effectiveness, (e) disruptiveness, and (f) affordability. There are also two items related to the severity of the child's behaviour and one item about the parent's understanding of the treatment which are not included in the total acceptability score. Each item is rated on a 7-point Likert-type scale.

Trustworthiness of the post-intervention interview data. The quality of the data gained from the semi-structured post-intervention parent interviews was depended upon the trustworthiness of the data collection process (Johnson & Christensen, 2012). Several steps were taken by the researcher in order to increase the trustworthiness of the data (Johnson & Christensen, 2012; Tracy, 2010). First, the data has "face validity" because it relates to similar constructs from other social validity research (e.g. Ogilvie & McCrudden, 2017; Reimers et al., 1992). Second, to increase the credibility of the data, the researcher attempted to establish trust and rapport with the participants by: (a) being warm, welcoming and accepting, (b) taking the time to understand the family and their goals for their child with ASD, (c) taking an interest in the child with ASD and focussing on his strengths, (d) finding areas of shared interest, and (e) being sensitive to cultural practices, such as accepting food

when offered (Kvale, 1996). This led to the collection of thick data (a lot of data) and rich data (many layered, detailed data; Fusch & Ness, 2015) enhancing the credibility of the data set (Shenton, 2004). Third, the researcher used memoing and a reflexive diary to increase transparency and keep track of her thoughts during the data analysis process (Johnson & Christensen, 2012). Fourth, the researcher checked and ensured the resonance of the data by discussing the results with her primary supervisor and fellow post-graduate students, who related to the themes and were able to identify ways to modify their practice in relation to these findings (Tracy, 2010). Last, the fact that the interviews were not conducted by the individual who delivered the therapy/parent training, reduces the likelihood that parents would have altered their description of their perceptions of the intervention in order to protect the researcher's feelings (Johnson & Christensen, 2012).

## **Interobserver Agreement**

IOA was collected by the same independent observer, in the same way as Study 1 (see Chapter 5) for all of the child outcomes variables. For IOA checks related to parent fidelity of implementation, however, an agreement was recorded any time the raters were within one point of each other on the Likert-scale for each procedural fidelity checklist item (adjacent agreement; Vismara et al., 2012). The percentage of agreement was calculated for each session using the formula: Agreements/(Agreement + Disagreements)  $\times$  100%. Table 6.4 indicates that the mean IOA was between 81% and 100% for all participants and outcomes. Table 6.4

Means (and Ranges) for IOA Percentages Across Children and Dependent Variables.

	Dean	Rick	Sean	Idris	Alex
Fidelity	88%	88%	92%	94%	92%
	(78 - 100%)	(72 - 100%)	(83 - 100%)	(77 - 100%)	(83 – 100%)
Imitation	88%	96%	89%	98%	97%
	(66 - 97%)	(93 - 97%)	(77 - 100%)	(95 - 100%)	(95 - 100%)
Total Utterances	92%	96%	89%	99%	100%
	(73 - 97%)	(90 - 100%)	(81 - 98%)	(98 – 100%)	
Engaged	85%	92%	89%	99%	100%
Utterances	(55 - 92%)	(87 - 97%)	(83 - 98%)	(98 – 100%)	
Intentional	N/A	N/A	N/A	94%	94%
Vocalisations				(88 - 97%)	(87 - 98%)

Partial	89%	95%	95%	83%	89%
Engagement	(72 - 97%)	(77 - 98%)	(83 - 97%)	(78 - 92%)	(80 - 100%)
Whole	86%	84%	81%	94%	86%
Engagement	(78 - 92%)	(75 - 92%)	(68 - 91%)	(88 - 100%)	(73 - 93%)

### **Procedural Integrity**

This is a measure of how well the trainer followed the procedures in each phase of the study. During the baseline and follow-up phases, procedural integrity (PI) was assessed using a checklist that was completed by an independent observer (see Appendix G). The observer watched the videos and scored them on a checklist that described each step of the procedures during that phase, for example "the session lasted ten minutes", and "trainer did not provide any feedback to the parent". PI was calculated for one randomly selected baseline and follow-up phase per child. The percentage of procedural integrity was calculated using the formula: steps correct/total steps × 100%. Mean PI during these phases was 100% for all participants.

One trainer (Waddington) implemented all of the parent training sessions. The trainer was a certified ESDM therapist for the duration of the study (see Study 1, Procedural Integrity). However, the trainer was not a certified ESDM parent coach and had not participated in any official training in coaching parents in the ESDM techniques.

Nevertheless, procedural integrity for the trainer's implementation of the parent training sessions was measured using an-18 item checklist that listed each of the steps in the parent training process (see Appendix F). Examples of items include "parent demonstrates previous skills for 10 minutes" and "parent and trainer discuss goals for the following week". An observer independently completed the checklist during two intervention sessions for Dean, Sean, Idris, and Alex, and one intervention session for Rick due to scheduling difficulties. The percentage of PI was calculated using the formula: steps correct/total steps × 100%.

Mean procedural integrity was 100% for Rick, Sean, Idris, and Alex, and 97% for Dean (range = 94 – 100%).

### **Results**

### **Fidelity of Implementation**

Figure 6.2 shows the percentage of ESDM fidelity items implemented correctly by each child's mother for each baseline, intervention, and follow-up session. Also shown are the results from the two generalisation probes that were conducted with each child.

# Fidelity of Implementation.

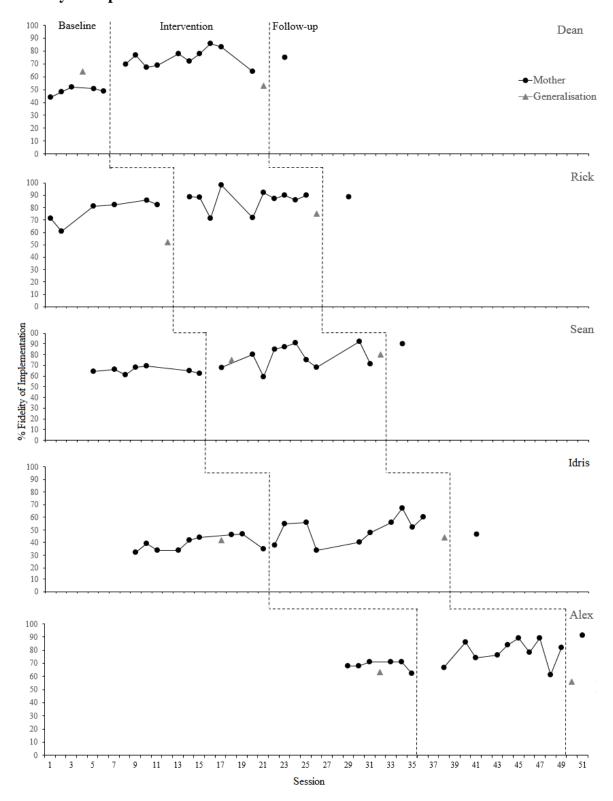


Figure 6.2. Percentage of fidelity items implemented correctly for each mother across phases. As indicated in Figure 6.2, Dean's mother had a mean of 48.8% fidelity of implementation during baseline which increased to a mean of 74.4% fidelity during the

parent training phase (NAP = 1.0). Visual analysis of the graph indicates a variable but increasing trend from the 3<sup>rd</sup> (1<sup>st</sup> intervention session in which data was collected) to 11<sup>th</sup> intervention session and then a decrease in fidelity on the last session. Table 6.4 shows the mean parent fidelity scores across each fidelity domain for baseline and intervention, and the mean change in fidelity from baseline to intervention. During intervention, Dean's mother's fidelity increased most in the transition, instructional techniques and management of attention domains and it decreased in the management of unwanted behaviours domain. Dean's mother's fidelity remained stable at follow-up at 75% (NAP = 1.0). Dean's father's fidelity decreased from 64% in baseline to 53% following intervention.

Rick's mother's mean fidelity increased from a 77.2% in baseline to 86.3% in intervention (NAP=0.85). Visual analysis of the graph indicates large variability in the data until the 8<sup>th</sup> intervention session when it became relatively stable. During intervention, Rick's mother's fidelity increased most in the management of attention, instructional techniques, and dyadic engagement domains and decreased slightly in the management of unwanted behaviours and sensitivity and responsivity domains. Rick's mother's fidelity at follow-up remained stable at 89% (NAP = 1.0). Rick's father's fidelity increased from 52% during baseline to 75% following intervention.

Sean's mother's mean fidelity increased from 65% to 77.6% in intervention (NAP = 0.86). Visual analysis of the graph indicates a variable but increasing trend from the 1<sup>st</sup> to 9<sup>th</sup> intervention session in which data was recorded and then a decrease in fidelity on the last session. During intervention, Sean's mother's fidelity increased most in the dyadic engagement, joint activity structure and elaboration, and management of attention domains and her fidelity decreased slightly in the management of unwanted behaviours, transition, affect and arousal, and sensitivity and responsivity domains. Sean's mother's fidelity remained high at 90% during the follow-up probe (NAP = 1.0). Sean's grandfather's fidelity increased from 75% in the generalisation probe in the 3<sup>rd</sup> week of intervention to 80% following intervention.

Idris' mother's mean fidelity increased from 39% in baseline to 50% in intervention (NAP = 0.82). Visual analysis of the graph indicates an increasing trend from the  $3^{rd}$  to  $5^{th}$  intervention session, which then decreased coinciding with moving house, before increasing again. During intervention Idris' mother's fidelity increased across all domains but it increased the most in the sensitivity and responsivity, multiple and varied communication, affect and arousal, and positive affect domains. Idris' mother's fidelity decreased slightly to 47% during the follow-up probe (NAP = 0.89). Idris' father's fidelity increased slightly from

42% during baseline to 44% following intervention.

Alex's mother's mean fidelity increased from 68.5% in baseline to 78.5% in intervention (NAP = 0.81). Visual analysis of the graph suggests that her fidelity increased sharply from the 3<sup>rd</sup> to 4<sup>th</sup> intervention session and then varied from that point. During intervention, Alex's mother's fidelity increased most in the multiple and varied communication, language, and management of attention domains and her fidelity decreased slightly in the instructional techniques, dyadic engagement, and management of unwanted behaviours domains. Alex's mother's fidelity increased to 91% in follow-up (NAP = 1.0). Alex's father's fidelity decreased from 63% in baseline to 56% in intervention.

Table 6.5

Average Fidelity Scores and Change in Fidelity for Each Fidelity Domain Across Baseline and Intervention for Each Child

-		Dean			Rick		Sean		
Fidelity Item	Baseline	Intervention	Change	Baseline	Intervention	Change	Baseline	Intervention	Change
Management of attention	3.0	8.1	+ 5.1	6.5	9.9	+ 3.4	4.7	8.1	+ 3.3
ABC Format	4.6	9.3	+ 4.7	10.7	10.7	0	7.1	8.4	+ 1.3
Instructional techniques	2.4	8.4	+ 6	8	11.1	+ 3.1	6	8.4	+ 2.4
Affect and Arousal	9	9.1	+ 0.1	9.5	10.2	+ 0.7	10.3	9.9	- 0.4
Management unwanted behaviours	11.4	8.5	- 2.9	11.5	11.1	- 0.4	11.6	10.8	- 0.8
Dyadic engagement	4.2	8.1	+ 3.9	7	9.6	+ 2.6	5.6	9.9	+ 4.3
Motivation	4.8	8.25	+ 3.5	8.75	9.75	+ 1	6	8.25	+ 2.3
Positive Affect	5.4	9.9	+ 4.5	9.5	11.1	+ 1.6	10.3	11.1	+ 0.8
Sensitivity and responsivity	6	10.8	+ 4.8	12	11.7	- 0.3	10.3	10.2	- 0.1
Multiple and varied communication	10.8	12	+ 1.2	11.5	11.7	+ 0.2	7.7	10.5	+ 2.8
Appropriateness of adult language	3	6	+ 3	6	8.4	+ 2.4	6	7.5	+ 1.5
Joint activity structure/elaboration	5.7	7.5	+ 1.8	8.3	10.15	+ 1.8	5.5	9	+ 3.5
Transition	1.8	8	+ 6.2	8	8.5	+ 0.5	7.7	7.2	- 0.5

Table 6.5, continued

Average Fidelity Scores and Change in Fidelity for Each Fidelity Domain Across Baseline and Intervention for Each Child

	Idris			Alex			
Fidelity Item	Baseline	Intervention	Change	Baseline	Intervention	Change	
Management of attention	3.0	5.7	+2.7	8.0	10.5	+2.5	
ABC Format	4.3	6.7	+2.4	7.7	9.2	+1.5	
Instructional techniques	2.3	4.2	+1.9	7.5	6.6	-0.9	
Affect and Arousal	4.0	6.9	+2.9	9.5	9.9	+0.4	
Management unwanted behaviours	8.0	9.9	+1.9	11.5	11.4	-0.1	
Dyadic engagement	3.3	4.8	+1.5	8.5	8.1	-0.4	
Motivation	3.0	5.1	+2.1	7.3	8.4	+1.2	
Positive Affect	4.3	7.8	+3.5	11.0	11.7	+0.7	
Sensitivity and responsivity	4.7	7.5	+2.8	10.0	10.2	+0.2	
Multiple and varied communication	4.7	7.5	+2.8	7.5	10.5	+3.0	
Appropriateness of adult language	2.3	4.2	+1.9	7.5	10.2	+2.7	
Joint activity structure/elaboration	1.8	4.1	+2.3	5.2	7.5	+2.3	
Transition	2.3	4.5	+2.2	6.5	8.4	+1.9	

## **Imitation**

Figure 6.3 shows the percentage of intervals containing independent (unprompted) instances of imitation for each child, session, and generalisation probe.

## Imitation.

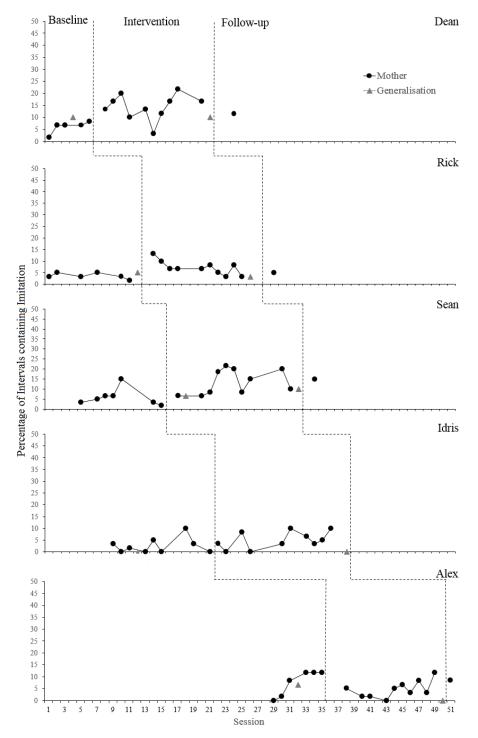


Figure 6.3. Percentage of 10-s intervals containing at least one independent (unprompted) instance of imitation per 10-min play sample for each child across phases.

As indicated in Figure 6.3, during the baseline phase, Dean had a mean of 6% of intervals containing imitation per 10-min play sample, which increased to 14.3% during intervention (NAP = 0.92). Visual analysis of the graph indicates an immediate increase in intervals containing imitation from the  $3^{rd}$  intervention session until the  $5^{th}$  session which decreased until the  $8^{th}$  session before increasing again until the final session. During followup, the number of intervals containing imitation decreased slightly from the mean in intervention to 11.7% intervals (NAP = 1.0). Dean imitated his father during 10.0% of intervals both before and after intervention.

During the baseline phase Rick had a mean of 3.6% of intervals containing imitation per 10-min play sample which increased to a mean of 7.2% of intervals during intervention (NAP = 0.86). Visual analysis of the graph indicates an immediate increase in intervals containing imitation in the  $3^{rd}$  intervention session which decreased until the  $5^{th}$  session, then remained relatively stable. During follow-up, the number of intervals containing imitation decreased slightly to 5% of intervals (NAP = 0.83). The number of intervals containing imitation with Rick's father decreased slightly from 5% of intervals during baseline to 3.3% of intervals following intervention.

During the baseline phase Sean had a mean of 6% of intervals containing imitation per 10-min play sample which increased to a mean of 13.5% of intervals during intervention (NAP = 0.89). Visual analysis of the graph indicates a predominantly increasing trend during intervention apart from decreases in the  $9^{th}$  and  $12^{th}$  sessions. During follow-up the number of intervals containing imitation increased slightly from the mean in intervention to 15% of intervals (NAP = 0.93). The number of intervals containing imitation with Sean's grandfather increased from 6.7% during the generalisation probe in the  $3^{rd}$  week of intervention to 10% following intervention.

During the baseline phase Idris had a mean of 2.6% of intervals containing imitation per 10-min play sample which increased to a mean of 5% of intervals in intervention (NAP = 0.69). Visual analysis of the graph indicates a variable but slightly increasing trend from the 3<sup>rd</sup> to the 12<sup>th</sup> intervention session. During follow-up there were no intervals containing imitation, which was a decrease from the mean in intervention (cannot calculate NAP). Idris did not imitate his father during baseline, or following intervention.

During the baseline phase Alex had a mean of 7.5% of intervals containing imitation per 10-min play sample which decreased to a mean of 4.7% of intervals in intervention (NAP = 0.34). Visual analysis of the graph suggests a decrease in Alex's imitation on the  $3^{rd}$ 

intervention session and then an increase until the 12<sup>th</sup> intervention session. During follow-up the percentage of intervals with imitation was 8.3% (NAP= 0.42) which is a slight decrease compared with the final intervention session. Alex imitated his father during 6.7% of intervals in baseline and no intervals following intervention.

#### **Functional Utterances/Intentional Vocalisations**

Figure 6.4 shows the percentage of 10-s intervals containing intentional vocalisations, functional utterances, and engaged functional utterances for each child, session and generalisation probe.

The figure indicates that during baseline Dean had a mean of 71.3% of intervals containing functional utterances per 10-min play sample and 41.3% of intervals included engaged utterances. Thus, 46% of all intervals containing utterances included engaged utterances. This remained relatively stable at 70% of intervals containing functional utterances during intervention (NAP = 0.44) and a mean of 65.7% intervals which included engaged utterances (NAP = 0.95). In this phase 93.8% of all intervals containing utterances included engaged utterances. Visual analysis of the graph indicates that the number of intervals containing utterances, including engaged utterances, remained relatively stable during intervention apart from a decrease in the 6<sup>th</sup> intervention session. In follow-up, the number of intervals containing utterances increased slightly to 73.3% (NAP = 0.3), and 70%of intervals included engaged utterances (NAP = 1.0). In this phase 95% of intervals containing utterances included engaged utterances. The percentage of intervals containing utterances with Dean's father increased from 78.3% in baseline to 85% following intervention. The percentage of intervals containing engaged utterances also increased from 66.7% in baseline to 81.7% following intervention. Table 6.6 shows the mean MLU, variety of utterances and frequency of utterances for Dean, Rick and Sean across each phase of the study. This table indicates that Dean's MLU varied between 1.8 and 2.1 across the study. His variety of utterances remained relatively stable during baseline and intervention before increasing to 53 at follow-up. His frequency of utterances increased from 61 in baseline to 70.5 in the 1<sup>st</sup> month of intervention. This remained stable for the following 2 months of intervention before increasing slightly to 75 at follow-up. Dean's father- The percentage of intervals containing engaged utterances also increased from 66.7% in baseline to 81.7% following intervention.

## **Functional utterances.**

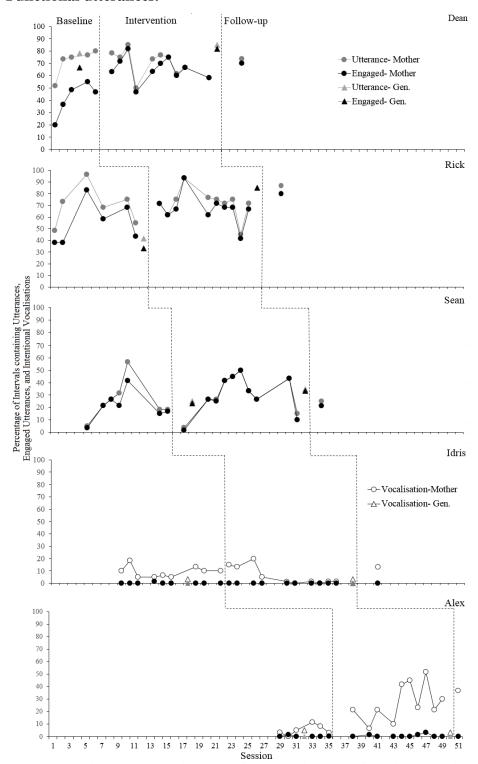


Figure 6.4. Percentage of 10-s intervals containing intentional vocalisations, functional utterances, and engaged functional utterances per 10-min play sample for each child across phases. Note: Gen. = Generalisation

During baseline Rick had a mean of 69.4% of intervals containing utterances and a mean of 55% containing engaged utterances. Thus, in this phase, 79.3% of all intervals containing utterances included engaged utterances. This increased slightly in intervention to a mean of 71.7% of intervals with functional utterances (NAP = 0.58) and a mean of 67.1%containing engaged utterances (NAP = 0.72). In this phase 92% of intervals containing utterances included engaged utterances. Visual analysis of the graph indicates a variable but increasing trend until the 6<sup>th</sup> intervention session before decreasing until the last intervention session. The number of intervals containing functional utterances increased to 86.7% (NAP = 0.83) in follow-up and 80% of intervals contained engaged utterances (NAP = 0.83). In this phase, 92% of intervals containing utterances included engaged utterances. For Rick, the percentage of intervals containing utterances with his father increased from 41.7% in baseline to 85.0% following intervention. Intervals containing engaged utterances also increased from 33.3% to 85%. Table 6.6 indicates that Rick's mean MLU and variety of utterances were highest during the follow-up probe and lowest in the 1<sup>st</sup> month of intervention. His frequency of utterances was similar in baseline, and the 1<sup>st</sup> and 3<sup>rd</sup> months of intervention (58.7, 71.5, and 59.8 respectively) and highest in the follow-up probe.

During baseline Sean had a mean of 25.5% of intervals containing utterances per 10min play sample and a mean of 21% of intervals containing engaged utterances. Thus, in this phase, 82% of intervals containing utterances included engaged utterances. During intervention this increased to a mean of 31.2% of intervals containing utterances (NAP = 0.64) and a mean of 30.3% of intervals containing engaged utterances (NAP = 0.72). In this phase, 97% of intervals containing utterances included engaged utterances. Visual analysis of the graph indicates a variable but predominantly increasing trend. At follow-up the percentage of intervals containing utterances decreased slightly to 25% in total (NAP = 0.57), and 21.7% for engaged utterances (NAP = 0.57). The number of intervals containing utterances with his grandfather increased from 25% during the generalisation probe in the 3<sup>rd</sup> week of intervention to 35% following intervention. Intervals containing engaged utterances also increased from 23.3% to 33.3%. Table 6.6 indicates that Sean's mean MLU varied between 1.1 and 1.3 during each phase of the study. His mean variety of utterances increased from 4.0 in baseline and the  $1^{st}$  month of intervention, to between 10.0 and 11.0 in the  $2^{nd}$  and 3<sup>rd</sup> months, and at follow-up. His mean frequency of utterances was highest in the 2<sup>nd</sup> month of intervention and lowest in the 1<sup>st</sup>.

Idris only had one functional utterance ("No") across all sessions and phases. This

occurred during the 4<sup>th</sup> baseline session. For Idris, the percentage of intervals containing intentional vocalisations decreased from a mean of 9.3% of intervals during baseline to 6% of intervals containing utterances in intervention (NAP = 0.29). Visual analysis of the graph indicates that utterances increased slightly during the 3<sup>rd</sup>, 4<sup>th</sup>, and 5<sup>th</sup> intervention sessions before decreasing after he moved house to between zero and three utterances for the remainder of intervention. During follow-up Idris produced vocalisations during 13.3% of intervals, which was an increase from the means in both baseline and intervention (NAP = 0.83). During sessions with Idris' father 3.3% of intervals contained vocalisations both during baseline and following intervention.

During baseline Alex had a mean of 0.3% of intervals containing utterances per 10-min play sample, all of which were engaged. This remained relatively stable at a mean of 0.7% of intervals containing functional utterances in intervention (NAP = 0.57). He did not produce any functional utterances with his father nor with his mother during follow-up. Alex's intentional vocalisations with his mother increased from a mean of 5.3% of intervals in baseline to a mean of 27.3% of intervals in intervention (NAP = 0.95). Visual analysis of the graph indicates an increasing but variable trend from the  $3^{\rm rd}$  to the final intervention session. The percentage of intervals containing intentional vocalisations increased again during follow-up to 36.6% (NAP = 1.0). The percentage of intervals containing intentional vocalisations with Alex's father decreased from 5% in baseline to 3.3% following intervention.

Table 6.6

Mean MLU, Variety and Frequency of Utterances (1 d.p.) for Dean, Rick, and Sean Across

Baseline, Intervention, and Follow-up.

	MLU	Variety	Frequency
Dean			
Baseline	1.8	34.4	61.0
Month 1 intervention	2.1	34.0	70.5
Month 2 intervention	1.8	36.0	69.3
Month 3 intervention	1.8	34.0	71.0
Follow-up	2.1	53	75
Rick			
Baseline	3.1	45.0	58.7
Month 1 intervention	2.4	35.5	61.5
Month 2 intervention	2.9	61.5	81.5
Month 3 intervention	2.8	43.25	59.8
Follow-up	3.3	75	95
Sean			
Baseline	1.1	4.0	17.3
Month 1 intervention	1.0	4.0	10.5
Month 2 intervention	1.2	11.0	30.5
Month 3 intervention	1.3	10.0	24.5
Follow-up	1.2	10.0	19

Note. The mean for MLU was only calculated for the sessions in which the child had one or more functional utterances.

# **Engagement**

Figure 6.5 shows the percentage of 10-s intervals containing partial or whole engagement for each child, session, and generalisation probe.

# Engagement.

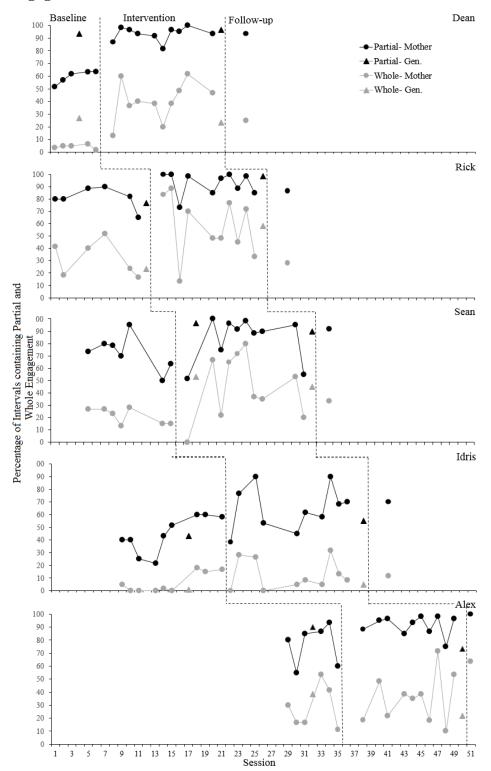


Figure 6.5. Percentage of 10-s intervals containing partial engagement and whole engagement per 10-min play sample for each child across phases.

As indicated in Figure 6.5, during baseline, Dean was partially engaged with his mother for a mean of 59.3% intervals and was engaged for the whole 10-s for a mean of 3.6% of intervals. This increased to a mean of 93.3% of intervals for partial engagement (NAP = 1.0) and 40.3% of intervals for whole engagement (NAP = 1.0) during intervention. Visual analysis of the graph indicates an immediate and relatively large increase in partial engagement in the  $3^{rd}$  intervention session and a variable but increasing trend for whole engagement. The increase in intervals containing partial engagement was maintained at 93.3% in follow-up (NAP = 1.0). The number of intervals containing whole engagement decreased to 25% in follow-up (NAP = 1.0). Partial engagement with Dean's father increased from 93.3% of intervals during baseline to 96.7% of intervals following intervention. Whole engagement with Dean's father decreased from 26.7% intervals during baseline to 23.3% of intervals following intervention.

During baseline, Rick was partially engaged with his mother for a mean of 80.9% of intervals and was engaged with his mother for the whole 10-s nterval for a mean of 31.9% of intervals. This increased to a mean of 92.5% of intervals for partial engagement (NAP = 0.83) and 57.6% of intervals for whole engagement (NAP = 0.80). Visual analysis of the graph suggests variable trends that were not increasing or decreasing for both partial and whole engagement. At follow-up, the percentage of intervals containing partial engagement decreased to 86.7% (NAP= 0.67) and the percentage of intervals containing whole engagement decreased to 28.3% (NAP = 0.5). Rick's partial engagement with his father increased from 76.7% of intervals in baseline to 98.3% of intervals following intervention and his whole engagement also increased from 23.3% to 58.3% of intervals.

During baseline, Sean was partially engaged with his mother during a mean of 72.9% of intervals and was fully engaged with his mother for the whole 10-s during a mean of 21.2%. This increased during intervention to a mean of 84.2% of intervals for partial engagement (NAP = 0.74) and 45% of intervals for whole engagement (NAP = 0.78). Visual analysis of the graph indicates a variable but increasing trend for partial engagement until the  $11^{th}$  intervention session, and then a decrease in the final intervention session. At follow-up the percentage of intervals containing partial engagement increased slightly to 91.6% of intervals (NAP = 0.86) and the number of intervals containing whole engagement decreased to 33.3% (NAP = 1.0). With Sean's grandfather, partial engagement decreased slightly from

96.7% of intervals in the 3<sup>rd</sup> week of intervention to 90% of intervals following intervention and his whole engagement also decreased from 53.5% to 45% of intervals.

During baseline, Idris was partially engaged with his mother for a mean of 44.4% of intervals and was engaged with his mother for the whole 10-s interval for a mean of 6.3%. This increased to a mean of 65.2% of intervals for partial engagement (NAP = 0.82) and 12.7% of intervals for whole engagement (NAP = 0.67) in intervention. Visual analysis of the graph indicates an increasing trend for both partial and whole engagement from the  $3^{rd}$  to  $5^{th}$  intervention session, before decreasing when his family moved house and then steadily increasing, with the highest engagement recorded in the  $10^{th}$  intervention session. At follow-up the number of intervals containing partial engagement increased slightly from the mean in intervention to 70% of intervals (NAP = 1.0) and the number of intervals containing whole engagement remained stable at 11.7% of intervals (NAP = 0.7). Idris' partial engagement with his father increased from 43.3% of intervals during baseline to 55% of intervals following intervention, and his whole engagement increased from 0% to 5%.

During baseline, Alex was engaged with his mother for part of the time during 76.7% of intervals and he was engaged with her for the whole 10-s for 28.3% of intervals. During intervention, this increased to a mean of 91.3% of intervals for partial engagement (NAP = 0.84) and 35.3% of intervals for whole engagement (NAP = 0.63). Visual analysis of the graph indicates that the intervention data for whole engagement was relatively stable and the intervention data for whole engagement was considerably more variable. At follow-up the number of intervals containing partial engagement increased to 100% (NAP = 1.0) and the number of intervals containing whole engagement increased to 63.3% (NAP = 1.0). Alex's partial engagement with his father decreased from 90% of intervals during baseline to 73.3% following intervention and his whole engagement decreased from 38.3% to 21.7%.

### **Correlations**

Pearson's correlation coefficient (r) was calculated to evaluate the relationship between changes in parent fidelity and child outcomes. It is a measure of the linear correlation between two variables (Evans, 1996). Negative correlations indicate that when one variable increases the other decreases, whereas positive correlations indicate that when one variable increases the other variable also increases. Scores between 0-0.39 indicate a very weak or weak correlation, scores between 0.4 and 0.59 indicate a moderate correlation, scores between 0.6 and 0.79 indicate a strong correlation and between 0.8 and 1.0 a very strong correlation (Evans, 1996).

Table 6.7 lists the correlations between parent fidelity and the child outcome variables. The correlation between parent fidelity and imitation was positive and weak for Alex and Rick, positive and moderate for Idris, positive and strong for Dean, and positive and very strong for Sean. The correlation between parent fidelity and total utterances was positive and weak for Dean and Rick, and positive and moderate for Sean. The correlation between parent fidelity and their child's engaged utterances was positive and strong for Dean, Rick and Sean. The correlation between parent fidelity and vocalisations was positive and strong for Alex and negative and weak for Idris. The correlation between parent fidelity and partial engagement was positive and moderate for Rick, positive and strong for Sean and Alex, and positive and very strong for Idris and Dean. The correlation between parent fidelity and whole engagement was positive and moderate for Rick, positive and strong for Sean, Idris, and Alex and positive and very strong for Dean.

Table 6.7

Pearson's Correlation Coefficients (2 d.p.) Between Parent Fidelity and Child Imitation,

Functional Utterances, Intentional Vocalisations, and Engagement for each Child, and Phase

	Imitation	Total	Engaged	Intentional	Partial	Whole
		Utterances	Utterances	Vocalisations	Engagement	Engagement
Dean	0.66	0.02	0.75	-	0.91	0.82
Rick	0.22	0.23	0.63	-	0.59	0.50
Sean	0.81	0.55	0.63	-	0.65	0.79
Idris	0.49	-	-	-0.19	0.84	0.69
Alex	0.07	-	-	0.71	0.74	0.76

### **Social Validity**

TARF-R. Table 6.8 summarises the results of the TARF-R social validity questionnaire. Results of this questionnaire suggest that all five mothers found the intervention to be socially acceptable, as indicated by total acceptability scores above the midpoint (60 or higher). Idris' mother gave the intervention the lowest total acceptability score. This was mainly due to her rating the intervention effectiveness and disruption lower than the other four mothers. All five mothers found the intervention to be highly reasonable and affordable. They were also all very willing to implement the procedures. All five mothers gave the lowest ratings on the disruption domain.

Table 6.8

Results from TARF-R.

Scales/Subscales			Child			Maximum Score	Mean (SD)	
	Dean	Rick	Sean	Idris	Alex	-		
Total acceptability	105	100	105	89	106	119	101 (7.1)	
Reasonableness	18	20	20	18	20	21	19.2 (1.1)	
Willingness	18	18	20	18	20	21	18.8 (1.1)	
Side-effects#	21	17	17	16	18	21	17.8 (1.9)	
Effectiveness	20	20	21	13	19	21	18.6 (3.2)	
Disruption/time#	14	12	13	10	15	21	12.8 (1.9)	
Affordability	14	13	14	14	14	14	13.8 (0.4)	
Severity#*	9	N/A	N/A	12	12	14	11 (1.7)	
Understanding*	6	7	7	6	6	7	6.4 (0.5)	

<sup>#</sup> Includes items that are reverse coded so that a higher score is indicative of a more favourable rating. \* Not included in Total Acceptability score.

### **Post-Intervention Interview**

The information gained from the semi-structured interview has been organised into four separate but related themes: Effect on child outcomes, model of intervention, parent training procedures, and relationship with the trainer. Each of these themes were further organised into the categories of strengths, weaknesses, and improvements (parent training procedures only). Each of these categories contained several more specific codes. See Appendix I for a table containing examples of the codes used for the model of intervention theme in the strengths category.

Effect on child outcomes. Strengths. Three of the five parents (all but Idris' and Alex's mother) stated that they found the parent training intervention to be effective overall. In fact, Sean and Rick's mothers both said that they found the training to be the most effective intervention they had done with their children. Rick's mother said "this is by far the most impact, in a good way, of any professional that we've interacted with, in our whole time since he was 3.5." Alex's mother also said that this intervention was more effective for her child's communication than a previous, similar parent training intervention.

The parents identified different positive outcomes for their children following the

intervention. Each parent mentioned improvements in expressive language, which included both spoken and non-verbal communication. For example, when asked about Dean's spoken language his mother said "he improve a lot, make me feel good" while Alex's mother said "it's good that he can communicate with us instead of getting upset..., he knows that he can touch something that he wants". Sean and Dean's mothers also commented on their child's improved ability to understand and follow instructions. Three parents (Sean, Rick, Idris) mentioned improvements in aspects of their child's social interaction and engagement. Idris' mother said "He's getting more aware of his environment now and looking at us and checking with us all the time". She also stated that her relationship with him had been strengthened since doing the training: "we had a good relationship but now we had a bonding, the bonding is getting more and more". Two parents also said that their children had become more tolerant of being around others "...at kindy... if they're doing a game or something he'll go and sit in the middle of them, where he never would've [before]" (Sean's mother). Four parents (all but Alex's mother) stated that aspects of their children's play skills had improved, including turn-taking, functional play, and pretend play. Sean's mother said "originally he wouldn't pick up a toy and automatically know what to do with it, whereas I've seen him picking up his brother's lego toys now and working out to race them across the table." Idris' mother identified improvements in his imitation skills and Dean's mother also stated that he had improved in some personal care skills such as getting dressed to leave the house.

Challenges. Each parent also identified specific child skills which they felt had not improved, or improved less, during the training. Two parents (Alex and Sean's mothers) mentioned that they would like their children to use their expressive language more consistently. Alex's mother said: "Just for him to be able to communicate comfortably, and you know for it to be, not just sometimes, I want it to be more consistent." Two parents (Dean and Sean's mothers) also wanted their children to interact more with other children of a similar age. Sean's mother stated that, for Sean, "making a friend or something like that would be really good because at the moment he's just happy to play by himself". Rick's mother stated that Rick's imitation skills and social interaction increased less than his other skills during the intervention. For example, "I think social interactions for him are very difficult and... if he's under any kind of stress and anxiety that is the first thing that will disappear, is that social kind of awareness so I would say we have made smaller gains in that area.". Alex's mother felt that she needed more time to target his play skills and Dean's

mother wanted to address his refusal to take off his jersey in hot weather. Idris' mother stated that it was difficult to achieve any of his goals and, particularly felt that the intervention did not improve his expressive language, stereotypy and ability to use greetings: "she asked us to, showing the hi, waving the hands for hi and bye but he was more into other kinds of hand movements like hand shaking and saying hi-five".

Elements of the intervention that may have contributed to these improvements, or lack of improvements, include (a) the ESDM model of intervention, (b) the procedures used to train the parents, and (d) the relationship between the parent/child and the trainer. These themes will be explored in the following sections.

Model of intervention. Strengths. The parents identified several strengths of the ESDM model of intervention. All of the parents liked that the model taught children skills through play and fun activities. Sean's mother said "You just don't think about how fun stuff for him will be really good learning opportunities" and Alex's mother said "it's teaching you to play with your child and enjoy it". Two parents also spoke about the ease of targeting skills within the context of daily routines. Rick's mother said "that's what's the beauty of it, you can put it in anywhere, and if you haven't seen your child 'cause they're in day-care you can do it while they're in the bath" and Alex's mother said "it feels really natural, we're not doing anything crazy different. It's just showing me how to put things into everyday activities". Four of the parents (not Idris' mother) commented on how the techniques were simple and easy to understand. Alex's mother said "you know it wasn't any big names or crazy name terms for things it was just simple". Rick's mother also liked the flexibility of the intervention and felt that it aligned well with her own parenting style: "I thought that looks gentle and nice and sits comfortably with how we already parent Rick".

Three parents identified specific ESDM techniques that they found particularly helpful. Sean's mother emphasised the importance of reinforcing communicative attempts as well as delivering a clear antecedent and waiting for the response: "you know just making him wait a bit longer... you realise, oh, he's going to respond to that." Alex and Idris' mothers appreciated the need to elaborate and use a variety of different toys in games. Finally, Alex's mother liked the focus on communication.

Challenges. There were also aspects of the model that some parents found challenging. Two parents (Rick and Idris' mothers) reported that they sometimes found it difficult to remember to implement the techniques. Rick's mother stated: "...sometimes it is challenging just to remember all the stuff that you've got to do". Rick's mother also said that

it was difficult to consistently implement the procedures when "in the course of every day and people being tired and your child being tired, and you being tired actually is more to the point". Alex's mother shared that it was hard to find time for all of the activities: "I think doing more of the activities, would be a bit more difficult for us, spending more time but you know you need to have time to succeed so, but it is hard for us". Although she added that "I think any sort of treatment probably would be [hard]". Idris' mother found it particularly difficult to find time to implement the procedures when they were moving house: "we were really busy moving the house and we couldn't give any time to him because we were settling in and stuff."

Alex and Idris' mothers reported having difficulties helping their child to achieve some of their goals. Alex's mother questioned whether teaching Alex's play skills were developmentally appropriate: "probably because he's not up to the playtime, I don't know if he is or not but it's harder for us the playing ones". She also found it difficult to reconcile her earlier learning with the learning from later chapters: "...so playing with him and then playing with him leaving him..., it's kind of hard to get a grasp on which one I'm doing". Idris' mother found imitating his vocalisations to be ineffective and also questioned the methods for teaching some non-verbal communication skills such as waving and pointing. She said:

I think especially I would say about the waving, yeah I couldn't keep on going waving all the time. I think he is giving attention when I am doing like "Hi" if I'm, like when I come out of a room or when I see him every time, he's responding to the "hi" or the waving by just seeing, he will look at me like what I'm doing like "what's the crazy thing that I am doing"

Parent training procedures. *Strengths*. The parents identified several strengths of the parent training techniques and the structure, timing and location of the training sessions. Four parents (all but Idris' mother) identified elements of the training techniques that they found to be particularly helpful. All of these parents felt that the trainer modelling the skill was a useful technique. Three of the parents reported that this was the most useful technique. Dean's mother found this technique to be the most effective because "especially for my English,... when she show, I just see it easy, what to copy it". Sean's mother also preferred a practical demonstration of the skills:

I think... just the practical parts were the better parts just because I could see it first-hand what I'm supposed to be doing. You know especially when you get a lot of

information, you know you're reading it and things it can just be overload, overwhelming and just go in one ear and out the other.

All of these parents valued the PowerPoint® presentations and the way the trainer used them as a guideline to the conversation. Rick's mother said "they were good from the aspect where we didn't have to go through everything on them." These parents also liked receiving feedback from the trainer both about positive aspects of their ESDM techniques and possible areas for improvement. Rick's mother said that the feedback was:

... very effective because it reinforced things that I was already doing so that's really good because I know I'm doing that properly. And then it helped me to focus on things I forget to do, or things I wasn't doing or things I could have been doing slightly better, so I found it very effective for me...

Sean's mother also appreciated the immediacy of the feedback: "it was good to get that feedback straight away rather than say the next week when you'd sort of forgotten what you'd done." Three parents reported that they found the ESDM training manual to be helpful. When asked which training technique was the most helpful, Alex's mother replied "I think obviously the manual, reading the manual is important, really important." Two parents also liked the "refrigerator notes" or summary that they received at the end of each session. Sean's mother said "by the time you've had all that information go in, it can be a bit overwhelming. So having that reference made it much easier 'cause you'd go right, that's what I'm doing this week". Rick's mother preferred a combination of teaching techniques:

I think for me the most helpful thing would be like the three step process where we talk about it, I do it, and then H. gives me feedback about what I did or didn't do that I should have, because then you can see whether you're putting into practice the way that you should the things that you've talked about.

Sean and Rick's parents felt that an hour was a good length of time for the training sessions. Sean's mother said "I think it was good because it covered everything". Rick's mother also thought that it suited her to have one session per week for 12 weeks. Alex, Sean, Idris and Dean's mothers stated that they did not find the training session to be disruptive. Alex's mother said "I mean I'm a stay-at-home mum so, we can do these things during the day". Sean and Dean's mother thought that having the training sessions at home suited them. Sean's mother explained:

I quite like it being here because there's no other distractions and he can just, he's safe to show her all his stuff that he wants to do so... And then if he needs to go and have some time out he can do that.

Challenges. There were also aspects of the parent training techniques and the structure, timing and location of the training sessions that parents found challenging. Three of the parents (Alex, Rick, and Idris' mothers) stated that some weeks they struggled to find time to read the ESDM parent manual. Idris' mother said: "Like the first few chapters I could read and follow up with her but after that I don't have time to read the book and I couldn't follow-up with her." Dean's mother did not read the book but her husband did: "Yeah he read because my English [makes it] hard to understand so yeah, he read."

Rick's mother also mentioned that the weekly sessions were disruptive in the sense that "when you have an appointment booked it means that you can't do things because you've got that person coming, so you can't go to the park because you need to be back in time to have the video. However, she felt that the disruption was worth it: "if I felt like we weren't getting enough out of it then I'd be concerned but the gains for us outweigh those inconveniences".

Improvements. Each of the parents suggested potential alterations and improvements to the parent training techniques and/or the structure, timing and location of the parent training sessions. Alex and Idris' mothers would have preferred more feedback from the trainer. Alex's mother said "even more feedback on the videos like what good things, bad things, would have been really good." Alex's mother also suggested that the trainer could have spent more time modelling the techniques. She explained "a good thing for me would be to be shown more techniques, I can learn by reading but I am better just by watching and then doing, it's a lot easier for me, it makes a lot more sense". Idris' mother suggested that the trainer could have more thoroughly explained each of the slides on the PowerPoint® presentation and given her a copy of the notes. She said "but for every slide she didn't explain the things completely, every detail, because I wanted to go through all the details. Because the PowerPoint® was not given to me, only the last sheet".

Three of the parents (Idris, Dean, and Alex's mothers) suggested that each training session could last for longer than 1 to 1.5 hours. Dean's mother said that this is because her son "likes to play", while Idris and Alex's mother said it was to provide more time for the teaching techniques. Four of the parents (Dean, Idris, Sean, and Alex's mothers) suggested that the training would have been more beneficial if it had continued for more than 12 weekly

sessions. Three of the parents did not suggest a precise number of additional sessions but Dean's mother referred to needing "more help". Sean's mother elaborated that although she was sad that the training was going to finish: "you sort of go, well what else can you teach you know what I mean. You've got to have something to be able to teach." Idris' mother suggested that "at least three stages, with 10 to 12 weeks would be good enough". Alex's mother suggested that she needed 2 weeks between sessions for the topics in the second half of the training: "...just to process it and practice it because for people like me it takes a little bit of time..."

Four of the parents (all but Idris' mother) suggested that "in an ideal world" their child's father could be included in the training. However, Alex' mother also said "that wouldn't have been able to happen" and Rick's mother said "that would have been nigh on impossible". Some parents also suggested that the trainer's supervisor (Idris' mother), teachers from the child's kindergarten (Rick's mother), and members of their extended family (Rick and Sean's mother) could be included in some training sessions. Idris' mother thought it would be helpful if some of the sessions were conducted in the kindergarten or at a playground and Alex's mother said: "I wouldn't mind going somewhere. Especially if they had, you know, different resources that I could see and use, that he would maybe like and show interest in."

Relationship with the trainer. *Strengths*. Four parents (all but Alex's mother) commented on strengths of both their relationship with the trainer and their child's relationship with the trainer. Three parents mentioned some of the personal qualities that they found beneficial. For example, Sean and Rick's mother stated that they appreciated that the trainer was non-judgemental and flexible. This was in regard to setting appointments, for example, Sean's mother said: "She certainly didn't make me feel terrible for having to cancel one week 'cause it just wasn't working that week you know, everyone has those moments" and around difficulties targeting the child's goals, for examples, Rick's mother said:

If the trainer is good then they're going to be flexible enough to understand that what they're trying to get you to do might not work with your child, or they can understand or see what you're not doing, so they might have to try and get you to do it in a different way.

Rick and Sean's mothers both commented on the importance of the trainer's knowledge and experience of working with children with ASD. Sean's mother said: "It's just nice hearing that from someone that's dealt with lots of kids because, like I say, I haven't had that

experience." Idris' mother also stated that it was good that the trainer was "really practical oriented." Sean's mother commented on the trainer's ability to clearly explain the concepts and Rick's mother on the trainer's ability to reflect on her own performance:

And she'll pull herself up later and say now I didn't do that thing 100 percent correctly so she, so that's good because it's a good dialogue to have to remember how to do things and be reminded about them and to appreciate that even an expert doesn't do them properly all the time so you don't feel so bad.

Four parents said that their child enjoyed interacting and playing with the trainer (all but Alex's mother). Two of these parents (Idris and Sean's mothers) shared that their child was initially hesitant or unhappy about the trainer's presence in their home, but that they then developed a positive relationship. For example, Sean's mother said: "At the beginning it was very much he didn't want her here because she was a strange person. But now, as soon as she walks in, he grabs her to take her to the trampoline." Rick's mother stated that the important qualities for someone interacting with her child included "someone kind, gentle, kind of calm but fun if that makes sense and someone who you can see is keeping an eye on your child's behaviour and reading that behaviour" and that, because the trainer had these qualities "that made me feel comfortable and that had a good outcome on the study".

*Challenges.* Rick's mother was the only parent to mention any challenges that arose because her child's relationship with the trainer. She stated that Rick could sometimes become upset when the trainer left the house:

sometimes when [the trainer] arrived he's very tired and having someone arrive and leave the house is sometimes quite, can upset him quite a lot so, a couple of weeks when [the trainer] would leave he'd have a meltdown and throw shoes and throw things.

Although none of the parents mentioned any challenges in terms of their or their child's relationship with the trainer, Rick's mother did state "that person has to be all of those things you want, approachable and kind of kind and gentle, and all of those kind of nice things." and "if she had not been that would have made me want to leave the study, if I'd got really anxious about her interacting with Rick then I would have wanted to pull out"

#### Discussion

One aim of this study was to evaluate whether a 12 week home-based and parent implemented version of the ESDM was effective in increasing imitation, functional utterances, intentional vocalisations, and engagement in young children with ASD. A second aim was to determine if the parent training programme was effective and viewed as socially valid for enabling parents to implement the ESDM with fidelity. A third aim was to assess if any treatment gains would maintain over time and generalise to a different family member other than the parent who received the training. The results of this study suggest that all five parents improved in their ability to faithfully/correctly implement the ESDM procedures with their children. Indeed, all parents, except Idris' mother, achieved an 80% or higher level of fidelity on at least one occasion. Their high level of fidelity was maintained at the 1 month follow-up after the training programme had been completed. In terms of child outcomes, each child showed some improvement on at least two of the child dependent variables during intervention and these improvements were generally positively correlated with their parent's level of fidelity of implementation. Most of the children's improvements were maintained at follow-up, however, three of the children did not maintain their improvement on one of the outcome measures at this time. In terms of generalisation, only two of the five family members increased their use of the ESDM procedures following intervention. Further, only Rick and Dean had increases in functional utterances and/or engagement with their fathers following intervention and none of the children imitated their fathers more at this time. All five mothers reported that they found the intervention to be socially valid and acceptable, although Idris' mother gave the intervention a lower acceptability rating than the other mothers. The mothers also identified strengths and challenges related to the effect of the intervention on outcomes for their child, the model of intervention (ESDM), the parent training procedures, and the relationship with the trainer.

## **Parent Fidelity of Implementation**

Most parents appeared to have learned to use the ESDM procedures with an acceptable level of fidelity (80% or above). These results are consistent with previous ESDM parent training research in that they suggest that most parents learned to use the techniques (Vismara et al., 2016; Vismara, Colombi et al., 2009; Vismara et al., 2012; Vismara, McCormick et al., 2013; Vismara & Rogers, 2008). The current results are also consistent with several literature reviews of parent training for young children with ASD, which generally concluded that most parents were able to learn to implement the various

intervention techniques (e.g. McConachie & Diggle, 2007; Meadan et al., 2009; National Autism Centre, 2016; Oono et al., 2013; Patterson et al., 2011). Thus, the findings of this study provide further support that many parents can learn to implement interventions with their young children with ASD. However, they contrast with the Rogers, Estes, et al. (2012) study, in which only 45% of parents in the ESDM parent training group reached fidelity. Rogers, Estes, et al. (2012) suggest that this may be because the parents in ESDM parent training had not had adequate time to practice and master the skills following the training.

Three of the four parents who had reached acceptable levels of EDSM fidelity (80% or above) during parent training maintained this level at follow-up. This is consistent with several ESDM parent coaching studies, which also found that all the parents who reached fidelity during intervention also maintained high levels of fidelity during follow-up (Vismara et al., 2016; Vismara, Colombi et al., 2009; Vismara et al., 2012, Vismara, McCormick et al., 2013; Vismara & Rogers, 2008). This is an important finding as it suggests that the parents continued to accurately implement the ESDM procedures without the continued support of the trainer. However, Dean's mother's fidelity decreased on the final intervention session, which suggests that she had not yet learned to consistently implement the ESDM procedures. In addition, 1 month after parent training, Dean's mother's fidelity was 10% lower than her highest fidelity score. Research on similar parent training programmes, such as PRT, has also found that some parents did not maintain adequate levels of fidelity after the training was completed (Cadogan & McCrimmon, 2015; Coolican, Smith, & Bryson, 2010; Randolph, Stichter, Schmidt, & O'Connor, 2011).

The findings of this study also differ from most previous ESDM parent training research, in that three of the parents who reached fidelity did so on, or before, the 4<sup>th</sup> intervention session. In contrast, other ESDM parent training studies found that, on average, parents reached fidelity between the 6<sup>th</sup> (Vismara, Colombi, et al., 2009; Vismara et al., 2012) and 8<sup>th</sup> (Vismara & Rogers, 2008) intervention session. These three parents also had higher baseline levels of fidelity than the majority of parents in the previous parent training studies. Similarly, the parents who had the highest baseline fidelity in the Vismara, McCormick et al. (2013) study also reached 80% ESDM fidelity or higher within the first few weeks of intervention. This may be because the first chapters of the ESDM parent manual describe the majority of the ESDM fidelity items, with the exception of explicitly addressing the ABC format, instructional techniques, and some aspects of language (Rogers, Dawson, et al., 2012). Therefore, it would seem plausible that parents who were using many of the

techniques before the training, would have the greatest increase in fidelity within the first few weeks.

Interestingly, there was considerable variation in parents' fidelity scores from session to session, such that, after parents had reached an acceptable level fidelity for the first time, there were at least two sessions in which their fidelity of implementation was under 80%. This variability appeared to be greater for the parents in this current study than those who participated in previous ESDM parent coaching research (Vismara et al., 2012; Vismara, Colombi et al., 2009; Vismara, McCormick et al., 2013; Vismara & Rogers, 2008). There are several potential explanations for this variation. First, the nature of the task may have affected the parents' ability to implement the procedures. Specifically, it may have been easier to for parents to implement the procedures during the child's most preferred activities. For example, Sean's mother's fidelity was highest during sessions when they were jumping together on the trampoline, which she described as one of Sean's most preferred and motivating activities. Further, some of the ESDM procedures, such as management of attention, may be easier to implement during sensory social, rather than object-focused activities (Rogers & Dawson, 2010). Parents may also have found the fidelity items more difficult to implement during the second half of the parent training, when they were also attempting to use the procedures to directly target child skills (Rogers, Dawson, et al., 2012). For example, during the posttreatment interview, Alex's mother stated that she found it very difficult to work on play skills with her son but that the first few chapters were easy and made sense. Further, the variation in treatment fidelity may be due to extraneous events that occurred before the treatment session (Johnson & Christensen, 2012). For example, in the post-treatment interview, several parent reported that it was harder to implement the procedures when they, or their child, was feeling unwell or tired. More research is needed to determine factors that may affect parents' ability to implement intervention procedures with their children with ASD, once they have already reached a high level of fidelity in at least one instance.

Rick's mother had over 80% ESDM fidelity during several baseline sessions, whereas in all of the previous parent training research, aside from the study by Rogers, Estes, et al. (2012), none of the parents met fidelity prior to training. Rogers and Vismara (2015) also suggest that "sometimes parents come into the treatment with well-developed play and interaction skills with their child and their initial P-ESDM fidelity of implementation scores are already at four or five in most areas" (p. 26). There are several potential reasons why Rick's mother had such high fidelity. First, Rick had been diagnosed with ASD for longer

than the other children who participated in the study, thus, Rick's mother had more time to find play and interactional strategies that worked well with him. She had also previously participated in a 1 day workshop on play-based intervention for young children with ASD. It is likely that there was considerable overlap between the skills covered in the workshop and those used in ESDM intervention (Rogers, Estes, et al., 2012). Further, despite the fact that Rick's mother had over 80% fidelity during baseline, her fidelity improved a similar number of percentage points to some of the other parents during the parent training programme (i.e. Idris and Alex's mothers). She also reported that she found the intervention to be highly effective and beneficial.

Dean's mother's fidelity increased the most, percentage-wise, during the intervention. This may be because she had lower baseline fidelity than the three mothers who also reached fidelity. There is evidence of a ceiling effect for the three remaining mothers as, due to their high baseline fidelity, there was limited "room to improve" during intervention (Johnson & Christensen, 2012). The fact that there was a large and immediate increase in Dean's mother's fidelity between baseline and the third parent training session suggests that the initial improvement was perhaps due to performance effects, rather than Dean's mother learning entirely new skills (Johnson & Christensen, 2012). In other words, once she had received the information about positive ways to interact with her child, she was easily able to implement these skills because she already knew how to do them.

Although Idris' mother's use of the ESDM procedures improved, she did not reach acceptable levels of ESDM fidelity during the parent training programme or 1 month later. This is consistent with several previous ESDM parent coaching studies, which found that at least one parent did not reach fidelity in the course of intervention (Rogers, Estes, et al., 2012; Vismara et al., 2016; Vismara, Colombi, et al., 2009; Vismara, McCormick, et al., 2013). There are several potential explanations for why the parent training programme was less effective for Idris' mother.

First, she had the lowest baseline level of fidelity and implemented very few of the ESDM techniques prior to the parent training study. She did show a similar percentage of improvement to most of the other parents during the intervention and it is possible that her fidelity would have continued to increase had the intervention continued for longer. In fact, the developers of the ESDM model do suggest that some parents may need additional, more intensive parent training in order to consistently implement the ESDM with fidelity (Dawson, Estes, et al., 2010; Ruppert et al., 2016; Vismara, McCormick, et al., 2013). During the post-

treatment interview, Idris' mother also stated that she would like to receive more training and intervention.

Second, Idris' mother was the only parent in this study who was working full-time. It is possible that due to her work commitments, she had less time than the other parents to practice and implement the ESDM techniques with her son, and, therefore, did not reach fidelity. Research suggests that mothers of children with ASD are less likely to work full-time than mothers of children without ASD or developmental disabilities, which could be partially due to their increased need to provide and co-ordinate intervention for their children (Cidav, Marcus, & Mandell, 2012). Thus, mothers who work full-time may struggle to provide the same level of intervention as mothers who work part-time or do not work.

Third, there was a large decrease in Idris' mother's fidelity coinciding with the family moving house, which also corresponded with a decrease in Idris' imitation, vocalisations, and engagement. Idris' mother stated that moving house was a particularly disruptive time for both herself and Idris. Research suggests that relocation is stressful for both typically developing children and their parents (Munton, 1990). Further, Davis and Finke (2015), also suggested that children with ASD in military families and their parent(s) experienced increased stress upon relocation. Bagner and Graziano (2013) found that the more stressors a parent faces, the more likely he or she is to withdraw from parent training programmes. It is possible that this disruptive time, and the stress it produced, may have further decreased Idris' mother's ability to focus on using the ESDM techniques with Idris. More research should be conducted on the effect of stressful life events, such as moving house, on parent's ability to implement interventions with their children with ASD. In addition, future parent training programmes should evaluate ways of reducing parent stress in the context of participation in parent training programmes.

Fourth, Idris did not receive a formal diagnosis of ASD until the 10<sup>th</sup> intervention session, despite the fact that he was referred for diagnosis more than 6 months prior. This is consistent with research which suggests there is a large gap between the age of a child when they are first referred and the age at which they receive a final diagnosis (Howlin & Moore, 1997; Shattuck et al., 2009). Qualitative research on parent's experiences of ASD diagnosis suggests that once "a label or diagnosis was given, [parents] were able to understand their child's behaviour, accept the condition, and plan for the future" (Midence & O'Neill, 1999, p.283). Thus, it is possible that, prior to his official diagnosis, Idris' mother did not fully recognise the importance or necessity of using intervention techniques which were developed

specifically for children with or at risk for ASD. This is supported by the fact that during the post-treatment interview Idris' mother stated that since his diagnosis she had been attempting to use the ESDM techniques with him for at least 1-hr each day.

Last, it is possible that some of Idris' characteristics made it harder for his mother to implement the ESDM procedures with him. For example, Idris' *Vineland-II* results suggest that his play skills were lower than those of the other children, although it should be noted that he was also the youngest child (Sparrow et al., 2005). His mother also reported that he mostly played with toys in a non-functional, repetitive way. As the ESDM is a play-based therapy, it may be harder to for parents to implement with children who do not play with toys in a functional way (Rogers & Dawson, 2010).

#### **Child Outcomes**

During parent training, children improved on at least two of the child outcome measures, although each child either had minimal improvements, did not improve, or decreased their performance on at least one outcomes measure. This is consistent with previous research which suggests that, while most parents can be taught to use intervention techniques, their improved use of these techniques does not always translate to improved outcomes for their children with ASD (Oono et al., 2013). Indeed, Vismara et al. (2016) found that while the majority of parents in their study learned to implement the ESDM techniques, there was no difference between their children's social communication skills and those of children who received treatment-as-usual. Vismara, McCormick, et al. (2012) similarly found that parents' correct use of ESDM techniques had a minimal effect on their children's joint attention initiations. The following sections will detail the effect of the parent training on each of the child outcome variables.

Engagement. All five children were more engaged with their mothers during parent training compared to baseline and, with the exception of Rick, maintained this improvement at follow-up. These results are consistent with previous ESDM parent coaching studies, which reported that children showed increases on a measure of attentiveness and social initiations during the intervention (CBRS; Mahoney & Wheeden, 1998). This measure included behaviours such as attending to the adult, cooperating with instructions, initiating play ideas, and sharing enjoyment and enthusiasm (Vismara et al., 2012; Vismara, Colombi, et al., 2009; Vismara & Rogers, 2008). Further, several additional studies which investigated the effectiveness of parent training for young children with ASD have also reported that the majority of children improved on various aspects of engagement including, but not limited to,

increases in child responsiveness to bids for joint attention (e.g. Kasari et al., 2010; Kasari et al. 2014; Rocha, Schriebmen, & Stahmer, 2007), increases in reciprocal social interaction (e.g. Aldred, Green, & Adams, 2004), increases in joint engagement (e.g. Kasari et al., 2014), and increased eye contact and directed positive affect (Vernon et al., 2012).

There were moderate to strong correlations between the increase in each parent's use of the ESDM procedures and the increase in both partial and whole engagement for all of the children. During intervention parents improved on fidelity items related to child motivation and attention, such as: management of attention, sensitivity and responsivity, use of positive affect, motivation, and management of affect and arousal; all of which could encourage a child to be more engaged with their parent (Rogers & Dawson, 2010). The link between higher ESDM fidelity and child engagement is further supported by the fact that Rick and Alex's mothers had the highest baseline use of these techniques and their children also had the highest levels of engagement during baseline.

During the parent training phase, Dean had the greatest improvements in both partial and whole engagement and these improvements were strongly correlated with his mother's increased implementation of the ESDM techniques. During the parent training programme Dean's mother improved the most in the fidelity items related to child attention and motivation, which provides a plausible explanation for Dean's increased engagement with her. Further, during baseline, Dean was considerably more engaged with his father than his mother, and his father was also using more of the ESDM fidelity techniques. This suggests that, prior to the parent training, Dean was already able to engage with, and attend to, an adult for an extended period of time, provided the adult was using techniques to gain and maintain his attention. Thus, his mother's increased use of ESDM fidelity techniques may merely have elicited attentional skills that were already within Dean's repertoire (Johnson & Christensen, 2012).

During parent training Rick and Sean's whole engagement improved more than their partial engagement. For Alex, the difference between the session with the highest number of intervals containing whole engagement between baseline and intervention was also greater than the corresponding difference in partial engagement. This may have been due to ceiling effects for partial engagement (Johnson & Christensen, 2012). On average, Rick, Alex, and Sean were engaged with their mothers for part of the time during about 75% of the 10-s intervals during baseline and therefore there may have been limited room for improvement. In contrast, during baseline Rick, Alex, and Sean were engaged with their parents for the

whole 10-s interval less than a third of the time (on average). This suggests that, while Rick, Alex, and Sean were minimally engaged with their parents for most of the 10-min play sample during baseline, their parents were not consistently maintaining their attention. This also suggests that whole engagement may be a better measure of intervention effects than partial engagement.

Idris' partial and whole engagement increased during parent training despite the fact that his mother did not reach fidelity. This may be because, during parent training, she had the highest fidelity scores on items such as management of affect and arousal, sensitivity and responsivity, and positive affect, and she had the lowest scores for items such as joint activity structure and elaboration, instructional techniques, language, and transition. There was also a strong correlation between her use of the fidelity procedures and Idris' partial engagement. This suggests that the improvements she did show may have meant she was able to better "entertain" Idris, and gain his attention, but she was not yet creating clear learning opportunities, or consistently teaching him new skills (Rogers & Dawson, 2010).

Imitation. Four of the five children showed some improvement in the percentage of intervals containing imitation during parent training. However, for Alex, imitation decreased during the intervention and follow-up phases, and for Idris, the improvement was minimal and also was not maintained at follow-up. In a previous ESDM parent coaching study, Vismara, Colombi et al. (2009) similarly found that although most children showed improvements in imitation, one child did not. However, in the remaining ESDM parent training studies which evaluated this outcome, imitation skills improved for all of the children (Vismara et al., 2012; Vismara & Rogers, 2008). There appears to be limited research evaluating the effect of other parent training programmes on teaching imitation to young children with ASD (Ingersoll & Gergans, 2007). Of the studies that do exist, Wainer and Ingersoll (2015) similarly found that imitation skills did improve for all but one of the children, while the remaining studies found that imitation improved for all children (e.g. Charlop-Christy & Carpenter, 2000; Ingersoll & Gergans, 2007).

Dean showed a large and immediate increase in imitation on the third intervention session. This immediate increase suggests that he was perhaps already able to imitate actions and vocalisations but was not doing so very frequently in baseline (Johnson & Christensen, 2012). It also suggests that his mother's improved use of the ESDM intervention techniques may have resulted in Dean using his already existing imitation skills with her, rather than learning new imitation skills. This may have been because his mother was using many more

techniques to gain his attention and increase his engagement during intervention compared to baseline. Thus, Dean may have imitated his mother more simply because he was attending to her, and found the actions that she was doing to be interesting.

In contrast, for Sean, there was a generally increasing trend for imitation during intervention and the greatest increase coincided with the chapter entitled "Do What I Do", which focused on imitation (Rogers, Dawson, et al., 2012). During the parent coaching phase, his mother also improved her use of teaching techniques such as correct use of ABCs and instructional techniques. This suggests that it may have been Sean's mother's use of clear antecedents, prompting, and reinforcement strategies that resulted in her son's increased use of imitation (Alberto & Troutman, 2009; Catania, 2017; Skinner, 1953)

For Rick, it is not clear whether the improvement in intervals containing imitation during intervention represented a true increase in the skill. First, there was only a weak correlation between his mother's improvements in fidelity and his use of imitation. This suggests that Rick's use of imitation was relatively unaffected by his mother's use of the ESDM procedures. During the post-intervention interview, Rick's mother also stated that Rick had shown minimal improvements in imitation during the intervention. Thus, the increase in imitation during intervention may have been due to factors other than the parent training. One such factor could be variation in the nature of the action to be imitated. Specifically, during the first few sessions of intervention, most instances of imitation involved Rick reaching towards his mother's outstretched hands during a well-practiced sensory social game. Although this behaviour met the criteria for imitation, it is not clear whether he was truly trying to copy his mother or whether he had learned through experience that he should reach towards her hands during this part of the game. Rick and his mother did not play this game during baseline, which could explain the increase in the mean number of intervals containing imitation during intervention.

There are several potential explanations for Alex's decrease in imitation during intervention, despite his mother's competent use of ESDM techniques. First, Alex's percentage of intervals containing imitation in the final intervention session was the same as the final baseline session. This might indicate that there was no change in his imitation ability between baseline and the end of the parent training, rather than a decrease. Second, research suggests that some types of imitation are easier for children with ASD than others (Ingersoll & Gergans, 2007; Rogers & Dawson, 2010). Therefore, the differences in imitation between baseline and intervention could be due to the nature of the action to be imitated. Specifically,

similar to Rick, many of the actions Alex imitated during baseline involved reaching towards his mother's outstretched hands during a familiar sensory social game. During this phase he and his mother also frequently rolled cars back and forth. During intervention, the most common activity was reading books, which did not give Alex an opportunity to engage in either of these actions. Further, at this time his mother wanted to increase his use of pointing so created many opportunities for him to imitate a point. However, each time she needed to prompt him to do so as he was still learning to perform this skill. Further, it is possible that teaching imitation was not a priority for Alex's mother. During the post-intervention interview she stated that her main goal for Alex was to improve his communication and she did not mention his imitation skills. It is possible that her focus on, and success in, improving Alex's use of intentional vocalisations meant that she was creating fewer opportunities for imitation during the intervention period.

During intervention, Idris had the lowest mean percentage of intervals containing imitation. In some respects, one would not expect Idris' imitation to increase because his mother did not learn to implement the ESDM procedures with a high degree of fidelity. In particular, she had not mastered the ESDM fidelity items related to teaching new skills such as use of the ABCs and instructional techniques. Behavioural learning theory suggests that in order to teach a child new skills one should provide a clear antecedent, prompt him or her to perform the desired behaviour (if necessary), and then provide reinforcement (Skinner, 1953). Idris' mother may not have made it clear to him when he was expected to imitate her, and, as she did not consistently provide reinforcement, he was perhaps not provided with incentive to continue to do so.

Functional utterances. This outcome was most applicable to Dean, Rick, and Sean as their ESDM goals included items related to spoken language. During intervention these three children had minimal improvements in the total percentage of intervals containing functional utterances, however, each child directed more utterances towards their mothers. This improvement in engaged utterances was maintained for Dean and Rick 1 month following intervention, but was not for Sean. Several ESDM parent coaching studies have reported that the frequency of spontaneous functional utterances improved for the majority of children (Vismara et al., 2012; Vismara et al., 2016; Vismara, Colombi, et al., 2009; Vismara, McCormick, et al., 2013; Vismara & Rogers, 2008). This measure appears to be similar to engaged utterances. A recent meta-analysis of parent training for young children with ASD found that all nine of the studies which included a measure of expressive language reported

positive outcomes following intervention (Meadan et al., 2009). It is not possible to directly compare these results with those of the current study due to the wide variety in measures, however, these findings seem to be generally more positive than those reported in the current study, due to the minimal improvement in the total percentage of utterances for each child.

Additional analyses revealed that Rick, Dean, and Sean's variety of utterances was higher at follow-up than baseline. Previous studies evaluating training for parents of young children with ASD have also found that children's variety of utterances improved following intervention (Kaiser, Hancock, & Nietfeld, 2000; Seung et al., 2006). However, all three children had minimal increases in MLU during intervention or follow-up. This contrasts with the findings of Kaiser et al. (2000) who trained parents to use enhanced milieu teaching strategies with six preschool children with ASD and found that each of these children had a higher MLU 6 months following intervention. This could be due to the longer follow-up period in the Kaiser et al. (2000) study compared to the current study.

It is likely that the increase in engaged utterances for Dean, Rick, and Sean is related to their parents increased use of ESDM techniques during intervention, particularly those related to gaining and maintaining attention. As previously mentioned, all three parents showed improvements in this area and maintained these improvements 1 month after the intervention. During baseline, Dean and Rick frequently commented on aspects of the task or environment without directing these comments towards their parents. For example, Dean would independently play with his fire truck and say words or phrases such as *fire truck* or *go faster*. Thus, although Dean, Rick, and Sean, were not using many more functional utterances during each session, it is likely that their parents were using ESDM techniques to encourage them use these utterances in the context of a joint interaction.

There are several potential reasons why the total percentage of intervals containing utterances did not improve for any of the children in this study. First, it is possible that this measure was not developmentally appropriate for Rick or Dean as they both had a high percentage of intervals containing utterances and a wide variety of utterances during the baseline phase. It also appears that both of these boys were using more spoken language than any of the participants in the previous ESDM parent training studies that included a measure of spontaneous functional utterances (Vismara et al., 2012; Vismara et al., 2016; Vismara, Colombi, et al., 2009; Vismara, McCormick et al., 2013; Vismara & Rogers, 2008). This may have been because Dean and Rick were older than the majority of participants in these studies. They also had expressive language age equivalencies of 2 or more on the *Vineland-II* 

which suggests that they were already able to use a wide variety of spoken language (Sparrow et al., 2005). Further, Dean and Rick's intervention goals related to increasing the quality and functionality of their spoken language, rather than the quantity of language. For example, one of Dean's expressive goals was to answer yes or no questions and one of Rick's goals was to use pronouns appropriately. Progress towards these goals would not have directly resulted in an improvement in the amount or variety of utterances. In fact, in the post-intervention interview both Rick and Dean's parents stated that the intervention had greatly improved their child's communication. Rick's mother said this was particularly the case for his use of pronouns.

For Sean, the percentage of intervals containing functional utterances only improved minimally during intervention and follow-up, however, the mean variety of utterances more than doubled from baseline to the end of intervention, and this improvement in variety was maintained at follow-up. His expressive goals also related to increasing his variety rather than frequency of utterances. In the post-intervention interview his mother stated that she found the intervention to be very effective in improving his spoken language, as prior to the intervention he would only say a few words such as *Go*. Again, it is possible that the variety of utterances was a more suitable measure of language development for Sean. In future, it may be more appropriate to measure children's progress towards their specific language goals rather than measuring the percentage of intervals containing utterances for all participants.

Intentional vocalisations. Idris and Alex had almost no spoken language and produced very few intentional vocalisations (purposeful vocal sounds that do not contain a functional word or word approximation) prior to parent training. Therefore, it was deemed developmentally appropriate to teach the parents to target intentional vocalisations rather than functional utterances during the 12 week intervention period (Rogers & Dawson, 2010; Rogers, Dawson et al., 2012). Alex's intentional vocalisations increased steadily during intervention and were maintained at follow-up. However, Idris' vocalisations decreased during intervention and returned to baseline levels at follow-up. Few studies have investigated the effect of training programmes for parents of children with ASD on vocalisations rather than spoken language (Meadan et al., 2009). However, Elder, Valcante, Yarandi, White, and Elder (2005) conducted a parent training programme for fathers of young children with ASD and found that the mean frequency of vocalisations improved following intervention. Further, Kashinath, Woods, and Goldstein (2004) targeted intentional gestures instead of spoken language for a minimally verbal child with ASD and found that

these increased during parent training. The results of the current study are similar in that intentional vocalisations improved for the child of the parent who learned to use the ESDM techniques with fidelity (Alex).

It is likely that Alex's increases in intentional vocalisations were due to his mother's increased use of ESDM techniques as there was a strong positive correlation between these two variables. Further, as Alex had low baseline levels of intentional vocalisations and these showed a gradual increasing trend during intervention, this suggests that his mother was teaching him to "use his voice" in order to access preferred items or continue preferred activities. In particular, his mother's use of ESDM techniques such as management of attention and ABC format increased during the intervention. Thus, she may have made it clear to Alex that he needed to vocalise and then provided reinforcement each time he did so.

While Alex's percentage of intervals containing intentional vocalisations increased, the percentage of intervals containing functional utterances did not. This suggests that intentional vocalisations may have been a more appropriate developmental target for Alex than functional utterances. Typically developing children begin to make intentional vocalisations many months before beginning to speak (Vihman et al., 1985). Research also suggests that children with ASD develop speech in a similar way to those without ASD, though are typically delayed in doing so (Tager-Flausberg et al., 1993). It is possible that, in time, Alex's intentional vocalisations may have been shaped into functional speech, although this remains unclear due to the lack of long-term follow-up in this study.

Idris' intentional vocalisations decreased during intervention. As with imitation, one would not expect Idris's vocalisations to increase as his mother did not learn to implement the ESDM techniques with a high degree of fidelity. There are several potential explanations as to why they may have decreased, rather than remaining stable. First, it is possible that his mother's incorrect use of the ESDM procedures may have resulted in a decrease in his use of vocalisations. Some authors caution that incorrect use of an intervention could potentially be more harmful to the child than providing no intervention at all (Koegel et al., 2016). However, this is unlikely as there was only a weak correlation between his vocalisations and his mother's use of the ESDM techniques. The decrease in Idris' use of intentional vocalisations coincided with his family relocating to a new home. It is possible that the relocation itself had a negative effect upon Idris' development as well as producing a decrease in his mother's fidelity. An insistence on sameness and difficulty with transitions are both defining characteristics of ASD (American Psychiatric Association, 2013). Moving

house may have been hugely disruptive to Idris and may have resulted in a temporary decrease in his use of intentional vocalisations as he adjusted to the new environment. This is supported by the fact that his vocalisations returned to baseline levels a couple of months after the move. More research is needed on the impact of stressful events such as relocation on outcomes for children with ASD.

### Generalisation

Rick's father's use of the ESDM techniques improved following parent training, whereas the remaining four family members either showed minimal improvements or decreased in their use of the techniques. To the author's knowledge no other study has examined generalisation of parent fidelity to a second family member. There are several reasons why Rick's father may have shown greater improvements in fidelity than the other family members. First, during the post-intervention interview Rick's mother specifically mentioned sharing information she had learned during the parent training sessions with Rick's father because it was important to her that they were "on the same page". On the other hand, Sean and Dean's mothers did not mention sharing any techniques and Alex's mother said that she did not have time to do so. Therefore, it is possible that Rick's mother was directly teaching her partner to use the techniques, which could have resulted in his improvement in fidelity. Further, Rick's mother had the highest mean fidelity during intervention. Thus, it is possible that she was modelling the correct techniques to Rick's father during her daily interactions with Rick.

Overall, the improvements in child outcomes did not generalise to the second family member. Specifically, Dean, Idris, and Alex's engagement with their mothers improved during intervention but engagement with their fathers did not improve. This suggests that they may have been more engaged with their mothers due to their increased use of ESDM techniques, but that their ability to attend to, and interact with, adults who were not explicitly using these techniques had not increased. Alex also did not use intentional vocalisations more with his father following intervention, despite the large increase in his use of vocalisations with his mother. This lack of generalisation suggests that children's improvement in skills may lack external validity, as they may have learned to use them only with their mothers, rather than any other individuals.

This finding highlights the importance of including fathers in parent training programmes. Research suggests that mothers of children with ASD participate in parent training research more often than fathers (Flippin & Crais, 2011). This may be because

mothers are generally more involved in caring for their children with ASD than fathers (Tehee, Honan, & Hevey, 2009). However, the inclusion of fathers in training could lead to further improvements in outcomes for children with ASD because it could result in more consistency between parents in their use of the techniques (Flippin & Crais, 2011). During the post-intervention interview each of the mothers stated that they would have liked their child's father to have participated in the parent training programme. However, several parents stated that this would have been very difficult due to the fathers' work commitments. More research is needed to find effective ways of including fathers, or parents who work full-time, in training programmes for parents of young children with ASD.

# **Social Validity**

The results of the TARF-R suggest that each of the mothers found the training programme to be socially valid and acceptable (Reimers et al., 1992). Specifically, each of the five parents: (a) found the intervention procedures to be reasonable, (b) stated that they would be willing to continue the intervention, (c) found that there were minimal side effects, and (d) found the intervention to be affordable. This is important as parents who find the goals, procedures, and outcomes of an intervention to be appropriate and meaningful may be more likely to continue to implement the intervention, and may recommend the intervention to others (Foster & Mash, 1999). Parents generally scored the intervention lowest on the disruptiveness domain, which indicates that they did find it to be somewhat disruptive. Further, four of the five parents found the intervention to be effective, but Idris' mother did not. These generally positive results align with the findings of several previous ESDM parent coaching studies. For example, in their 2012 study, Vismara et al. stated that parents found distance training to be "informative and... valuable" (p.8). In 2013, Vismara, McCormick, et al. also found that parents were generally satisfied with the distance coaching intervention in terms of the information provided, the effectiveness of the intervention, and the support provided by the trainer. Vismara et al. (2016) also reported that parents in the ESDM group were more satisfied and confident following training than parents who were receiving treatment-as-usual in the community. Thus, the current findings provide further evidence that parents do find ESDM parent coaching to be an acceptable intervention.

The qualitative post-intervention interview provided several explanations for why parents found the intervention to be acceptable and socially valid. First, parents identified key aspects of the ESDM philosophy that contributed to their positive view of the intervention. This included an appreciation of the play-based nature of the model and also the ability to

provide intervention in the context of the child's daily routines. This finding supports the validity of the theoretical basis for the ESDM, in terms of the emphasis on teaching through play and fun activities, and also including learning opportunities throughout the child's day (Rogers & Dawson, 2010). All of the parents also felt that the intervention led to significant gains for their child in at least one area. Research suggests that individuals are more likely to find an intervention to be acceptable when changes in their child's skills or behaviours are meaningful to them (Wolf, 1978). Further, all of the parents found the parent training procedures including direct instruction, modelling, practice, and feedback to be helpful, although in some cases they would have preferred the trainer to use more of a particular technique (e.g. modelling). Their positive view of these techniques may have influenced their perception of the intervention as a whole.

Idris' mother gave the intervention the lowest acceptability scores and in the post-intervention interview she also stated that the intervention had not worked and suggested the most areas for improvement. Idris's mother was the only parent who did not reach an acceptable level of fidelity during the intervention and Idris also showed the least improvement during intervention. Her perception of the intervention aligns with research that suggests that parents will find an invention to be less acceptable if it does not result in meaningful changes in the child's behaviour (Wolf, 1978).

# **Implications**

The results of this study suggest that home-based ESDM parent training may be equally as effective as clinic-based parent training. This is an important finding given the potential advantages of teaching skills in a child's natural environment, such as increased generalisation of skills and convenience for busy families (Rogers, Estes, et al. 2012; Oosterling et al., 2010). These findings suggest that despite the additional distractions that can occur in a child's home environment including the presence of siblings, and less control over materials (Rogers & Vismara, 2015), most parents still learned to implement the ESDM procedures with a high degree of fidelity. Further, several of the parents who participated in this study stated that they preferred to conduct the sessions at home, either because it was more convenient for them or because they felt that their child was most comfortable at home. This suggests professionals should consider discussing with parents whether they would prefer to receive training in a clinic setting or at home.

Another key difference between this study and the majority of previous ESDM parent coaching research was that the training was implemented by a trainer who did not help to

develop the model of intervention. In addition, although the trainer had several years of experience working with children with ASD, she had been a practicing ESDM therapist for less than a year and had not received formal training in ESDM parent coaching. Thus, as the majority of parents were able to learn the to implement the ESDM procedures, this suggests that the ESDM may be effective in real-world contexts, not just when implemented in clinic settings by highly-trained professionals (Smith et al., 2007). In other words, it suggests that a trainer with experience typical of those within community organisations can also train parents to use ESDM techniques.

Although only five parents participated in the study, they represented four different ethnicities: New Zealand European, New Zealand Māori, Indian, and Cambodian. Further two of the parents had immigrated to New Zealand as adults. This is reflective of the multicultural nature of New Zealand's population (Statistics New Zealand, 2013a). All five of the parents found the intervention to be acceptable, and four of the five parents learned to implement the ESDM procedures with a high degree of fidelity. The participants in this current study represent a wider variety of ethnicities than participated in most previous ESDM parent coaching studies, in which the majority of participants were either white Americans or Latino/Hispanic (Rogers, Estes et al., 2012; Vismara et al., 2012). The results of this study suggest that the ESDM techniques may be perceived as acceptable and valid across several different cultures. However, it is possible that adapting the training to each parent's specific cultural values and parenting beliefs may have further increased both the acceptability and effectiveness of the intervention (Forehand & Kotchick, 2016). Much more research is needed into culturally appropriate methods of coaching parents of children with ASD.

While the teaching procedures used in this study were very similar to those used in the initial ESDM parent coaching studies (i.e. Rogers, Estes, et al., 2012; Vismara et al., 2012; Vismara, Colombi, et al., 2009; Vismara, McCormick, et al., 2013), they are quite different from those used in the most recent study (Vismara et al., 2016). This 2016 study had a much greater emphasis on coaching strategies such as parent reflection, collaborative discussion, and joint planning (Rogers & Vismara, 2015; Hanft, Rush, & Sheldon, 2004). Parents also had a greater degree of control over their child's goals, and the information to be covered in each of the teaching sessions (Vismara et al., 2016). It could be argued that the teaching procedures used in the current study and the majority of the previous ESDM parent coaching studies were aligned with an "expert model" of teaching (Brookman-Frazee, 2004).

This is because procedures like modelling and direct feedback suggest that the trainer is an expert in the content area and is transferring their expertise and knowledge to the parents. On the other hand, procedures such as reflection, discussion, and joint planning indicate that the trainer and parent are working together in an equal partnership to help the child. Research suggests that a collaborative approach to working with parents may lead to better outcomes in terms of parent use of intervention techniques, increased parent confidence, and reduced parent stress, as well as greater child outcomes (Brookman-Frazee, 2004; Steiner et al., 2012). Indeed, the most recent ESDM parent coaching study also reported better improvements in parent fidelity than the previous parent coaching study which also used a group design (Rogers, Estes, et al., 2012). Therefore, although this study had generally positive results and the parents found the intervention procedures to be acceptable, it is possible that collaborative parent training strategies may have resulted in even better outcomes for both the children and their parents. Future research should compare the effectiveness of ESDM parent training using an "expert" teaching approach to ESDM parent coaching using a collaborative approach in terms of both parent perceptions of the intervention and child and parent outcomes.

Several themes emerged from the post-intervention interview that have implications for the implementation of parent training programmes. First, the parents differed in terms of the training procedures that they found to be most helpful. Specifically, several parents found trainer modelling of the techniques to be most helpful, while another two preferred a combination of techniques such as learning about the skill, practicing, and receiving feedback. Research suggests that adults may learn better when the teaching techniques align with their preferred method of learning (Knowles, 1980). Future research should assess whether taking these parental preferences into account will increase the effectiveness of a parent training programme.

Four of the five parents stated that they would have preferred that the parent training sessions lasted for longer than an hour and/or that the programme continued for longer than 12 weeks. This finding was interesting considering that three of these parents had reached an acceptable level of fidelity during at least one intervention session. It is possible that these parents were aware that they were not implementing the ESDM procedures correctly 100% of the time, and wanted to continue the training until their fidelity was at a higher level. This is supported by the fact that Rick's mother was the one parent who did not feel that she needed additional training and she was also the parent with the highest level of fidelity. Further, the

variation in fidelity from week to week suggests that parents were not equally able to implement the techniques in all contexts. Thus, their desire for additional training could have indicated that they did not yet feel confident implementing the ESDM procedures across all of their child's daily activities and routines. Further, Alex's mother stated that she needed more time between sessions in the second half of the parent training programme and that she found it very difficult to teach her son play skills. This suggests that, while she was confident in using the ESDM techniques successfully to teach Alex certain skills, she found it more difficult to use these techniques to target other, potentially more difficult, skills. These findings suggest that parents may benefit from additional training even after they have reached fidelity. Future research should investigate the effectiveness of a longer parent training programme or allowing the parents to choose the length of the training programme and the amount of time between sessions.

In the post-intervention interview Idris' mother stated that, because the training had not worked, she felt that she and Idris would benefit from at least two more 12 week cycles of intervention. This suggestion aligns with a tiered approach to intervention for parents who do not learn to consistently implement techniques during the initial parent training (Ruppert et al., 2016). Adults who do not respond to the initial intervention may respond to additional, more intensive or targeted training. In future, researchers should examine the most effective approaches to modifying and intensifying parent training for those for whom the initial training procedures are less effective.

Three of the parents identified personal characteristics of the trainer that they felt had a positive impact upon both their relationship and the intervention. Specifically, parents identified characteristics such as flexibility, practicality, and being non-judgemental as positive trainer qualities. Research from similar fields also suggests that the stronger the "therapeutic alliance" or professional relationship between a trainer and a client, the more likely it is that the client will respond positively to treatment (Diamond et al., 2006; Martin, Garske, & Davis, 2000). This finding is also consistent with much previous research which suggests than the trainer's personal characteristics and ability to develop positive relationships influence the effectiveness of parent coaching interventions (Dinnebeil, Hale, & Rule, 1996; Hanft et al., 2004). However, it is possible that the parents viewed the intervention more positively simply because they got on well with the trainer (Ogilvie & McCrudden, 2017).

Four of the parents also emphasised the importance of their child's relationship with

the trainer. Specifically, parents commented on their child's desire to play with the trainer and the importance of their child developing relationships with unfamiliar adults. The ability to develop strong relationships with the child with ASD is one of the key elements of ESDM and other relationship-focused interventions (Dawson & Bernier, 2013), and it is interesting to note that it is also an element that is important and valuable to parents. In future, researchers should assess the impact of the child's relationship with the trainer on the outcome of the therapy. One should also consider the possibility that parents perceive therapy to be more acceptable if their child appears to be enjoying it, regardless of whether or not they show additional improvements.

#### Limitations

This study is limited in several ways that Study 1 was not. First there was only one post-intervention data point for both generalisation and follow-up. In single-case research at least three data points are needed to establish a clear trend (Johnson & Christensen, 2012). Further, there was a large degree of variability in both parent and child outcomes in the current study. It is possible that these single data points were not representative of typical parent-child interactions and, therefore, only limited conclusions can be drawn both about the generalisation of child skills to a second parent and the maintenance of both parent and child outcomes over time.

Further, the presence of the trainer during the 10-min play samples may have had an influence on both the parent and the child, meaning that the samples may not have been representative of their interactions when the trainer was not present (Johnson & Christensen, 2012). Specifically, parents may have felt nervous to use some of the strategies in front of the trainer, or may have used strategies that they felt the trainer wanted to see, rather than interacting with the child as they typically would. Further, the presence of the trainer may have distracted the child during some of the play samples. For example, both Rick and Sean would try to engage the trainer in preferred activities such as chase and jumping on the trampoline, which may have made it more difficult for their parents to implement the ESDM techniques. In future researchers could address this problem by asking parents to video baseline and post-intervention themselves without the trainer present.

In previous ESDM parent coaching studies, parents were required to read a chapter in the parent ESDM manual each week (Rogers, Dawson, et al., 2012). However, in the current study, parents were not required to do so. Thus, some parents reported that they had read the parent manual and some did not. Specifically, Dean's mother did not read the manual, Rick,

Idris and Alex's mothers read some of the chapters, and Sean's mother read all of them. It is possible that the parents who read the manual had a greater understanding of the treatment than those who did not and the outcome of the study may have been better if it had it been compulsory to read the parent manual. On the other hand, several parents reported that they did not read the manual because they did not have time to do so. Thus, they may have found the treatment less acceptable or socially valid if they had felt increased pressure to read a chapter of the manual each week.

In this study parents were not required to document the number of hours each week in which they practiced the ESDM techniques with their children. This is consistent with the majority of parent training studies, as a systematic review by Lieberman-Betz (2014) found that only 14% of studies evaluating the effectiveness of parent-mediated communication intervention measured the dosage of parent delivered intervention. Due to this lack of data, it is not clear how many hours of intervention children were actually receiving each week and whether or not it was consistent between families. It is possible that some parents were practicing the techniques for many hours each week, whereas some parents were not, which would have provided valuable insight into the reasons why some parents and children responded better to the intervention than others. Further, it remains unclear how many hours of parent delivered intervention were required to improve child outcomes.

The social validity findings are, by nature, limited in terms of their reliance on parent report (Alberto & Troutman, 2009). Specifically, it is possible that parents might have reported that the intervention was effective even if it were not because of extraneous factors, such as not wanting to disappoint the presumed wishes of the therapist. This may particularly be the case when the family has limited access to services, as some families may be grateful to receive any type of intervention. For example, in an RCT evaluating the effectiveness of EIBI for children with ASD the parents of children in the control group also rated the quality of the intervention as very high, despite the fact that children in the EIBI group had significantly better outcomes (Smith et al., 2000). Chapter 7: General Discussion, will include further general limitations that apply to both Studies 1 and 2.

In summary, the results of this study suggest that home-based parent training based on the ESDM may be effective in teaching some parents to implement ESDM strategies with their young children with ASD and that parents may find this this intervention to be socially valid and acceptable. Further, parent use of these strategies may improve some outcomes for their children with ASD when interacting with their mothers, however, these improvements

may not generalise to a second parent or family member. Chapter 7 will discuss the main findings of Studies 1 and 2, and will relate these findings to the research on methods of early intervention and different early intervention delivery approaches. It will also discuss the overall implications and limitations of the two studies.

#### **CHAPTER 7**

#### GENERAL DISCUSSION

### **Main Findings**

The results of several studies suggest that intensive early intervention programmes can be effective for improving social, communication and more general adaptive behaviour functioning of young children with ASD (Bibby et al., 2002; Flanagan et al, 2012; Granpeesheh et al., 2009; Harris & Handleman, 2000; Perry et al., 2011). However, it is likely that many families may not be able to access or afford these types of intensive early intervention services. The purpose of the two studies conducted for this thesis was to evaluate the effectiveness of two less intensive versions of the ESDM early intervention programme. In Study 1 a therapist delivered ESDM therapy to four young boys with ASD. This consisted of 3 hours of intervention per week for 12 weeks. Study 2 involved training the parents of five different young boys with ASD to use the ESDM techniques. This consisted of one 60-min training session per week for 12 weeks. Both studies occurred in each family's respective home.

The results of Study 1 suggested that, following the 12 weeks of direct ESDM therapy, all four children improved their engagement with the therapist and imitated the therapist more often. Three of the four children also increased their use of functional utterances, whereas the fourth child increased his use of intentional vocalisations. These improvements were maintained at a follow-up completed 1 month after the completion of the 12<sup>th</sup> week of the therapy programme and generalised to some degree to each child's mother.

The results of Study 2 suggested that, following the 12 weeks of parent training, all five parents improved in their ability to implement the ESDM procedures. Specifically, four of these parents achieved an 80% or higher level of fidelity. These improvements were maintained 1 month after the completion of the parent training programme. Following the 12 weeks of parent training, the children were also observed to have shown improvements on at least two of the child-related dependent variables (i.e. imitation, functional utterances/intentional vocalisations, engagement). Most of these improvements were maintained 1 month after the completion of the 12 week programme. Except for one parent (Rick's father), the family members who did not participate in the training programme did not improve in their ability to implement the ESDM procedures with fidelity. Additionally, most child improvements did not appear to generalise from their (trained) mothers to the

second (untrained) family member. Finally, social validity data suggested that parents found the parent training programme to be acceptable and beneficial.

#### **Evidence-Based Practice**

The findings from Studies 1 and 2 contribute to the evidence base for the effectiveness of ESDM as an early intervention for young children with ASD. Specifically, the results of Study 1 suggested that a low-intensity, therapist delivered version of the ESDM intervention was associated with several positive outcomes for the four participating children. These findings are consistent with two previous studies that evaluated the effects of low-intensity, therapist delivered versions of ESDM (Colombi et al., 2016; Devescovi et al., 2016). Further, Study 1, with its multiple-probe design, could be seen as providing a high degree of internal validity and thus offering additional evidence for a positive intervention effect. However, the Devescovi et al. (2016) study provides only suggestive evidence due to the absence of a control group. According to Reichow's evaluative criteria (2011; Reichow et al., 2008), "Promising EBPs" must be supported by at least three single-case studies of a least adequate research rigor/quality or at least two group experimental designs of at least adequate research needs to be conducted to determine whether or not low-intensity one-on-one ESDM therapy is a promising EBP.

The results of Study 2 suggested that the 12 week ESDM parent training programme was effective in teaching most of the participating parents to implement the ESDM procedures with a high degree of fidelity. Their implementation of the ESDM procedures, in turn, was associated with several positive outcomes for the children. In respect of the parent improvements in their implementation of ESDM techniques, the results of Study 2 are consistent with five of the six previous ESDM parent-training studies (Vismara et al., 2012; Vismara et al., 2016; Vismara, Colombi et al., 2009; Vismara, McCormick et al., 2013; Vismara & Rogers, 2008). In the remaining study, Rogers, Estes et al. (2016) reported less impressive outcomes, in that only 45% of parents in the ESDM parent training group reached fidelity. With respect to the child dependent variables, the results of Study 2 are better than those reported by Rogers et al. (2012) and Vismara et al. (2016), as in both of these studies the children in the ESDM parent-training group did not show greater improvements on any of the dependent variables than the children in the TAU group. The results for engagement in Study 2 are consistent with several previous ESDM parent-training studies, which also found that all children improved on a measure of engagement during intervention (Vismara et al.,

2012; Vismara, Colombi, et al., 2009; Vismara & Rogers, 2008). However, in Study 2, the children had less impressive outcomes for imitation than in several of the other previous ESDM parent training studies that reported improvements in imitation abilities for all participating children (Vismara et al., 2012; Vismara & Rogers, 2008). The children in Study 2, also had less impressive gains in functional utterances compared to five of the six previous studies (Vismara et al., 2012; Vismara et al., 2016; Vismara, Colombi, et al., 2009; Vismara, McCormick, et al., 2013; Vismara & Rogers, 2008). Overall, outcomes for the children who participated in Study 2 could be seen as mixed. Taken as a whole, this new data from Study 2, when placed in the context of the existing ESDM parent-training literature, suggests that this approach cannot yet be classified as promising or as an evidence-based practice (Reichow, 2011; Reichow et al., 2008). Consequently, there would seem to be a need for more research to determine if there might be a more effective method of training parents in the use of ESDM techniques. Future research could also focus on exploring whether there might be certain parent and/or child variables that might influence the efficacy of parent delivered ESDM procedures.

In terms of original contributions made in this thesis, Studies 1 and 2 are among the few studies investigating the effectiveness of the ESDM that have been conducted by a researcher who was independent of the developers of the ESDM. Specifically, it appears that, of the 20 studies published on the ESDM, only four were conducted by researchers who were independent of the team of researchers who developed the ESDM programme (i.e. Colombi et al., 2016; Eapen et al., 2013; Ogilvie & McCrudden, 2017; Vinen et al., 2017). In order for a model to be deemed to be an established EBP, it must be shown to be effective by multiple research teams in multiple locations (Reichow, 2011; Reichow et al., 2008). Further, once research suggests that models are effective when implemented in clinic settings, by highly trained clinicians (efficacy research), one must also evaluate whether the intervention is effective in community settings, when implemented by community practitioners who may not be as highly trained and may be less closely monitored (effectiveness research; Smith et al., 2010). Study 1 could be considered to be example of research that lies mid-way along the efficacy-effectiveness continuum in that the intervention was conducted in children's homes, but was implemented by a trained therapist. The fact that Study 1 found generally promising results adds to the evidence base supporting the effectiveness of a home-based, low-intensity and therapist delivered version of the ESDM (Smith et al., 2010). Study 2, in contrast, could be seen as falling more towards the effectiveness end of the efficacy-effectiveness research

continuum in that the study was conducted in the children's homes and implemented by the parents. Study 2 could also be seen as more of an effectiveness study because it appears to be the first ESDM parent coaching study conducted by a researcher who was not affiliated with the development of the ESDM programme. The positive results of Study 2 in terms of parent fidelity and some of the child outcomes could therefore be seen as providing some tentative support for the effectiveness of the ESDM (Smith et al., 2010).

# **Methods of Early Intervention**

# Naturalistic Developmental Behavioural Intervention Techniques

As discussed in Chapter 2, the ESDM has been conceptualised as a naturalistic developmental behavioural intervention (NDBI), which is one of the four main methods of early intervention for young children with ASD as described by Schreibman et al. (2015). The results of the present two studies provide additional evidence in support of the use of NDBIs for young children with ASD. There are several possible reasons why NDBIs in general, and the versions of the ESDM evaluated in Study 1 and Study 2 of the present thesis appear to be generally effective. First, it is possible that the increases in imitation, engagement, and/or functional utterances/intentional vocalisations shown by each of the children in Studies 1 and 2 were at least partially attributable to the therapist's/parents' use of the specific behavioural principles and behavioural teaching tactics (Alberto & Troutman, 2009; Catania, 2017; Skinner, 1953) that have been incorporated into the ESDM. Specifically, research suggests that behavioural principles and tactics such as (a) providing clear discriminative stimuli; (b) use of response prompting, response shaping, and behavioural chaining; and (c) skilful provision of reinforcement are key elements to successful interventions aimed at promoting social and communication skills and more general adaptive behaviours (National Autism Centre, 2015).

Further, it is likely that the techniques that the trainer used in Study 2 to teach the parents how to implement the ESDM procedures—techniques that were also based on a number of behavioural principles, were effective in helping the parents to learn how to implement the ESDM techniques with a high degree of fidelity. These techniques (e.g., modelling, practice with feedback, and praise) have been consistently shown to be effective in teaching parents how to implement early intervention procedures with their children with ASD (McConachie & Diggle, 2007; Meadan et al., 2009; Ruppert et al., 2016).

It is also possible that the use of naturalistic teaching techniques may have contributed to the generally positive child and parent outcomes obtained in Study 1 and Study 2.

Specifically, techniques drawn from PRT, an established EBP (National Autism Centre, 2015), such as providing children with choices, reinforcing attempts, varying tasks and interspersing maintenance and acquisition tasks, may have increased the children's motivation to participate in the teaching activities (Koegel et al., 2016). This increased motivation may have, in turn, led to an increase in child engagement, which could also have contributed to children's learning/increases of functional utterances and improved imitation abilities. Further, teaching the child within his or her natural environment could have promoted the generalisation of the improvements from intervention (Stokes & Baer, 1977).

There is limited amount of high-quality research on the effectiveness of developmental/relationship-focused intervention techniques when used in isolation with young children with ASD (Eikeseth, 2008; Reichow, 2011; Reichow et al., 2008). Therefore it is not clear if the use of these relationship-building procedures contributed to the positive child outcomes obtained in Studies 1 and 2. It is possible that the use of developmentally appropriate teaching targets meant that the goals were achievable for each child and this might have thus increased the likelihood of success (Rogers & Dawson, 2010). It is also possible that use of relationship-focused techniques, such as the therapist/parent giving positive affect and using sensory social games, may have increased the children's interest/motivation and thus increased their engagement and learning (Dawson, 2008; Kuhl, 2007).

The ESDM and other NDBIs are, by definition, comprised of several different intervention techniques, which are intended to be used together in a package. When these techniques are used together, as was the case in Studies 1 and 2, it is not possible to determine which components were responsible for the effectiveness of the intervention nor is it possible to determine the relative contribution of each technique within the overall package. Logically, it could be that: (a) all of the ESDM intervention techniques were equally necessary and contributed equally to achieving a positive intervention effect for all children, (b) only some of the ESDM intervention techniques were necessary and contributed to a positive intervention effect for all children, or (c) different ESDM techniques contributed to varying degrees to the intervention effect and the precise contribution of the different techniques varied across children. For example, due to the limited evidence for the effectiveness of developmental/relationship-focussed intervention, it is possible that these techniques do not generally lead to an increase in skills or engagement for young children with ASD. If this were the case, it would seem unnecessary for therapists and parents to

continue to implement these techniques as part of the ESDM intervention package. On the other hand, it is possible that developmental/relationship focused techniques do in fact increase the efficacy of ESDM intervention. For this reason it is essential that researchers directly investigate the effective components of ESDM intervention. This could include comparing the effectiveness of: (a) the naturalistic behavioural intervention techniques with the developmental/relationship focused techniques, (b) object play with sensory social play, and (c) adult-led activities with activities that involve many opportunities for child choice.

# **Comprehensive Intervention**

The ESDM can be described as a comprehensive intervention as it targets numerous child development areas (Odom et al., 2010; Rogers & Vismara, 2008). Comprehensive interventions are generally implemented intensively (e.g., 20 or more hours per week for 10 or more months; Odom et al., 2010). In Study 1, however, the therapist only implemented the intervention for 3 hrs per week for 12 weeks. It is possible that comprehensive interventions may be less effective than targeted interventions when implemented at a low-intensity as increasing the number of goals logically decreases the amount of time that can be used to target each goal. However, in Study 1, all children showed improvement on each of the dependent variables, which spanned three developmental domains (imitation, expressive language, and engagement). This suggests that it may be possible to target more than one developmental domain within a low-intensity intervention for young children with ASD. However, it is not clear whether the children in Study 1 would have shown greater increases in each of these developmental domains had these domains had been the sole target of the intervention and/or if the "dosage" of the intervention was greater (e.g., 10 hrs per week rather than 3 hrs per week). More research is needed into the effectiveness of comprehensive interventions versus interventions targeting a more narrow range of skills or developmental areas. Future research could also examine the relative efficacy of differing intervention doses.

In Study 2, parents were not required to log the number of hours that they used the intervention techniques with their children. Therefore, it is not possible to determine how intensively they delivered the intervention. However, evidence from post-intervention interviews with the parents suggested that some parents might not have used the procedures often due to not remembering to do them and/or not having time to implement the intervention techniques and teach all of the goals. This might help to explain why none of the children in Study 2 showed mastery of all of their goals following their 12 weeks of intervention. It is also possible that some parents may have prioritised some developmental

areas over others. Specifically, research suggests that communication development is one of the highest priorities for parents (Pituch et al., 2011; Rodger et al., 2004; Whittaker, 2007). Thus, some parents may have spent more time targeting expressive language than, for example, imitation. If so, this could contraindicate the use of a comprehensive intervention in favour of an intervention that initially targets only a few high priority areas. More research is needed to determine whether training parents to implement comprehensive interventions is as effective as training parents to implement a more narrow or targeted intervention. Of course, it might also be a useful future direction for researchers to develop new and more effective ways of supporting parents in using implementation of comprehensive interventions given that ASD and other types of developmental disorders and delays are generally associated with a fairly wide range of behavioural deficits and excesses (American Psychiatric Association, 2013).

# **Early Intervention Delivery Methods**

Studies 1 and 2 involved different ESDM delivery methods. Specifically, in Study 1, a PhD student who was trained in the ESDM (Waddington) delivered the intervention directly to four young children with ASD. In Study 2, this same PhD student trained five parents to deliver the ESDM intervention to their children with ASD. The results of Study 1 suggest that the therapist delivered intervention had a positive effect on all of the children and all of the child-related dependent variables, except for Alan's functional utterances, for which the intervention effect was relatively small. In Study 2, in contrast, while the parent-implemented intervention had a strong positive effect on imitation, engaged functional utterances, and engagement for Dean; and intentional vocalisations for Alex, the effects were less for the remaining children and for the other child-related dependent variables. These results suggest that the therapist delivered intervention in Study 1 may have been more effective for improving child outcomes than the parent training intervention in Study 2. However, it should be noted that the design of the studies in this thesis does not allow for direct comparison between the two delivery approaches.

In some respects it would seem counterintuitive that the therapist delivered intervention was more effective than the parent delivered intervention because the parents were theoretically able to implement the ESDM procedures throughout the day and thus for more than 3 hrs per week. However, several studies have also found that, while parent training can be effective in improving parent implementation of the intervention techniques, this approach does not always lead to a corresponding increase in child outcomes (Oono et

al., 2013). Relatedly, a meta-analysis on the effectiveness of language intervention for young children with a variety of disabilities found that there were minimal differences in the effectiveness of parent delivered intervention compared with therapist delivered intervention for improving child language outcomes (Roberts & Kaiser, 2011). The total amount of therapist delivered language intervention in these studies varied from 9 to 27 hrs. This suggests that although parents were theoretically able to deliver the language intervention for many more hours per week than the therapists could, this did not generally lead to greater child language improvements. There are several possible reasons why the therapist delivered intervention in Study 1 appears to have been more effective than the parent-training programme in Study 2, at least in terms of improving child outcomes.

First, in Study 1, the therapist had received considerable training and practice with the ESDM model of intervention and fidelity checks showed that she consistently implemented the procedures with a high degree of fidelity (80% or above) from the first week of intervention. In contrast, the majority of the parents in Study 2 only reached an acceptable degree of fidelity after receiving at least four weeks of parent training. This means that the children in Study 1 were probably receiving a higher quality/fidelity of intervention from the very beginning of the 12 week intervention session, whereas the children in Study 2 were not receiving this high level of quality/fidelity of intervention until the end of the 1<sup>st</sup> month of the 12 week programme and, even then, there was considerable variation in the extent to which individual parents' correctly implemented the ESDM techniques across the course of the 12 week programme. This may explain the more immediate increase in child-related dependent variables in Study 1 as compared to Study 2. It also suggests that in parent training studies it might be more reasonable to measure child outcomes only after the parents have attained a high level of fidelity and used the procedures consistently at this high level of fidelity for several weeks. Inclusion of a longer follow-up period would also strengthen future research studies (Kaiser & Roberts, 2013).

Second, it is possible that the therapist was more effective at working directly with the children than she was at training the parents to use the ESDM techniques. Providing direct therapy involves a different set of skills than training/coaching parents (Rogers & Vismara, 2015). Therefore, although parents did show an improvement in their implementation of the ESDM techniques during the intervention, it is possible that these improvements would have been greater if the therapist/trainer had been more experienced in training parents or if she had used other training/coaching techniques such as reflection, discussion, and joint planning

(Brookman-Frazee, 2004). Greater and more consistent improvements in parent implementation of the ESDM techniques may have led to greater corresponding improvements in the child-related dependent variables.

Third, it is also possible that the therapist delivered intervention in Study 1 resulted in the children receiving more concentrated and intensive therapy than that received by the children in Study 2, at least in terms of the quality/fidelity of implementation and frequency/density of learning opportunities. This might have been due to parents generally having to deal with many daily distractions, which were generally not present or which were more easily controlled during direct therapy sessions. Such distractions included the need for parents, but not the therapist, to complete household tasks (e.g. cooking, cleaning, laundry etc.), perform job tasks (e.g., reply to work-related emails) and attend to other children (Rogers & Vismara, 2015). It is therefore possible that an intervention that involves more concentrated therapy, yet is lower in dosage in terms of total hours, might be more effective than a less concentrated, higher-dosage intervention. This hypothesis could be tested in future research.

Fourth, the ESDM approach to the training of therapists places considerable emphasis on directly targeting child goals and collecting data on these goals. The ESDM approach to parent training, in contrast, does not place as much emphasis on these aspects of the programme (Rogers, Vismara et al., 2012; Rogers & Dawson, 2010). Specifically, ESDM therapists generally choose at least two developmentally appropriate goals for each child across each of the developmental domains and then record the child's progress on steps towards these goals every 15 min during each intervention session (Rogers & Dawson, 2010). In contrast, during ESDM parent training, especially during the initial few weeks, the focus is on teaching parents to implement certain ESDM techniques with their children (e.g. use of positive affect, joint activity routines) rather than getting the parents to target specific child goals. These differences could mean that a therapist-directed intervention is perhaps more likely to be responsive to the child's progress on specific outcome measures, whereas a parent might be more focused on his or her own reactions during joint activity routines with the child.

Last, it is possible that some child skills were more difficult for parents to target than others. Specifically, in Study 2, engagement was the only child-related dependent variable for which all five children had some improvement. Thus, it may be harder for parents to teach their children to use utterances and to imitate than it is for them to increase their engagement.

It is possible professionals are better equipped to teach these skills or that parents need more training in order to successfully increase their children's engagement and utterances. More research is needed in this area.

The child outcomes in the parent training intervention (Study 2) should also be compared to the generalisation of child outcomes to their mothers in the therapist delivered intervention (Study 1). This is because skills that a child only uses with his or her therapist may have a limited impact on his or her overall functioning in daily life (Rogers & Dawson, 2010). The results of Study 1 suggest that, aside from Charlie, all of the children improved on all of the child-related dependent variables with their parents following the intervention compared with baseline. Therefore, the children in Study 1 generally had greater improvements with their mothers than the children in Study 2, despite the fact that the parents of the children in Study 1 had not received any direct parent training. Instead, they had only observed how the therapist ran the ESDM therapy sessions. However, the child improvements with their mothers in Study 1 were generally not as great as the child improvements with the therapist. Further, three of the four parents in Study 1 did not attain an acceptable level of fidelity (i.e., 80% or better) with respect to implementing the ESDM procedures. Of course, it is possible that parents in Study 1 did learn to successfully implement some critical elements of the ESDM by simply watching the therapist-directed sessions. The comparison of the generalisation of child outcomes from Study 1 to child outcomes with their mothers in Study 2 suggest that a useful direction for future research would be to evaluate a programme that combined therapist delivered intervention with a parent-training programme.

Overall, the results of Studies 1 and 2 suggest that researchers should conduct a more rigorous comparison of ESDM delivery methods in order to determine the relative efficacy of a parent delivered versus a therapist delivered versus a combined parent delivered and therapist delivered ESDM intervention. If a combination of parent delivered and therapist delivered intervention proved to be relatively more efficacious, then researchers should also investigate the most effective relative dosage of each delivery method (e.g., low versus medium versus high dosage).

### **Implications**

The findings of the two studies reported in this thesis would seem to have some potentially important implications for the delivery of ESDM intervention when professional input is limited, as often seems to be the case in many areas of New Zealand. Specifically, the

results of Studies 1 and 2 suggest that a low-intensity therapist delivered version of the ESDM was associated with consistently positive child outcomes, whereas the parent delivered version was associated with more mixed child outcomes. This could indicate that the present application of a parent delivered version of the ESDM might not be as consistently effective for improving child outcomes. This also suggests the potential value of using a combination of parent delivered therapy and therapist delivered therapy.

All of the participants in both of these studies were under the age of 5 at the start of the baseline phase. The consistently positive outcomes reported in Study 1 and the generally positive outcomes reported in Study 2 appear to suggest that early intervention may be an effective approach for young children with ASD. Due to the design of the study and the sample size, it is not possible to determine whether older children (over 5 years) would not have responded equally well, or better, to these same interventions. However, it is possible that the children's improvements were due in part to increased brain "plasticity" compared to older children (Holland et al., 2014). This could have meant that they were able to learn new skills more rapidly than older children. Further, the "social motivation hypothesis" suggests that early intervention approaches, such as the ESDM, that focus on increasing children's attention to faces and developing positive social interactions may alter the brain and increase the reward value of social stimuli (Fava & Strauss, 2014; Rogers et al., 2014). Specifically, in Studies 1 and 2 the children's increased engagement could be attributed in part to changes within the social reward areas of the brain. More research is needed into the effect of early intervention on the brain development of children with ASD.

Regardless of whether or not younger children are more responsive to ESDM intervention, there are several potential advantages of providing intervention early. First, exposure to an increased number of learning opportunities at a young age may prevent children with ASD from falling further and further behind their typically developing peers across all areas of development (Lang et al., 2016; Rogers & Dawson, 2010). Some of the skills that the children learned in Studies 1 and 2 may mean that they will be better able to access learning opportunities within their natural environments. Specifically, imitation skills improved for all of the children in Study 1 and three of the children in Study 2, and engagement improved for all of the children in both of the studies. Depending on their generalisation of skills, this ability to attend to others, and to copy them, may mean that these children will be better able to learn new skills through modelling other adults and peers. Again, more research is needed to determine the degree to which children generalise their

engagement and imitation skills from one environment/person to another.

Some research suggests that infants (i.e. children under the age of 2) with ASD may be even more responsive to early intervention than children with ASD who are between the ages of 3 and 5 (Bradshaw et al., 2016). However, in the current research, this was not the case, as Idris was the only participant who was under the age of 2 (1 year 11 months old) at the start of the study and he had limited improvements during the intervention. The reasons for this minimal improvement compared to the other children are discussed in Chapter 6, but include the fact that his mother was the only parent who did not learn to implement the ESDM techniques with a high degree of fidelity. In contrast, Charlie was the youngest participant in Study 1 (2 years 3 months old at the start of the study) and he had particularly large improvements in functional utterances compared to the other children in both Studies 1 and 2. These findings reiterate that age is only one of many factors which may influence a child's responsiveness to intervention and more research is needed into moderators of treatment effectiveness (Fava & Strauss, 2014; see section on Moderators of Treatment Effect in Chapter 2).

Both Studies 1 and 2 included participants who were between the ages of 4 and 5 years at the start of the intervention (Alan, Study 1; Sean and Rick, Study 2). In contrast, all of the previous low-intensity ESDM therapy and ESDM parent-training research has only included children with ASD who were under the age of 4 at the start of intervention (Colombi et al., 2016; Devescovi et al., 2016; Rogers, Estes et al., 2012; Vismara et al., 2016; Vismara, Colombi et al., 2009; Vismara, McCormick et al., 2013; Vismara & Rogers, 2008). This may be because the ESDM curriculum only includes developmental skills that are typically learned in the first 4 years of life (Rogers & Dawson, 2010). However, some older children with ASD may still be learning skills that children without ASD or developmental disabilities learn typically before the age of 4 (Kennedy & Courchesne, 2008). Indeed, all three of the children over the age of 4 in the current thesis had increases in imitation, functional utterances/intentional vocalisations, and/or engagement during the intervention. This suggests that the current applications of the ESDM intervention may be appropriate for children with ASD who are over the age of 4. This is particularly important as research suggests that estimates of the average age of diagnosis in the United States range from 48 to 58 months (Centres for Disease Control, 2014; Shattuck et al., 2009) and around 49 months in Australia (Bent, Dissanayake, & Barbaro, 2015).

The children and parents who participated in Studies 1 and 2 varied in terms of their prior exposure to intervention. Specifically, in Study 1, Alan and his parents had received more intervention and parent training, respectively, than the other children and parents in the study. Further, in Study 2, Rick and Alex's mothers had participated in parent training programmes that were more similar to ESDM parent training than the other parents in the study. Given that all parents improved in their use of the techniques and all children improved on at least two outcome measures, this would suggest that the intervention was effective regardless of individual's prior exposure to intervention. However, it is also possible that prior exposure to intervention does moderate individuals' response to intervention. Further, it may affect parents' perceptions of the intervention. For example, Alan's parents may have perceived the ESDM intervention to be more socially acceptable because of their reported negative perceptions of his ABA therapy. More research is needed to determine the effect of prior exposure to intervention on children and parents' response to ESDM therapy and parent training.

The results of Studies 1 and 2 suggest that the children did not fully generalise their use of skills that they learned with the therapist (Study 1) or with their mother (Study 2) to a second person. This may be because a "train and hope" approach towards generalisation was used (Stokes & Baer, 1977). In hindsight, it is perhaps not surprising that generalisation was limited in that children were only taught to perform skills with one person (i.e., either with the therapist in Study 1 or with the mother in Study 2). It has been recommended that intervention should occur in multiple settings and with multiple people as a way of programming for generalisation (Stokes & Baer, 1977). However, this was not possible in Study 1 due to limited resources. The present approach to intervention might have been improved by actively programming for generalisation, such as by having multiple people provide the intervention in multiple environments (e.g. home, school, community).

### Limitations

There were several limitations that were common to Studies 1 and 2, and indicate that the findings of these studies should be interpreted with caution. First, the generalisability of the findings from this study is limited due to the small number and limited diversity of participants. Specifically, only nine children participated in these two studies and all of these children were male, which is consistent with research which suggests that males are 4.5 (CDC, 2014) times more likely to be diagnosed with ASD than females. However, it is not clear how effective this treatment would have been for young girls with ASD. Further,

each of the parents in these studies, apart from Chris' mother, was married or in a long-term relationship, which is not representative of many New Zealand families (Statistics New Zealand, 2013b). In Study 1, three of the children were New Zealand European and the other child was Indian. Therefore, it is not clear how effective this intervention would have been for children of other cultural backgrounds or ethnicities, such as with Māori or Pacifica children. Thus, the findings of this study need to be replicated with a larger, and more diverse sample of children with, or at risk for, ASD. Further, Studies 1 and 2 were only implemented by one therapist/trainer (Waddington), therefore, it is not clear whether other therapists/trainers would obtain similar results with similar types of children. Future research could examine the influence of therapist training and experience on outcomes from early interventions such as the ESDM.

Jacobson, Roberts, Berns, and McClinchy (1999) argue that for any improvement to be considered clinically significant, an individual must develop to the point where their behaviour approaches "normal functioning". In Studies 1 and 2, the changes in the child dependent variables were not measured in relation to the average, typical or normative levels of performance, but rather in terms of gains from the child's performance observed during the baseline phase. These gains cannot be compared to any existing norms as it is not clear, for example, how often a typically developing young child would imitate or be engaged with an adult within a 10-min activity period. Therefore, it is not evident whether the children's improvements seen in Study 1 and Study 2 reached what would be viewed as the normal range of functioning. The lack of such norms limits the conclusions which can be drawn about the clinical significance of the behavioural changes obtained in Study 1 and Study 2 (Jacobson et al., 1999).

All of the dependent variables used in this study were proximal rather than distal predictors of intervention success (Rogers, Estes, et al., 2012). Proximal outcomes are behaviours that were directly targeted by the intervention such as utterances, engagement, and imitation, whereas distal outcomes are more general measures of a variety of behaviours such as intelligence, adaptive behaviour, and autism symptomology. Rogers, Estes et al. (2012) stated that in order to show improvement on distal intervention outcomes, a child must show "many behavioral changes, integrated into overall performance, generalized across environments, and performed in a structured situation..." (p.1062). Therefore, as distal measures were not used in Studies 1 and 2, it is not clear whether this level of behaviour

change occurred for the children in these studies. This further limits the conclusions which can be drawn about the clinical significance of the findings.

Further, it is possible that some of the outcome measures were not developmentally appropriate for some of the children. Specifically, the percentage of intervals containing functional utterances may not have been the most developmentally appropriate measure for Chris and Jeevan in Study 1 or Dean and Rick in Study 2. This is because these four participants already had relatively high levels of [unengaged] functional utterances during baseline, and they also had intervention goals related to the quality (e.g. improvements in grammar, learning different communicative functions) rather than the quantity of speech. This may have been because these four children were older than the majority of children who participated in previous ESDM parent training research which also used similar measures of expressive language (Vismara et al., 2012; Vismara, Colombi, et al., 2009; Vismara, McCormick et al., 2013; Vismara & Rogers, 2008). Thus, in order to more accurately measure each child's response to the intervention, it may have been more appropriate to measure their progress towards specific [individualised] language goals, rather than adopting a more generic measure related to the general increase in functional utterances. Indeed, several other early intervention studies for children with ASD have used an individualised approach to measuring expressive language (e.g. Kaiser et al., 2002; Kashinath et al., 2006) On the other hand, certain well-validated generic measures, such as MLU or variation in utterances, might be considered more appropriate for some children, such as children who enter intervention with more advanced expressive language (Kaiser et al., 2000; Kasari, Kaiser, et al., 2014).

The decision was made to adopt an interval recording procedure to measure child engagement because engagement, as defined in this thesis, could have potentially occur for extended time periods. In light of this and based on guidelines offered by Alberto and Troutman (2009), it was deemed to be more appropriate to use interval recording as this procedure is known to provide a good estimate of the duration of engagement, rather than frequency of occurrence. However, functional utterances and imitation generally occur for short periods of time, and therefore, it could be more appropriate to adopt a frequency measure for these dependent variables, rather than using a partial interval recording method. Although data were collected on the frequency of utterances, this was not the case for imitation. Therefore, it is not possible to report the number of times (frequency) that each

child imitated the therapist/parent during each session. That is because each interval may have contained one or more instances of imitation (Alberto & Troutman, 2009).

Both partial-interval and whole-interval recording were used to measure child engagement. It appears that partial-interval recording might have led to an overestimation of engagement, while whole-interval recording might have led to an underestimation (Alberto & Troutman, 2009). For example, if the child was engaged with their parent for 1 s of a 10-s interval, then this would be coded as an instance or occurrence of partial engagement. Using this method, the total percentage of partial engagement during a session could be very high even if the child were only briefly engaged with their parent during a majority of 10-s intervals of that session. On the other hand, if a child was engaged with their parent for 8 or 9-s during several 10-s interval, then this would not be coded as whole engagement. Therefore, the total percentage of whole engagement for each session could be quite low, even if the child was engaged with their parent for most of the time. As most of the children (all except Idris and Charlie) had high partial engagement during the majority of intervention sessions, this led to ceiling effects for this variable. In other words, the percentage of intervals containing partial engagement was frequently between 90 and 100%, which indicated limited "room for improvement". This suggests that whole interval recording may be a more appropriate measure of child engagement. This was also the case for functional utterances for Chris, Jeevan, Dean, and Rick, as they spoke many times during each session.

Research suggests that the Non-overlap of All Pairs (NAP) metric is a valid measure of effect size for interventions that are evaluated using single case experimental designs (Parker & Vannest, 2009). The NAP can discriminate amongst different sets of data and correlates well with other statistical measures of variance. However, any form of statistical analysis in single case research is limited because there is a lack of independence within the data points (Parker & Vannest, 2009). Further, this type of analysis provides an indication of whether or not data points overlap between the baseline and intervention phases, but it does not provide an indication of the size of the increase or decrease. For example, if Child A and B both had between one and three functional utterances per session in baseline and this increased to between four and five functional utterances per session in intervention for Child A, and to between six and ten functional utterances in intervention for Child B, then the NAP statistic for both Child A and Child B would be 1.0. Research also suggests effect size estimates using partial and whole interval recording may have questionable validity (Ledford, Ayres, Lane, & Lam, 2015). This could occur if the target behaviour did not have the same

average duration during baseline versus intervention. In such cases it could be that the method of recording may be more accurate in one phase compared to the other, which could possibly result in a less accurate calculation of effect size.

There are also limitations to the multiple probe across participants design that was used in Studies 1 and 2. Each participants' baseline phase involved probes that did not occur every week. This was because it was deemed to be unreasonable to ask families to participate in baseline sessions for many weeks before the intervention was implemented. For example, had Study 2 used a multiple baseline, rather than a multiple probe design, Rick's family would have participated in 11 weekly baseline sessions rather than six probes. However, the intermittent nature of data collection in a multiple probe design means that it is generally considered to be less rigorous than a multiple baseline design in which there is continuous data collection during baseline (Kennedy, 2005). Specifically, multiple probe designs are reported to be less sensitive to rapid increases or decreases in behaviour and are less able to detect cyclical changes in behaviour (Kennedy, 2005).

The ESDM parent/therapist fidelity checklist that was used in Studies 1 and 2 is an adapted, simplified version of the rating system used in previous ESDM research (Rogers & Dawson, 2010). A simplified version was used because the observer who conducted the PI and IOA checks in Studies 1 and 2 was less versed in the ESDM programme and thus needed a simplified checklist in order to be able to accurately record fidelity. The observer had completed the ESDM introductory workshop and had read the ESDM therapist manual, but she was not a certified ESDM therapist. With the simplified checklist, a high level IOA on the rating of parent fidelity of implementation was obtained. This high level of IOA suggests that the use of the simplified version of the fidelity rating system resulted in a reliable measure of this dependent variable. However, because this simplified version was used for Study 1 and Study 2, it is not possible to directly compare the fidelity results to those reported in other ESDM studies that used a different fidelity checklist. Thus, this fidelity data should be interpreted with caution. It is possible that a score of 80% fidelity on the simplified checklist may have represented lower or higher quality of ESDM implementation than a score of 80% fidelity on the original ESDM fidelity checklist. Also, due to the simplified nature of the fidelity checklist used in Study 1 and Study 2, it is possible that important elements of ESDM intervention were not measured. Future research could compare the original ESDM fidelity rating system with the current simplified version.

The results of this study may also have been influenced by researcher bias (Johnson & Christensen, 2012). Specifically, the observers who analysed the videos were not blind to the study phase or the aims of the study. Therefore, the results may have been influenced by what the observers expected to see, rather than what they actually observed. For example, if they expected the child to speak more during the intervention phase than the baseline phase, they may have been more attentive to functional utterances when coding the intervention phase. This would, therefore, artificially inflate the number of functional utterances in intervention compared to baseline. However, the IOA checks indicated that there was high agreement between observers, which suggests that the data collection was accurate. Nevertheless, in future, the individuals who code the data should be blind to the study phase and, when possible, the aims of the study.

The follow-up phase in Studies 1 and 2 only occurred 1 month after the intervention. Therefore, it is not clear whether the children or parents maintained any gains from the intervention in the longer term. It is particularly important to measure long-term follow-up (e.g. 6 months to several years after intervention) as some research suggests that children with ASD do not always maintain their improvements following early intervention, including EIBI (Starr, Popovic, & McCall, 2016). Therefore, it is essential that more research is conducted into the longer-term maintenance of child outcomes following low-intensity therapist delivered intervention and parent maintenance of ESDM fidelity following parent training.

#### Conclusion

There is evidence to suggest that early intervention may be particularly effective for young children with ASD. The ESDM is one promising early intervention approach for young children with ASD which combines aspects of behavioural, naturalistic and developmental/relationship focused intervention approaches. Some research suggests that the ESDM is an effective intervention approach when delivered intensively either one-on-one and in groups. However, this type of intensive intervention is often prohibitively expensive and there is a limited number of professionals who are trained to provide such interventions. Studies 1 and 2 in this thesis evaluated ESDM delivery methods which could feasibly be provided to a large number of children at a limited cost to families or service providers. Specifically, Study 1 evaluated the effectiveness of 3 hours a week of home-based therapist delivered ESDM intervention for 12 weeks for four young children with ASD. The results of this study suggest that all four children had improvements in imitation, functional

utterances/intentional vocalisations, and engagement during the intervention which were maintained four weeks after treatment and generalised to some degree to their mothers. Study 2 evaluated the effectiveness of 1 hour a week of home-based parent training based on the ESDM for 12 weeks for the parents of five young children with ASD. The results of this study suggest that four of the five parents learned to implement the techniques with a high degree of fidelity and that these improvements were generally maintained one month after treatment. The children in this study also improved on at least two of the child-related outcome measures and most of these improvements were maintained one month after treatment. However, these improvements did not generalise to a second family member. These generally promising results suggest that methods of ESDM intervention delivery that require relative few hours of professional input may be effective in improving some outcomes for young children with ASD. In future, researchers could focus on replicating and extending these findings and further evaluating other low-intensity methods of delivering early intervention to young children with ASD. In addition, more research is needed into the relative effectiveness of parent training versus direct therapy.

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# ${\bf APPENDIX~A}$ Summary of Studies Evaluating the Effectiveness of the Denver Model and DIR/Floortime $^{\rm TM}$

Table 7.1

Participant Characteristics, Intervention Characteristics, Quality/Rigor, and Study Design for the Included Denver model and DIR/Floortime<sup>TM</sup> Studies.

	Child Characteristics				Intervention characteristics			_
Study and Intervention	N	Age (Mo		Parent/Therapist Characteristics	Туре	Intensity and Duration	Rigor	Design
Denver Rogers et al., 1986	26	24-72	ASD	Teacher and TAs as therapists, training ns.	Centre-based group intervention	11 hrs/week for 6 to 8 months	Weak	Quasi-experimental one group pre-post test
Rogers et al., 1987	11	36-72 (M=56)	ASD	20 therapists (teachers, TAs, SWs, SLPs, OTs)	Therapist" in-vivo training Child: centre-based group intervention	Therapist: 40 hr. training week, 2 x follow-up training days Child: 12 to 26 hours per week for approx. 4 months	Weak	Quasi-experimental one group pre-post test
Rogers and Lewis, 1989	41	39-72	ASD, DD	Teacher and TAs as therapists, training ns.	Centre-based group intervention	22.5 hrs/week for 12 months	Weak	Quasi-experimental one group pre-post test
Rogers and DiLalla, 1991	74	24-72	ASD, DD, behavioural/emoti -onal disorders	Primary therapist ns., training, ns.	Centre-based group intervention	15 to 22.5 hrs/week for ~18 months	Weak	Quasi-experimental one group pre-post test
DIR/Floortime™ Solomon et al., 2007	68	18-72	ASD	Mother and father participated	Clinic- and home-based parent training	1 day workshop, 3 to 4 hrs/month for 8 to 12 months	Weak	Quasi-experimental one group pre-post test

Pajareya and Nopmaneejumru- slers, 2011	16-int. 16- TAU	24-72	ASD	1 parent (mother) per child	Home-based parent training	3 hour workshop, 1.5 hour one-on-one training	Adequate	Randomised controlled trial
Liao et al., 2014	11	45-69 M=56	ASD	1 parent (mother) per child	Home-based parent training	2-3 hrs/week for 3 weeks then 2 hrs/2 weeks for 10 weeks	Weak	Quasi-experimental one group pre-post test
Solomon et al., 2014	61-int. 63- TAU	32-71 M=50	ASD	1 parent per child	Home-based parent training	3hrs/month for 12 months	High	Randomised controlled trial

Note: DS= Down syndrome, ns. = not specified, Int.= intervention group, TA= Teacher's aide TAU= treatment-as-usual group

Table 7.2 Child, Parent, and Therapist Outcomes of the Included Denver Model and  $DIR/Floortime^{TM}$  Studies.

Study	Child Outcomes	Parent/Therapist Outcomes
Denver	Sig. increase in cognition, language and social	No parent/therapist outcomes
Rogers et al.,	emotional development (EIPPP).	reported.
1986	Sig. increase in symbolic play and social	
	communication (POS).	
	Sig. increase in interactions involving positive	
	affect and social initiations to the mother	
Rogers et al.,	Sig. increase in fine motor, gross motor,	Staff found intervention to be socially
1987	cognitive, language, social/emotional	valid.
	development (EIPPP).	Sig. improvement in implementation
		of five key components of the model
		(POS).
Rogers and	Sig. increase in fine motor, gross motor,	No parent/therapist outcomes
Lewis, 1989	cognitive, language, social/emotional	reported.
	development (EIPPP).	
	Sig. increase in symbolic and social	
	communicative play (POS).	
	Sig. reduction in autism symptomology (CARS)	
Rogers and	Sig. increase in fine motor, gross motor,	No parent/therapist outcomes
DiLalla, 1991	cognitive, language, social/emotional	reported.
	development (EIPPP). Children with ASD	
	diagnosis showed similar improvements to those	
	with behavioural/emotional and developmental	
	disorders	
	Sig. increase in language development (SICD	
	and other measures used for a small number of	
	children)	
DIR/Floortime <sup>TM</sup>	Sig. increase in functional developmental	80% of parents were satisfied or very
Solomon et al.,	progress (FEAS)	satisfied with the intervention
2007		No change in parent behavior when
		interacting with the child (FEAS)
Pajareya and	Sig. better increase in functional developmental	No parent/therapist outcomes
Nopmaneejumru-	progress (FEAS and FEDQ) than TAU.	reported.
slers, 2011	Sig. better decrease in autism symptomology	
	(CARS) than TAU.	

Liao et al., 2014	Sig. better increase in functional developmental	Sig. reduction in parenting stress
	progress (FEAS) and total score for adaptive	(Parenting Stress index)
	behavior (VABS-II) than TAU	
Solomon et al.,	Sig. better improvements in pivotal	Sig. better improvements in parental
2014	developmental behaviours (CBRS), ASD	interaction style (MBRS) for the DIR
	diagnostic severity (ADOS) and socioemotional	intervention group.
	behavior (FEAS) for the DIR intervention	No sig. differences between groups in
	group.	parenting stress (Parenting Stress
	No sig. differences between groups on	index)
	development (MSEL), language (MCDI), or	
	autism symptomology (SCQ)	

Note: CARS= Childhood Autism Rating Scale; CBRS=Child Behavior Rating Scale; EIPPP= Early Intervention Profile and Preschool Profile; FEAS=Functional Emotional Assessment Scale; FEDQ=Functional Emotional Developmental Questionnaire; MCDI= MSEL= Mullen Scales of Early Learning; MBRS= Maternal Behavior Rating Scale; POS= Play Observation Scale; SCQ= Social Communication Questionnaire; SICD= Sequenced Inventory of Communicative Development; VABS-II= Vineland Adaptive Behavior Scales, second edition

### APPENDIX B

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#### REVIEW PAPER

## Effectiveness of the Early Start Denver Model: a Systematic Review

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Abstract The present review identified 15 studies that have evaluated the Early Start Denver Model (ESDM) as an early intervention program for a total of 209 children with autism spectrum disorder. The articles were summarized in terms of participant characteristics, intervention characteristics, study design, quality of the study/research rigor, and outcomes. ESDM intervention characteristics included provision of training to parents or therapists, intensive one-to-one intervention, and group intervention. Most studies reported positive child, parent, and therapist outcomes. The included articles used a range of group and single-case designs, but nearly half of the studies were rated as having weaknesses in terms of quality/rigor. We conclude that the ESDM is a potentially promising intervention, but the limited number of highquality studies indicates the need for additional research to evaluate its effectiveness.

Keywords Autism spectrum disorder · Early start Denver model · Early intervention · Systematic review

Autism spectrum disorder (ASD) is a neurodevelopmental disorder characterized by deficits in social-communicative functioning and the presence of repetitive and restricted behaviors, interests, and activities (American Psychiatric Association 2013). The prevalence of ASD has increased dramatically in the past 20 years and is now estimated to be approximately 1 in 68 children (Wingate et al. 2014). The

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School of Education, Victoria University of Wellington, PO Box 17-310, Karori Campus, Wellington, New Zealand increase in prevalence appears to be largely due to earlier and more accurate diagnosis (Dawson and Bemier 2013). In fact, screening tools such as the Modified Checklist for Autism in Toddlers (M-Chat; Robins et al. 2001), the Autism Diagnostic Observation Scale (ADOS-2)-toddler module (Lord et al. 2012), the Baby and Infant Screen for Children with autism Traits (BISCUIT; Matson et al. 2007), and the First Year Inventory (Baranek et al. 2003) allow for infants as young as 12 months of age to be assessed as at risk for ASD. It is likely that new scientific techniques for identifying biomarkers of ASD will further increase the accuracy of diagnosis in infants and toddlers (Dawson and Bernier 2013).

Research suggests that ASD is caused by a combination of genetic and environmental risk factors (Dawson 2008). Positive interactions between an infant or toddler and his or her social environment-such as those that might occur in a high-quality early intervention program-might, for example, "help guide brain and behavioral development back towards a normal pathway" (Dawson 2008, p. 776). Several studies have found that younger children respond better to treatment than older children, which suggests the value of early intervention and the possibility of a critical period of development in which early interventions are most effective (Granpeesheh et al. 2009; Harris and Handleman 2000). The increased number of children being identified with ASD, combined with earlier diagnosis and the potentially increased responsiveness of younger children to early intervention, suggest that there will be an increasing need for effective early intervention

A number of studies have evaluated early intervention programs for children with ASD. This research began in the 1960s, with initial studies focused on teaching a range of behaviors in children with ASD (e.g., Ferster and DeMyer 1961; Hingtgen et al. 1967; Leff 1968). However, the first attempt to develop and evaluate a manualized early



intervention program was reported by Lovaas (1987). In that study, Lovaas reported that a form of early intensive behavioral intervention (EIBI), based on the principles of applied behavior analysis (including extensive use of discrete-trial training), could produce large improvements in scores on a standardized assessment of intellectual functioning (i.e., IQ scores) and increases in many areas of adaptive behavior functioning for about half of the participating children with ASD. A 4-year follow-up study reported that these gains were largely maintained (McEachin et al. 1993).

Since then, numerous additional studies have provided further evidence to support the benefits of EIBI for improving IQ and adaptive behavior functioning in young children with ASD (Cohen et al. 2006; Howard et al. 2005; Sallows and Graupner 2005; Smith et al. 2000). Based on these studies, it would seem that EIBI could be classified as a well-established treatment for young children with ASD (Dawson and Bemier 2013). However, several reviewers have expressed concerns with the methodology used in some of these EIBI studies. In addition, not all of the children participating in these studies have responded as positively to the treatment (Dawson and Bernier 2013).

In light of these somewhat mixed results, variations of the EIBI program developed by Lovaas (1987) have been developed and evaluated. For example, the group of approaches termed Naturalistic Developmental Behavioral Interventions (NDBIs; Schreibman et al. 2015) incorporate aspects of both the discrete-trial training approach described by Lovaas (1987) with more naturalistic teaching strategies. While NDBIs often include discrete-trial and therapist-initiated training, they also include more incidental and child-initiated training opportunities (e.g., following the child's lead). Examples of NBDIs include Incidental Teaching (Hart and Risley 1975), Social Communication/Emotional Regulation/Transactional Supports (SCERTS; Prizant et al. 2006), Enhanced Milieu Teaching (Yoder and Warren 2001), and Early Achievements (Landa et al. 2011).

Although there is evidence to suggest that discrete-trial training can be effective, its potential limitations include a possible lack of generalization, prompt dependency, a lack of spontaneity, and an increase in challenging behaviors associated with escape and avoidance of the training (Smith 2001). NBDIs, in contrast, may be more likely to promote generalization and spontaneity and more likely to preempt challenging behavior and prevent prompt dependency (Schreibman et al. 2015).

The present review focuses on summarizing studies that have evaluated one specific type of NBDI, that is, the Early Start Denver Model (ESDM; Rogers and Dawson 2010). The ESDM could be classified as a type of NBDI, in that it involves the use of behavioral teaching techniques (e.g., response prompting, shaping, reinforcement) as well as a number of naturalistic and developmentally oriented strategies,

such as ensuring the child receives positive affect responses from adults and incorporating the use of play and sensory/social routines into the therapy sessions (Rogers and Dawson 2010). Additional specific techniques incorporated into the ESDM include intensive teaching, a positive behavior approach for problematic behaviors, and creating opportunities for family involvement (Rogers and Dawson 2010).

The ESDM is based upon two previous naturalistic approaches: pivotal response treatment (PRT) and the Denver Model. In line with PRT, the ESDM targets a number of skills that are considered pivotal to learning and development, such as increasing child motivation, promoting spontaneity, and focusing on social initiations (Koegel et al. 1999a, b). In addition, similar to the Denver Model, the ESDM places emphasis on teaching imitation, which is thought to be key for learning new skills, as well as being important for developing positive relationships with children (Rogers et al. 1986; Rogers and DiLalla 1991). The ESDM can be provided to toddlers as young as 12 months of age. Specific target skills covered in the ESDM curriculum include receptive and expressive communication, joint attention, imitation, social skills, play skills, and motor skills.

ESDM intervention is usually delivered by parents or therapists within a child's home environment or in group settings such as early childhood education centers. Another key aspect of ESDM intervention involves training parents and therapists to implement ESDM techniques with fidelity (\*Vismara et al. 2009a, b)

Although several literature reviews of intervention programs for children with ASD have included studies that implemented the ESDM (e.g., Bradshaw et al. 2015; Warren et al. 2011), there appear to be no published reviews that have focused explicitly on studies that have evaluated the ESDM. Given that the ESDM appears to be an increasingly popular form of early intervention for children with ASD (Penner et al. 2015; \*Vivanti et al. 2014), a review of this specific early intervention model would seem timely.

#### Method

#### Search Procedures

We searched (a) PsycINFO, (b) Education Resources Information Centre (ERIC), (c) Cumulative Index to Nursing and Allied Health Literature (CINAHL), (d) Scopus, and (e) PubMed. These databases were selected in order to find relevant articles published in the areas of health, education, and psychology. The search was limited to English language papers appearing in peer-reviewed journals from inception to June 2015. The following search terms were entered into the Anywhere field, which covers the articles' titles, abstracts, and/or full-text: Denver model OR ESDM AND ASD OR



Autism. We also conducted ancestral searches using the reference lists from studies identified via the database search.
Additional database searches were conducted for all authors of studies identified in the initial database search.

#### Inclusion and Exclusion Criteria

Articles were included for review if they met the following inclusion criteria: (a) empirical research evaluating the effects of an intervention that was described as involving the ESDM; (b) children as the recipients of the intervention with an average age of below 60 months (as this is the oldest recommended age for ESDM intervention); (c) children who had a diagnosis of ASD, or Pervasive Developmental Delay-Not Otherwise Specified (PDD-NOS), or who were described as being at at risk for a diagnosis of ASD or PDD-NOS; (d) the results of the study included at least one objective child, parent, or therapist outcome measure. All study designs, including case studies were included in this review.

Articles were excluded from the literature review if they related to the Infant Start or Denver Models of intervention, rather than the ESDM. Infant Start is a recent intervention focusing on children under the age of 12 months (Rogers et al. 2014), whereas the Denver model is a precursor to the ESDM developed in the 1980s (Rogers et al. 2006).

#### Data Extraction

Included articles were coded according to the following categories: (a) child characteristics as reported immediately prior to ESDM intervention (number, age, and diagnosis), (b) parent/therapist characteristics (number, training, and years of experience), (c) the model of providing intervention (i.e., training parents or therapists to deliver the intervention or direct delivery of one-on-one or group intervention by parents, therapists or experts/research staff), (d) intensity and duration of the intervention (i.e., the number of weeks and hours per week of intervention), (e) type of study design (e.g., randomized control trial, single-case multiple-baseline design), (f) quality of the study/research rigor, (g) child outcome measures (e.g., scores on cognitive, language, and/or adaptive behavior assessments), and (h) parent/therapist outcomes measures (e.g., the extent to which the parents or therapists implemented the intervention as intended, that is, treatment integrity).

With respect to item (f), we rated each study using a metric of research quality/rigor (i.e., The Evaluative Method for Determining Evidence-Based Practices in Autism; Reichow 2011; Reichow et al. 2008). This method can be used to evaluate the quality of both group and single-case research designs. With this approach, each study is given a rating of "strong," "acceptable," or "weak" depending on the number of primary and secondary quality indicators that it meets. Primary quality indicators include the quality of the

description/definition of: (a) participant characteristics, (b) independent variables, (c) comparison/baseline condition, (d) dependent variables, (e) link between research question and data analysis, and (f) statistical analysis/experimental control. Secondary quality indicators were the extent to which the study addressed issues related to: (a) random assignment (when relevant), (b) interobserver agreement, (c) blinding, (d) treatment integrity, (e) attrition, (f) generalization or maintenance, (g) effect size, and (h) social validity. The rating scale has been reported to have good reliability and validity (face, concurrent, and content validity; Reichow et al. 2008).

#### Inter-rater Agreement

The first author conducted the initial search of the five databases, which yielded 44 results. An additional article for possible inclusion was identified though the author search. From these 45 possible articles, the first author identified 15 articles that were judged to meet the inclusion criteria based on her reading of the title, abstract, and as much of the full text as necessary to make a decision. The second author replicated the search and nominated 14 of the 15 articles identified for inclusion by the first author (93 % agreement). Consultation, resulted in the additional article (i.e., \*Vismara et al. 2012) being included. The first author then extracted data from each of the 15 studies and created a summary of the articles (Tables 1 and 2). To check the accuracy of these summaries, the second author independently reviewed the summaries in Tables 1 and 2 and compared these to the original articles using a 12-point checklist. Completing the checklist involved noting whether the summary provided by the first author accurately matched the information contained in the original article (e.g., Is the number of participants correct? Is the type of intervention delivery model correct?). From this checklist, a type of inter-rater agreement could be computed by comparing the number of checklist items that were noted as correct with the total number of checklist items. Overall agreement was 97 % and consensus was then reached to resolve the few discrepancies.

#### Results

Tables 1 and 2 provide a summary of each study in terms of (a) participant characteristics, (b) intervention characteristics, (c) quality/rigor, (d) study design, and (e) outcomes.

#### Child Characteristics

The 15 articles evaluated the effectiveness of the ESDM from 12 individual studies. These 12 studies provided ESDM intervention to a total of 209 children with ASD or at risk for an ASD diagnosis. The control groups (children who did not



concurrent multiple multiple baseline Single-subject nonmultiple-baseline Quasi-experimental Quasi-experimental Quasi-experimental Quasi-experimental multiple group one group preone group precontrolled trial controlled trial controlled trial controlled trial one group precompanison Single-subject Adequate Single-subject Randomized Randomized Randomized Randomized post-test post-test post-test baseline Case study Strong Strong Strong 15 h/week therapist intervention and 16 h/ Strong Weak Weak Weak Weak Weak Weak Weak 15-25 h of group intervention (therapist to Therapist Self-instruction, 10 h training 1.5 h/week parent coaching for 12 weeks, (therapist-to-child ratio = 1:4) and 1 h/ seminar (direct therapy), 3 h training week parent intervention for 2 years child ratio = 1:3) per week for 1 year seminar (parent coaching), 4 h team week one-on-one for 10 months Child: 1 h/week for min. 9 weeks 15-20 h/week group intervention 1 workshop lasting 4 full days and self-guided website 1.5 h/week for 12 weeks 1 h/week for 12 weeks Intervention characteristics as \*Dawson et al. 2010 1 h/week for 12 weeks 1 h/week for 12 weeks Intensity and duration Intervention characteristics as \*Rogers et al. 2012 supervision A summary of participant characteristics, intervention characteristics, quality/rigor, and study design of the included studies Intervention characteristics Home-based therapist and based distance learning parent coaching (clinic-Center-based group and parent-delivered oneon-one intervention Telehealth" delivered Telehealth" delivered Center-based group ESDM intervention vs. live instruction) home-based parent workshop in direct education sessions education sessions education sessions education sessions Therapist training in Clinic-based parent direct ESDM and Father primary participant Clinic-based parent Clinic-based parent parent education Therapist training intervention one-on-one sessions Type 10 therapists (5 per group) experience Number of 2 months training and As \*Dawson et al. 2010 24 community-based El trained in ESDM and As \*Rogers et al. 2012 minimum of 5 years practitioners with a Therapists received 1-2 parents per child 1-2 parents per child demonstrate d.onparents not stated trained in ESDM Therapists formally Therapists formally Clinical diagnosis 1 father, 7 mothers 2.5 to 30 years 1 parent per child reached fidelity 1 parent per child going fidelity Parent/therapist Characteristics experience Clinical diagnosis Clinical diagnosis Child Characteristics as \*Dawson et al. 2010 and Clinical diagnosis 4. \*Dawson 24 (int.) 24 18-30 M=23 Clinical diagnosis Clinical diagnosis Clinical diagnosis following study) Atrisk of ASD Atrisk of ASD of ASD (one of ASD of ASD Diagnosis N=24 typically developing controls Age (months) M = 21M = 2836-58 M=49 22–58 M=38 16-38 42 10-36 24-51 Child characteristics (48-63 months) 0 Characteret al. 2012 \*Rogers istics as et al. 2012 49 (TAU) (JAC) 49 (int.) 10.\*Vismara N/A 23 8 5 \*Eapen et al. 2013 \*Vivanti \*Vismara ct al. 2010 \*Vismara \*Vismara ct al. 2012 Vismara et al. 2014 et al. 2008 7. \*Vismara ct al. 2012 ct al. 2013 \*Rogers Dawson 12. \*Estes 2013a 2009a 2009b 2013b Table 1 ct al g g g g et al. Study



Table 1 (continued)	ntinued)							
Study	Child characteristics	cteristics		Parent/therapist	Intervention characteristics		Rigor	Design
	N	Age (months) Diagnosis		CHILDRES	Type	Intensity and duration		
13. *Fulton 38 et al. 2014	38	38-63 M=52.2	Clinical diagnosis of ASD or PDD-NOS	Clinical diagnosis 6 therapists formally of ASD or trained in ESDM and PDD-NOS reached fidelity	Center-based group ESDM intervention	15-20 h/week group intervention (therapist-to-child ratio = 1:4) and 1 h/ week one-on-one for II.8 months	Weak	Quasi-experimental one group pre post-test
14. *Vivanti 27 (int.) et al. 2014 30 (TAU)	27 (int.) 30 (TAU)	≤60 M =40	iagnosis	Therapists formally trained in ESDM and reached fidelity	Center-based group ESDM intervention	15-25 h/week group ESDM for 12 months Strong	Strong	Quasi-experimental multiple-group comparison
15. *Estes et al. 2015	Long-term f 2010	ollow-up (2 years	s) of *Dawson et al.	As *Dawson et al. 2010	<ol> <li>*Estes Long-term follow-up (2 years) of *Dawson et al. As *Dawson et al. 2010 Intervention characteristics as *Dawson et al. 2010 et al. 2015</li> </ol>	s as *Dawson et al. 2010	Strong	Randomized controlled trial

receive ESDM intervention) included a total of 103 ASD, or at risk, children and 24 typically developing children. All of the children in the study of \*Vivanti et al. (2013) also participated in the study of \*Vivanti et al. (2014) and 10 (36 %) of the children in the study of \*Fulton et al. (2014) also participated in the \*Eapen, Črnčec, and Walter (2013) study. The results from children who participated in multiples studies were included in the analysis. Participants ranged from 9 to 63 months of age at the start of intervention. The study of \*Vismara et al. (2013b) did not have any children as participants as this study reported on a four day therapist training intervention.

In nine studies (\*Dawson et al. 2010; \*Eapen et al. 2013;

In nine studies (\*Dawson et al. 2010; \*Eapen et al. 2013; \*Fulton et al. 2014; \*Vismara et al. 2012; \*Vismara et al. 2009a, b; \*Vismara et al. 2013a; \*Vivanti et al. 2013; \*Vivanti et al. 2014), the children had a diagnosis of ASD, although one child in the \*Fulton et al. (2014) study had a PDD-NOS diagnosis. In seven of these studies (\*Dawson et al. 2010; \*Vismara et al. 2012; \*Vismara et al. 2009a, b; \*Vismara et al. 2013a; \*Vivanti et al. 2013; \*Vivanti et al. 2014), participants' ASD diagnosis was based on the Autism Diagnostic Observation Scale (ADOS; Lord et al. 2002) and the Toddler Autism Diagnostic Interview (T-ADI; Lord et al. 1994). Six studies (\*Dawson et al. 2010; \*Eapen et al. 2013; \*Fulton et al. 2014; \*Vismara et al. 2009a, b; \*Vismara et al. 2013a) included a clinical diagnosis of ASD based on the criteria from the Diagnostic and Statistical Manual-fourth edition (DSM-IV; American Psychiatric Association 2000). In the remaining two studies (\*Rogers et al. 2012; \*Vismara et al. 2008), the included children were described as being at risk for an ASD diagnosis based on clinical judgment or a clinical assessment using the ADOS-toddler (Lord et al. 2012), the Early Screening of Autistic Traits Questionnaire (Swinkels et al. 2006), the Infant Toddler Checklist (Wetherby and Prizant 2002), or the M-CHAT (Robins et al. 2001). All included studies except those by \*Vismara et al. (2012), \*Vismara et al. (2013a), and \*Vivanti et al. (2014) reported excluding participants on the basis of significant other medical, physical, genetic, or neurological conditions.

Several studies did not report on the gender of the participants (\*Vismara et al. 2009a, b; \*Vismara et al. 2013a), but in all the remaining studies, at least 76 % percent of participants were male.

#### Setting

nt intervention group, TAU treatment as usual group

Three of the studies took place primarily within clinics in Sacramento, California, Seattle, Washington, or Ann Arbor, Michigan in the USA (\*Vismara et al. 2009a; \*Vismara et al. 2008; \*Rogers et al. 2012). The study by \*Dawson et al. (2010) took place in the homes of children who lived within 30 min of the University of Washington. The studies that



Table 2 A summary of the child, parent, and therapist outcomes of the included studies

Study	Child Outcomes	Parent/Therapist outcomes
1.*Vismara et al. 2008	Child's diagnosis changed from ASD to PDD-NOS	The parent reached fidelity (85 %) on the ESDM fidelity scale on the 8th week, this level was maintained at follow-up
2.*Vismara et al. 2009a	Imitative behaviors increased for 7 of 8 children during intervention and this level was maintained for the children who participated in follow-up	5 of 8 parents reached fidelity on the ESDM fidelity scale by the 6th week. For these parents, that level of fidelity was maintained at follow-up
3 *Vismara et al. 2009b	There was no significant increase in imitative behaviors	Therapist scores on the ESDM fidelity scale, therapist-parent coaching fidelity scale and satisfaction and understanding survey increased significantly during the didactic training and team supervision conditions. Parent scores on the ESDM fidelity scale were significantly higher than baseline during all treatment conditions. There were no significant differences between the two delivery methods
<ol> <li>*Dawson et al. 2010</li> </ol>	The ESDM group were significantly more likely to have improved diagnosis (ASD→PDD-NOS)	No parent/therapist outcomes reported
	The ESDM and typically developing groups had significantly higher attention engagement and active processing of faces than the ASD TAU group. These two groups also had significantly higher cortical activation when viewing faces rather than objects, whereas the ASD TAU group had higher activation for objects. There were no significant differences between groups for orienting to the stimulus or perceptual processing of faces	No parent/therapist outcomes reported
6. *Rogers et al. 2012	Following intervention, here were no significant group differences in cognitive ability (MSEL), receptive and expressive language (MCDI), imitation, or orienting to stimuli. The TAU group had a significantly greater decrease in social affect scores (ADOS) than the ESDM group	Parents in the ESDM group had significantly better working alliances with their primary therapist than those in the TAU group. There were no significant group differences in parent ESDM fidelity
7.*Vismara et al. 2012	Spontaneous and prompted functional verbal utterances, spontaneous imitation, and attentiveness and social initiations (CBRS) increased significantly during intervention and follow-up.	There was a significant increase in parent ESDM fidelity scores during intervention. On average, parents reached fidelity (80 %) by the 6th week. Parental responsivity, affect, and achievement oriented behaviors (MBRS) increased during intervention,
	Receptive and expressive vocabulary (MCDI) increased significantly during intervention. There was also a slight increase in adaptive behavior (VABS) scores	parent directive behavior did not increase. Parents gave positive ratings on a feasibility and acceptability questionnaire
8. *Eapen et al. 2013	Overall cognitive skills (MSEL), receptive language, expressive language and visual reception increased during intervention. Receptive communication and gross motor skills increased significantly during intervention but overall adaptive behavior (VABS-II) did not. There was a significant decrease in autism specific features (SCQ) during intervention	No parent/therapist outcomes reported
9.*Vismara et al. 2013a		6 of 8 parents reached fidelity (80 %) on the ESDM fidelity scale during intervention, and 7 of 8 parents during follow-up. On average parents reached fidelity in the 7th week. Parent engagement (MBRS) increased during intervention and follow- up. Parents gave positive ratings on a satisfaction survey
10. *Vismara et al. 2013b	No child outcomes reported	Therapist direct ESDM fidelity increased significantly during intervention and follow-up. Self-evaluated ESDM fidelity was significantly higher than baseline during intervention but not follow-up. Therapists gave positive ratings on a satisfaction survey
11. *Vivanti et al. 2013	Overall cognitive skills (MSEL), including visual reception, fine motor, receptive language and expressive language improved following intervention. There was no significant decrease in autism severity (ADOS). More functional use of objects, goal understanding and imitation was associated with greater intervention gains	No parent/therapist outcomes reported
12. *Estes et al. 2014	Same as *Rogers et al. 2012	Following intervention, the parents in the ESDM group had significantly lower parenting stress (QRS) scores than the TAU group. There was no significant difference between groups for parent sense of competence (PSOC)



Table 2 (continued)

Study	Child Outcomes	Parent/Therapist outcomes
13. *Fulton et al. 2014	Following intervention, there was a significant decrease in maladaptive behavior (ESDM behavior rating) and a significant increase in cognitive skills (MSEL). There was no significant change in adaptive behavior (VABS-II) or autism- specific features (SCO)	Parents gave positive ratings on a satisfaction survey
14. *Vivanti et al. 2014	Following intervention, the ESDM group had significantly higher cognitive ability (MSEL), and receptive language (MSEL) scores than the TAU group. There were no group differences in adaptive behavior (VAB-II), or autism severity (ADOS)	No parent/therapist outcomes reported
15. *Estes et al. 2015	2 years after intervention, the ESDM group had significantly higher adaptive behavior (VABS) including socialization than the TAU group. They also had significantly lower overall symptom severity (ADOS) including restricted and repetitive behaviors. There were no significant differences between groups for diagnostic categorization, intellectual functioning (DAS), hyperactivity and irritability (ABC), repetitive behaviors (RBS), or peer relationship outcomes (ADI-R)	No parent/therapist outcomes reported

ABC Aberrant Behavior Checklist, ADOS Autism Diagnostic Observation Scale, ADI-R Autism Diagnostic Interview-Revised, CBRS Child Behavior Rating Scale, MCDI MacArthur-Bates Communicative Development Inventory, DAS Differential Ability Scale, MBRS Maternal Behavior Rating Scale, MSEL Mullen Scales of Early Learning, PSOC Parent Sense of Competence questionnaire, QRS Questionnaire on Resources and Stress, RBS Repetitive Behavior Scale, SCQ The Social Communication Questionnaire, VABS Vineland Adaptive Behavior Scales, VABS-II Vineland Adaptive Behavior Scales-Second edition

focused on training therapists took place in varying locations in the USA, including a children's hospital, a university-based clinic, a private clinic, and a public school (\*Vismara et al. 2009b, 2013b). Four studies took place in group early child-hood education centers, two in Melbourne, Australia (\*Vivanti et al. 2013; \*Vivanti et al. 2014) and two in Sydney, Australia (\*Eapen et al. 2013; \*Fulton et al. 2014). The final two studies used a "telehealth" or virtual teaching approach to deliver training to parents across the United States of America (\*Vismara et al. 2012, 2013a).

#### Parent/Therapist Characteristics

Five studies had a parent-training component and either one or two parents were present during intervention sessions (\*Rogers et al. 2012; \*Vismara et al. 2012; \*Vismara et al. 2009a; \*Vismara et al. 2013a; \*Vismara et al. 2008). However, it was not always specified whether it was the mother or the father who received training. Two studies focused on the training of therapists; one included therapists with a minimum of 5 years of experience working with young children with ASD (\*Vismara et al. 2013b), while the other included therapists with 2.5 to 30 years of experience (\*Vismara et al. 2009b). Of the five studies that involved direct intervention by therapists (\*Dawson et al. 2010; \*Eapen et al. 2013; \*Fulton et al. 2014; \*Vivanti et al. 2013; \*Vivanti et al. 2014), it was the case that all of these therapists had first received formal ESDM training and had reached fidelity (80 to 85 %) on an

ESDM implementation fidelity measure (Rogers and Dawson 2010) prior to implementing the intervention.

#### Intervention Characteristics

With respect to the five studies (reported in six papers, \*Estes et al. 2014; \*Rogers et al. 2012; \*Vismara et al. 2013; \*Vismara et al. 2019; \*Vismara et al. 2009a; \*Vismara et al. 2013a; \*Vismara et al. 2008) that involved training parents to implement the intervention (i.e., p-ESDM), the training provided to parents involved parent education, coaching, and feedback based on the principles of the ESDM. The intensity of these p-ESDM training sessions ranged from 60 to 90 min per week over a 12-week period. Three of these p-ESDM studies, the results of which were reported in four papers, took place in a clinic (\*Estes et al. 2014; \*Rogers et al. 2012; \*Vismara et al. 2009a; \*Vismara et al. 2012; \*Vismara et al. 2013a) used a telehealth approach, which involved weekly video conferences with parents.

Therapist training in the study of \*Vismara et al. (2009b) used a self-instructional model, a 10-h didactic training session for direct therapy, a 3-h session for parent coaching, and 4 h of team supervision across 10 months. The training was delivered via a distance-learning model and in vivo. Therapist training in the \*Vismara et al. (2013b) study involved a 4-day therapist training workshop.



In one study, the results of which were reported in three publications (\*Dawson et al. 2010; Dawson et al. 2012; \*Estes et al. 2015) evaluated intensive parent and therapist one-on-one intervention within the same group of children. In these papers, the children received a reported average of 15.2 h per week of therapist intervention and 16.3 h per week of parent intervention over 2 years.

The remaining four studies (\*Eapen et al. 2013; \*Fulton et al. 2014; \*Vivanti et al. 2013; \*Vivanti et al. 2014) investigated the effects of an early childhood education centerbased group ESDM intervention. The intensity varied between 15 and 20 h per week (\*Eapen et al. 2013; \*Fulton et al. 2014; \*Vivanti et al. 2013) and 15 to 25 h per week (\*Vivanti et al. 2014). In the study by \*Eapen et al. (2013), participants also received 1 h per week of one-on-one intervention.

#### Study Designs and Quality Ratings

These 15 articles reported using a variety of research designs. Specifically, there was one case study design (\*Vismara et al. 2008) and three studies that used a single-case, multiple-baseline across participants design (\*Vismara et al. 2012; \*Vismara et al. 2009a; \*Vismara et al. 2013a). Of the group studies, two were randomized controlled trials, the outcomes of which were reported in five articles (\*Dawson et al. 2010; Dawson et al. 2012; \*Estes et al. 2014; \*Estes et al. 2015; \*Rogers et al. 2012), four used a one group pre-post-test design (\*Eapen et al. 2013; \*Fulton et al. 2014; \*Vismara et al. 2013b; \*Vivanti et al. 2013) and another two used quasi-experimental multiple-group comparison designs (\*Vismara et al. 2009b; \*Vivanti et al. 2014).

In terms of rated quality, three studies (the outcomes of which were published in six articles; \*Dawson et al. 2010; Dawson et al. 2012; \*Estes et al. 2014; \*Estes et al. 2015; \*Rogers et al. 2012; \*Vivanti et al. 2014) were rated as having a "strong" research design according to the criteria developed by Reichow et al. (2008) and Reichow (2011). One study (\*Vismara et al. 2012) was rated as having an "adequate" research design, whereas eight studies were rated as "weak" due to the absence of an adequate control group (\*Eapen et al. 2013; \*Fulton et al. 2014; \*Vismara et al. 2013b; \*Vivanti et al. 2013), inadequate statistical power (\*Vismara et al. 2009b), failure to replicate the experimental effect (\*Vismara et al. 2008), insufficient baseline data (\*Vismara et al. 2009a, or too few high quality primary quality indicators (\*Vismara et al. 2013a).

#### Child Outcomes

Positive results are defined as an improvement in the outcome measure for the majority of participants during intervention (single case), a significant improvement during intervention (one group pre-post-test) or significantly better results for the ESDM treatment group compared to the TAU group following intervention (multiple group comparison).

Negative results are defined as no improvement or deterioration in the outcome measure for the majority of participants during intervention (single case), no significant difference or a significant deterioration during intervention (one group prepost-test), or no difference between groups or greater improvement for the TAU group compared to the ESDM group following intervention (multiple group comparison).

#### Child Behavioral Functioning and Development

Twelve articles reported on measures of child behavioral functioning and development (\*Dawson et al. 2010; \*Eapen et al. 2013; \*Estes et al. 2015; \*Fulton et al. 2014; \*Rogers et al. 2012; \*Vismara et al. 2012; \*Vismara et al. 2009a, b \*Vismara et al. 2013a; \*Vismara et al. 2008; \*Vivanti et al. 2013; \*Vivanti et al. 2014). These measures were collected using a variety of instruments, including the Child Behavior Rating Scale (CBRS; Mahoney and Wheeden 1998), the Differential Ability Scale (DAS; Elliot 1990), the MacArthur-Bates Communicative Development Inventory (MCDI; Fenson et al. 2007), the Mullen Scale of Early Learning (MSEL; Mullen 1995), the Vineland Adaptive Behavior Scales (VABS; Sparrow et al. 1984; Sparrow et al. 2005), the Aberrant Behavior Checklist (ABC; Aman and Singh 1986; Aman et al. 1985), the Repetitive Behavior Scale (Bodfish et al. 1999), a standardized measure of peer relationships (Lord et al. 1994), and the ESDM behavior rating scale (Rogers and Dawson 2010). The four articles using the CBRS (\*Vismara et al. 2009a, b; \*Vismara et al. 2013a; \*Vismara et al. 2008, and the one article (\*Fulton et al. 2014) using the ESDM behavior rating scale reported positive results on these measures. Results from articles using the VABS, the MSEL, and the MCDI were mixed. Two articles found positive results for the overall VABS (\*Dawson et al. 2010; \*Estes et al. 2015), while three articles reported negative results (\*Eapen et al. 2013; \*Fulton et al. 2014; \*Vivanti et al. 2014). Five articles reported positive results for the overall MSEL (\*Dawson et al. 2010; \*Eapen et al. 2013; \*Fulton et al. 2014; \*Vivanti et al. 2013; \*Vivanti et al. 2014), while one (\*Rogers et al. 2012) reported negative results. Two articles reported positive results for the MCDI (\*Vismara et al. 2012; \*Vismara et al. 2013a), while one article reported negative results (\*Rogers et al. 2012). Results for the remaining measures were negative.

Observations of Social Interaction and Communication Skills

Six articles reported at least one child outcome measure that was based on direct observation of the child's social interaction and communication skills (\*Rogers et al. 2012; \*Vismara



et al. 2012; \*Vismara et al. 2009a; \*Vismara et al. 2013a; \*Vismara et al. 2008; \*Vismara et al. 2012). These measures included spontaneous verbal utterances, imitation skills, social orienting, and joint attention. All articles reported positive results for these outcome measures, with the exception of \*Vismara et al. (2009b) and \*Rogers et al. (2012) who found negative results on a measure of imitation skills. \*Rogers et al. (2012) also found negative results on a measure of social orienting and joint attention following intervention.

#### Physiological Measures

One article (Dawson et al. 2012) included electroencephalogram (EEG) activity as an outcome measure. They reported mixed results as brain areas corresponding with active cognitive processing and attention to faces showed significantly more activation in the ESDM intervention group than the TAU group. However, there were no significant differences between groups in activation of areas associated with perceptual processing of faces or orienting towards the stimulus.

#### Autism Severity and Diagnostic Outcomes

Seven articles (\*Dawson et al. 2010; \*Estes et al. 2015; \*Eapen et al. 2013; \*Fulton et al. 2014; \*Rogers et al. 2012; \*Vivanti et al. 2013; \*Vivanti et al. 2014) reported on autism severity using either the ADOS (Lord et al. 2002) or the Social Communication Questionnaire (SCQ; Berument et al. 1999). Of the five articles using the ADOS (\*Dawson et al. 2010; \*Estes et al. 2015; \*Rogers et al. 2012; \*Vivanti et al. 2013; \*Vivanti et al. 2014), only one reported positive results following intervention (\*Estes et al. 2015). Of the articles using the SCQ, one reported positive results (\*Eapen et al. 2013), and the other negative results (\*Fulton et al. 2014) following intervention. Three articles (\*Dawson et al. 2010; \*Estes et al. 2015; \*Vismara et al. 2008) reported on participants' change in diagnosis from ASD to PDD-NOS or "no diagnosis" following intervention. Two articles reported positive results in this regard (\*Dawson et al. 2010; \*Vismara et al. 2008), while \*Estes et al. (2015) reported negative results.

#### Parent and Therapist Outcomes

Six articles (\*Rogers et al. 2012; \*Vismara et al. 2013; \*Vismara et al. 2009a, b; \*Vismara et al. 2013a; \*Vismara et al. 2013a; \*Vismara et al. 2008) included a measure related to the extent to which parents implemented ESDM therapy correctly. Five articles (\*Vismara et al. 2012; \*Vismara et al. 2009a, b; \*Vismara et al. 2013a; \*Vismara et al. 2008) reported positive results in that the majority of parents (at least five out of eight) achieved an acceptable level of treatment integrity (80 % correct implementation) within six to eight sessions. However

\*Rogers et al. (2012) reported negative results with no significant differences in scores for treatment integrity in the ESDM group compared with the TAU group.

Three articles (\*Estes et al. 2014; \*Vismara et al. 2012; \*Vismara et al. 2013a) reported on additional parent outcome measures including the Maternal Behavior Rating Scale (MBRS; Mahoney et al. 1998), the Questionnaire of Resources and Stress (QRS; Konstantareas et al. 1992), and the Parenting Sense of Competence Questionnaire (PSOC; Johnston and Mash 1989). Positive results were reported for the MBRS and the QRS, but negative results were reported in the PSOC (\*Estes et al. 2014).

Two articles (\*Vismara et al. 2009b; \*Vismara et al. 2013b) included a measure of therapist implementation of the ESDM therapy procedures. Both articles reported positive results with respect to treatment integrity after the therapist had received the training. However, \*Vismara et al. (2013b) noted that while self-reported treatment integrity increased with training, this was not maintained at follow-up.

#### Social Validity

Five articles (\*Fulton et al. 2014; \*Vismara et al. 2012; \*Vismara et al. 2013a, b; \*Vismara et al. 2009b) reported positive results on a measure of feasibility, acceptability, or satisfaction with the intervention. \*Rogers et al. (2012) also reported positive results on a measure of working alliances between parents and therapists.

#### Generalization

None of the studies assessed generalization of child, parent, or therapist outcomes.

#### Moderators of Outcome

Several articles identified variables prior to intervention that could have a moderating effect on child and parent outcomes. \*Rogers et al. (2012) found that higher number of hours of intervention when both the ESDM and TAU groups were combined, predicted significantly better scores on the ADOS, MSEL, MCDI and Nonsocial orienting measures. They also found that chronological age negatively predicted MSEL scores after intervention. These results suggest that higher intervention hours and lower chronological age predicted better intervention outcomes in this sample. However, \*Vivanti et al. (2013) found that intensity of treatment and chronological age did not predict treatment outcomes in a group of 21 children receiving group ESDM therapy.

\*Rogers et al. (2012) found that nonsocial orienting was a significant predictor of increased scores on the MSEL and decreased ADOS social affect scores when both the ESDM and TAU groups were combined and that social orienting was



a significant predictor of reduced ADOS restricted and repetitive behavior scores. However, contrary to their hypothesis, imitation skills were not a significant predictor of treatment outcomes. In contrast, \*Vivanti et al. (2013) found that scores in an imitation task were a significant predictor of positive intervention outcomes, as well as functional object use, and goal understanding. They also found that cognitive ability and social attention were not significant predictors of intervention outcomes. \*Estes et al. (2014) found that the number of negative life events predicted increased parenting stress and decreased sense of competence in both the ESDM and TAU groups.

#### Discussion

The purpose of this review was to evaluate the ESDM as an early intervention approach for children with ASD. This review identified 15 relevant articles, all of which were published since 2008. The manner in which the ESDM program was delivered in these studies included (a) training parents to implement the program, (b) training therapists to implement the program, (c) having already trained therapists provide intensive one-on-one intervention, while also training parents to intensively provide intervention, and (d) having already trained therapists provide group intervention. The study designs included one case study, three multiple-baseline across participants designs, four quasi-experimental one-group prepost-test designs, two quasi-experimental multiple-group comparisons, and two randomized controlled trials, reported in five publications. The interventions varied significantly in intensity and duration; parent training was generally the shortest and least intensive form of intervention (approximately 1 h per week over 12 weeks), whereas all studies investigating group ESDM intervention and intensive delivery involved at least 15 h per week of therapy over a minimum of 10 months.

#### Outcomes

All studies reported at least some positive outcomes for the trained parents, therapists, and/or the participating children following the delivery of the training to the parents or therapist or receipt of the intervention by the children. All studies that investigated parent training reported that treatment integrity and parent engagement increased for the majority of parents and that this increase was maintained during follow-up. The two studies that examined therapist training also reported significant increases in treatment integrity (\*Vismara et al. 2009b; \*Vismara et al. 2013b). Most studies also reported positive child outcomes, particularly for cognitive skills, language ability, imitation, attentiveness, and social initiations. Results for other adaptive behaviors and severity of autism symptoms were mixed. Indeed, about 50 % of studies that assessed these latter types of dependent variables reported

negative results. Two studies reported positive changes in autism diagnosis following intervention (\*Dawson et al. 2010; \*Vismara et al. 2008). However, 2-year follow-up for one of these studies indicated that while the ESDM group had a significant reduction in autism symptoms compared to the TAU group, statistical differences in diagnostic categorization between the intervention and TAU groups were no longer evident (\*Estes et al. 2015). This suggests that while the ESDM group had reduced autism symptoms the majority still met the criteria for ASD.

Interestingly, the study by \*Rogers et al. (2012) was the only comparison study to find that the p-ESDM group did not have significantly better outcomes following intervention than the TAU group. They found that there were no significant differences in child outcomes between groups except that the TAU group had significantly lower social affect scores, which could perhaps indicate lower autism symptom severity. These results are consistent with several studies investigating other types of parent training that also failed to find a significant improvement in child outcomes for the parent-training group compared with the TAU group (Drew et al. 2002; Jocelyn et al. 1998). However, parents in the p-ESDM intervention group reported significantly higher working alliances (\*Dawson et al. 2010) with their therapists as well as significantly lower parenting stress than the TAU group (\*Estes et al. 2014). This suggests that the p-ESDM intervention might have some positive impact upon parents compared with the standard treatment that the other children received. It should be noted that, in this study, the TAU group actually received more hours of intervention per week than the p-ESDM group. It is possible that better outcomes for the p-ESDM group would become evident if they had received the same intensity and duration of intervention as the TAU group.

The study by \*Dawson et al. (2010) was the only study in this review to investigate the effects of intensive ESDM intervention on brain activity (via EEG scans). Their findings suggest normalized functioning in areas of the brain associated with active cognitive processing and attention to faces for the children in the intensive ESDM intervention group compared to the TAU group. This is one of the first studies to investigate the impact of early intervention on brain development, and although it is limited in terms of the small sample size and high level of unusable data, these findings suggest that early intensive intervention might improve brain functioning. While this is an intriguing suggestion, the effect reported by Dawson et al. (2012) would need to be replicated to verify this possibility.

Although this review suggests that the ESDM is a promising treatment for young children with or at risk for an ASD diagnosis, the results from most of these studies must be interpreted with caution due to the relatively low ratings on research rigor. Indeed, eight of the 12 studies were rated as having methodological weakness that reduced the certainty of evidence. It is also the case that one of the studies with the



strongest ratings did not report significant improvements for children in the ESDM group (e.g., \*Rogers et al. 2012). According to criteria developed by Reichow et al. (2008; Reichow 2011), specific numbers of group and single-case studies with adequate or strong research rigor are needed for an intervention to be considered an evidence-based practice (e.g., at least two group designs of strong research report strength conducted in different geographic locations). Using these criteria, at this stage, parent and therapist ESDM training cannot be described as evidence-based practices. Still, the positive results from several of these studies do support classification of the ESDM as a promising intervention for young children with or at risk for an ASD diagnosis.

This conclusion is based in part on the mostly positive outcomes from some of the studies reviewed, such as the generally very strong studies conducted by \*Dawson et al. (2010) and \*Estes et al. (2015). The results of these applications of the ESDM are comparable to those found in many studies investigating EIBI interventions that have involved providing at least 25 h of discrete trial intervention per week for more than two years (e.g., Smith et al. 2000; Cohen et al. 2006). This provides preliminary evidence that intensive ESDM intervention might be comparable to the effects of EIBI. In light of this, the ESDM might be seen as a more practical option given that it would seem to require less intensity of intervention and a less structured approach, which might be more acceptable to some parents and therapists. In addition, the more naturalistic orientation of the ESDM compared to some applications of EIBI as described by Lovaas (1987) could have the potential advantages outlined earlier (i.e., the ESDM might be more likely to promote generalization and spontaneity and more likely to preempt challenging behavior and prevent prompt dependency). Further comparative research is needed before such conclusions can be drawn.

The recommended steps for systematically validating and disseminating psychosocial interventions, such as the ESDM, include (a) some initial small scale efficacy studies, (b) manualization of the treatment if the results of the small scale studies are positive, (c) larger-scale efficacy studies such as RCTs using the manualized program, and (d) community effectiveness studies, in which the training might be delivered by the usual personnel, such as parents and/or teachers (Smith et al. 2007). This logical progressive pattern can be seen in the 15 papers reviewed herein. Specifically, the first few studies were small scale evaluations involving a case study (\*Vismara et al. 2008), single-case design studies (\*Vismara et al. 2009a), and a small scale multiple-group comparison (Vismara et al. 2009a) all of which provided some preliminary evidence of efficacy. The ESDM manual was then developed (Rogers and Dawson 2010) and followed by a larger RCT evaluating intensive parent and therapist intervention (\*Dawson et al. 2010) and an RCT evaluating a program for training parents to implement the ESDM (\*Rogers et al. 2012).

More recent studies have extended the evaluation of the ESDM to the wider community (\*Eapen et al. 2013; \*Fulton et al. 2014; \*Vivanti et al. 2013; \*Vivanti et al. 2014). This shift in study design and type suggests that the research on ESDM aligns well with the suggested stages for the validation and dissemination of psychosocial interventions. This overall program of research suggests the ESDM to be promising. However, given the limitations in some of these study's design, we would argue that there is a need for more research. Future research could be both small scale, to isolate the active mechanisms and necessary components of the intervention, and larger scale research to evaluate its effectiveness for use in the community. In addition, there may be value in comparative research. Indeed, while the ESDM appears promising, it is unclear whether it is more effective than other manualized treatments for young children with, or at risk for, an ASD diagnosis, such as EIBI (Lovaas 1987), Early Achievements (Landa et al. 2011) or PRT (Koegel et al. 1999a, b).

Another area of future research could be to evaluate the ESDM within other countries and with more diverse parent groups as there may well be cultural, educational, and socioeconomic differences that impact upon the acceptability and effectiveness of this program. Future research could also seek to investigate whether there are parent or child variables and/ or specific procedures that predict the success of the ESDM. The research into such issues has to date been limited and has revealed mixed results (\*Rogers et al. 2012; \*Vivanti et al. 2013). A final area for future research could be to consider additional dependent variables as outcome measures. Many studies in the present review examined imitation ability, for example, but relatively few have examined other measures of joint attention, such as eye contact, pointing, showing, and orienting towards stimuli. This is surprising considering that most research into early intervention for toddlers with ASD includes a measure of joint attention (Schertz et al. 2012), and the ESDM emphasizes the importance of nonverbal communication and attending to the communication partner (Rogers and Dawson 2010). Other potentially important dependent variables, which were underrepresented in this review, might include direct measures of problem behavior, functional play skills, appropriate interactions with peers and siblings, and successful transition into preschool or other early childhood settings.

#### Limitations of this Review

This review used the Evaluative Method for Determining Evidence-Based Practices in Autism (Reichow et al. 2008; Reichow 2011) to assess the quality of the included studies and to determine whether the interventions qualified as evidence-based practice. Other reviews have used different criteria (e.g., Horner et al. 2005). The use of different criteria may have resulted in different evaluations of research report



rigor, and thus, a different evaluation of the overall strength of evidence for the effectiveness of ESDM intervention. Furthermore, the decision to include only peer-reviewed studies, which were published in English, may have limited the overall scope of studies identified for this review.

#### Conclusion

The ESDM is a naturalistic behavioral/developmental intervention for young children with, or at risk for, an ASD diagnosis. The procedures used in the ESDM include a range of naturalistic teaching procedures that are based on the principles of applied behavior analysis and a developmental orientation. Based on our review of 12 studies, reported in 15 separate articles, the ESDM appears to be a promising treatment for young children with, or at risk for, an ASD diagnosis. However, there are methodological limitations that reduce the certainty of evidence for eight of the 12 studies. Given the generally promising results, the ESDM can be recommended as a practical approach to delivering a sensible early intervention program to children with, or at risk for, an ASD diagnosis. Still, more research is needed on all aspects of this model, particularly independent replications of the findings.

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#### Compliance with Ethical Standards

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Conflict of Interest The authors declare no conflicts of interest.

Ethical Approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed Consent Informed consent was obtained from all individual participants included in the study.

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# **APPENDIX C**

#### Parent Information Sheets and Consent Forms for Studies 1 and 2

# **Direct Therapy Information Sheet**

Project Title: Developmental, Relationship-Based Early Intervention for Children with Autism This research has been assessed and approved by Victoria University of Wellington Human Ethics Committee (Reference Number 22085).

Dear Parent/Legal Guardian,

We would like to invite you and your child to participate in a research study. The purpose of this study is to evaluate the effect of researcher-implemented developmental, relationship-based early intervention on outcomes for children with a diagnosis of Autism Spectrum Disorder (ASD). This intervention will be based upon Rogers and Dawsons' Early Start Denver Model (ESDM). This intervention will involve assessing your child's current skills across all areas of development and then providing naturalistic intervention in the context of play-based, relationship-focused routines. This includes both joint activity routines involving objects and sensory social routines such as "peekaboo", "chase", and "hide and seek".

If you give consent for you and your child to participate in this intervention, we will deliver three, approximately one hour, therapy sessions per week (3 hours in total). This excludes the time taken to set up, tidy up and discuss any progress the child has made. This intervention will last for 12 weeks and will take place in your home. Following intervention we will assess your child's generalisation of his or her skills interactions with you (the parent).

Prior to the start of the intervention, we will meet with you to complete several pre-assessments to evaluate your child's skills across developmental domains. These will take approximately two hours to complete. Assessments might include, for example, the MacArthur-Bates Communicative Development Inventories (MCDI) to assess your child's receptive and expressive language, the Vineland Adaptive Behaviour Scales, second Edition (VABS-II) to measure your child's adaptive behaviour, and the Social Communication Questionnaire (SCQ) to assess your child's severity of autism symptoms. We will also use an intervention satisfaction questionnaire, to assess how acceptable and effective you expect the treatment to be. The satisfaction questionnaire will then be repeated after the intervention has finished.

During the first session of intervention we will also directly assess your child's developmental skills using an adapted version of the ESDM Curriculum Checklist for Young Children with Autism. This will be used to determine developmentally appropriate goals for intervention.

In addition to these pre- and post-assessments, we will video record weekly 10-minute play interactions between you and your child as well as your child and the researcher. These recordings will take place weekly three to five weeks before intervention, each week during intervention, and several weeks afterwards. These videos will be coded for the frequency of intentional communication (gestures, vocalisations, words, signs etc.), functional play, and joint attention behaviours. The generalisation videos of you and your child will be also be coded using these measures. The videos will be viewed by the research team only and will be used solely for these data collection purposes. You may request access to receive copies of these videos at any time. We anticipate that this research will commence mid-2015 and will continue until the end of 2017. Our involvement with each family will last approximately 8 to 10 months, which includes the time for pre-assessment, intervention (12 weeks) and follow-up for the direct therapy intervention.

# **Confidentiality**

Any information that is obtained in connection with this study and that can be identified with you or your child will remain confidential and will be disclosed only with your permission.

The results of this project will be presented in written and verbal reports, but we will not use your name or the name of your child in any oral or written reports and we will not provide any personal information that would enable anyone to identify you or your child in any reports.

Please note that you are under no obligation to give consent to allow your child to participate in this study. Your decision about whether or not you want to participate will not affect your present or future relationship with Victoria University of Wellington.

If you decide to participate, you have the right to withdraw your consent and discontinue the child in you care's participation until 31 December 2017. Your decision to discontinue participation will not affect your present or future relationship with Victoria University of Wellington.

#### **Ethics**

This research has been assessed and approved by Victoria University of Wellington Human Ethics Committee (Reference Number 22085). If at any time you have any questions or concerns about your treatment as a research participant in this study, contact Dr. Susan Corbett, Chair of the Victoria University of Wellington Human Ethics Committee (Phone: +64 4 463 5480; Email: susan.corbett@vuw.ac.nz).

# **Data Storage and Deletion**

All data will be stored in a locked filing cabinet/password protected computer in a locked office at Victoria University of Wellington. Only members of the research team will have access to this data.

The data will be stored for 5 years after publication and then shredded and thrown away after the 5-year storage period.

# Reporting/Dissemination

The results of this study will be submitted for publication in research and or professional journals and may be presented at a conference. A brief report will be sent to you and all other participants acknowledging participation and outlining overall findings. However, if at any time you would like more detailed feedback, we would be more than happy to provide this either in person, or via the telephone, letter, or email.

# **Direct Therapy Consent Form**

Project Title: Developmental, Relationship-Based Early Intervention for Children with Autism This research has been assessed and approved by Victoria University of Wellington Human Ethics Committee (Reference Number 22085).

To indicate your agreement with each statement below, please tick the corresponding box.

I have read and understood the Information Sheet for this study.

I understand that my involvement in this project includes completing assessments related to my child's skills as well as a questionnaire about my satisfaction with the intervention.

I understand that the involvement of the child in my care includes participation in developmental, relationship-based early intervention delivered by trained researchers and that data will be collected on my child's intentional communication, functional play, and joint attention behaviours.

I understand that the investigators do not foresee any potential physical, psychological, social, legal, or other risks to me or the child in my care as a result of participating in this study.

I understand that the sessions will be videotaped in order for the researchers to collect data on the child in my care's behaviour and that I may request access to these videos.

I understand that my identity and that of the child in my care will not be disclosed in any way stemming from this research.

I understand that all research data will be securely stored at Victoria University of Wellington premises for at least five years, and will be destroyed when no longer required.

I understand that research data gathered for the study may be published provided that my identity and the identity of the child in my care are not disclosed.

I understand that I will receive feedback acknowledging participation and outlining overall findings and that I can request additional feedback at any time.

Any questions that I have asked have been answered to my satisfaction.

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☐ I agree to participate in this investigation and understand that I may withdraw my permission until
31 December 2017 without any negative effect.
☐ I agree to allow the child in my care to participate in this investigation and understand that I may
withdraw my permission until 31 December 2017 without any negative effect.

Name of Parent/Legal Guardian:	Name of Child:
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Parent/Legal Guardian's Signature: Phone Number/Email:

Date:

# Parent Coaching Information Sheet

Project Title: Developmental, Relationship-Based Early Intervention for Children with Autism This research has been assessed and approved by Victoria University of Wellington Human Ethics Committee (Reference Number 22085).

Dear Parent/Legal Guardian,

We would like to invite you and your child to participate in a research study. The purpose of this study is to evaluate the effect of a parent-implemented developmental, relationship-based early intervention on outcomes for children with a diagnosis of Autism Spectrum Disorder (ASD). This intervention will be based on Rogers and Dawsons' Early Start Denver Model (ESDM). This intervention will involve assessing your child's current skills across all areas of development and then providing naturalistic intervention in the context of play-based, relationship-focused routines. This includes both joint activity routines involving objects and sensory social routines such as "peekaboo", "chase", and "hide and seek".

If you give consent for you and your child to participate in this intervention, this will involve one hour of parent coaching per week for 12 weeks each and will take place in the home. This intervention will involve coaching based on the principles of developmental, relationship-based therapy including instruction of the techniques, modelling by the trained researcher, direct practice and feedback. Following the parent coaching we may assess your child's generalisation of his or her skills to an early childhood education context.

Prior to the start of the intervention, we will meet with you to complete several pre-assessments to evaluate your child's skills across developmental domains. These will take approximately two hours to complete. Assessments might include, for example, the MacArthur-Bates Communicative Development Inventories (MCDI) to assess your child's receptive and expressive language, the Vineland Adaptive Behaviour Scales, second Edition (VABS-II) to measure your child's adaptive behaviour, and the Social Communication Questionnaire (SCQ) to assess your child's severity of autism symptoms. We will also use an intervention satisfaction questionnaire, to assess how acceptable and effective you expect the treatment to be. The satisfaction questionnaire will then be repeated after the intervention has finished. Following intervention you will also be invited to participate in an interview (approximately one hour) about your perceptions of the impact of the treatment.

During the first session of intervention we will also directly assess your child's developmental skills using an adapted version of the ESDM Curriculum Checklist for Young Children with Autism. This will be used to determine developmentally appropriate goals for intervention.

In addition to these pre- and post-assessments, we will video record weekly 10-minute play interactions between you and the child. These recordings will take place weekly three to five weeks before intervention, each week during intervention, and several weeks afterwards. We may also film several 10-minute videos of your child interacting with his or her early childcare teacher both before and after intervention. These videos will be coded for the frequency of intentional communication (gestures, vocalisations, words, signs etc.), functional play, and joint attention behaviours. In addition, the videos of you (or the early childcare teacher) and your child will be coded using the ESDM Fidelity Checklist to assess the fidelity of your implementation of the developmental, relationship-based intervention techniques. The videos will be viewed by the research team only and will be used solely for these data collection purposes.

We anticipate that this research will commence mid-2015 and will continue until the end of 2017. Our involvement with each family will last approximately 8 to 10 months, which includes the time for pre-assessment, intervention (12 weeks) and follow-up for both the parent training and direct therapy interventions.

# **Confidentiality**

Any information that is obtained in connection with this study and that can be identified with you or your child will remain confidential and will be disclosed only with your permission.

The results of this project will be presented in written and verbal reports, but we will not use your name or the name of your child in any oral or written reports and we will not provide any personal information that would enable anyone to identify you or your child in any reports.

Please note that you are under no obligation to give consent to allow your child to participate in this study. Your decision about whether or not you want to participate will not affect your present or future relationship with Victoria University of Wellington.

If you decide to participate, you have the right to withdraw your consent and discontinue the child in you care's participation until 31 December 2017. Your decision to discontinue participation will not affect your present or future relationship with Victoria University of Wellington.

#### **Ethics**

This research has been assessed and approved by Victoria University of Wellington Human Ethics Committee (Reference Number 22085). If at any time you have any questions or concerns about your treatment as a research participant in this study, contact Dr. Susan Corbett, Chair of the Victoria

University of Wellington Human Ethics Committee (Phone: +64 4 463 5480; Emailsusan.corbett@vuw.ac.nz).

# **Data Storage and Deletion**

All data will be stored in a locked filing cabinet/password protected computer in a locked office at Victoria University of Wellington. Only members of the research team will have access to this data. The data will be stored for 5 years after publication and then shredded and thrown away after the 5-year storage period.

# Reporting/Dissemination

The results of this study will be submitted for publication in research and or professional journals and may be presented at a conference. A brief report will be sent to you and all other participants acknowledging participation and outlining overall findings. However, if at any time you would like more detailed feedback, we would be more than happy to provide this either in person, or via the telephone, letter, or email.

# Parent Coaching Consent Form

Project Title: Developmental, Relationship-Based Early Intervention for Children with Autism This research has been assessed and approved by Victoria University of Wellington Human Ethics Committee (Reference Number 22085).

To indicate your agreement with each statement below, please tick the corresponding box.

I have read and understood the Information Sheet for this study. I understand that my involvement in this project includes receiving parent coaching to implement developmental, relationship-based early intervention for the child in my care and that data will be collected on my implementation of the intervention procedures.

I understand that the involvement of the child in my care includes participation in developmental, relationship-based early intervention delivered by myself and trained researchers and that data will be collected on my child's intentional communication, functional play, and joint attention behaviours.

I understand that the investigators do not foresee any potential physical, psychological, social, legal, or other risks to me or the child in my care as a result of participating in this study.

I understand that the sessions will be videotaped in order for the researchers to collect data on the child in my care's behaviour and my implementation of the intervention techniques.

I understand that my identity and that of the child in my care will not be disclosed in any way stemming from this research.

I understand that all research data will be securely stored at Victoria University of Wellington premises for at least five years, and will be destroyed when no longer required.

I understand that research data gathered for the study may be published provided that my identity and the identity of the child in my care are not disclosed.

I understand that I will receive feedback acknowledging participation and outlining overall findings and that I can request additional feedback at any time.

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This questions that I have asked have been answered to my satisfaction.
☐ I agree to participate in this investigation and understand that I may withdraw my permission until
31 December 2017 without any negative effect.
☐ I agree to allow the child in my care to participate in this investigation and understand that I may
withdraw my permission until 31 December 2017 without any negative effect.

# Parent Coaching Information Sheet- Family Member

Project Title: Developmental, Relationship-Based Early Intervention for Children with Autism This research has been assessed and approved by Victoria University of Wellington Human Ethics Committee (Reference Number 22085).

Dear Parent/Family member,

We would like to invite you to participate in a research study. The purpose of this study is to evaluate the effect of a parent-implemented developmental, relationship-based early intervention on outcomes for children with a diagnosis of Autism Spectrum Disorder (ASD). This intervention will be based on Rogers and Dawsons' Early Start Denver Model (ESDM).

This intervention will involve assessing your child/family member's current skills across all areas of development and then providing naturalistic intervention in the context of play-based, relationship-focused routines. This includes both joint activity routines involving objects and sensory social routines such as "peekaboo", "chase", and "hide and seek".

One of your child/family member's parents/legal guardians has already consented to participate in this parent coaching study. This will involve one hour of parent coaching per week for 12 weeks and will take place in the home. This intervention will involve coaching based on the principles of developmental, relationship-based therapy including instruction of the techniques, modelling by the trained researcher, direct practice and feedback. Following the parent coaching we will assess your child/family member's generalisation of his/her skills to you (a novel person).

To assess this generalisation of skills we will film one 10-minute video of the child interacting with you before the parent coaching begins and another 10-minute video when the parent coaching ends. These videos will be coded for the frequency of intentional communication (gestures, vocalisations, words, signs etc.), functional play, and joint attention behaviours. In addition, the videos of you and your child/family member will be coded using the ESDM Fidelity Checklist to assess the fidelity of your implementation of the developmental, relationship-based intervention techniques. The videos will be viewed by the research team only and will be used solely for these data collection purposes. We anticipate that this research will commence mid-2015 and will continue until the end of 2017. Our involvement with each family will last approximately 8 to 10 months, which includes the time for pre-assessment, intervention (12 weeks) and follow-up for both the parent training and direct therapy interventions.

# **Confidentiality**

Any information that is obtained in connection with this study and that can be identified with you or your child will remain confidential and will be disclosed only with your permission.

The results of this project will be presented in written and verbal reports, but we will not use your name or the name of your child/family member in any oral or written reports and we will not provide any personal information that would enable anyone to identify you or your child/family member in any reports.

Please note that you are under no obligation to give consent to participate in this study. Your decision about whether or not you want to participate will not affect your present or future relationship with Victoria University of Wellington.

If you decide to participate, you have the right to withdraw your consent and discontinue the child in you care's participation until 31 December 2017. Your decision to discontinue participation will not affect your present or future relationship with Victoria University of Wellington.

#### **Ethics**

This research has been assessed and approved by Victoria University of Wellington Human Ethics Committee (Reference Number 22085). If at any time you have any questions or concerns about your treatment as a research participant in this study, contact Dr. Susan Corbett, Chair of the Victoria University of Wellington Human Ethics Committee (Phone: +64 4 463 5480;

Email: mailto: susan.corbett@vuw.ac.nz).

# **Data Storage and Deletion**

All data will be stored in a locked filing cabinet/password protected computer in a locked office at Victoria University of Wellington. Only members of the research team will have access to this data. The data will be stored for 5 years after publication and then shredded and thrown away after the 5-year storage period.

# Reporting/Dissemination

The results of this study will be submitted for publication in research and or professional journals and may be presented at a conference. A brief report will be sent to you and all other participants acknowledging participation and outlining overall findings. However, if at any time you would like more detailed feedback, we would be more than happy to provide this either in person, or via the telephone, letter, or email.

# Parent Coaching Consent Form- Family Member

Project Title: Developmental, Relationship-Based Early Intervention for Children with Autism This research has been assessed and approved by Victoria University of Wellington Human Ethics Committee (Reference Number 22085).

To indicate your agreement with each statement below, please tick the corresponding box.

I have read and understood the Information Sheet for this study. I understand that my involvement in this project includes playing with my child/family member to allow the research team to assess his/her generalisation of skills learned in the parent coaching programme to a different person and that data will be collected on my implementation of the intervention procedures.

I understand that the investigators do not foresee any potential physical, psychological, social, legal, or other risks to me as a result of participating in this study.

I understand that the sessions will be videotaped in order for the researchers to collect data on my child/family member's behaviour and my implementation of the intervention techniques.

I understand that my identity and that of my child/family member will not be disclosed in any way stemming from this research.

I understand that all research data will be securely stored at Victoria University of Wellington premises for at least five years, and will be destroyed when no longer required.

I understand that research data gathered for the study may be published provided that my identity and the identity of my child/family member are not disclosed.

I understand that I will receive feedback acknowledging participation and outlining overall findings and that I can request additional feedback at any time.

Any questions that I have asked have been answered to my satisfaction.

☐ I agree to participate in this investigation and understand that I may withdraw my	permission unti
31 December 2017 without any negative effect.	

Name:	
Name of Child/Family Member:	
Phone Number/Email:	
Signature:	
Date:	

#### APPENDIX D

#### **ESDM Curriculum Assessment Materials**

- Small table and two straight wooden chairs
- Large beanbag
- Carts with drawers and other containers for holding toys
- Small rugs for floor areas
- A variety of small, clear containers with lids
- Small box with bubbles, balloons, slinkies, animal picture book
- Set of coloured blocks of different sizes
- Set of coloured markers and paper
- Set of farm animals and two identical pictures of farm animals
- Children's book with farm animals, children's book with vehicles
- Two or three cars and trucks
- Bucket that holds two to five balls and beanbags of different sizes
- Nesting cups
- Ring stacker
- Several inset puzzles
- Shape sorter with lid
- Fat pegs and peg boards
- Set of plastic eating objects
- Large doll with clothes
- Baby blanket and small bed or box to use for bed
- Set of personal grooming objects
- Set of popbeads
- Set of large Duplo
- Toy involving a hammer and pegs or balls etc.
- Pop-up toy with various types of buttons to open
- Preferred child snacks
- Fat beads with a string or cord
- Photos of family members and self

# **APPENDIX E Data Collection Studies 1 and 2**

0.00-0.30	□None	□None	□None
	Imitation	Imitation	Imitation
	Utterance (E)	Imitation Utterance (E)	Imitation Utterance (E)
	Utterance (Not E)	Utterance (Not E)	Utterance (Not E)
	☐ Intentional Vocalisation	☐ Intentional Vocalisation	☐ Intentional Vocalisation
	☐ Engagement-partial	☐ Engagement-partial	☐ Engagement-partial
	☐ Engagement- whole 0:10	☐ Engagement- whole 0:20	☐ Engagement- whole 0:30
0.30-1.00	□None	□None	□None
	Imitation	Imitation	Imitation
	Utterance (E)	Utterance (E)	Utterance (E)
	Utterance (Not E)	Utterance (Not E)	Utterance (Not E)
	☐ Intentional Vocalisation	☐ Intentional Vocalisation	☐ Intentional Vocalisation
	☐ Engagement-partial	☐ Engagement-partial	☐ Engagement-partial
	☐ Engagement- whole 0:40	☐ Engagement- whole 0:50	☐ Engagement- whole 1.00
1.00-1.30	□None	□None	□None
	Imitation	Imitation	Imitation
	Utterance (E)	Utterance (E)	Utterance (E)
	Utterance (E) Utterance (Not E)	Utterance (Not E)	Utterance (E) Utterance (Not E)
	☐ Intentional Vocalisation	☐ Intentional Vocalisation	☐ Intentional Vocalisation
	☐ Engagement-partial	☐ Engagement-partial	☐ Engagement-partial
	☐ Engagement- whole 1:10	☐ Engagement- whole 1:20	☐ Engagement- whole 1:30
1.30-2.00	□None	□None	□None
	Imitation	Imitation	Imitation
	Utterance (E)	Utterance (E)	Utterance (E)
	Utterance (Not E)	Utterance (Not E)	Utterance (Not E)
	☐ Intentional Vocalisation	☐ Intentional Vocalisation	☐ Intentional Vocalisation
	☐ Engagement-partial	☐ Engagement-partial	☐ Engagement-partial
	☐ Engagement- whole 1:40	☐ Engagement- whole 1:50	☐ Engagement- whole 2:00
2.00-2.30	□None	□None	□None
	Imitation	Imitation	Imitation
	Utterance (E)	Utterance (E)	Utterance (E)
	Utterance (Not E)	Utterance (Not E)	Utterance (Not E)
	☐ Intentional Vocalisation	☐ Intentional Vocalisation	☐ Intentional Vocalisation
	☐ Engagement-partial	☐ Engagement-partial	☐ Engagement-partial
	☐ Engagement- whole 2:10	☐ Engagement- whole 2:20	☐ Engagement- whole 2:30
2.30-3.00	□None	□None	□None
	Imitation	Imitation	Imitation
	Utterance (E)	Utterance (E)	Utterance (E)
	Utterance (Not E)	Utterance (Not E)	Utterance (Not E)
	☐ Intentional Vocalisation	☐ Intentional Vocalisation	☐ Intentional Vocalisation
	☐ Engagement-partial	☐ Engagement-partial	☐ Engagement-partial
	☐ Engagement- whole 2:40	Engagement- whole 2:50	☐ Engagement- whole 3:00
3.00-3.30	□None	□None	□None
	Imitation	Imitation	Imitation
	Utterance (E)	Utterance (E)	Utterance (E)
	Utterance (Not E)	Utterance (Not E)	Utterance (Not E)
	☐ Intentional Vocalisation	☐ Intentional Vocalisation	☐ Intentional Vocalisation
	☐ Engagement-partial	☐ Engagement-partial	☐ Engagement-partial
	☐ Engagement- whole 3:10	☐ Engagement- whole 3:20	☐ Engagement- whole 3:30

3.30-4.00	□None	□None	□None
	Imitation	Imitation	Imitation
	Utterance (E)	Utterance (E)	Utterance (E)
	Utterance (Not E)	Utterance (Not E)	Utterance (Not E)
	☐ Intentional Vocalisation	☐ Intentional Vocalisation	☐ Intentional Vocalisation
	☐ Engagement-partial	☐ Engagement-partial	☐ Engagement-partial
	☐ Engagement- whole 3:40	☐ Engagement- whole 3:50	☐ Engagement- whole 4:00
4.00-4.30	□None		
4.00-4.30			
	Imitation	Imitation	Imitation Utterance (E)
	Utterance (E)	Utterance (E)	
	Utterance (Not E)  ☐ Intentional Vocalisation	Utterance (Not E)  ☐ Intentional Vocalisation	Utterance (Not E)  ☐ Intentional Vocalisation
	☐ Engagement-partial	☐ Engagement-partial	☐ Engagement-partial
	☐ Engagement- whole 4:10	☐ Engagement- whole 4:20	☐ Engagement- whole 4:30
4.30-5.00	□None	□None	□None
	Imitation	Imitation	Imitation
	Utterance (E)	Utterance (E)	Utterance (E)
	Utterance (Not E)	Utterance (Not E)	Otterance (Not E)
	☐ Intentional Vocalisation	☐ Intentional Vocalisation	☐ Intentional Vocalisation
	☐ Engagement-partial	☐ Engagement-partial	☐ Engagement-partial
	☐ Engagement- whole 4:40	☐ Engagement- whole 4:50	☐ Engagement- whole 5:00
5.00-5.30	□None	□None	□None
	Imitation	Imitation	Imitation
	Utterance (E)	Utterance (E)	Utterance (E)
	Utterance (Not E)	Utterance (Not E)	Utterance (Not E)
	☐ Intentional Vocalisation	☐ Intentional Vocalisation	☐ Intentional Vocalisation
	☐ Engagement-partial	☐ Engagement-partial	☐ Engagement-partial
	☐ Engagement- whole 5:10	☐ Engagement- whole 5:20	☐ Engagement- whole 5:30
5.30-6.00	□None	□None	□None
	Imitation	Imitation	Imitation
	Utterance (E)	Utterance (E)	Utterance (E)
	Utterance (Not E)	Utterance (Not E)	Utterance (Not E)
	☐ Intentional Vocalisation	☐ Intentional Vocalisation	☐ Intentional Vocalisation
	☐ Engagement-partial	☐ Engagement-partial	☐ Engagement-partial
	☐ Engagement- whole 5:40	☐ Engagement- whole 5:50	☐ Engagement- whole 6:00
6.00-6.30	□None	□None	□None
	Imitation	Imitation	Imitation
	Utterance (E)	Utterance (E)	Utterance (E)
	Utterance (Not E)	Utterance (Not E)	Utterance (Not E)
	☐ Intentional Vocalisation	☐ Intentional Vocalisation	☐ Intentional Vocalisation
	☐ Engagement-partial	☐ Engagement-partial	☐ Engagement-partial
	☐ Engagement- whole 6:10	☐ Engagement- whole 6:20	☐ Engagement- whole 6:30
6.30-7.00	□ None	□None	□None
0.50-7.00	Imitation	Imitation	Imitation
	Utterance (E)	Utterance (E)	Utterance (E)
	Utterance (Not E)	Utterance (Not E)	Utterance (Not E)
	☐ Intentional Vocalisation	☐ Intentional Vocalisation	☐ Intentional Vocalisation
	☐ Engagement-partial	☐ Engagement-partial	☐ Engagement-partial
	☐ Engagement whole 6:40	☐ Engagement whole 6:50	☐ Engagement whole 7:00

7:00-7:30	□None	□None	□None
7:00-7:30			
	Imitation	Imitation	Imitation
	Utterance (E)	Utterance (E)	Utterance (E)
	Utterance (Not E)	Utterance (Not E)	Utterance (Not E)
	☐ Intentional Vocalisation	☐ Intentional Vocalisation	☐ Intentional Vocalisation
	☐ Engagement-partial	☐ Engagement-partial	☐ Engagement-partial
	☐ Engagement- whole 7:10	☐ Engagement- whole 7:20	☐ Engagement- whole 7:30
7:30-8:00	□None	□None	□None
	Imitation	Imitation	Imitation
	Utterance (E)	Utterance (E)	Utterance (E)
	Utterance (Not E)	Utterance (Not E)	Utterance (Not E)
	☐ Intentional Vocalisation	☐ Intentional Vocalisation	☐ Intentional Vocalisation
	☐ Engagement-partial	☐ Engagement-partial	☐ Engagement-partial
	☐ Engagement- whole 7:40	☐ Engagement- whole 7:50	☐ Engagement- whole 8.00
8.00-8.30	□None	□None	□None
	Imitation	Imitation	Imitation
		Utterance (E)	Utterance (E)
	Utterance (E) Utterance (Not E)	Utterance (E) Utterance (Not E)	Utterance (E) Utterance (Not E)
	☐ Intentional Vocalisation	☐ Intentional Vocalisation	☐ Intentional Vocalisation
	☐ Engagement-partial	☐ Engagement-partial	☐ Engagement-partial
	☐ Engagement- whole 8:10	☐ Engagement- whole 8:20	☐ Engagement- whole 8:30
8.30-9.00	□None	□None	□None
	Imitation	Imitation	Imitation
	Utterance (E)	Utterance (E)	Utterance (E)
	Utterance (Not E)	Utterance (Not E)	Utterance (Not E)
	☐ Intentional Vocalisation	☐ Intentional Vocalisation	☐ Intentional Vocalisation
	☐ Engagement-partial	☐ Engagement-partial	☐ Engagement-partial
	☐ Engagement- whole 8:40	☐ Engagement- whole 8:50	☐ Engagement- whole 9.00
9.00-9.30	□None	□None	□None
	Imitation	Imitation	Imitation
	Utterance (E)	Utterance (E)	Utterance (E)
	Utterance (E) Utterance (Not E)	Utterance (E) Utterance (Not E)	Utterance (E) Utterance (Not E)
	☐ Intentional Vocalisation	☐ Intentional Vocalisation	☐ Intentional Vocalisation
	☐ Engagement-partial	☐ Engagement-partial	☐ Engagement-partial
	☐ Engagement- whole 9:10	☐ Engagement- whole 9:20	☐ Engagement- whole 9:30
9.30-10.00	□None	□None	□None
	Imitation	Imitation	Imitation
	Utterance (E)	Utterance (E)	Utterance (E)
	Utterance (Not E)	Utterance (Not E)	Utterance (Not E)
	☐ Intentional Vocalisation	☐ Intentional Vocalisation	☐ Intentional Vocalisation
	☐ Engagement-partial	☐ Engagement-partial	☐ Engagement-partial
	☐ Engagement- whole 9:40	☐ Engagement- whole 9:50	☐ Engagement- whole 10.00

# APPENDIX F

# **Modified ESDM Parent/Therapist Fidelity Checklist**

1. Management of attention	
- Adult attracts attention to face and auditory cues (e.g., holds items near the face, waits until child is paying	$\square$ Never (0) $\square$ Occ. (3) $\square$ Sometimes (6) $\square$ Usually (9) $\square$ Con. (12)
attention to give the cue):	
2. ABC Format	
-Teaching opportunities occurred more than once every 30s	$\square$ Never (0) $\square$ Occ. (1) $\square$ Sometimes (2) $\square$ Usually (3) $\square$ Con. (4)
-There were clear antecedents (the child knows what behaviour is expected, cues are not repeated)	$\square$ Never (0) $\square$ Occ. (1) $\square$ Sometimes (2) $\square$ Usually (3) $\square$ Con. (4)
-Correct behaviours and attempts were reinforced	$\square$ Never (0) $\square$ Occ. (1) $\square$ Sometimes (2) $\square$ Usually (3) $\square$ Con. (4)
3. <u>Instructional techniques</u>	
Least-to-most prompting was used (Verbal and gestural before physical, follow through on antecedents):	$\square$ Never (0) $\square$ Occ. (3) $\square$ Sometimes (6) $\square$ Usually (9) $\square$ Con. (12)
4. Affect and Arousal	
Problems with affect and arousal (e.g. child too excited, too high energy, or bored/low energy) are managed	$\square$ Never (0) $\square$ Occ. (3) $\square$ Sometimes (6) $\square$ Usually (9) $\square$ Con. (12)
skilfully (score full marks if no problems with affect/arousal)	
5. Management of unwanted behaviours	
Unwanted behaviours (e.g. crying, screaming, aggression, property destruction) are managed using positive	$\square$ Never (0) $\square$ Occ. (3) $\square$ Sometimes (6) $\square$ Usually (9) $\square$ Con. (12)
techniques to redirect the child, elicit a more positive behaviour and re-establish engagement and positive affect	
(score full marks if no unwanted behaviours)	
6. Dyadic engagement	
-Both partners (child and therapist) lead and follow e.g. Child cues adult turns, provides play ideas etc.	$\square$ Never (0) $\square$ Occ. (3) $\square$ Sometimes (6) $\square$ Usually (9) $\square$ Con. (12)
7. Motivation	
-Child is given a choice of materials and appears highly motivated by the task (sometimes=motivated no choice,	$\square$ Never (0) $\square$ Occ. (1.5) $\square$ Sometimes (3) $\square$ Usually (4.5) $\square$ Con. (6)
or choices but difficult to motivate)	$\square$ Never (0) $\square$ Occ. (1.5) $\square$ Sometimes (3) $\square$ Usually (4.5) $\square$ Con. (6)

-There is a mix of maintenance and acquisition tasks	
8. Positive Affect	$\square$ Never (0) $\square$ Occ. (3) $\square$ Sometimes (6) $\square$ Usually (9) $\square$ Con. (12)
Adult has rich genuine positive affect which is matched by child positive affect	
9. Sensitivity and responsivity	□Never (0) □Occ. (3) □Sometimes (6) □Usually (9) □Con. (12)
The adult reads the child very well and makes every effort to interpret the child's meaning	
10. Multiple and varied communication	$\square$ None (0) $\square$ One (3) $\square$ Two (6) $\square$ Three (9) $\square$ Four+ (12)
Opportunities were given for: requesting, commenting, naming, protesting, seeking help, finishing, greeting,	
imitating voice or gesture	
11. Appropriateness of adult language	$\square$ Never (0) $\square$ Occ. (3) $\square$ Sometimes (6) $\square$ Usually (9) $\square$ Con. (12)
Adult generally uses the one-up rule (using one more word than the child)- (Sometimes= roughly same number of	
words, Occasionally/never=far too many or few)	
12. Joint activity structure and elaboration	
Activity has a clear set-up phase (N/A if set up phase is not included in the video)	$\square$ N/A $\square$ Never (0) $\square$ Occ. (1) $\square$ Sometimes (2) $\square$ Usually (3) $\square$ Con. (4)
Activity include sufficient elaborations	$\square$ Never (0) $\square$ Occ. (1) $\square$ Sometimes (2) $\square$ Usually (3) $\square$ Con. (4)
Activity has a clear clean up phase (N/A if clean up phase is not included in the video)	$\square$ N/A $\square$ Never (0) $\square$ Occ. (1) $\square$ Sometimes (2) $\square$ Usually (3) $\square$ Con. (4)
13. Transition between activities	$\square$ N/A $\square$ Never (0) $\square$ Occ. (3) $\square$ Sometimes (6) $\square$ Usually (9) $\square$ Con.
When a child finishes an activity he/she is quickly given a choice of a new one (Sometimes=quick no choice)	(12)
<u>Total Score</u>	/156

 ${\bf APPENDIX}~G$  Procedural Integrity Baseline/Follow-up Checklists Studies 1 and 2

Procedures- Baseline Study 1	Procedural i	ntegrity
1. a. box of toys was made available to participant at the start of each session	/1	
b. If no toy was taken after 10-s then toys were removed from the box and	/1	N/A
given to the participant		
c. If participant did not take toy then research chose toy for participant	/1	N/A
2. Researcher included naturalistic probes of imitation and/or language on	/20	
average every 30s		
(-1 for each probe above this number)		
3. Researcher responded appropriately to any child attempts to initiate play	/1	
or interaction		
4. No prompting was used and no specific developmental skills were	/1	
targeted		
5. Session lasted 10 minutes from child taking the toy	/1	

NB/ steps were taken to block/prevent any challenging or dangerous behaviours e.g. putting materials in the child's mouth, hitting the therapist or throwing the materials. Also, if child was missing materials from the activity, the therapist would provide the child with the materials, e.g. pens for paper, pieces for a puzzle.

Procedures Generalisation-Study 1	Procedural integrity
1. Box of toys was made available to participant at the start of each session	/1
2. Session lasted 10 minutes from child taking the toy	/1

Procedures Intervention- Study 2	
Parent and therapist discuss progress from previous week (can occur	•
after demonstration of previous week's skills)	
Parent demonstrates previous weeks skills for ten minutes	•
Therapist provides feedback on previous weeks skills including	
1. At least two positive comments	•
2. At least one area to work on	•
3. Modelling any of the skills with the child if necessary	•
Therapist covers PowerPoint presentation of new chapter including:	
1. Briefly discusses the overall topic and its importance	•
2. Covers all information on each slide	•
3. Places emphasis on child's target skills	•
4. Ensures that parent understands the content	•
5. Recaps the content	•
6. Discussion lasts for approximately 20-30 minutes	•
Parent and therapist discuss goals for the following week	•
Therapist gives parent checklist of goals for the following week	•
Therapist models new skills with child for approximately 3-10	•
minutes	
Parent practices new skills with child for approximately 3-10 minutes	•
Therapist provides feedback on the play including	
1. At least two positive comments	•
2. At least one area to work on	•
3. Modelling any of the skills with the child if necessary	•

Procedures Baseline/Generalisation/Follow-Up-Study 2	Procedural integrity
1. The mother was present at the start of filming	/1
2. Session lasted 10 minutes	/1
3. Researcher did not provide any feedback to the parent	/1
4. Additional parents/adults did not interact or play with the child for more	/1
than 30s	, 1
uiaii 508	

#### **APPENDIX H**

# **Post-intervention Interview Questions Study 2**

# **Background**

Can you please tell me a little bit about your child?

Why did you want to get involved in this parent training?

Compared to other children with autism, how high do you think your child's needs are?

What are your main goals for your child?

*If unsure prompt (goals related to behaviour, communication social skills)* 

# **Understanding**

What parts of this training do you feel you've understood well?

Are there any parts you feel you don't understand as well as the other parts?

How can we make it more understandable for you or other parents in the future?/any particular improvements to what we could do?

# **Effectiveness**

How effective was this training on your ability to play and interact with your child?

How effective was this training on your child's play skills? Communication? Interaction with others? Overall behaviour?

What aspects of this training have you found challenging or ineffective?

Do you have any suggestions for how to make the training more effective in the future?

Specifically:

Do you think 12 weeks was a good length of time? If not, would more or less be better? Why?

Do you think an hour to an hour and a half a week was a good amount of time? If not, would more or less be better? Why?

Do you think anyone else should have been included in the training? If so who and why?

Do you think the training should have happened in any other places? If so, where and why?

# Teaching procedures

Now we are going to talk about the teaching procedures of training. How did you find the way the therapist worked with you?

Are there any things about the way the therapist worked with you that you would change?

Specifically:

Would you make any changes to the PowerPoint?

Use of the manual?

Therapist modelling the skills with your child?

Practicing the skills with your child?

Therapist providing feedback on your skills?

Were any of these teaching procedures particularly helpful?

Were there any that you didn't find very helpful?

Were there any chapters in the book/topics that you found particularly useful for yourself and/or child?

Were there any chapters in the book that were not useful for yourself/your child?

# Willingness

What are the reasons that you would be willing to continue with this training?

Are there reasons why you would not be willing to continue this training?

What would make you more willing in the future?

# Recommendation

Would you recommend this training to other people? Why or why not.

Are there any aspects of this training that you would change before recommending it to someone else?

# Side-effects

Were there any negative side-effects on your child's behaviour as result of this training?

Whats could be done in the future to minimise these side-effects?

# **Disruptiveness**

Did you find this training too disruptive in your daily life. If so, why? Both the actual training and implementing the training

If it was not disruptive, can you tell me why?

How can we make it less disruptive in the future?

Do you have any questions for me?

# APPENDIX I

Theme	Category	Code	Example of significant statement
Effect on child outcomes	Strengths	Overall effectiveness	Rick's mother: "this is by far the most impact, in a good way, of any professional that we've interacted with, in our whole time since he was 3.5."
			Sean's mother: "and to be honest we actually think that this [the training] is the best thing that's happened so far, because"
			Dean's mother: "I think he's okay, he's okay. He's better."
		Expressive language	Rick's mother: "he talked about himself as 'Rick' all the time and we did that too, we would say, oh 'It's Rick's turn, Rick does this', so once [the therapist] made me aware that we were doing that and we stopped, it only took him like a week he started saying 'you'"
			Alex's mother: "it's good that he can communicate with us instead of getting upset, he knows that he can touch something that he wants".
			Dean's mother, when asked about his talking: "he improve a lot, make me feel good".
			Sean's mother: "because we've got him, he's talking, he's saying things now"
			Idris' mother: "he's taking the ball to us and pulling our hand and asking him to jump on that."
		Receptive language	Sean's mother: "But yeah a lot of it's still not guaranteed that you're going to get an answer when you try to talk to him but there's definitely a better chance."
			Dean's mother: "Like he can understand more, yeah"

#### **APPENDIX J**



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# MEMORANDUM

то	Larah van der Meer			
COPY TO	TO Jeff Sigafoos, Hannah Waddington, Laura Roche, Amarie Carne Catriona Barclay, Jessica Sutherland			
FROM	AProf Susan Corbett, Convener, Human Ethics Committee			
DATE	19 June 2015			
PAGES	1			
SUBJECT	Ethics Approval: 22085 Developmental, Relationship-Based Early Intervention for Children with Autism			

Thank you for your application for ethical approval, which has now been considered by the Standing Committee of the Human Ethics Committee.

Your application has been approved from the above date and this approval continues until 31 March 2018. If your data collection is not completed by this date you should apply to the Human Ethics Committee for an extension to this approval.

Best wishes with the research.

Kind regards



Susan Corbett

Convener, Victoria University Human Ethics Committee