Fostering Development in Young Children with Autism Spectrum Disorder and Intellectual Disability

A Center-based Discrete Trial Approach

Nienke C. Peters-Scheffer

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Surrounded by walls

that would not only guard us

but lock us inside

eventually

(Kayak, 2008)

Opgedragen aan mijn vader:

Karel Herman Scheffer

1953 - 2009

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Chapter 1

General Introduction

Preamble

With a prevalence rate of six to seven in thousand children, Autism Spectrum Disorder (ASD) is one of the most common childhood developmental disorders (Fombonne, 2009). Although during adolescence and adulthood the core symptoms of ASD seem to decrease over time, ASD is seen as a chronic disability with a poor prognosis pertaining development. Most individuals with ASD require professional care throughout their lives (Howlin, Goode, Hutton, & Rutter, 2004; Mordre et al., 2012; Seltzer et al., 2003).

ASD and intellectual disability (ID) often co-occur and greater severity of one of these two disorders seems to affect the other disorder on a multitude of elements including the presentation of core symptoms (communication, social interaction, stereotypic behavior) and associated features such as gender, challenging behavior, medical conditions as well as the course of the disorder (Matson & Shoemaker, 2009; O'Brien & Pearson, 2004; Shattuck et al., 2007). Furthermore, these co-occurring disorders have a negative impact on treatment efficacy and overall development (Ben-Itzchak, Lahat, Burgin, & Zachor, 2008; Eikeseth, 2009). Diagnosing ASD in individuals with ID is sometimes challenging as symptoms of both disorders overlap since individuals with ID often display autistic behaviors such as absent or delayed speech, stereotyped movements and poor social connectedness (Hartley & Sikora, 2010).

Although persons with ASD and ID distinctly differ from persons with only ASD or only ID and may have other needs, much of the recent advances in ASD research have been with persons without ID limiting the ability to generalize study findings to children with ASD and ID (Matson & Shoemaker, 2009). Research regarding intervention for this population, as well as research aiming to understand these co-occurring disorders is warranted.

Several studies on early intensive behavioral intervention (EIBI) have yielded encouraging results in improving children's cognitive, adaptive, and social functioning (e.g., Lovaas, 1987; Sallows & Graupner, 2005). Although EIBI research in children with ASD and ID is limited (see for an exception: Smith, Eikeseth, Klevstrand, & Lovaas, 1997), currently EIBI seems the treatment of choice for children with ASD with and without ID (Eikeseth, 2009). This thesis focuses on the development and behavioral treatment of children with ASD and ID. The first part centers on the behavioral characteristics of children with ASD and ID, while in the second and third part the effectiveness of early behavioral treatment and its current state of the art are examined.

1.1 Children with ASD and ID

ASD is used as an umbrella term referring to individuals with autistic disorder (AD), Asperger syndrome, and Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS). Childhood Disintegrative Disorder is sometimes excluded from ASD due to its distinct developmental course and Rett due to its distinct etiology.

ASD is characterized by restricted patterns of behavior and interests and qualitative impairments in communication and social interaction. These impairments emerge early and persist in development even though their precise manifestation changes over the course of development. A large variability in behavioral and cognitive characteristics between individuals with ASD is seen. Currently, no biological marker exists and ASD is diagnosed based on the behavioral phenotype (American Psychiatric Association, 2000; Fombonne, 2009).

In approximately 50 to 70 percent of the individuals with ASD an ID is present (Matson & Shoemaker, 2009), which is characterized by a significant impairment in cognitive (i.e., an IQ below 70) and adaptive behavior with an onset before 18 years of age (American Psychiatric Association, 2000). Between 17 to 40 percent of the individuals with ID has ASD (Bryson, Bradley, Thompson, & Wainwright, 2008; De Bildt, Sytema, Kraijer, & Minderaa, 2005). Beside the fact that ASD and ID are associated, both conditions severely affect learning and development (O'Brien & Pearson, 2004). Associated medical conditions, particularly neurological or chromosomal disorders, are estimated to be present in approximately 10 to 25 percent of all cases with ASD and this prevalence is probably higher in individuals with ASD and ID (Gillberg & Bildstedt 2000; O'Brien & Pearson, 2004).

The presence and severity of ASD and ID are both associated with the core deficits of ASD. In both individuals with ASD and individuals with ID verbal and non-verbal communication is impaired and development of speech and language is highly correlated with IQ. In addition, ASD as well as ID are associated with impairments in social interaction and individuals with ASD and ID show greater impairments than those with ASD or ID only (Matson, Mayville, Lott, Bielecki, & Logan, 2003; Wilkins & Matson, 2009). Severity of ID is associated with the degree of social impairment, with individuals with severe ID displaying greater social impairments (Sevin et al., 1995). Stereotypic behavior is associated with ASD as well as ID and research suggests distinct symptom profiles based on IQ. Children with an IQ below 70 display more motor stereotyped behavior (e.g., hand flapping) and sensory abnormalities, while children with an IQ above 70 display more complex repetitive behaviors such as circumscribed interests or rituals (O'Brien & Pearson, 2004).

Adaptive behavior (i.e., behavior that promotes independence, social acceptability and quality of life; Matson et al., 2003) is impaired in individuals with ASD and in individuals with ID. In general, individuals with ASD and ID display less adaptive behavior across all domains (i.e., communication, daily living skills, and socialization) than individuals with ID. Level of adaptive behavior decreases as severity of ASD symptoms increases (Carpentieri & Morgan, 1996; Matson, Dempsey, & Fodstad, 2009; Matson, Rivet, Fodstad, Dempsey, & Boisjoli, 2009). Adaptive behavior profiles show that social skills are more impaired in individuals with ASD and ID than in individuals with ID only (Smith & Matson, 2010).

The presence and severity of ASD and ID are associated with an increased risk of psychiatric and behavioral conditions (Lovullo & Matson, 2009). For instance, Totsika, Hastings, Emerson, Berridge, and Lancaster (2011) found that behavioral and emotional problems were more frequent among children with ASD and ID, followed by children with ASD and children with ID only as compared to their typically developing peers.

Bradley, Summers, Wood, and Bryson (2004) found higher rates of psychopathology (e.g., anxiety, mood disorders, sleep problems, stereotyped behavior and tics) and organic syndromes in persons with ASD and ID than in persons with ID only. Furthermore, ASD and ID are seen as major risk factors for challenging behavior such as aggression, self-injurious behavior, tantrums and stereotyped behavior. The incidence of challenging behavior increases as IQ decreases and a diagnosis of ASD in addition to ID increases the risk of challenging behavior. This is worrisome as challenging behavior has a tendency to persist over time and puts the individual at risk for abuse, poor social adjustment, exclusion from social situations, inappropriate treatment including medication overuse, referral to facility, and results in more caregiver stress than the core symptoms of ASD and ID (Emerson et al., 2001; Matson & Horovitz, 2010; Murphy et al., 2005).

In sum, individuals with ASD and ID show greater impairments as compared to individuals with ASD or ID only. Consequently, their long-term prognosis is poor and an even worse prognosis is found in individuals with lower IQ. Prognosis is further influenced by physical disability, self-perception and environmental factors such as parental and family coping, service engagement, and societal acceptances (see for a review: O'Brien, 2001). Given the symptoms of individuals with ASD and ID and the additional psychiatric and behavioral problems seen in this population, it is not surprising that many parents report heightened levels of parental stress (e.g., Dabrowska & Pisula, 2010; Eisenhower, Baker, & Blacher, 2005; Griffith, Hasting, Nash, & Hill, 2010).

Distinguishing between ASD specific symptoms and ID is difficult and it is challenging to attribute the absence of social and communication skills and stereotyped and repetitive behavior in children with for example profound ID (IQ < 20) to their overall cognitive impairment or to additional ASD. As stated by Howlin (2000) their care and outcome is dominated by the severity of ID, making the diagnosis to some extent a theoretical matter. However, in general additional ASD affects the prognosis and intervention of individuals with ID. Their intervention should be even more individualized, specialized and structured than those of individuals with the same severity of ID but without ASD.

Given the severity of symptoms and their effect on development, early and intensive treatment for children with ASD and ID is required. As younger children with ASD and/or ID may not have fallen as far behind their peers and have more behavioral and neural plasticity than older children with ASD and/or ID, intervention should commence as early as possible. In addition, learned skills such as those related to imitation and social interaction may facilitate social learning and have a cascading effect on the development of children with ASD and ID.

1.2 Treatment options for children with ASD and ID

A large number of treatments have been developed for individuals with ASD (e.g., applied behavior analysis, diets and vitamins, floor time, holding, medication, neurofeedback, Options, Picture Exchange Communication System, sensory integration, speech and music therapy, special education and visual schedules; Green, Pituch, et al., 2006; Hess, Morrier, Heflin, & Ivey, 2008).

For most treatments, little empirical evidence or even clinical consensus is provided (Foxx, 2008; Schechtman, 2007) and many children with ASD receive ineffective treatments or no treatment at all. Since interventions are costly in both time and money for families and society and may have adverse effects on the development and wellbeing of the children with ASD (see for an example Chelation therapy: Sinha, Silove, & Williams, 2006), research investigating effective treatment options for children with ASD is warranted to assist parents and professionals in making informed decisions regarding the treatment of their child. Building on research from the 1960s, EIBI is the most frequent researched treatment for children with ASD and currently enjoys the strongest research validation for effectiveness in children with ASD with and without ID (Eikeseth, 2009; Matson & Smith, 2008; Vismara & Rogers, 2010).

1.2.1 Early Intensive Behavioral Intervention

In two studies, Ferster and DeMyer (1961; 1962) found that instead of social stimuli such as praise and attention, tangible reinforcers such as food could be used to teach children with ASD new behaviors. These studies showed that ASD may be seen as a failure to learn from the social cues in the environment and that principles derived from learning theory may be applicable in the education of children with ASD. Over the next decades, multiple studies using principles of learning theory have been conducted to teach individuals with developmental disabilities new behaviors or reduce challenging behavior (see for reviews: Granpeesheh, Tarbox, & Dixon, 2009; Matson, Hattier, & Belva, 2011).

A basic assumption in the studies on behavioral treatment is that everything that people do can be considered behavior (including verbal and nonverbal communication) and that the consequences of behavior can either strengthen or weaken it. Behavior will be strengthened if it is followed by a desirable consequence, while behavior weakens if it is followed by an aversive consequence or by withholding a desirable consequence. By systematically arranging the environment by presenting specific consequences after behavior, the probability of future behavior increases or decreases depending on the type of consequence following that behavior (Granpeesheh, Tarbox, et al., 2009). This is called operant conditioning.

Based on these principles, several early intervention programs have appeared and although slight differences between programs exist, all programs use operant conditioning and other learning principles derived from the learning theory including discrete trial instruction, error-correction, fading, functional analysis, generalization, incidental teaching, modeling, prompting, reinforcement, shaping, stimulus control, stimulus and response generalization, and task analysis (see e.g., Duker, Didden, & Sigafoos, 2004). To address pre academic skills (e.g., attending, imitation, matching), daily living skills, language, social skills and behavioral problems (e.g., stereotypy, self-injury) in these programs comprehensive developmentally sequenced curricula are used which are individualized to each child's strengths and deficits.

Treatment is provided in a one-to-one setting and is intensive, that is 20 to 40 hours of treatment per week with additional informal instructions and practice during the other waking hours for a minimum of two years (Green, Brennan, & Fein, 2002; Lovaas, 2003; Love, Carr, Almason, & Petursdottir, 2009; Matson & Smith, 2008).

An important teaching technique used in many EIBI programs and particularly in the early stages of treatment is Discrete Trial Teaching (DTT), in which skills are broken down into smaller (sub) skills, and taught in a restricted operant arrangement called a discrete trial. During a DTT session many learning opportunities (e.g., up to 10 per minute) are provided to the child and the structured teaching format aides the child in isolating the key components of the learning situation by breaking down the complex behavior into units which can be easily discriminated. The skills learned need to be actively generalized to other persons, materials and situations. Therefore, DTT is usually combined with more naturalistic approaches such as incidental teaching (Harris & Delmolino, 2002; Jensen & Sinclair, 2002; Lovaas, 2003; Smith, 2001).

1.3 The present research

In this thesis, results are presented of studies on (1) behavioral characteristics of children with ASD and ID, (2) effectiveness of early behavioral treatment, and (3) health care conditions required for effective behavioral treatment for children with ASD and ID. One meta-analysis and several studies in children with ASD and ID are conducted and aims and methods are shortly described below.

1.3.1 Behavioral characteristics of children with ASD and ID

During social interaction, typically developing children learn by imitation of others, which creates more learning opportunities than individual learning by trial and error. Anticipating other people's goals and intentions, children select from whom, when and what kind of behaviors to imitate and mix imitation and self-discovery to learn new behavior (Meltzoff, Kuhl, Movellan, & Sejnowski, 2009).

Among the earliest signs of ASD are deficits in processing social and emotional information (e.g., American Psychiatric Association, 2000; Baron-Cohen, 2001; Woods & Wetherby, 2003). For example, around their first birthday infants later diagnosed with ASD look less often at others than typically developing children (Osterling & Dawson, 1994; Osterling, Dawson, & Munson, 2002) or children with general learning difficulties (Osterling et al., 2002). As opposed to typically developing children, eye-tracking studies show that children with ASD fixate more on body parts and objects instead of faces (Klin, Jones, Schutlz, Volkmar, & Cohen, 2002; Speer, Cook, McMahon, & Clark, 2007).

In contrast to typically developing children and children with other developmental disabilities, children with ASD do not show a differential brain response to their mother's face as compared to unfamiliar faces.

However, their brain response after seeing a favorite object differs from brain responses to unfamiliar objects. This may indicate a selective impairment in social attention in ASD (Dawson, Carver, et al., 2002).

In addition, children with ASD show a strong preference for highly contingent, non-variable contingency feedback instead of the imperfect contingent feedback that is characteristic for social interactions (Gergely & Watson, 1999). Greene et al. (2011) showed that children and adolescents with ASD and typical development show similar social orienting behavior, but that in individuals with ASD social cues do not receive the same privileged status as these cues have in individuals with typical development. According to the authors, individuals with ASD use non-social mechanisms to process social cues and as these non-social mechanism may not function efficiently this may explain the altered social processing. Using fMRI, Scott-Van Zeeland, Dapretto, Ghahremani, Poldrack, and Brookheimer (2010) found that in contrast to typically developing children, children with ASD do not experience social stimuli as rewarding.

Results of these studies are in line with the social motivation theory (Dawson, Webb, & McPartland, 2005; Schultz, 2005), which states that the lack of reward associated with social stimuli causes children with ASD to spend reduced time attending to faces, speech and other social stimuli and thereby acquiring less knowledge in processing non-verbal behavior and speech (Dawson, Meltzoff, Osterling, Rinaldi, & Brown, 1998; Kuhl, Coffey-Corina, Padden, & Dawson, 2005). As a result, children with ASD may become less proficient in recognizing and understanding the intentions of other people, which places them at increased risk for misinterpreting others' social cues. This might explain the difficulties that children with ASD have in explaining, predicting and influencing others' behavior and communication and why a subset of children with ASD fail to learn from their natural environment (Bredekamp & Copple, 1997). Beginning in the first year of life, these impairments in social attention and processing may result in a deprivation in social learning and have a cascading effect on the development of children with ASD, especially as the understanding of others as intentional beings is seen as a necessary precursor for acquiring a Theory of Mind (Dawson, Carver, et al., 2002; Tomasello, 1995). Results from both behavioral as fMRI studies support the presence of a selective impairment in social processing in children with ASD (e.g., Dawson et al., 1998, Dawson, Carver, et al., 2002; Greene et al., 2011; Klin et al., 2002). However, few studies have focused on how children with ASD and ID interpret social cues and research addressing the understanding of intentions in children with ASD and ID is warranted.

Next to difficulties in communication and social interaction, individuals with ASD show a restricted repertoire of activities and interests (American Psychiatric Association, 2000). In natural settings, deficits in flexibility are frequently reported in individuals with ASD (Gioia, Isquith, Kenworthy, & Barton, 2002), but studies using neuropsychological tests (e.g., Wisconsin Card Sorting Test and the Trail Making Test) or other experimental cognitive paradigms have yielded inconsistent findings (Corbett, Constantine, Hendren, Rocke, & Ozonoff, 2009; Hill & Bird, 2006; Lopez, Lincoln, Ozonoff, & Lai, 2005; South, Ozonoff, & McMahon, 2007). A deficit in flexibility is one of the core features of ASD.

However, our knowledge base on the nature and extent of flexibility is scarce, especially about situations in which children with ASD and ID show a lack of flexibility (Geurts, Corbett, & Soloman, 2009).

One of the few measures to assess behavioral flexibility is the Behavior Flexibility Rating Scale (BFRS), which was developed by Green and her colleagues (Green, Sigafoos et al., 2006; 2007; Pituch et al., 2007) to identify specific situations in which individuals with developmental disabilities show an insistence on sameness. In 968 individuals with AD, Asperger and Down syndrome, Pituch and colleagues (2007) assessed the factor structure of the BFRS and found two factors: interruption/disruption and position/location.

The BFRS was administered in individuals with autism, Asperger syndrome, Down syndrome, Angelman syndrome and non-specific ID. Results of these studies (Green, Sigafoos et al. 2006; Didden et al., 2008) showed that individuals with AD and Asperger syndrome showed significantly more problems in behavioral flexibility than individuals with other diagnoses. However, both studies used the same participants with ASD, were not able to ensure the diagnosis of the sample due to limitations with the data collection methods and no data were collected on child factors that might predict and/or be associated with behavioral flexibility.

In contrast to many other studies published in the area of ASD, studies described in this thesis focus on the behavioral characteristics of children with ASD and ID instead of cognitive features that may underlie their functioning. Three major cognitive theories underlying the behavioral symptoms of ASD have been developed, namely: Theory of Mind deficit, executive dysfunctioning and a weak central coherence (Rajandran & Mitchell, 2007). Theory of Mind (TOM) refers to the everyday ability to infer what others think, believe, and desire in order to explain and predict their behavior, and it is assumed that individuals with ASD fail to understand the mental states of others. This may explain difficulties in joint attention, pretend play, and social interaction (Baron-Cohen, Tager-Flusberg, & Cohen, 1993). As opposed to typically developing individuals who show a preference for integrated and global processing, individuals with ASD show disturbed information processing characterized by a focus on details. This theory is referred to as the central coherence theory (CC) and may be used to explain a preoccupation with details and parts (Happé, 1997). The executive dysfunctioning theory (EF) proposes that individuals with ASD are impaired in several higher-level capacities necessary for the control of actions such as planning and monitoring of behavior, set shifting, inhibiting automatic action and holding information on-line in working memory. These impairments in executive functioning may underlie several social and non-social impairments including the repetitive and restrictive behaviors seen in ASD (Hill, 2004).

Applying these theories to young children with ASD and ID is challenging given the difficulties in distinguishing the unique contribution of ASD and ID to the impairments associated with TOM, EF and CC. For example, in a meta-analysis Yirmiya, Eral, Shaked, and Solomonica-Levi (1998) conclude that individuals with ASD and individuals with ID both show a limited understanding of TOM as compared to typically developing children, but that this impairment is more severe in ASD.

According to the authors, this suggests that the severity of the impairment rather than the impairment itself is unique in ASD.

Although these theories are valuable in interpreting behavioral manifestations in individuals with ASD and ID, applying these theories to (very) young children is controversial. Given the limited cognitive development of young children with ASD and ID, it is expected that the skills associated with TOM, CC, and EF are not present in their fully developed form and it is unknown to which extend findings on TOM, CC and EF in older and/or higher functioning populations can be generalized to preschool children with ASD and ID. For example, Happé (1995) found that typically developing 4-5 year-old children succeed in TOM tasks as compared to a developmental age of 9 years in children with ASD. Hence, the developmental age in which children with ASD succeed in TOM tasks is substantially higher than the developmental age of preschool children with ASD and ID.

In preschoolers with ASD and ID deficits related to EF may be seen as a result of a general developmental delay rather than being specific to ASD. For example, Dawson, Munson, et al. (2002) compared children with ASD to children with developmental delays and typically developing children with a comparable developmental age and found no significant differences on six EF tasks. In a longitudinal study, Griffith, Pennington, Wehner, and Rogers (1999) found neither group differences between preschoolers with ASD and a matched control group on eight EF tasks, nor differences in the performance on the Spatial Reversal task over the course of a year.

In sum, assessment of impairments related to TOM, EF, and CC is complex in preschool children with ASD and ID, particularly as in this population a variable development is seen and children are not able to participate in extensive and structured tests. Only few standardized tasks are available and evidence for their validity and reliability is lacking (Isquith, Crawford, Espy, & Gioir, 2005).

Our focus on behavioral characteristics is in line with the behavioral approach studied in Part II of this thesis and in which treatment addresses the behavioral characteristics of children with ASD with and without ID. While most other interventions focus on the underlying mechanisms of ASD by manipulating the variables that precede behavior (stimulus control), behavioral treatments manipulate the antecedents and consequences of behavior to shape new behavior to lessen the severity of ASD and ID on the child's daily functioning.

In Part 1, we will focus on two behavioral characteristics that may have a severe impact on the development of preschool children with ASD and ID. Our first aim is to determine whether children with ASD and ID see others as intentional beings who initiate and react to goal-directed actions. This will be assessed by measuring the understanding of other people's intentions and exploring its associated variables. Next, we examine the potential of the revised Behavioral Flexibility Rating Scale (BFRS-R) in assessing behavioral flexibility in every day behavior of children with developmental disability, including children with ASD and ID. In addition, we determine to which extent children with ASD and ID experience difficulties regarding behavioral flexibility and explore its associated variables.

Behavioral characteristics of children with ASD and ID do not only affect their own development, but also that of their parents. Parents of children with ASD and ID experience more parental stress than parents of typically developing children or children with other disabilities (Griffith et al., 2010; Osborne et al., 2008). However, not all parents experience heightened levels of stress. Furthermore, their level of stress may be related to parental characteristics as well as child characteristics. Therefore, using a longitudinal design, we determine which child characteristics are associated with increased levels of maternal stress.

1.3.2 Effectiveness of behavioral treatment

The first study addressing the effectiveness of comprehensive behavioral treatment in children with ASD was conducted by Lovaas (1987). A group of 19 children younger than 46 months received 40 hours of behavioral treatment per week for at least 2 years and was compared to a control group receiving ten hours or less behavioral treatment and a control group receiving treatment as usual. At pretreatment, the groups were comparable on several variables; however, after two years the experimental group outperformed both control groups on educational placement and IQ. Results were maintained at follow-up (McEachin, Smith, & Lovaas, 1993).

Lovaas' approach (1987) has been replicated in older children (Eikeseth, Smith, Jahr, & Eldevik, 2002; 2007), children with ASD and ID (Smith et al., 1997), and in other settings, such as home (Cohen, Amerine-Dickens, & Smith, 2006; Sheinkopf & Siegel, 1998) and school (Eikeseth et al., 2002). These studies show that EIBI results in a significant improvement in cognitive, social and communication skills and in reductions in challenging behavior (Eldevik et al., 2009; Makrygianni & Reed, 2010; Reichow & Wolery, 2009; Spreckley & Boyd, 2009; Virués-Ortega, 2010). Also, EIBI increases the likelihood of being included in mainstream settings later on (Eikeseth, 2009; Matson & Smith, 2008; Smith, Groen, & Wynn, 2000). When compared to other treatments Eikeseth et al. (2002) and Howard, Sparkman, Cohen, Green, and Stanislaw (2005) show that against the popular notion that any intensively implemented intervention can produce meaningful benefits for children with ASD, only EIBI results in large and lasting improvements.

Great variability in children's gains within and between studies is seen with a proportion of the children making rapid and remarkable progress, while other children's gains are limited (Eikeseth, 2009; Reichow & Wolery, 2009). Differences in their response to EIBI can be partially predicted by: (1) children's characteristics and level of functioning at treatment onset such as chronological age (Granpeesheh, Dixon, Tarbox, Kaplan, & Wilke, 2009; Harris & Handleman, 2000), IQ (Granspeesheh, Dixon, et al., 2009), co morbid conditions such as ID and Rett syndrome (Smith et al., 1997; Smith, Klevstrand, & Lovaas, 1995), severity of autism (Ben-Itzchak & Zachor, 2007; Smith et al., 2000), social skills (Ben-Itzack & Zachor, 2007) and behavioral profiles (Sallows & Graupner, 2005), (2) parental characteristics such as stress and coping skills (Grindle, Kovshof, Hastings, & Remmington, 2009; Osborne, McHugh, Saunders, & Reed, 2008),

(3) treatment characteristics such as the intensity (i.e., number of hours per week and duration; Eldevik, Eikeseth, Jahr, & Smith, 2006; Granpeesheh, Dixon, et al., 2009; Lovaas, 1987; Reed, Osborne, & Corness, 2007; Reichow & Wolery, 2009), and the quality of the intervention (i.e. treatment integrity; Sallows & Graupner, 2005), and (4) the amount and quality of the supervision received (Davis, Smith, & Donahoe, 2002; Eikeseth et al., 2009).

Since the recommended 30 to 40 hours of behavioral intervention per week is a major burden to families enrolled in EIBI and might be too stressful for the child and family, low intensity behavioral treatment programs (LIBT) in which children receive fewer hours of intervention (i.e., 0 – 15 hours per week) are frequently implemented in clinical practice (Love et al., 2009; Mudford, Martin, Eikeseth, & Bibby, 2001). Since research on the effectiveness of LIBT and the conditions in which significant progress is found is scarce (see for an exception Eldevik et al., 2006), studies comparing the effectiveness of LIBT to treatment as usual are timely.

The treatment described in this thesis was implemented by Stichting De Driestroom, a service provider for individuals with ID in the eastern part of the Netherlands. Within their preschools, a treatment model was developed in which children with ASD and ID receive a combination of (pre)school services supplemented with four to ten hours of one to one training for two years or more with an emphasis on DTT. The treatment format is based on Lovaas (2003) and no aversives are employed.

Teaching new skills is only valuable if after treatment the child uses these skills in a variety of environments. Therefore, several procedures are included in the treatment model to generalize and maintain the behavior over time and across settings. Functional adaptive behaviors producing positive consequences for the child after treatment are selected for instruction. Generalized responding of the child is stimulated by exposing the child to several objects (e.g., different sizes, colors, materials) that the child will be confronted with in the daily setting. During treatment, tangible reinforcement is paired with social stimuli (e.g., praise) to prevent the child from too much dependency on tangible reinforcement. Treatment is provided by therapists, teachers at the pre-school and when possible by parents in the child's natural environment in order to increase the likelihood of the child using the skills acquired (Lovaas, 1993). Finally, generalization is actively addressed during monthly meetings about the progress of the child and parents and teachers receive guidelines how to generalize and maintain the learned behavior.

Aim of Part II is to synthesize the results of studies published on the effectiveness of EIBI in children with ASD on IQ, adaptive behavior and language. Then, in two studies the effectiveness of LIBT in children with ASD and ID will be determined.

1.3.3 Health care conditions required for early behavioral treatment

Albeit effective, in the Netherlands only few children receive EIBI and implementing such a program can be burdensome for many families both emotionally and financially (Johnson & Hastings, 2002; Trudgeon & Carr, 2007). For the implementation of high quality behavioral treatment, several health care conditions are required. However, most cited conditions are related

to educating parents and professionals about autism and EIBI, obtaining funding of the treatment and the quality of treatment including concerns related to delivery models (e.g., home based vs. school based), appropriate staff training, and difficulties in recruiting and maintaining a treatment team (Jacobson & Mulick, 2000; Johnson & Hastings, 2002; Trudgeon & Carr, 2007).

In EIBI programs, a clinical training pyramid is used in which under supervision of a senior therapist, several undergraduate or bachelor level therapists provide treatment. The senior therapist is supervised by a program supervisor or consultant with a master degree and at the top of the pyramid is the program director, who has significant experience in the field of Applied Behavior Analysis and ASD.

With yearly costs estimated between € 24,000 and € 48,000, EIBI is an expensive treatment. Gaining funding is hard and frequently through a tribunal process (Mulick, 1999). Some families are unable to access any funding and as children with ASD already put an additional strain on parents due to additional costs and income losses, most parents are not able to finance the treatment themselves. This lack of funding may result in programs of lesser quality or parents delivering part of the treatment themselves (Sharpe & Baker, 2007; Trudgeon & Carr, 2007).

The costs associated with ASD for society are high and the lifetime costs for caring for an individual with ASD are estimated at approximately three million Euro (Järbrink & Knapp, 2001). Increasingly more individuals are classified as having ASD and costs for society are increasing. However, although costly in the short term, EIBI may result in increased independence after treatment reducing the costs associated with ASD.

The few cost-offset studies regarding the costs and benefits of EIBI found that the costs of the programs overweigh the estimated long term savings due to more independence of the individuals with ASD after treatment (Chasson, Harris, & Neely, 2007; Jacobson, Mulick, & Green, 1998; Motiwala, Gupta, & Hon, 2006). For example, based on a success rate of 47 percent of the children mainstreaming in regular education (Lovaas, 1987; Sallows & Graupner, 2005), Jacobson et al. estimated savings between the € 149,000 - € 162,000 per child for ages 3-22 years and € 523,000 - € 863,000 per person for ages 3-55 years.

Next to concerns regarding the funding of the treatment, concerns about the quality of programs implemented in the community are expressed. Quality of treatment depends on the capacity of the therapist to reproduce the intervention during daily sessions (e.g., procedural fidelity) and the extent to which treatment is implemented as designed (e.g., treatment integrity; Symes, Remington, Brown, & Hastings, 2005). Several studies have investigated the effectiveness of behavioral intervention when implemented with lower levels of treatment fidelity and most studies found better outcome when integrity was high as opposed to when it was low (Downs, Conley Downs, & Rau, 2007; Groskreutz, Groskreutz, & Higbee, 2011; Grow et al., 2009; Sarakoff & Sturmey, 2008).

Studies show that therapists can be effectively trained to implement DTT with high procedural fidelity using written instructions, lectures, videotaped modeling, role-play and feedback (e.g., Catania, Almeida, Liu-Constant, & Digennaro Reed, 2009; Sarokoff & Sturmey, 2004; 2008; see for a review: Thomson, Martin, Arnal, & Fazzio, 2009) and new approaches such as e-Learning (Granspeesheh et al., 2010). However, to maintain high levels of procedural fidelity, long-term and ongoing support and feedback such as on-site coaching with performance feedback is necessary (LeBlanc, Ricciardi, & Luiselli, 2005).

Despite the crucial role therapists play in providing treatment, little research has been undertaken to determine the contribution of individual differences between therapists affecting their performance during EIBI. A notable exception is the study by Symes et al. (2005) who interviewed 19 therapists about factors enhancing or reducing the correct delivery of DTT. According to the therapists, therapist's patience, child factors (e.g., likeability, motivation, compliance and IQ) and training in the theory underlying ABA, behavioral management and instruction techniques, enhanced effective treatment delivery, while treatment delivery is hindered by the child's challenging behavior, a lack of treatment progress, reinforcer issues, being observed during treatment delivery and therapists' reactions toward challenging behavior. In addition, teaching advanced skills and ambiguity of treatment goals complicate effective treatment. Symes et al. (2005) did not observe procedural fidelity and they based the conclusions on therapist's perceptions only. Therefore, results need to be interpreted with caution.

In sum, concerns related to the funding and quality of EIBI may hinder implementation of effective EIBI in the community. Therefore, these issues will be addressed in the studies described in Part III of this thesis. Aims are to provide a cost-benefit analysis including a broader range of outcome studies and to estimate to which extent benefits of EIBI programs may overweigh the initial costs of these programs. Then, the contribution of individual differences between therapists to the quality of EIBI will be investigated.

1.4 Outline of the thesis

1.4.1 Behavioral characteristics of children with ASD and ID

Part I consists of five studies related to the behavioral characteristics of children with ASD and ID and how these impairments affect the development of these children. The studies described in Chapter 2 and 3 focus on the understanding of other's intentions. Given the potential impact of impaired understanding of other's intentions on the communicative and social development of children with ASD, the studies described in Chapter 2 and 3 explore the understanding of intentions in children with ASD and ID. In the study described in Chapter 2 the understanding of communicative intent is assessed during a hiding game, while in the study described in Chapter 3 an adapted version of the behavioral re-enactment task of Meltzoff (1995) is used. Data on standardized measures regarding cognitive development, severity

of ASD, adaptive behavior, behavioral flexibility, early social communication and language are collected to determine which variables predict and are influenced by the understanding of other's intentions.

Next to impairments in communication and social interaction, a lack of behavioral flexibility is often seen in individuals with ASD (Wahlberg & Jordan, 2001). Studies focusing on the assessment of behavioral flexibility in individuals with ASD and ID are scarce, as are instruments for measuring behavioral flexibility in specific situations in individuals with developmental disabilities. Therefore, two studies were designed addressing behavioral flexibility. In Chapter 4, the psychometric properties (i.e., factor analysis, internal consistency, and intra- and inter rater reliability and convergent validity) of the behavior flexibility rating scale-revised (BFRS-r), a new scale for assessing behavioral flexibility in individuals with developmental disabilities are examined. Next, the BFRS-R is used to determine to which extent children with ASD and ID experience difficulties regarding behavioral flexibility and explore which variables are associated with behavioral (in) flexibility in children with AD and ID, PDD-NOS and ID, and ID only. This study is described in Chapter 5.

Parents of children with ASD experience more parental stress than parents of typically developing children or children with other development disabilities. Several studies show that parental stress and treatment outcome are interrelated (Birnbauer & Leach, 1993; Eisenhower et al., 2005; Griffith et al., 2010; Osborne et al., 2008). Next to parental characteristics, as gender, age, coping style and (perceived) levels of social and professional support, child variables may be linked to heightened levels of parental stress as well. Therefore, using a longitudinal design the study described in Chapter 6 investigates if and how child characteristics influence maternal stress. Data are collected on several variables (i.e., cognitive functioning, adaptive behavior, autism subtype and severity, behavioral flexibility, communication and behavioral problems), and an attempt is made to provide a comprehensive view of child characteristics and their relative contribution to maternal stress.

1.4.2 Effectiveness of behavioral treatment

In Part II, three studies address the effectiveness of EIBI and LIBT. The first study published on EIBI (Lovaas, 1987) reported "recovery from ASD" in 47 percent of the participants and reduced ASD severity in an additional 42% of the cases. However, a number of methodological problems in the sampling, design and analysis of the study have been reported (see for example: Gresham & MacMillan, 1998). The controversy surrounding the study resulted in an increasing body of research with some of the studies replicating the results of Lovaas, while other studies reported more limited gains. To synthesize the outcome of the individual studies on EIBI and investigate the effectiveness of EIBI in young children with ASD with and without ID, a meta-analysis is conducted. Included studies collected data on (non-verbal) IQ, adaptive behavior and receptive and expressive language and all used a group design. Experimental groups of 11 studies that received EIBI are compared to groups of children receiving less

intensive treatment, parent managed treatment, eclectic treatment or treatment as usual. Quality of the studies will be assessed using the Downs and Black Checklist (1998). This meta-analytic study is presented in Chapter 7.

Since research determining the effectiveness of LIBT is scarce (see for an exception: Eldevik et al., 2006), the effectiveness of LIBT and the conditions in which significant progress is found will be examined. Therefore, two studies with a pre-test-post-test-control group design are conducted. Chapter 8 describes a pilot study, in which 12 children with ASD and ID received on average 6.5 hours LIBT per week supplementing their preschool services in the community. They are compared to 22 children with ASD and ID receiving regular treatment at preschools in the Netherlands. At pre-treatment and eight months into treatment, outcome is measured on IQ, developmental age, adaptive behavior, autism symptom severity, and emotional and behavioral problems.

As the sample size of the experimental group in the study described in Chapter 8 is small, the assessment battery rather restricted, the intervention period relatively short and no measures on treatment fidelity were collected, a second study was conducted in which 40 children with ASD and ID participate. As in the first study, children in both groups received community services at (pre)schools for children with ID that their families selected. The children in the treatment group receive additional four to ten hours LIBT per week. Over a period of two years, standardized data are collected on developmental age, adaptive behavior, interpersonal relations, play, language development, autism severity, early social communication skills, maternal stress, behavioral flexibility, and emotional and behavioral problems. Also, data of the treatment program and on treatment fidelity are collected. Results of this experimental and longitudinal study are presented in Chapter 9.

1.4.3 Health care conditions required for behavioral treatment

Albeit effective, only few children are engaged in EIBI programs in the Netherlands and existing programs are faced with difficulties in obtaining funding and concerns about recruiting and maintaining a suitable treatment team (Johnson & Hastings, 2002; Trudgeon & Carr, 2007). Studies investigating the costs and benefits of EIBI conclude that long-term cost savings may overweigh the short-term intervention costs. However, these studies based their analyses on best outcome studies, in which approximately 50 percent of the children mainstreamed in regular education and many studies report less favorable outcome after EIBI (Eikeseth, 2009). Consequently, results of existing cost-benefit studies may not be generalized to other EIBI programs. Therefore, a cost offset analysis of EIBI relative to treatment as usual for children with ASD in the Netherlands will be conducted. Effectiveness will be based on recently published meta-analyses. In Chapter 10 potential cost savings based on several national and international studies and reports are presented per child and extended to the ASD population in the Netherlands.

Next to treatment, child and family variables (Eikeseth et al. 2009; Granpeesheh, Dixon, et al., 2009; Lovaas, 1987; Osborne et al., 2008), quality of treatment delivery predicts treatment outcome (Allen & Warzak, 2000; Bibby, Eikeseth, Martin, Mudford, & Reeves, 2002). Quality of treatment is dependent on the capacity of the therapist to reproduce the intervention as intended. Only one study investigated the factors that enhanced and hindered effective treatment delivery of DTT (see Symes et al., 2005). Therefore, the study described in Chapter 11 investigates the relationship between therapist characteristics and the therapist-child relationship and treatment integrity using objective measures of treatment integrity.

In Chapter 12 the implications of above studies are discussed. Finally, a summary in English and Dutch and the curriculum vitae and publication list of the author are provided.

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Part I
Behavioral Characteristics of Children with Autism Spectrum Disorder and Intellectual Disability

Chapter 2

Comprehension of Communicative Intent in Children with Autism Spectrum Disorder and Intellectual Disability

Submitted for publication

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Abstract

Purpose: Aim of this study was to assess the ability of children with autism spectrum disorder (ASD) and intellectual disability (ID) to infer communicative intent as expressed through pointing and eye-gaze shift.

Method: In the context of a hiding game, 100 children who were between two and ten years old were given eight experimental trials in which they had to locate a toy or candy after a point or gaze shift cue. Also, standardized data were collected on adaptive behavior, cognitive functioning, subtype and severity of ASD, early social communication and language in order to assess associations between these variables and communicative intent.

Results: Only 56% of the participants passed the warm-up condition (3 or 4 trials out of 4 correct) and were included in the analyses. Despite large individual differences, on average children performed above chance in the point condition, while the children performed under chance in the gaze condition. Children's search performance was associated with developmental age, early social communication skills, as well as their receptive and expressive language.

Conclusion: When compared to their performance in the warm-up condition, results might suggest that children with ASD and ID experience difficulty in inferring communicative intent as expressed through pointing and eye gaze. Findings are discussed in relation to the literature.

2.1 Introduction

Next to language, individuals use non-verbal signals (e.g., eye contact, eye gaze and gestures) to communicate with others. As early as three to five month of age, infants begin to follow other's gaze direction to nearby targets within their visual field and when they are one year old children follow other's gaze to more remote targets (Corkum & Moore, 1995). At the same age, children reliably fixate to targets instead of the pointing hand (Butterworth & Grover, 1988) and begin to point to objects and activities (Leung & Rheingold, 1981).

Pointing and gazing just focus the infant's attention in a certain direction, but to derive to which object and with which motive the adult is directing the attention, it needs to comprehend the underlying communicative intent. Beyond individual's goals, these intentions are generally comprised of three levels: (a) a social intention or motive (i.e., what the adult wants the infant to do, feel or know), (b) a communicative intention (i.e., knowing that the adult is trying to communicate, causing the infant to attend to and interpret the adult's social intention), and (c) a referential intention (i.e., attending to a specific object or event; Tomasello, Carpenter, & Liszkowski, 2007).

Between six to nine month of age, typically developing infants begin to understand that other individuals are goal-directed. For example, 9-month old infants respond more patiently to an adult when the adult is trying but unable to give them an object than when the adult is unwilling to give them a toy (Behne, Carpenter, Call, & Tomasello, 2005a) and 12- and

14-month old infants imitate an unusual action (e.g., switching on the light by touching the lamp with the forehead) more often when the adult freely chooses that action, than when the adult was forced to use the unusual action by some constraint (e.g., when the adult's hands were occupied; Gergely, Bekkering, & Kiraly, 2002). Next, 14- to 18-month old infants are more likely to copy intentional actions of others than accidental actions (Carpenter, Akhtar, & Tomasello, 1998).

As stated, around their first birthday children follow others' eye gaze and pointing (Butterworth & Grover, 1988; Corkum & Moore, 1995). However, this does not necessarily indicate that the child infers the communicative intent underlying these actions. Therefore, several studies (Behne, Carpenter, & Tomasselo, 2005; John & Mervis, 2010; Povinelli, Reaux, Bierschwale, Allain, & Simm, 1997; Tomasello, Call, & Gluckman, 1997) used a hiding game to assess whether children understand the communicative intent underlying non-verbal communication. Tomasello and colleagues indicated which of the three containers contained a reward by (a) holding up a replica of the correct container, (b) pointing to the correct container, or (c) placing a wooden block on the correct container. In contrast to great apes, children of 2.5 to 3 years were capable of finding the hidden reward.

In studies of Povinelli and colleagues (1997) and Behne and colleagues (2005b), an adult indicated the hidden reward by pointing to or gazing at one of the two containers. In the study of Povinelli and colleagues children between 2 and 2.5 years reliably used the pointing gesture to guide their search, but performed not above chance when the experimenter only gazed at the baited container. However, in the study of Behne and colleagues typically developing children of 14-, 18- or 24-month old found the hidden toy significantly more often than would have been expected by chance with both types of cues. Differences in the set up of the tasks may explain inconsistencies between studies as in the study of Povinelli and colleagues the adult only turned his head to look at the correct container, while in the study of Behne and colleagues the adult alternately gazed between the child and the correct container. Also, in study of Povinelli and colleagues the adult was positioned closer to the incorrect container, while in the study of Behne and colleagues the distance between the adult and the correct and incorrect container was similar. Finally, the Povilenni study only used one gaze trial in twelve participants, while in the Behne study sixty children participated who were all exposed to eight trials.

Behne and colleagues (2005) found a significant effect of age on search performance with a success rate of 84% on both point and gaze trials for the 24-month old, while the 18-month old (60% success rate) performed better on the point trials than on the gaze trials and the 14-month old (7% success rate) made a number of errors on both types of trials. The improvement in search performance took place earlier for pointing than for gazing. By successfully retrieving the reward the children showed they were not only capable of following the point or gaze cues, but also inferred why in this specific context the experimenter was directing their attention (i.e., understand that they had to find the toy, while the experimenter informed them about the location of the hidden reward).

As suggested by Carpenter, Nagell, and Tomasello (1998) inferring communicative intent allows children to explain, predict and influence others' behaviors. A deficit at this fundamental level may hinder the development of higher-level social understanding and have severe consequences for the child's social functioning. For example, Colonessi, Rieffe, Koops, and Peruchini (2008) found that in typically developing children the ability to understand intentions at 12 and 15 month of age predicted the ability to explain others' actions at 38 month. This might indicate that the understanding of intentions can be seen as one of the first steps towards a Theory of Mind. Responding to other's pointing and gazing is also associated with more complex social-communicative skills and language (Baldwin, 1991).

Delays and impairments in the use of non-verbal communication including difficulties in following eye gaze and pointing are frequently reported in young children with autism spectrum disorder (ASD) and intellectual disability (ID). Stone, Ousley, Yoder, Hogan, and Hepburn (1997) found that two and three-year-olds with ASD were less likely to point, show objects or use eye gaze to communicate than children with comparable chronological and developmental age. Children with ASD seem more impaired in protodeclaratives (behaviors to comment or share attention) than in protoimperatives (tangibly maintained behaviors; Goodhart & Baron-Cohen, 1993). Furthermore, children with ASD attend less to social cues of others. For example, analysis of home-video's of first birthdays has revealed that infants later diagnosed with ASD looked less frequently at others than typically developing infants or infants later diagnosed with general learning difficulties (Osterling & Dawson, 1994; Osterling, Dawson, & Munson, 2002). Several eye-tracking studies reported that individuals with ASD fixate less on faces (especially the eyes) and more on body parts and objects than typically developing children (Klin, Jones, Schutlz, Volkmar, & Cohen, 2002; Speer, Cook, McMahon, & Clark, 2007).

By attending less to social cues of others, children with ASD may acquire less proficiency in interpreting the social cues and intentions of others than typically developing children. Consequently, children with ASD are at increased risk for misinterpreting others' intentions and may experience difficulties in explaining, predicting and influencing others' behavior and communication (Dawson, 1991; Dawson, Meltzoff, Osterling, Rinaldi, & Brown, 1998). Support for a selective social impairment in children with ASD is provided by a study of Dawson et al. (2002), in which they compared brain responses of typically developing children, children with ASD and children with intellectual disability (ID) to photos of familiar and unfamiliar persons and objects. All groups showed different brain responses to photos of familiar objects than to photos of unfamiliar objects. However, in contrast to the typically developing children and children with ID, children with ASD did not show a different response to photos of their mother as compared to photos of unfamiliar persons. These results are in line with a fMRI study by Greene et al. (2011) who found that children and adolescents with and without ASD showed similar social orienting behavior. However, as opposed to typically developing individuals who showed greater brain activity for social cues than for non-social cues, children and adolescents with ASD did not distinguish between social and non-social cues. Results seem to indicate that while typically developing children treat social cues with a privileged status, individuals with ASD process social cues using the same mechanism as non-social cues.

An explanation for the reduced time children with ASD spent attending to social cues may be provided by a fMRI study conducted by Scott-Van Zeeland, Dapretto, Ghahremani, Poldrack, and Bookheimer (2010) who found that in contrast to typically developing children, children with ASD do not experience social stimuli as rewarding. In addition, social stimuli are unpredictable and complex, while children with ASD prefer the less variable feedback of non-social stimuli instead of the variable feedback, which is characteristic for social stimuli (Gergely & Watson, 1999). Other people's goals and intentions guide children in selecting from whom, when and what kind of behaviors they learn. The lack of understanding others' intentions and goals is suggested as underlying deficit in early social cognitive skills and language acquisition in children with ASD (e.g., joint attention, communication, and imitation) and may have important implications for the understanding of ASD (Tomasello, 2000; Meltzoff, Kuhl, Movellan, & Sejnowski, 2009). Since children with ASD and ID might not benefit sufficiently from socially mediated learning opportunities, this may have a cascading effect on their development (Meltzoff et al., 2009).

The present study used a hiding game similar to the one used by Behne et al. (2005b) to assess whether children with ASD and ID comprehend the communicative intent behind gaze shifting or pointing. It is expected that children who comprehend the communicative intent behind gaze shifting or pointing retrieve the reward more often than may be expected by chance (e.g., higher than 50%). To address heterogeneity amongst individuals with ASD and ID with respect to degree in cognitive functioning and severity of ASD and which factors might predict and are influenced by the understanding of communicative intent, associations between the ability to infer communicative intent and developmental age, early social communication, language and diagnosis, severity and subtype of ASD are also explored.

We expected that (1) children with a lower developmental age are less successful in interpreting the communicative intent (i.e., retrieve the reward significantly less often) than children with higher developmental age. Next, we expected that (2) as the severity of ASD increases, understanding of communicative intent decreases. As the process of language acquisition is highly guided by social input (Meltzoff et al., 2009) and the understanding of others' communicative intent may be related to the emergence of early social communication skills (e.g., joint attention, behavioral requests and social initiations) and language, it is (3) expected that children with less ability to infer communicative intent have less advanced social communication skills and receptive and expressive language abilities.

2.2 Method

2.2.1 Participants and Setting

Participants were 100 children (77 males) aged between 2;7 and 9;11 years (*M* = 72.19 month; *SD* = 19.89) with ASD and ID. All children visited a preschool or school for children with ID located throughout the Netherlands. Exclusion criteria for participation were the

presence of neurological disorders of known etiology and somatic factors interfering with data collection (*n* = 13, e.g., severe visual impairment and severe epilepsy). Written consent was obtained for all children.

Prior to their inclusion to the study all children received the diagnosis autistic disorder (AD; *n* = 83) or Pervasive Developmental Disorder – not otherwise specified (PDD-NOS; *n* = 17) according with the DSM-IV or ICD-10 criteria and the diagnosis was made by a clinician who was independent of the study. The diagnosis was confirmed by the Autism Diagnostic Observation Schedule (Lord, Rutter, DiLavore, & Risi, 2006) and the Childhood Autism Rating Scale (Schopler, Reichler, & Rochen Renner, 2007) assessed by the first author. The Wing subgroups questionnaire (WSQ; Castelloe & Dawson, 1993) indicated that 71 children had the aloof subtype, while 18 children had the passive subtype and 11 the active-but-odd subtype.

In addition to ASD, all children had a diagnosis of ID set by a psychologist, special educator or psychiatrist. An assessment of the Mullen Scales of Early Learning (Mullen, 1995) and the Vineland Adaptive Behavior Scales (Sparrow, Balla, & Cicchetti, 1984) administered by the first author confirmed this diagnosis. Two children were excluded due to the absence of an ID.

2.2.2 Measures and Procedure

Communicative intent

An experiment modeled after the first experiment of Behne et al. (2005b) was used to measure the response of participants to both pointing and gazing. During the experiment, the experimenter and the child sat at a table in a distraction-free environment at the preschool or school the child visited. In the context of a hiding game, four pairs of containers were used to hide a reward. A motivational screening of ten potential rewards (candy, cookie, chips, balloon and six small toys) was conducted between other assessments and based on the child's responses (reaching or asking for the object) four rewards were selected for each child. Each of the three conditions consisted of four trials and each pair of containers and reward was used during one trial in each condition. A small white blanket covered the containers during the hiding in the experimental conditions. A camera facing the child recorded the hiding game. In all trials a response time of 20 seconds was used.

In the warm-up phase, the experimenter presented four pairs of identical containers and rewards to the child by briefly showing them. As the child watched, the experimenter hid a reward in a container of the first pair. After the containers were closed, the experimenter stimulated the child to retrieve the reward by moving the containers in the direction of the child. If the child found the reward, the experimenter responded enthusiastically and permitted the child to eat the candy or play briefly with the toy. When needed, the experimenter helped the child to retrieve the reward. However, in this situation an incorrect response was noted. This warm-up condition was repeated for all four pairs of containers.

During the experimental trials, the experimenter presented the open and empty containers to the child and placed them on her lap. Then the experimenter showed the reward briefly to the child and hid the reward in one of the containers, covering the hiding process with a blanket so the child did not know which box contained the reward. Following the hiding procedure, the experimenter placed the boxes on the table and moved the containers in the direction of the child to stimulate the child to retrieve the toy. Depending on the condition, the experimenter pointed to or gazed at the box, which contained the reward.

In the point-condition, the experimenter demonstrated a point (held at her midline) across her body at the box in which the reward was hidden and smiled briefly to the child, while in the gaze condition the experimenter turned her head, gazing twice between the container with the reward and the child. During all conditions, cues were provided non-verbally, although the experimenter reacted with excitement when the child found the reward. Each child participated in both experimental conditions (four trials per condition) and the location where the box with the reward was placed (right or left) was chosen randomly. However, the reward was never hidden more than twice in a row on the same side. When the child had made a clear choice, but experienced difficulty opening the box, the experimenter held her hand up and asked if she should help opening the box. If the child could not find the hidden reward (e.g., opened the wrong box), the experimenter opened the correct container and showed the reward to the child. Since in the study of Behne et al. (2005b) no effect of order of cue presentation was found, in all children the warm-up condition was followed by the point and then the gaze condition.

All trials were videotaped and coded by three undergraduate special education students unaware of the exact characteristics of the children. If a child chose the container the experimenter was pointing to or gazing at, this was scored as a correct response; choosing the other container was scored as an incorrect response. Before the students started coding, the first author explained the procedure and trained them in coding the trials. Before coding the experimental data, an average kappa of .84 (range: .83-.86) was obtained between all combinations of raters including the first author, indicating an excellent interrater reliability (Cicchetti, 1994). To assess interrater reliability during coding, 29% of the videotapes were randomly selected and independently coded by another rater. With an average kappa of .93 interrater reliability was excellent (Cicchetti, 1994).

Standardized assessments

Several instruments were administered to the children and their parents to measure adaptive behavior, cognitive functioning, subtype and severity of ASD, early social communication and language. The Autism Diagnostic Observation Schedule, the Mullen Scales of Early Learning, the Early Social Communication Scales and the language tests were administered in a distraction-free room with two chairs and a table at the (pre) school the child visited. Within one month from the assessments, the first author interviewed the parents in their home regarding the adaptive behavior oftheir child. The CARS was completed based on the assessments and parental report.

Adaptive behavior

The Vineland Adaptive Behavior Scales – survey form (VABS; Sparrow et al., 1984; Dutch version by De Bildt & Kraijer, 2003) is a semi-structured parent interview that assesses adaptive behavior on a total scale and on the subscales communication, daily living skills and socialization. Age-equivalents for the composite score and the subscales are provided.

Cognitive functioning

The Mullen Scales of Early Learning (Mullen, 1995) is a standardized developmental test for children from birth to 68 month of age. Four of the five subscales were administered: fine motor, visual reception, expressive language and receptive language. Due to the chronological age of the children not all standardized scores could be calculated. Therefore, developmental age and ratio IQ were used in our analyses. Ratio IQ was calculated by dividing the mean developmental age equivalence score on the visual reception, fine motor, receptive language and expressive language subscales by the chronological age. A mean developmental age was calculated based on the developmental ages on all four scales.

Severity and subtype of ASD

The Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2006) is a semi-structured standardized observation that measures ASD symptoms in social relatedness, communication, play and repetitive behaviors. The Childhood Autism Rating Scale (CARS; Schopler et al., 1988) was administered as a measure of autism symptom severity. The Wing Subgroups Questionnaire (WSQ; Castelloe & Dawson, 1993) is a questionnaire with 13 behavioral domains (e.g., communication, social approach, play, imitation, motor behavior, resistance to change) on which parents rate their child's behavior on a scale from zero (never) to six (always) for each domain. A summary score is calculated for each subtype and the highest summary score is considered to indicate the child's subtype.

Early social communication and language

The Early Social Communication Scales (ESCS; Mundy et al., 2003) is a videotaped semi-structured observational instrument. The scale measures how the child initiates and responds to tasks involving joint attention, behavioral requests and social interaction. Toys and activities are used to elicit social and communicative behavior in an ecologically valid context. The first author administered the ESCS, and videotapes were scored by four raters who were unaware of the exact aim of the study (including the other scores of the participants). Interrater reliability was assessed using videotaped data from 28.7% of the children and intraclass correlation coefficients between the paired ratings of the six subscales ranged from .66 to .73, suggesting good reliability (Cicchetti, 1994).

Receptive language (vocabulary) was measured by the Peabody Picture Vocabulary Test (PPVT; Dunn & Dunn, 1997), while expressive language (spoken language) was measured by the vocabulary test of the Schlichting Test for Language Production (Schlichting, Van Eldik, Lutje Spelberg, Van der Meulen, & Van der Meulen, 1995).

2.3 Results

2.3.1 Search performance

First, the percentage of successful search performances was calculated for each condition (i.e., warm-up, point and gaze). On average, the correct container was chosen in 65% (*SD* = 32) of trials in the warm-up condition. Despite that children saw the object being hidden, 44% of the children performed on or under chance (i.e., zero, one or two trials correct) in the warm-up condition and were therefore excluded from the analysis. Characteristics of the included and excluded children are displayed in Table 1.

An ANOVA with repeated measures was conducted to test for differences in search performance between conditions in the children who passed minimally three trials in the warm up condition. As Mauchly's test indicated that the assumption of sphericity had been violated ($\chi^2(2) = 8.76$, $p = .01$), degrees of freedom were corrected using Greenhouse-Geisser estimates of sphericity ($\varepsilon = .87$). There was a significant main effect of the condition on search performance ($F(1.74, 95.67) = 76.93$, $p < .001$). Repeated contrasts revealed that children retrieved the reward significantly more often in the warm-up condition ($M = 88.54$; $SD = 12.54$) than in the point condition ($M = 63.84\%$; $SD = 35.33$; $F(1, 55) = 28.52$; $p < .001$) and that the reward was significantly more often found in the point condition than in the gaze condition ($M = 39.29\%$; $SD = 26.7$; $F(1, 55) = 41.86$; $p < .001$). If children comprehended the communicative intent behind the pointing or gazing, they would be expected to find the toy more often than expected by chance (i.e., more than 50% of the trials). A one-sample *t*-test (test value = 50) revealed that on average children performed significantly above chance in the point condition ($t(55) = 2.93$, $p < .01$), while the children performed under chance in the gaze condition ($t(55) = 2.98$, $p < .01$). In the warm-up condition all participants performed above chance (3 or 4 out of 4 trials correct), while these percentages are lower in the point condition (57.1%) and gaze condition (17.9%). Based on binomial tests (7 out of 8 trials correct, $p = .03$) 14.29% of the children with ASD performed above chance in both conditions. In Figure 1 the percentages of children who successfully retrieved the reward in zero, one, two, three or four trials are presented for each condition.

To further address individual differences in search performances within the sample and to explore which variables are associated with the understanding of communicative intent, several analyses were conducted exploring relationships between the understanding of communicative intent and cognitive functioning, severity and subtype of ASD, early social communication skills and language.

Table 1. Characteristics of the included and excluded participants.

	Included children			Excluded children		
	Mean	*SD*	*Range*	*Mean*	*SD*	*Range*
Chronological age	68.21	16.21	34-108	77.25	22.97	31-119
Cognitive age						
Developmental age in months	25.74	8.10	12.00-52.00	21.90	9.34	4.75-43.50
IQ	39.27	14.16	15.48-78.78	31.59	17.73	10.59-71.05
Verbal IQ	34.11	16.20	12.50-79.23	27.44	18.10	7.23-71.93
Non Verbal IQ	44.47	14.01	18.45-80.77	35.74	18.39	11.18-74.07
Adaptive behavior in months						
Composite	21.07	5.97	13-41	20.02	8.12	11-45
Communication	26.76	10.50	13-53	24.00	11.11	12-53
Daily Living Skills	24.11	7.87	11-45	22.68	10.34	11-47
Socialization	23.55	6.74	13-46	21.61	7.86	11.48
ASD						
ADOS total	15.30	3.83	7-22	15.84	4.01	7-24
ADOS communication	5.84	1.94	2-9	6.14	1.65	2-10
ADOS social interaction	9.55	2.51	5-16	9.77	3.21	2-16
CARS	41.59	5.61	30-53	41.00	5.03	30-50
Early social communication scales						
Initiating joint attention	6.96	8.53	0-37	7.70	8.38	0.28
Responding to joint attention	97.07	64.82	0-200	85.79	73.18	0-200
Initiating behavioral requests	24.52	6.66	11-43	22.07	7.83	2-40
Responding to behavioral requests	70.73	30.46	0-100	59.19	35.35	0-100
Initiating social initiations	2.64	1.61	0-6	2.70	2.04	0-7
Responding to social initiations	6.70	2.82	1-15	6.67	3.55	0-16
Language in months						
Receptive language (PPVT)	26.21	5.02	21-24	18.91	9.20	14-48
Expressive language (WO)	22.09	10.23	14-56	24.61	4.71	21-43

Note. Included children; *N*= 56, except for adaptive behavior (*n*= 55); Excluded children *N*= 44, except for the early social communication scales (*n*= 43).

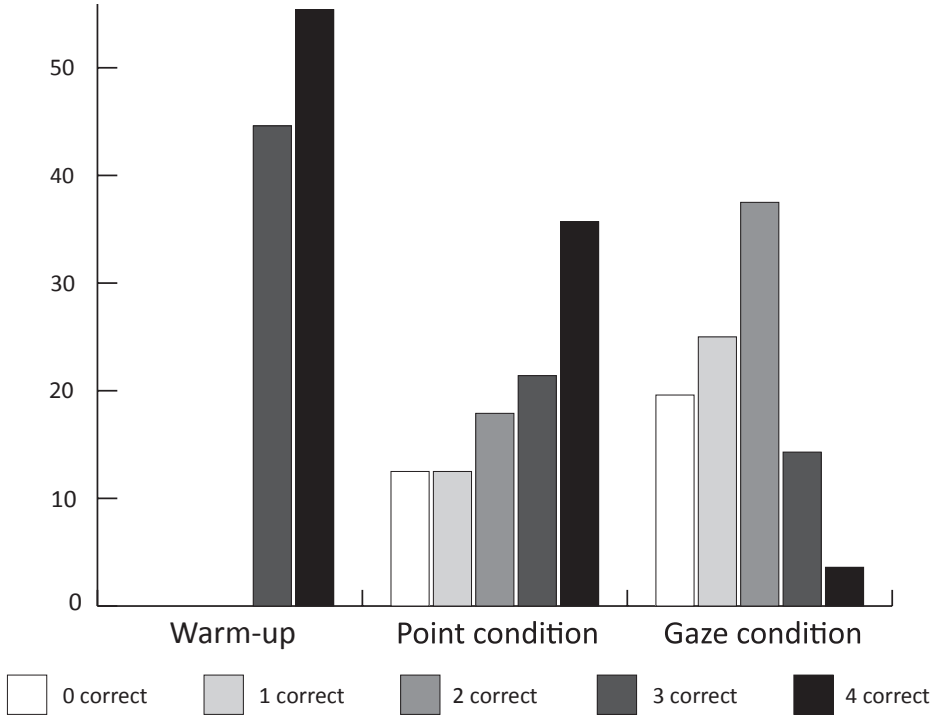

Figure 1. Percentage of children who had respectively none, one, two, three or all four trials correct in the warm-up, point or gaze condition (N=56).

2.3.2 Associations between search performance and cognitive functioning, severity of ASD and language

Pearson correlations for the search performance in both conditions were computed. Results are displayed in Table 2. Based on the criteria of Cohen (1992) associations with a large effect were found between search performance in the point condition and developmental age and responding to behavioral requests, associations with a moderate effect between search performance in the point condition and IQ, the communication and total scale of the ADOS, responding to joint attention, receptive and expressive language, while associations with a small effect were found for the social interaction scale of the ADOS.

For the gaze condition, associations with large effects were found between search performance and responding to behavioral requests, associations with moderate effects between search performance and developmental age, IQ, the social interaction and total scale of the ADOS, responding to joint attention, initiating social initiations and receptive and expressive language. An association with a small effect was found between search performance and the communication scale of the ADOS.

Table 2. Pearson correlations between search performance in the point and gaze condition and cognitive functioning, severity of ASD, early social communication and language.

	Point condition	Gaze condition
	r	*r*
Understanding of communicative intent		
Warm up condition	.20	.35**
Point condition		.61**
Gaze condition	.61**	
Chronological age	.18	.08
Cognitive age		
Developmental age in months	.55**	.41*
IQ	.38**	.33*
Verbal IQ	.40**	.38**
Non Verbal IQ	.30*	.23
ASD		
ADOS total	-.34**	-.35**
ADOS communication	-.31*	-.28*
ADOS social interaction	-.26*	-.32*
CARS	-.06	.03
Early social communication scales		
Initiating joint attention	.15	.21
Responding to joint attention	.40**	.37**
Initiating behavioral requests	.16	.10
Responding to behavioral requests	.66**	.52**
Initiating social initiations	.16	.37**
Responding to social initiations	-.02	.09
Language in months		
Receptive language (PPVT)	.38**	.33*
Expressive language (WO)	.38**	.30*

Note. * p <.05; ** p <.01 (2-tailed).

2.3.3 Developmental age and severity of ASD on search performance

As ASD severity (ADOS-total score) and developmental age were significantly correlated with the search performance in both the point and gaze condition (see Table 2),

hierarchican regression analyses were conducted between ASD severity and developmental age as independent variables and search performance in the point or gaze condition as dependent variable. In model 1, the average developmental age on the Mullen Scales of Early Learning was entered in the first step, while ASD severity was entered in the second step. Only developmental age contributed significantly to the model. To assess the direct effect of severity of ASD on search performance, two other regression analyses (model 2) were conducted, in which in the first step the severity score of the ADOS was entered, while developmental age was entered in the second step. Although severity of ASD significantly predicted search performance in the first step, it no longer contributed significantly to the model when developmental age was entered. Table 3 displays the unstandardized regression coefficient (*B*) and standard error of the unstandardized regression coefficient (*SE B*) and the standardized regression coefficient (*β*) of both regressions. R^2 was significantly different from zero at the end of each step for each model and all models significantly improved the ability to predict search performance (for the point condition model 1 and 2: $F(2,53) = 12.48$, $p < .001$; for the gaze condition model 1 and 2: $F(2,53) = 6.95$, $p < .01$).

Table 3. Multiple regressions to predict search performance for children with ASD plus ID (N=56).

		Search performance in the point condition			Search performance in the gaze condition		
		B	*SE B*	*β*	*B*	*SE B*	*β*
Model 1	Step 1:						
	Constant	2.30	13.38		4.67	11.14	
	Developmental age	2.39	0.50	.55**	1.35	0.41	.41**
	Step 2:						
	Constant	30.52	26.44		36.83	21.73	
	Developmental age	2.13	0.54	.49**	1.05	0.44	.32*
	Severity of ASD	-1.41	1.14	-.15	-1.60	0.94	-.23
Model 2	Step 1:						
	Constant	112.46	18.59		77.13	14.12	
	Severity of ASD	-3.18	1.18	-.34	-2.47	0,90	-.35**
	Step 2:						
	Constant	30.52	26.44		36.83	21.73	
	Severity of ASD	-1.41	1.14	-.15	-1.60	0.94	-.23
	Developmental age	2.13	0.54	.49**	1.05	0.44	.32

Note. For the point condition: model 1: *R^2 = .30 for step 1 and Δ R^2 = .02 for step 2; and model 2: R^2 = .12 for step 1; Δ R^2 = .20 for step 2. For the gaze condition: model 1: R^2 = .16 for step 1 and Δ R^2 = .04 for step 2; and model 2: R^2 = .12 for step 1; Δ R^2 = .08 for step 2; * p < .05, ** p <.01.*

At the end, both models explained 32% of the variance in search performance in the point condition and 20% in the gaze condition. Hence, the association between severity of ASD and search performance can be explained in terms of developmental age. There is a negative linear relation between developmental age and severity of ASD (r = -.39; p < .01) and between developmental age and search performance. When developmental age is not controlled for, significant correlations were found between severity of ASD and search performance (see Table 2).

To determine the effect of search performance on receptive and expressive language two multiple regressions were conducted with search performance in the point and gaze condition as independent variables and receptive or expressive language as dependent variable. However, as only search performance in the point condition significantly contributed to the model (B = 22.74; $SE\ B$ = 1.30; β = .38; p < .01 for receptive language and B = 15.09; $SE\ B$ =2.66 ; β =.38; p <.01 for expressive language), search performance in the gaze condition was excluded from the analyses (t = 0.93; p = .36 for receptive language; t = 3.01; p <.01 for the gaze condition). R^2 was significantly different from zero for both receptive and expressive language and both models significantly improve the ability to predict receptive and expressive language from children's search behavior (receptive language: $F(1, 54) = 9.29$, p < .01; expressive language: $F(1 ,54) = 9.04$, p < .01. For receptive language 14.7% and for expressive language 14.3% of the variance was explained.

2.4 Discussion

The present study assessed the ability to infer communicative intent as expressed through pointing and eye-gaze shift in 56 children with ASD and ID. The understanding of communicative intent was measured using a hiding game, in which children had to locate a toy or small edible after a point or eye gaze shift provided by the experimenter during eight experimental trials. Also, standardized data were collected on adaptive behavior, cognitive functioning, subtype and severity of ASD, early social communication and language in order to assess associations between these variables and communicative intent.

Although large individual differences in search performance appeared, on average the correct container was chosen in 64% of the trials if the experimenter pointed at the correct container and in 39% of the trials as the experimenter gazed at the correct container. This was significantly less than in the warm-up condition (88%) in which the children saw where the experiment hid the object. This might indicate that children with ASD and ID experience difficulty in inferring communicative intent as expressed through pointing and eye gaze. Children's search performance was predicted by developmental age. Significant correlations between the search performance and severity of ASD can also be explained in terms of developmental age.

As no control group was included, it seems premature to conclude that children with ASD have a deficit in the understanding of communicative intent. However, taken into account the difference between participants, conditions, materials and sample sizes, results of the current study can be compared to two other studies that used the same experiment (Behne et al., 2005b; John & Mervis, 2010). Based on binomial tests (this study and Behne et al.: 7 or 8 out of 8 correct, p = .03; John & Mervis: 10, 11, or 12 out of 12 correct, p = .02) respectively 7%, 60% and 84% of typically developing children of 14-, 18- and 24-month old found the hidden object at a rate expected above chance, while 60% children with Down syndrome and 26.7% Williams syndrome (chronological age: 3-5 years) were able to do so. In the present study, 14.29% of the children with ASD and ID performed above chance in both conditions. As expected, all three studies showed better search performance as the developmental age increased and that children displayed better search performance in the point condition than in the gaze condition. Probably inferring communicative intent through pointing is easier than through eye gaze as the topography of pointing is more explicit than of eye gaze. In addition, children with ASD focus more on body parts than on the eyes of others. However, this explanation seems not valid for typically developing children or children with other developmental disablities (Klin et al., 20002; Speer et al., 2007).

Results may suggest a delayed development of the understanding of communicative intent behind gestures and gaze in children with ASD and ID relative to typically developing children and children with Williams and Down syndrome. This finding is in accordance with a study by Peters-Scheffer, Didden, Korzilius, Sigafoos, and Verhoeven (submitted) who found a delayed development of intention understanding in children with ASD and ID during an imitation task. Tomassello (1995) suggests that the understanding of intentions might be seen as one of the first steps toward a Theory of Mind. Hence, results of the current study seem in line with studies who suggest that children with ASD require a higher developmental age than other subjects to succeed on Theory of Mind tasks (Happé, 1995). However, further research in children with ASD and ID should investigate how and whether a delay in understanding the intentions of others may predict a delay in Theory of Mind development. Future studies should include a more heterogenic sample of children with ASD and ID and a control group of typically developing children or children with ID only to further investigate the influence of ASD on intention understanding. When including a control group, children should be carefully matched to the experimental group on developmental age, as this seems to be related to the understanding of communicative intent.

Non-verbal behaviors such as gazing and pointing are crucial in social interactions and help to establish relationships between the child and other individuals. For example, the importance of eye gaze in (early) social development is already visible in newborns who prefer to look at a face with open eyes as opposed to a face with closed eyes. This preference provides newborns with an adaptive orientation mechanism that ensures that infants will fixate on and learn about the most relevant social stimuli in their environment (Batki, Baron-Cohen, Wheelwright, Connelan, & Ahluwalia, 2000).

Since most children in this study were non-verbal or had only limited speech, gazing and pointing seem essential for their social communication and understanding of communicative intent necessary to participate in social interaction. Although more research is needed, this study suggests that a delay in understanding communicative intent may complicate language acquisition in children with ASD and ID. However, many children with ASD and ID use non-vocal strategies such as pictures or manual signs (Mirenda, 2001) and results of this study need to be interpreted with caution as their speech is frequently affected by difficulties in motor-speech and other specific impairments.

Evidence for an association between variables cannot be considered as evidence of causation and research addressing that changes in the causal variable precedes changes in the outcome is warranted. Using a longitudinal design, research needs to address which variables influence the development of understanding communicative intent and are affected by a lack of understanding.

Given the potential impact of impaired understanding of communicative intent on the development of children with ASD, further research should explore how to teach and incorporate the intention understanding in the current early interventions for children with ASD. Moreover, parents and professionals working with young children with ASD and ID need to realize that these children are not always capable of understanding the communicative intent behind a gaze or point cue and possibly adapt their daily routines to inform and teach children.

In sum, this study showed that children with ASD and ID might have difficulty in understanding communicative intent and that the lack of understanding is influenced by the developmental age and may predict difficulties in acquiring early social communication skills and receptive and expressive language. Clearly, further research is necessary to extend these preliminary findings to enhance the comprehension of professionals and parents about the functioning and behavior of individuals with ASD and ID and to improve the early intervention for individuals with ASD and ID.

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Chapter 3

Understanding of Intentions in Children with Autism Spectrum Disorder and Intellectual Disability

Pending minor revisions accepted for publication

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Abstract

This study investigates the understanding of intentions in 100 children with autism spectrum disorder (ASD) and intellectual disability (ID; mean IQ: 36) between 2 and 10 years of age. A within subject design with two conditions was used. In the target condition, the adult performed the target behavior; while in the unsuccessful attempt condition, the experimenter unsuccessfully demonstrated the target behavior. Children with ASD and ID displayed significantly more target behavior and less off-task behavior in the target condition than in the unsuccessful attempt condition, which might indicate that children with ASD and ID experience difficulty in understanding the intentions of others. Target behavior seen after an unsuccessful attempt is predicted by imitation of target behavior and behavioral inflexibility and is associated with better performance on receptive and expressive language tests. The latter results are in line with several other studies suggesting a relation between the understanding of intentions and early social communication and language.

3.1 Introduction

Long before verbal instruction is possible, imitation of other people is one of the first fundamental means by which children learn a wide range of new behaviors and it serves a social function by facilitating interpersonal interactions early in life (Tomasello, Carpenter, Call, Behne, & Moll, 2005; Uzgiris, 1981). Even young children are predisposed to focus on other people and learn through social interaction. Learning by observation multiplies learning opportunities as it is generally more efficient than learning by trial-and-error and/or individual exploration (Meltzoff, Kuhl, Movellan, & Sejnowski, 2009).

Imitation is associated with joint attention (Ingersoll & Schreibman, 2006; Whalen, Schreibman, & Ingersoll, 2006), play skills (Fiese, 1990; Ingersoll & Schreibman, 2006), language (Charman et al., 2000; Charman et al., 2003; Toth, Munson, Meltzoff, & Dawson, 2006), socialization and enculturation (Tomasello, Savage-Rumbaugh, & Kruger, 1993; Tomasello et al., 2005). In typical development, imitation appears early with some imitative responses apparently present at birth (Meltzoff & Moore, 1983). From six to nine month of age, social interactions become more complex as triadic interactions (agent to object to agent) emerge and from late infancy onwards children are able to imitate actions of other persons (Carpenter, Nagell, & Tomasello, 1998; Tomasello, 1999). For example, Meltzoff (1988) showed that 9-month old infants are able to imitate simple actions of an adult with novel objects immediately and after a 24-hour delay.

Learning through observation is manifested on several levels. For instance, newborns with a mean age of 32 hours may mimic superficial behavior of an adult without understanding the intention of the behavior (Meltzoff & Moore, 1983), while in emulation, the goal of the behavior and the object is understood without attending to the behavior or intentions of the

demonstrator (Tomasello, 1999). In contrast to emulation, when only the final outcome is copied, imitation is the ability to re-enact the goal and method of the observed behavior by understanding the actor's intention (Tomasello et al., 1993). This kind of imitation is seen, for example, when infants reproduce intentional, but not accidental behavior of adults (Carpenter, Akhtar, & Tomasello, 1998) or produce an action that an adult intended, but did not actually produce (Meltzoff, 1995).

In different cultures, individuals use objects in various ways and objects have several properties and affordances. Consequently, by simply observing an object the child may not learn or perform the proper action. In accordance to Tomasello's theory of social learning (Tomasello, 1999; Tomasello et al., 2005), children need to select the appropriate action based on the comprehension of another person's intentional, goal-directed action and thus attend to both the means and the end result of the behavior to acquire the conventional use of objects, linguistic symbols and many other aspects of their culture. Research indicates that even infants comprehend intentions underlying actions. For example, infants from 9 months of age respond more patiently when an adult is unable to give them a toy than when the adult is unwilling to give the toy (Behne, Carpenter, Call, & Tomasello, 2005).

Further evidence of infants' understanding of intentions is provided by tasks with failed (unsuccessful) attempts or mistakes that create a mismatch between the goal of the individual and the demonstrated behavior. To succeed on such tasks, the infant needs to understand the actor's goal instead of simply observing the surface behavior. Using the behavioral re-enactment technique, Meltzoff (1995) presented 18-month old children with an actor either performing a goal-directed action on an object or an actor who was trying, but failing to perform a certain action on an object (unsuccessful attempt). Two additional groups of 18-month old infants participated in one of the two control conditions in which objects were randomly manipulated. In contrast to the children in the two control groups, children were able to perform the intended target behavior after seeing an unsuccessful attempt as often as the children who saw the successful demonstration of the complete target action. A second experiment confirmed that children were guided by the experimenter's intentions instead of the experimenter's movements. Additional evidence of infants' ability to differentiate the goal of the action from the surface behavior was provided in another study by Meltzoff (1996) in which he showed 18-month old children an adult trying, but failing, to pull apart a mechanical device (i.e., pulling apart a dumbbell). Because the object was too big for the children's hands, infants used different means to reach the same end goal (i.e., to pull apart the dumbbell) instead of just imitating the surface behavior.

Since then, several authors have used this type of behavioral re-enactment task to replicate and extend Meltzoff's (1995) study. Bellagamba and Tomasello (1999) used the behavioral re-enactment task with four experimental conditions in 80 children of 12- or 18-month old. In the first two conditions the target behavior or an intention to perform the target behavior was demonstrated, while in the first control condition the experimenter modeled a random act. The other control condition of Meltzoff (1995) was replaced for an end state condition in which the end state of the object set was demonstrated without showing the manipulation to the child.

Results replicated those of Meltzoff (1995) for the 18-month old infants and subsequently no differences were found in the frequency of target behavior between the conditions in which the target behavior or the intentional behavior was demonstrated. However, both conditions differed significantly from the end state condition in which less target behavior was displayed by the infants. Compared to the 18-month old infants, the 12-month old infants imitated less target acts and the 12-month old children performed significantly better in the demonstration of the target behavior than in the other conditions. A study of Johnson, Booth, and O'Hearn (2001) showed that 15-month old infants were able to re-enact the target behavior as a stuffed animal performed only the intention. This finding was confirmed in a longitudinal study of Bellagamba, Camaioni, and Colonnesi (2006) who found that 12-month olds produced less target behavior than 15-month olds. However, the children with the highest intention understanding at 12 months, tended to remain high at 15 month of age.

A study of Huang, Heyes, and Charman (2002) provided a different interpretation of the behavioral re-enactment task by introducing an emulation condition in which the children were presented with the initial and the end state of the target behavior without the actual manipulation being demonstrated by the experimenter. Also a spatial contiguity condition was added in which parts of the object were moved into close proximity. Although performance in the latter two conditions could not be the result of intention reading, children produced the same amount of target behavior in all conditions. The authors conclude that performance on the behavioral re-enactment task could be explained in terms of observational learning and general knowledge of the objects. Although the study of Huang et al. (2002) provides an alternative explanation, suggesting that understanding of intentions is an important part of imitation and that the understanding of others' intentional action is not innate, but develops between 9 to 15 months of age.

It is well know that children with autism spectrum disorder (ASD) experience difficulty in tests related to other people's thoughts, knowledge, and beliefs (Baron-Cohen, 2001) and numerous studies have reported impaired social learning and imitation in children with ASD (see for reviews: Rogers & Pennington, 1991; Smith & Bryson, 1994; Vanvuchelen, Roeyers, & De Weerdt, 2011; Williams, Whiten, & Singh, 2004). On average, children with ASD perform better on meaningful rather than on non-meaningful gesture imitation tasks, better on motor imitation tasks with objects than on gestural imitation tasks and better on structured-elicited than naturalistic spontaneous tasks (Ingersoll, 2008; Stone, Ousley, & Littleford, 1997). Although evidence is scarce and long-term studies are lacking, (subtle) imitation problems in ASD seem to persist into adolescence and adulthood (Hobson & Lee, 1999).

In preschool children, specific aspects of neurocognitive functioning appear to be important predictors of severity of ASD symptoms associated with social learning including orienting to social stimuli, immediate and deferred motor imitation, shared attention and responses to emotional stimuli (Dawson, Meltzoff, Osterling, & Rinaldi, 1998). Munson, Faja, Meltzoff, Abbott, and Dawson (2008) found that reward learning as measured on neurocognitive tasks predicted the rate of social and communication growth between ages 4 to 6.5 years. One interpretation of these results is that better performance on reward learning and novel preference tasks increases the likelihood that the child will detect and learn from interactions in their social environment.

Only a few studies report results on the behavioral re-enactment procedure in children with ASD. Aldridge, Stone, Sweeney, and Bower (2000) compared 10 children with ASD aged between 2;2 – 4;2 (years;months) with 10 normally developing infants aged 0;5 to 1;10. Participants were matched on an object concept task and presented with three gestural imitation tasks and three tasks of the intention condition of the behavioral re-enactment procedure. Against expectations, 50% of the children with ASD completed all behavioral re-enactment tasks, 30% completed two tasks and 20% one task, while of the typically developing children only 20% completed one task. On the other hand, none of the children with ASD succeeded in any of the gestural tasks, while 50% of the typically developing children completed all tasks, 30% completed two tasks and 10% one task. However, all but three infants in the control group were younger than 12 months and therefore not expected to pass the behavioral re-enactment tasks (Bellegamba & Tomasello, 1999). Further, apart from performance on an object performance task and the absence of language, no information was provided on the developmental ages of the children with ASD, making results difficult to interpret.

Carpenter, Pennington, and Rogers (2001) used a within subject design and a control group of 11 children with developmental disabilities to assess the understanding of unfulfilled intentions in eleven children with autism. Children were 2;6 to 5 years old and had a nonverbal developmental age of 2;2 to 4;7 years. In addition to the four conditions used by Meltzoff (1995), the authors added an additional 'end-state' condition, in which the experimenter showed the object in the target end state without showing how that end state was achieved (see also Bellagamba & Tomasello, 1999). Results indicated that children with developmental disabilities and children with ASD showed the target behavior more often in the intention and target condition than in the manipulation, end state, or baseline conditions. However, children with developmental delays performed the target behavior significantly more often in the intention and end state condition than in the manipulation condition, but this difference was not significant for the children with ASD. A significant positive correlation between chronological age and the relative performance of children with ASD in the intention condition was found, but this correlation was not significant for developmental age. When compared to the frequencies of target behavior displayed by typically developing children of 30 to 40 month old (Charman & Huang, 1999), Carpenter et al. (2001) concluded that the children with ASD and the children with developmental disabilities in their study display generally less target behavior than typically developing children. So, the relatively poor performance of the children with developmental delay could explain why an autism specific deficit was not found. Furthermore, included children had a relatively high (developmental) age and could have outgrown the problems in intentions understanding. Although studies of Aldridge et al. (2000) and Carpenter et al. (2001) provide preliminary evidence on the ability of intention understanding in children with ASD, results are not conclusive.

Therefore, the aim of the present study was to asses (a) the understanding of intentions in children with ASD and ID using a within subject design in a larger sample with a broader range of (developmental) age, (b) which skills (including developmental age and severity of autism) are associated with the performance on the unsuccessful intention task, and (c) explore the relation between the understanding of intentions and language.

We hypothesized that children with ASD and ID would show more target behavior when the experimenter demonstrated the target behavior than when an unsuccessful attempt was demonstrated and that more off-task behavior (no response/other behavior) would be shown after the unsuccessful attempt is demonstrated than when the experimenter demonstrated the target behavior. Second, given that several studies have found that chronological and/or developmental age is associated with the understanding of intentions (e.g., Bellagamba & Tomassello, 1999), we expected that children with a lower developmental age would display less target behavior in the unsuccessful attempt condition than children with a higher developmental age and larger differences between both conditions were expected for children with a lower developmental age.

Third, it was hypothesized that as the severity of autism increases, understanding of intentions decreases. In individuals with ASD, a large variability in behavioral and cognitive characteristics is seen. As the three subtypes of social interaction and communication (i.e., aloof, passive, and active-but-odd) may refer to distinct subgroups of children with ASD, they were included in this study. Fourth, given their behavioral characteristics, children classified as active-but-odd were expected to have a better understanding of intentions, while those classified as aloof were expected to have the least understanding of intentions (Castelloe & Dwason, 1993; Wing & Gould, 1979).

Because children with ASD are reported to experience difficulty in being flexible in their behavior towards their use of objects (Didden et al., 2008; Peters-Scheffer et al., 2008), behavioral flexibility was also assessed. Extracting relevant information from interactions in changing social contexts and persons requires flexibility. Preservation on familiar and predictable objects, persons and routines may lead to a failure to attend to and comprehend social and communicative information (Munson et al., 2008). Fifth, we hypothesized that children with greater behavior inflexibility would experience more difficulty in displaying the target behavior as the experimenter models an unsuccessful attempt than children who show greater behavioral flexibility.

Considering the interplay between imitation, early social communication, and language acquisition, data were collected on early social communication skills (i.e., joint attention, behavioral requests and social interaction) and receptive and expressive language. As described in Meltzoff et al. (2009), the process of language learning tends to be highly guided by social input. During the first year, infants are able to discriminate sounds from all languages, but with development this ability decreases and by one year of age the ability to differentiate sounds of foreign languages deteriorates. Five-month olds imitate adults' vowel sounds, but no longer acoustically matched non-speech sounds, and 9-month olds may learn a foreign language during social interchanges, but not when they are exposed to television or audiotapes. Since children with ASD might not benefit sufficiently from social mediated learning opportunities, this may have a cascading effect on their language development. Sixth, we expected that children with less early social communication skills would perform less accurately on the behavioral re-enactment tasks. Finally, we predicted that performance on the behavioral re-enactment tasks would be associated with scores on the language acquisition tests with lower tests scores associated with poorer task performance.

3.2 Method

3.2.1 Participants and setting

The participants were 100 children diagnosed with ID and autism (*n* = 83) or PDD-NOS (*n* = 17). The sample consisted of 77 boys and 23 girls. The children were between 3:7 to 9:11 (*M* = 5:10 years; *SD* = 19.76 months) participated. All children attended a preschool or school for children with ID in the Netherlands. Children with neurological disorders of known etiology and major physical impairments interfering with data collection (e.g., severe visual impairments and severe epilepsy) were excluded from the study.

Prior to their inclusion, all children received a diagnosis of ASD (i.e., autism or PDD-NOS) and ID by a clinician who was independent of the study and in according with the DSM-IV or ICD-10 criteria. The diagnoses were confirmed by the Autism Diagnostic Observation Schedule (ADOS; Lord, Rutter, DiLavore, & Risi, 2006), the Childhood Autism Rating Scale (CARS; Schopler, Reichler, & Rochen Renner, 2007), Mullen Scales of Early Learning (MSEL; Mullen, 1995), and the Vineland Adaptive Behavior Scales (VABS; Sparrow, Balla, & Cicchetti, 1984). An IQ below 80 in combination with an age equivalent score on the VABS that was at least 6 months lower than the child's chronological age was used as a criterion to confirm the classification of ID. The Wing subgroups questionnaire indicated that 72 children had the aloof subtype, while 18 children had the passive subtype, and 10 the active-but-odd subtype. Characteristics of the participants are listed in Table 1.

3.2.2 Measures and procedure

The children were identified by local preschools and schools for children with ID in the Netherlands. They distributed letters to the parents of children who met the following intake criteria: (a) chronological age between 2 and 10 years, (b) diagnosis of ID and ASD by an independent psychiatrist or psychologist and supported by psychometrically reliable and valid measures, such as the Autism Diagnostic Interview-revised, ADOS, Bailey Scales of Infants Learning, SON-r 2.5-7, and/or the Vineland Adaptive Behavior Scales (Lord, Rutter, & Le Couteur, 1994; Lord et al., 2006; Snijders, Tellegen, Winkel, & Laros, 1996; Sparrow et al., 1984; Van der Meulen, Ruiter, Lutje Spelberg, & Smrkovsky, 2002), and (c) the child lived at home so that parents were able to provide information about their child. All parents gave their written consent and did not receive any honorarium for their participation.

Table 1. Characteristics of the participants at baseline (N=100).

	Mean	*SD*	*Range*
Cognitive functioning			
Developmental age in months	23.49	8.56	4.75-52
IQ	35.97	16.02	10.59-78.78
Visual reception	25.78	8.94	5-54
Fine motor	27.48	7.79	6-51
Receptive Language	20.64	10.81	5-55
Expressive Language	20.05	10.12	3-48
Adaptive behavior in months			
Composite	19.96	6.52	11-45
Communication	24.67	10.27	12-53
Daily Living Skills	22.67	8.53	11-47
Socialization	21.96	6.79	11-46
Behavior flexibility[2]			
Behavior flexibility (total)	10.16	6.69	0-29
Behavior flexibility: objects	6.33	3.87	0-16
Behavior flexibility: environment	2.22	2.11	0-9
Behavior flexibility: persons	0.72	0.99	0-4
Autism			
ADOS total	15.51	3.90	7-24
ADOS communication	5.92	1.86	2-10
ADOS social interaction	9.67	2.79	2-16
CARS	41.23	5.69	30-53
Early social communication scales[1]			
Initiating joint attention	7.38	8.44	0-37
Responding to joint attention	94.12	68.04	0-200
Initiating behavioral requests	23.41	7.34	2-43
Responding to behavioral requests	65.99	32.73	0-100
Initiating social initiations	2.62	1.82	0-8
Responding to social initiations	6.65	3.35	0-18
Language			
Receptive language (PPVT)	25.21	4.85	21-43
Receptive language (Reynell)	19.98	8.01	14-43
Expressive language (WO)[1]	20.47	9.54	14-56

Note. [1] *n*= 97; [2] *n*= 92

Once participants were selected, the first author scheduled in-home interviews with the parents to administer the VABS (Sparrow et al., 1984) and the CARS (Schopler et al., 2007). The latter was completed both on parental interview and behavioral observations conducted by the first author. A week before the parental interview, parents completed the Behavioral Flexibility Rating Scale-revised (BFRS-R; Peters-Scheffer et al., 2008) and the Wing Subgroups Questionnaire (WSQ; Castelloe & Dawson, 1993), which was sent out by mail along with an information letter with contact information. Parents returned the completed BFRS-R to the first author during the interview. If not returned during the interview, the first author sent a reminder within 6 weeks with an opportunity to return the completed questionnaires.

In the same month as the interview, five assessments were completed at the preschool or school. These assessments were: (a) Mullen Scales of Early Learning, (b) Autism Diagnostic Observation Schedule, (c) Early Social Communication Scales (Mundy, Delgado, Block, Venezia, Hogan, & Seibert, 2003), (d) the behavioral re-enactment tasks, and (e) three language tests. The language tests were the Reynell Developmental Language Scales, the Peabody Picture Vocabulary Test and the Schlichting Test for Language Production. Testing occurred in a separated room at the child's preschool or school and were videotaped.

Re-enactment tasks

The re-enactment tasks were administered with five sets of two objects in line with Meltzoff (1995). The sets of objects and the tasks are described in Table 2. After a warm-up period in which the experimenter and the child played with some toys, the child was seated at a table facing the experimenter. The objects were placed within reach of the experimenter, but outside the visual field of the child.

Table 2. Behavioral re-enactment tasks used in this study resembling the tasks of Meltzoff (1995). Each task was repeated 3 times. Operational definitions of the child's behavior are provided in italics.

Pair	Objects	Target behavior	Unsuccessful attempt
1.	Dumb-bell	Pull halves apart. *The child pulls the dumbbell apart.*	Try to pull halves apart, but one hand slips of (alternating ends). *When holding the dumbbell, the child's hand or hands slip of.*
	Plug	Pull a straight four pin plug and contra plug apart. *The child pulls the plug apart.*	Try to pull a straight four pin plug and contra plug apart, but one hand slips off (alternating ends). *When holding the plug, the child's hand or hands slip of.*
2.	Light	Use the stick to push a button attached to a wooden rectangular to activate a light. *The child activates the light by using the stick to push a button.*	Move the stick towards the button attached to a wooden rectangular, but miss each time. *The child moves the stick in a horizontal position in the direction of the light, but misses the button to activate the light. Hence, the light is not activated.*

Continuation Table 2.

Pair	Objects	Target behavior	Unsuccessful attempt
	Buzzer	Use the stick to push a button attached to a wooden rectangular to activate a buzzer. *The child activates the buzzer by using a stick to activate the buzzer.*	Move the stick towards the button attached to a wooden rectangular, but miss each time. *The child moves the stick in a horizontal position in the direction of the buzzer, but misses the button to activate the buzzer. Hence, the buzzer is not activated.*
3.	Cube with beads	Drop the beads into an opening of the cube. *The child lowers the beads vertically all the way into the cube.*	Moves the beads towards the opening of the cube but miss each time. *The child lowers the beads vertically onto the table beside the cube.*
	Cylinder with scarf	Drop scarf into the cylinder. *The child lowers the scarf vertically all the way into the cylinder.*	Move the scarf towards the opening of the cylinder but miss each time. *The child lowers the scarf vertically onto the table besides the cylinder.*
4.	Rectangle	Place a cord loop over a horizontally protruding peg attached to a woorden rectangle. *By holding the cord between the tumb and indexf inger, the child puts the cord loop over the peg so that the peg protrudes through it.*	Move the cord loop to the horizontally protruding peg, but miss and drop it on the table. *The child holds the cord between the thumb and index finger and moves the cord toward the horizontally protruding peg, without the peg protruding the cord.*
	Triangle	Place a flat plastic teddy bear with a hole in the middle over a horizontally protruding peg attached to a wooden triangle. *The child puts the hole of a flat plastic teddy bear over a horizontally protruding peg attached to a wooden triangle so that the peg protrudes through it.*	Move the flat plastic teddy bear with a hole in the middle towards the horizontally protruding peg, but miss and drop it on the table. *The child moves the teddy bear towards the horizontally protruding peg attached to a wooden triangle so that the peg protrudes through it.*
5	Two blocks	Place one block with a hole over a vertically protruding peg on a similar block. *The child places the block on a similar block with the hole of the first block over the vertically protruding peg of the other block so that the peg protrudes through the hole of the block.*	Try to place one block with a hole over a protruding peg on a similar block but let the block slip of. *The child places the block on a similar block with the hole of the first block next to the vertically protruding peg of the other block, making the first block slip of the other.*
	Block and ball	Place a ball with a hole over a vertically protruding peg on a block. *The child places the hole of the ball over the vertically protruding peg on a block so that the peg protrudes through it.*	Try to place a ball with a hole over a protruding peg on a block, but let the ball slip of. *The child places the ball on the vertically protruding peg with the hole on the side letting the ball fall down on the table.*

Tasks were presented one at a time in a counterbalanced order to prevent an interaction effect between tasks and conditions. Since more orders (120 per condition) than participants were available, for each condition an order was selected at random. Each order was used only once. Because a within subject design was used, each child received five objects in the target behavior condition and five objects in the unsuccessful attempt condition.

In the target behavior condition the experimenter demonstrated the target behavior three times in approximately 20 seconds, while in the unsuccessful attempt condition during which the experimenter attempted, but did not successfully complete the task three times during an (approximately) 20-second trial. Therefore, the child did not see the end stage of the task. After demonstrating each task (i.e., not for the total block of 5 tasks), the materials were placed on the table in front of the child for a response period of 20 seconds. No additional verbal cues were provided. After the response period (either in the intention or target behavior condition), the object was removed and the next object was presented using the same procedures described until all 10 objects/tasks had been presented.

Five undergraduate students coded the videotapes. Before the students started coding, the materials were shown and all tasks in the unsuccessful attempt and target behavior condition were demonstrated. First, the students identified the task (e.g., dumbbell) and the condition (e.g., whether the experimenter demonstrated the target behavior or the demonstrated an unsuccessful attempt) to determine whether the experimenter used the right task and condition for the child. There was a 99.9% agreement between the condition scored by the students and the condition assigned by the experimenter. Subsequently, the student indicated whether the child displayed the target behavior, performed the task that was supposed to occur in the unsuccessful attempt, showed no response, or engaged in some other behavior. After a brief training period, in which several videos were shown to the students, inter-observer reliability was assessed on five videos of five children who were not participating in the current study. A mean kappa (Cicchetti, 1994) of 0.90 (range: 0.83 to 1.0) was obtained on determining the condition (i.e., unsuccessful attempt vs. demonstration of the target behavior) and a kappa of .85 (range: 0.64 to 1.0) on whether the child performed the target behavior. To assess inter-observer reliability during the coding, 28.7% of the videotapes were randomly selected and independently coded by a second observer. The resulting kappa was 0.91.

Standardized assessments

The standardized instruments were designed to assess a broad range of skills for both typically and atypically developing children and were chosen for their adequate psychometric properties.

Autism

The Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2006) is a semi-structured, interactive protocol designed to assess social and communicative functioning in children with ASD. Based on a parental interview and observations of the first author, the Childhood Autism Rating Scale (CARS; Schopler et al., 2007) was completed. This scale consists of 15 items on social emotional and interaction skills, response to sensory information and language and communication skills to distinguish children with autism from developmentally handicapped children without autism. The Wing Subgroups Questionnaire (WSQ; Castelloe & Dawson, 1993) is a questionnaire with 13 behavioral domains (e.g., communication, social approach, play, imitation, motor behavior, resistance to change) on with parents rate their child's behavior on a scale from zero (never) to six (always) for each domain. A summary score is calculated for each subtype and the highest summary score is considered to indicate the child's subtype.

Developmental age

The Mullen Scales of Early Learning (MSEL; Mullen, 1995) were used to evaluate cognitive ability in the domains of visual reception, fine motor, expressive language and language comprehension. The MSEL provides a standardized score for children between 0 to 60 months of age, but due to their chronological ages not all standardized scores could be calculated for the children participating in the current study. Therefore, developmental ages and ratio IQ were used in our analyses. A mean developmental age was calculated based on the developmental ages on the four subscales of the MSEL, while ratio IQ was calculated by the mean developmental age divided by the chronological age of the child and multiplied by 100.

Adaptive behavior

The Vineland Adaptive Behavior Scales – survey form (VABS) is a semi-structured parent interview that was administered to assess the adaptive behavior on adaptive behavior composite and in the domains: communication, daily living skills and socialization (Sparrow et al., 1984). The Vineland provides age equivalency scores for each domain.

Early communication and language

The Early Social Communication Scales (ESCS) is a videotaped semi-structured observational instrument that is used to measure initiating and responding to joint attention, behavioral requests, and social interaction. The protocol involves the presentation of a set of toys and activities in an attempt to evoke social and communicative behavior in an ecologically valid context (Mundy et al., 2003). The ESCS was administered by the first author and videotapes were scored by four raters, unaware of the purpose of the study. Inter-observer reliability was assessed using videotaped data from 28.7% of the children and Pearson's correlations between the paired rating of the six subscales ranged from 0.98 to 1.0, suggesting excellent reliability.

Receptive language was measured by the comprehension scales of the Dutch version of the Reynell Developmental Language Scales (Van Eldik, Schlichting, Lutje Spelberg, Van der Meulen, & Van der Meulen, 1995) and the Peabody Picture Vocabulary Test (PPVT; Dunn & Dunn, 1997), while expressive language was measured by the vocabulary test of the Schlichting Test for language production (Schlichting, Van Eldik, Lutje Spelberg, Van der Meulen, & Van der Meulen, 1995).

Behavioral flexibility

The Dutch version of the revised Behavior Flexibility Rating Scale (BFRS-R; Peters-Scheffer et al., 2008) was used to assess severity of behavioral flexibility. One of the parents completed a 3-point Likert-type scale of 16 items referring to extend to which a child reacts to unexpected events and changed routines. The BFRS-R yields a total score and scores for three subscales: (a) flexibility towards objects, (b) flexibility towards the environment, and (c) flexibility with respect to interpersonal interactions.

3.3 Results

First, frequencies of target behavior, unsuccessful attempts and other behavior were calculated for both conditions. Using paired sample *t*-tests, participants showed significantly more target behavior in the target behavior condition (M = 2.71; SD = 1.83) than in the unsuccessful attempt condition (M = 1.56; SD = 1.29), $t(99) = 8.33$; $p < .001$. Additionally, more off-task behavior (e.g., no reaction/other behavior) was shown in the unsuccessful attempt condition (M = 2.65; SD = 1.65) than in the target behavior condition (M = 2.20; SD = 1.81, $t(99) = 3.95$; $p < .01$).

An additional paired sample *t*-test showed that unsuccessful attempts were not evoked by the tasks. On average, participants engaged in the unsuccessful attempt significantly more frequently in the unsuccessful attempt condition (M = 0.75; SD = 1.05) than in the target behavior condition (M = 0.04; SD = 0.24; $t(99) = 6.78$; $p < .001$). Hence, children's performance was influenced by the adult model (i.e., the condition) and were thus less likely to be due to the materials used.

As potential differences in difficulty between the parallel tasks of each pair might explain differences found between conditions, a multivariate analysis with the frequency of target behavior displayed in both conditions as dependent variables was performed. Using Wilks' Lamba, there was no significant effect of the condition and the parallel tasks used in pair 1 (Λ = .97; $F(2,69) = 1.01$; $p = .37$); pair 2 (Λ = 1.00; $F(2,69) = 0.03$; $p = .97$); pair 3 (Λ = .99; $F(2,69)$ = 0.40 $p = .67$), pair 4, (Λ = .97; F (2,69) = 1.05; $p = .36$), and pair 5, (Λ = 1.00; $F(2,69) = 0.13$; p = .88). This indicates that the parallel tasks within each pair were equally difficult (e.g., lowering the beads into the cube was as difficult as lowering the scarf into the cylinder). Thus, assignment of a specific condition to a specific task did not appear to contribute to the frequency of correct target behavior displayed either in the target condition or in the unsuccessful attempt condition.

3.3.1 Impact of developmental age on the understanding of intentions

Considering the interplay between developmental age and imitation abilities and results of studies indicating an effect of age on the performance of infants on the behavioral re-enactment task, the effect of developmental age was assessed using a repeated measures analysis. Children with a developmental age below 9 months (n = 1) and above 39 months (n = 3) were excluded from the analysis due to small sample sizes. Figure 1 displays the target behavior shown in the target behavior and the unsuccessful attempt condition specified by developmental age. Results of a One-way ANOVA show that the performance of the children on tasks was significantly affected by the condition and the developmental age of the child, $F(4, 91) = 4.73$; $p < .01$.

A repeated measures analysis for each developmental age group revealed that the differences between the target behavior condition and unsuccessful attempt condition was significant in each developmental age group; $F(1,17) = 5.67$; $p = .03$ for 9 to 14 months, $F(1,18) = 11.50$; $p < .01$ for 15 to 20 months, $F(1,28) = 11.38$, $p < .01$ for 21 to 26 months, $F(1,18) = 41.23$; $p < .01$ for 27 to 32 months, and $F(1,10) = 26.67$, $p < .01$ for 33 to 38 months.

Planned contrasts (repeated) revealed significant changes in the frequency of target behavior in the target behavior condition between children with a developmental age of 9 to 14 months and from 15 to 20 months ($t(4) = - 4.13$; $p < .01$) and between 21 to 26 months and 27 to 32 months ($t(4) = -3.14$; $p < .01$) were significant, while differences between 15 to 20 months and 21 to 26 months ($t(4) = -1.38$; $p = .17$) and between 27 to 32 months and 33 to 38 months ($t(4) = -1.11$; $p = .27$) were not significant.

In the unsuccessful attempt condition differences in children with a developmental age between 9 to 14 months and from 15 to 20 months ($t(4) = - 3.14$; $p < .01$) between 15 to 20 and 21 to 26 months ($t(4) = -2.10$; $p = .03$) and between 27 to 32 and 33 to 38 months, ($t(4) = -2.91$; $p < .01$) were significant. However, the difference between children with a developmental age of 21 to 26 months and 27 to 32 months, ($t(4) = -0.11$; $p = .78$) was not significant.

3.3.2 Other variables associated with understanding goals

Partial correlation coefficients controlling for developmental age were calculated to explore which variables were associated with the target behavior displayed in the unsuccessful attempt condition. Results are displayed in Table 3. There were associations with small effects between the target behavior displayed in the unsuccessful attempt condition and behavioral flexibility (total scale), receptive language (PPVT) and the ADOS (total scale and subscales) and associations with moderate effects between the target behavior displayed in the unsuccessful attempt condition and responding to social interaction, behavioral flexibility towards objects and imitation (Cohen, 1988).

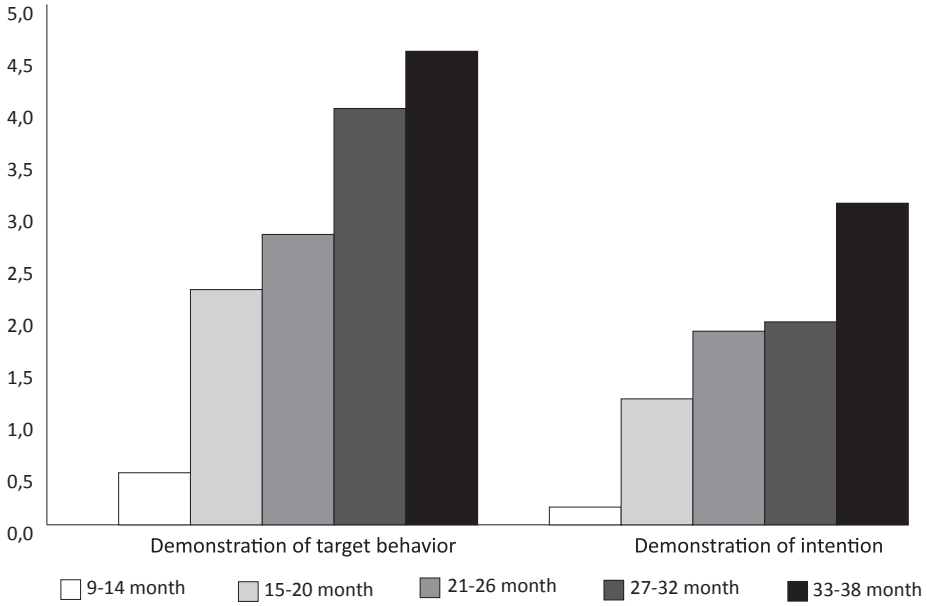

Figure 1. Target behavior performed in the target behavior and the unsuccessful attempt condition broken down by developmental age.

3.3.3 Impact of subtype and severity of ASD on the understanding of intentions

Autism severity (as measured by the ADOS), controlled for developmental age, was significantly correlated with the frequency of target behavior displayed in the unsuccessful attempt condition (r = -.25; p = .0), indicating that as the severity of autism increased, the frequency of target behavior displayed in the unsuccessful attempt condition decreased. A visual representation of the association between severity of autism and developmental age is presented in Figure 2. A one-way ANOVA displayed an overall significant effect of autism subtype on the target behavior displayed in the unsuccessful attempt condition (F(2, 97) = 5.82; p < .01). Planned contrasts revealed that children with the aloof subtype displayed significantly less target behavior in the unsuccessful attempt condition (M = 1.32; SD = 1.27) than children with the active-but-odd subtype (M = 2.60; SD = 1.17; t (97) = -3.08; p < .01), but not less than children with the passive subtype (M = 1.94; SD = 1.11; t(97) = -1.93; p = .06). There was no significant difference in the frequency of target behavior displayed in the unsuccessful attempt condition between children with the active-but-odd and the passive subtype (t(97) = 1.35; p = .18).

Table 3. Partial correlations with target behavior displayed in the unsuccessful attempt condition controlling for developmental age.

	r	*p*
Chronological age	-.09	.39
Behavior flexibility[2]		
Behavior flexibility	.27	.01
Behavior flexibility: objects	.32	.00
Behavior flexibility: environment	.11	.29
Behavior flexibility: persons	.12	.24
Autism		
ADOS total	-.25	.01
ADOS communication	-.22	.03
ADOS social interaction	-.20	.05
CARS	.05	.62
Early social communication scales[1]		
Initiating joint attention	.10	.32
Responding to joint attention	.20	.05
Initiating behavioral requests	.20	.06
Responding to behavioral requests	.18	.07
Initiating social initiations	.05	.64
Responding to social initiations	.31	.00
Language		
Receptive language (PPVT)	-.18	.02
Receptive language (Reynell)	-.07	.51
Expressive language (WO)[1]	-.23	.08
Frequency target behavior in target behavior condition (imitation)	.47	.00

Note. df= 97; [1] *df = 94;* [2] *df*= 89

However, the difference between the aloof children and the children with the active-but-odd subtype might be explained by the developmental age of the participants, as children in the aloof group had a significantly lower developmental age (*M* = 20.19; *SD* = 6.34) than children with the passive (*M* = 31.08 ; *SD* = 8.67) or active-but-odd subtype (*M* = 31.08; *SD* = 8.67).

3.3.4 Predictors of understanding other's goals

To determine whether the target behavior displayed in the unsuccessful attempt condition can be predicted from other child characteristics, a hierarchical regression was employed with the frequency of target behavior shown in the unsuccessful attempt condition as the dependent variable and developmental age, target behavior in the target behavior condition, autism severity (total score of the ADOS), behavioral flexibility towards objects and responding to social interaction as independent variables.

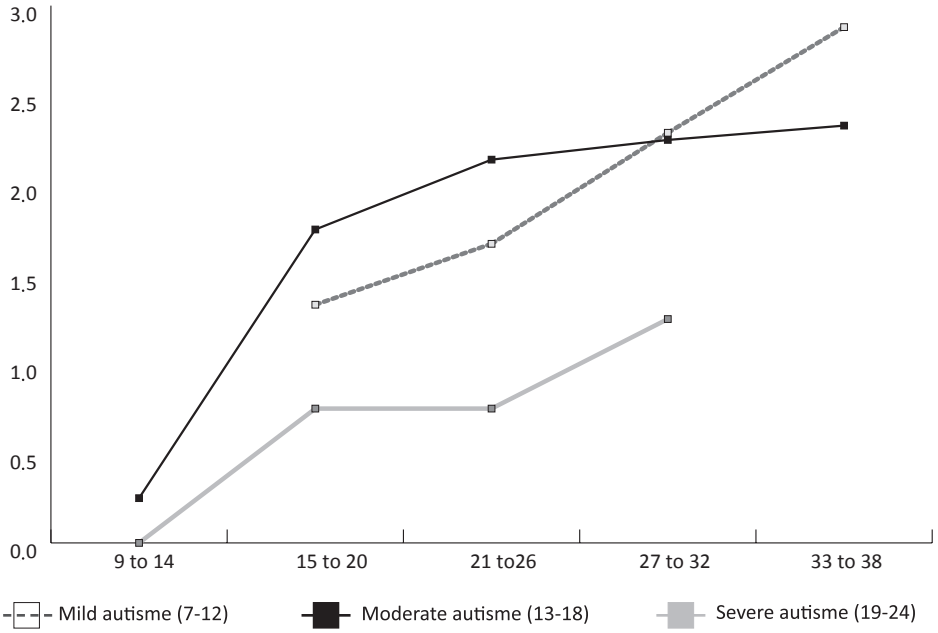

Figure 2. Frequency of target behavior displayed in the unsuccessful attempt condition by developmental age broken down by autism severity group.

As (developmental) age seems a consistent predictor of the understanding of intentions, this variable was entered in the first step. The ability to imitate seems a precursor to display the target behavior in the unsuccessful attempt condition and therefore the frequency of target behavior shown in the target behavior condition was entered in the second step of the regression. Finally, using a stepwise method autism severity, behavioral flexibility towards objects and responding to social interaction were added in the third step to explore whether any of these variables contributed significantly to the model. Table 4 displays the unstandardized regression coefficient (*B*) and standard error of the unstandardized regression coefficient (*SE B*) and the standardized regression coefficient (*β*) at each step.

Table 4. Multiple regression with frequency of target behavior in the unsuccessful attempt condition as dependent variable.

	Step 1			Step 2			Step 3		
	B	*SE B*	*β*	*B*	*SE B*	*B*	*B*	*SE B*	*β*
Constant	-.32	.34		-.10	.30		-.21	.30	
Developmental age	.08	.01	.55*	.03	.02	.17	.02	.02	.10
Target behavior (imitation)				.39	.08	.55*.	.35	.08	.49*
Behavioral flexibility: objects							.08	.03	.23*
	$F(1,87) = 36.71$; $p < .001$			$F(2,87) = 36.02$; $p < .001$			$F(3,85) = 27.48$; $p < .001$		

Note. $R^2 = .30$ for step 1; $\Delta R^2 = .16$ for step 2; $\Delta R^2 = .04$ for step 3; $^*p < .001$.

R^2 was significantly different from zero at the end of each step. Although a significant predictor when entered alone, in combination with the frequency of target behavior displayed in the target condition (i.e., the ability to imitate) developmental age does not longer contribute significantly to the model in Steps 2 and 3. All three models significantly improved the ability to predict the frequency of target behavior displayed in the unsuccessful attempt condition. As the predictors responding to social interaction, $t = 1.61$; $p = .11$ and severity of autism, $t = -1.64$; $p = .11$ did not significantly contribute to the models, they were excluded from the analyses. After Step 3, in which developmental age, the frequency target behavior in the target behavior condition (imitation), and behavioral flexibility towards objects were included, 49.2% of the variance was accounted for.

3.3.5 Association between the understanding of intentions and language

Finally, the relation between target behavior displayed in the unsuccessful attempt condition was assessed. As the measures of language strongly correlate (*r's* range from .73 to .85) a multivariate analysis was performed with the frequency of target behavior in the unsuccessful attempt condition as independent variable and receptive language (both on the Reynell and PPVT) and expressive language as dependent variables. Target behavior displayed in the unsuccessful attempt condition has a significant effect on language, $\Lambda = .60$; $F(15, 246.09) = 3.30$; $p < .01$, even when controlled for developmental age, $\Lambda = .60$; $F(15, 243.33) = 3.36$; $p < .01$.

The multivariate analysis was followed up with an univariate analysis for expressive language and both tests of receptive language. As expected, there was a significant effect of the target behavior displayed in the unsuccessful attempt condition on all dependent variables; $F(5, 94) = 10.27$; $p < .01$ for the Reynell, $F(5, 94) = 7.65$; $p < .01$ for the PPVT and $F(5, 91) = 5.90$; $p < .01$ on expressive language. Subsequently, per variable planned contrasts (repeated) were used to compare the language scores of children in each category (e.g., displaying 0, 1, 2, 3 or 4 target behaviors) to the previous category. As only one child displayed five target behaviors in the unsuccessful attempt condition, this category was not interpreted in the contrasts. Planned contrasts did not reveal any significant differences in scores between each category compared to the previous category. Descriptives are presented in Table 5.

Table 5. Means and standard deviations of developmental age in months on the language test specified by the frequency of target behavior performed in the unsuccessful attempt condition.

	PPVT		Reynell		Schlichting	
	M	*SD*	*M*	*SD*	*M*	*SD*
0	23.08	4.43	15.38	4.00	15.27	4.54
1	24.92	4.83	18.42	7.17	21.21	10.61
2	25.87	5.15	22.52	8.25	21.95	8.99
3	26.29	4.24	22.24	8.20	20.29	8.98
4	29.29	4.35	29.86	9.30	34.00	9.56

3.4 Discussion

This study addressed the understanding of intentions in children with ASD and ID aged between 2 and 10 years of age. A within subject design with five pairs of parallel tasks based on Meltzoff's (1995) behavioral re-enactment procedure was used. All children completed five tasks in a condition in which the experimenter modeled the target behavior successfully (target behavior condition) and five tasks in which the experimenter tried, but failed to perform the target behavior (unsuccessful attempt condition), thereby creating a mismatch between the adult's goal and the demonstrated surface behavior.

Participants showed significantly more target behavior in the target behavior condition than in the unsuccessful attempt condition, which might suggest that children with ASD have difficulty in interpreting other's intentions. This is in contrast with the two other studies in which evidence for an autism specific deficit was not found (Aldridgde et al., 2000; Carpenter et al., 2001). However Carpenter et al. (2001) also noted that within group patterns suggested that children with ASD might have a more simplistic understanding of intentions than other children. The discrepant findings, might stem from the fact that these two prior studies had

relatively small samples and the children in the study of Carpenter et al. (2001) had a higher (developmental) age than the children in our study. Because developmental age is correlated to the understanding of intentions, this might explain differences in outcome between studies. Still, because we did not include a control group, it would be premature to conclude that children with ASD have a deficit in intention understanding. However, when compared to 12 to 18-month old infants (Bellegamba & Tomasello, 1999; Bellegamba et al., 2006; Meltzoff, 1995), our data suggest that children with ASD with a comparable developmental age, might be expected to show less target behavior in both conditions. In typically developing children, intention understanding is reported to develop between 9 to 15 months and in 18-month old infants, the same amount of target behavior is seen after demonstration of the target behavior as after demonstration of the intention of such behavior (see e.g., Bellegamba & Tomasello, 1999; Meltzoff, 1995). Our study shows that the frequency of target behavior increases as developmental age increases, but that differences between the frequencies of target behavior displayed in the two conditions remain significant in all age groups. This might suggest a delayed development of intention understanding in children with ASD and ID. Further research with a longitudinal design would thus be indicated to further investigate whether children with ASD and ID have a deficit in intention understanding as compared to children with ID only, whether the understanding of intentions is developed in children with ASD, and which variables influence the development of intention understanding and are affected by a lack of intention understanding.

In addition to developmental age, severity of autism - as measured by the ADOS - seemed to influence the understanding of intentions. This relation was not found for autism severity on the CARS. However, the ADOS composite is comprised of items related to communication and social interaction, while the CARS focuses relatively less on communication and social interaction, as it also contains items related to emotional responses, body and object use, adaption to change, responses to sensory stimuli, fear, and activity level (Schopler et al., 2007). These CARS items might be less associated with intention understanding than communication and social interaction.

Similarly, children with the aloof subtype seemed more impaired in intention understanding than children of the passive or active-but-odd subtype. This seems in accordance to the relation between severity of autism and intention understanding and typically a lower IQ is found in the aloof subgroup (Castelloe & Dawson, 1993). While the present study seems to be the first to explore the relation between intention understanding and severity of autism, other imitation studies have reported a relation between autism severity and imitation abilities (Zachor, Ilanit, & Ben Itzchak, 2010).

Contrary to our expectations, behavioral inflexibility towards objects was positively associated with intention understanding. As the superficial movements in both conditions are fairly equal (put the ball on the stick versus trying to, but not succeeding to put the ball on the stick), experimenter's movements might have served as an effective model for the child's response to some extent.

Children who scored lower on the BFRS-R (i.e., had less behavioral flexibility) were more impaired in terms of exhibiting alternative behavior (e.g., throwing the ball away) and might thus have been more influenced by the experimenter's model (e.g., perform a task with the ball and the stick; not only with the ball or the stick).

A relation between the understanding of intentions and early social communication and the acquisition of language is suggested in the literature and by the data of our study. For example, infants' gaze following behavior at 10 to 11 months of age significantly predicts accelerated vocabulary growth through to 2 years of age (Brooks & Meltzoff, 2008). In addition, Heimann et al. (2006) showed that in typically developing children visual recognition memory, deferred imitation and turn taking skills predicted communicative gestures, while deferred imitation at 14 months also predicted vocal comprehension.

The associations between variables found in this study cannot be interpreted as evidence of causation and research addressing that changes in the understanding of intentions precedes changes in early social communication and language acquisition is warranted. Using fMRI, recent research (Scott-Van Zeeland, Dapretto, Ghahremani, Poldrack, & Bookheimer, 2010) found that — in contrast to typically developing children — children with ASD do not seem to experience social stimuli as rewarding. According to the social motivation theory (Dawson, Webb, & McPartland, 2005; Schultz, 2005), this deficit in the reward mechanism is hypothesized to result in children with ASD spending less time attending to faces, speech, and other social stimuli. This in turn may lead to problems in terms of acquiring knowledge related to the processing of facial expressions and speech (Dawson, Meltzoff, Osterling, Rinaldi, & Brown, 1998; Kuhl, Coffey-Corina, Padden, & Dawson, 2005). Furthermore, while typically developing children prefer variable feedback which is characteristic for social stimuli, children with ASD prefer the less variable feedback of non-social stimuli (Gergely & Watson, 1999).

Such impairments in social processing might also complicate the interpretation of other people's behavior and goals and consequently the selection of relevant behaviors to imitate. In this study, children who responded better to social interaction, performed better in intention understanding. This trend was also seen in responding to joint attention and to behavioral requests. Due to misinterpretation of other's goals, children with ASD might select irrelevant behaviors to imitate and thereby they would be expected to be less successful with observational learning approaches compared to typically developing children. There are also data to suggest that children with ASD imitate significantly better in a structured-elicited setting than in a naturalistic-spontaneous condition in which selection of the relevant behavior is necessary (Ingersoll, 2008). However, more research is needed to explore whether understanding intentions plays a significant role in selecting relevant behavior to imitate and whether this affects development of social learning in children with ASD. Lack of intention understanding might also influence the social aspect of imitation.

In a sample of 35 typically developing children, Colonessi, Rieffe, Koops, and Perucchini (2008) found that the abilities to understand intentions at 12 and 15 months of age predicted the later ability to explain other's actions in a psychological way at 39 months. This suggests that understanding of intentions is one of the first steps towards a theory of mind (see also: Meltzoff, 1995; 2007; 2011). As many children with ASD are shown to have deficits in performing theory of mind tasks (Baron-Cohen, 2001), the main results of this study are in line with theory of mind and executive function theories of autism, which posit early deficits in the understanding of intentionality. However, longitudinal research in children with ASD measuring both the understanding of intentions and theory of mind deficits would be necessary to further investigate this possible relation.

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Chapter 4

The Behavior Flexibility Rating Scale-Revised (BFRS-R): Factor Analysis, Internal Consistency, Inter-rater and Intra-rater Reliability, and Convergent Validity

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Abstract

We examined the psychometric properties of the behavior flexibility rating scale-revised (BFRS-R), a new scale intended for assessing behavioral flexibility in individuals with developmental disabilities. Seventy-six direct care staff members and 56 parents completed the BFRS-R for 70 children with developmental disabilities. Factor analysis revealed three factors (i.e., Flexibility towards objects, Flexibility towards the environment, and Flexibility towards persons) and results of several analyses indicated an excellent internal consistency and good intra-rater and inter-rater reliability of the total scale. These data suggest that the BFRS-R may provide a reliable rating of behavioral flexibility when used by direct-care staff and parents of children with developmental disabilities.

4.1 Introduction

Children with autism and related developmental disabilities are often said to insist on sameness and resist change (Rutter, 2005). The insistence on sameness and resistance to change may be viewed as a lack of behavioral flexibility (Wahlberg & Jordan, 2001). Thus many such children appear to have substantial difficulty in being flexible in their behavior and appear unable to cope with unpredictable changes in the environment (e.g., Lewis & Bodfish, 1998). Recent studies that have focused explicitly on the assessment of behavioral flexibility are lacking, as are contemporary instruments for measuring behavioral flexibility in specific situations in individuals with developmental disabilities.

Recently, Green et al. (2006) and Pituch et al. (2007) developed and evaluated the discriminant validity and factor structure of the behavior flexibility rating scale (BFRS). Based on the results of these two initial studies, a revised version of the scale (i.e., behavior flexibility rating scale-revised [BFRS-R]) was developed, which includes some wording and item changes (Green et al., 2007). Given these revisions, it would seem timely and important to assess the reliability and validity of the BFRS-R.

The BFRS and BFRS-R were developed as research tools for exploring issues related to behavioral flexibility and as clinical tools for identifying specific situations in which children with autism and related developmental disabilities insist on sameness or resist change. Systematic identification of these situations may be helpful in enabling clinicians to target interventions to scenarios that are problematic for the child. In an initial study, Green et al. (2006) showed that the BFRS has good discriminant validity. In this study, 726 parents used the BRFS to assess severity of behavioral inflexibility in children with autism, Asperger syndrome, and Down syndrome. Results showed that individuals with Asperger syndrome showed more problems in relation to the insistence on sameness as measured by the BFRS, followed by individuals with autism, and Down syndrome, respectively.

A subsequent factor analysis (Pituch et al., 2007) revealed that the BFRS appears to comprise two main factors: (a) Interruption/Disruption, and (b) Position/Location, with the remaining items comprising a third potential factor. The above studies suggest the instrument has potential for research and possibly also for clinical assessment. While these data are promising, further research is required to evaluate various psychometric properties of this instrument for assessing problems in behavior flexibility. The present study, therefore, was designed to examine the internal consistency, intra- and inter-rater agreement and convergent validity of the revised version of the BFRS.Data of this type may assist in evaluating the clinical utility of this measure.

4.2 Method

4.2.1 Participants and respondents

Respondents were 76 direct care staff members working in the Dutch day care centers where the children received early intervention services. At the time of the study, primary (*n* = 43) and secondary (*n* = 33) respondents had cared for participants for a mean of 2;2 years (range: 0.6-11 years) and 2.3 years (range: 0.7-11 years), respectively. Also parents (of whom 7 were fathers) of 56 children participated in this study. The scale was completed for 70 children (of whom 41 were boys) with developmental disabilities who attended two Dutch day care centers for children with disabilities. Their mean age was 6.7 years (range: 2-17 years). In addition to severe to mild intellectual disability, the children had a range of other diagnoses, including: (a) autism/PDD (*n* = 26), (b) Angelman syndrome (*n* = 4), (c) ADHD (*n* = 1), (d) Down syndrome (*n* = 3), (e) physical impairment (*n* = 11), and (e) other (*n* = 6).

4.2.2 Instruments

The instruments included the BFRS-R and the Sameness Questionnaire which served for the assessment of problems in behavioral flexibility. The BFRS-R is a revised version of the BFRS (Green et al., 2007). Compared to the original BFRS, the revised version includes several wording changes on individual items, the removal of one item and the inclusion of two new items. The BFRS-R is a 16-item rating scale for measuring behavioral flexibility in children with autism and related developmental disabilities in specific situations. (A copy of the BFRS-R is included in the Appendix A). Items in the BFRS-R refer to a number of specific and unexpected events and changed routines that may prove problematic for the individual. The severity of each potentially problematic situation is rated on a three-point Likert scale, ranging from '0' ('Not a problem at all') to '2' ('The situation causes severe problems'). The scale is completed by proxies who have known the individual for at least 6 months.

The Sameness Questionnaire is a 28-item questionnaire for assessing a child's resistance to change (Prior & MacMillan, 1973). Examples of items are: 'Does he become very upset if interrupted in what he is doing?', and 'Does he object to visiting new places?' Each items is rated on a 3-point Likert scale, ranging from '0' ('Behavior is not shown by child') to '2' ('Behavior is present to a considerable degree'). In a sample of 32 children (who were between 3 and 11 years of age) with various types of developmental disabilities Prior and MacMillan (1973) found that children with autism had a significantly higher mean flexibility score than children without autism indicating that these children had more problems with behavioral flexibility than those without autism.

4.2.3 Procedure

The BFRS-R was provided to the respondents with a letter explaining the aim of the study. Each respondent independently completed a Dutch-language translation of the BFRS-R without the involvement of an interviewer. However, they were informed that they could contact the first author in case they had any questions on how to fill in the scale. Three weeks after the completion of the first assessment, each respondent was asked to complete a second BFRS-R as well as the Sameness Questionnaire.

4.3 Results

4.3.1 Exploratory factor analysis

An exploratory factor analysis to identify possible subscales was conducted using all 132 BFRS-Rs completed by parents and staff members at the first assessment. A principle axing analysis revealed three factors, which were labeled as (a) Flexibility towards objects, (b) Flexibility towards the environment, and (c) Flexibility towards persons. Due to the correlation between factors one and three (i.e., r = .59), and between factors two and three (i.e., r = .46), direct oblimin rotation was used. In the pattern matrix factor loadings needed to be .30 or higher, and an item was retained on the factor on which it loaded most strongly. The percent of variance explained by the factors was 37.11, 3.16, and 7.54 for the factors 1, 2 and 3, respectively. Eigen values were 6.42 for factor 1, 1.04 for factor 2 and 1.76 for factor 3. Factor loadings are described in Table 1.

4.3.2.Internal consistency

Cronbach's α coefficient was calculated for internal consistency of the total scale using the data from all the respondents of the first assessment (α = .90). Alpha values for two of the subscales (i.e., Factor 1 [Items 1-3, 5-7, 10, 14 and 16] and Factor 2 [Items 4, 11-13, and 15]) were .90 and .73, respectively.

The alpha value of the third [Factor 3] subscale (Items 8 and 9) was .58. The alpha value for the total scale was excellent, suggesting that the total scale was homogeneous in content. Mean item-total correlation for the total scale was .55 (*SD* = 0.13) with a range from .28 to .74.

Table 1. Factor loadings for each item

		Factor		
		1	*2*	*3*
1.	A commenly used object is misplaced and cannot be found	.50		
2.	A planned event is delayed or cancelled with little warning because of unforseen circumstances	.67		
3.	The person is required to move from their current location and go to another location	.38		
4.	An object in the environment has been moved or repositioned from its usual location or position		.79	
5.	The person wants something what is not available	.82		
6.	An object or some materials that the person was using breaks or malfunctions	.65		
7.	A usual routine is altered or changed, for example the parent takes a new route home from school	.63		
8.	An unexpected interaction occurs with another person, for example a stranger tries to talk to the person			.67
9.	The person becomes momentarily separated from his/her family or group			.57
10.	Materials break, causing a premature end to an activity	.72		
11.	Another person is doing something annoying, for example making noise		.54	
12.	Objects or materials are not returned to their proper place at the end of an activity		.45	
13.	A new object, item, or person has been added to the environment		.42	
14.	An activity is interrupted before the person was able to finish the task	.83		
15.	A new activity is introduced into the person's routine		.60	
16.	Another person tries to use favorite possessions of the person		.68	

Mean item-total correlations for the subscales (1) Flexibility towards objects, (2) Flexibility towards the environment, and (3) Flexibility towards persons were .66 (*SD* = 0.07; range: .50-.75), .50 (*SD* = 0.04; range: .37-.61), and .41 (*SD* = 0.00; range: .41-.41), respectively. Intraclass correlation coefficients (calculated on the total sum of scores) for each item ranged from .34 to .79 (*M* = .55; *SD* = .11) and were .87, .65, and .54, for the three subscales, respectively (all correlations were statistically significant with $p < .01$). Intraclass correlation was considered excellent for 1 item, good for 3 items, fair for 11 items and poor for 1 item.

4.3.3 Intra-rater reliability

Intra-rater reliability was calculated for the BFRS-R, the three factors and the items for all respondents as well as for both parents and staff members. For each rater, percentage of exact and adjacent agreement between the first and second assessment was calculated (see Table 2). Mean percentage of exact agreement for the total scale was 74.64 (*SD* = 4.82; range: 68.6-88.6%) and mean percentage of adjacent agreement was 99.55 (*SD* = 0.7; range: 98.6-100%). For the staff members, mean percentage of exact agreement was 75.3 (*SD* = 6.9, range: 64.4-91.1%) and mean percentage of adjacent agreement was 100. Mean exact agreement and adjacent agreement for parents were 73.5 (*SD* = 8.3; range: 60-84%) and 99 (*SD* = 1.8; range: 96-100%), respectively.

Table 2. Mean intra-rater reliability and internal consistency for staff members and parents

Item	Total				Staff members				Parents			
	EA	*AA*	*K*	*SR*	*EA*	*AA*	*K*	*SR*	*EA*	*AA*	*K*	*SR*
1.	75.7	100	.53	.62	71.1	100	.47	.61	84.0	100	.64	.65
2.	88.6	98.6	.80	.84	91.1	100	.84	.91	84.0	96.0	.72	.72
3.	75.5	100	.53	.57	77.8	100	.55	.55	72.0	100	.59	.50
4.	75.7	98.6	.50	.50	75.6	100	.50	.54	76.0	96.0	.50	.43
5.	74.3	98.6	.52	.59	71.1	100	.47	.64	80.0	96.0	.59	.49
6.	78.6	100	.56	.67	75.6	100	.51	.62	84.0	100	.67	.76
7.	75.7	100	.53	.61	80.0	100	.61	.64	68.0	100	.41	.59
8.	68.6	100	.400	.50	64.4	100	.32	.40	76.0	100	.55	.70
9.	71.4	98.6	.47	.51	68.9	100	.39	.47	76.0	96.0	.57	.53
10.	71.4	98.6	.45	.51	77.8	100	.59	.58	60.0	100	.18	.38
11.	75.7	100	.56	.70	84.4	100	.70	.82	60.0	100	.32	.54
12.	77.1	100	.48	.48	80.0	100	.54	.55	72.0	100	.38	.38
13.	71.4	100	.34	.34	75.6	100	.49	.50	64.0	100	.14	.18
14.	70.0	100	.41	.53	73.3	100	.49	.59	64.0	100	.23	.41
15.	68.6	100	.38	.41	64.4	100	.28	.33	76.0	100	.52	.52
16.	75.7	100	.43	.54	73.3	100	.44	.58	80.0	100	.36	.46

NB. EA = exact agreement; AA = adjacent agreement; K = kappa; SR = Spearman Rank.

Mean Cohen's kappa coefficient for exact agreement was .49 (*SD* = .10; range: .34-.80) for all respondents, .51 (*SD* = .14; range: .28-.84) for staff members, and .46 (*SD* = .18; range: .14-.72) for parents. For each item, Spearman rank correlation coefficients were calculated (see Table 2). Mean Spearman correlation was .56 (*SD* = 0.12; range .34-.84) for all respondents, .58 (*SD* = 0.14; range .33-.91) for staff members and .51 (*SD* = 0.15; range .18-.77) for parents. Based on the criteria established by Cicchetti (1994), intra-rater agreement for the total scale was fair and ranged from poor to good for the items. Total severity score was calculated as the sum of the severity ratings for the 16 items for each assessment. Spearman correlation for total severity score was .81 ($p < .01$) for all respondents, .82 ($p < .01$) for staff members and .87 ($p < .01$) for parents. Severity score for each factor was also calculated. Spearman correlations ($p < .01$) for Factors 1, 2 and 3 were .84, .67, and .55 for all respondents, .82, .74 and .49 for staff members and .92, .54, .67 for parents, respectively.

4.3.4 Inter-rater reliability

On each item, percentages of both exact and adjacent agreement were calculated between pairs of staff members and between pairs of a staff member and a parent (see Table 3). Between staff members, mean percentage of exact agreement was 62.5 (*SD* = 10.07; range: 36.4-72.7%) and mean percentage of adjacent agreement was 99.8 (*SD* = 0.76; range: 97-100%).

Table 3. Inter-rater reliability and intraclass correlation for staff members and parents

Item	Staff members					Staff members - parents				
	EA	*AA*	*K*	*SR*	*ICC*	*EA*	*AA*	*K*	*SR*	*ICC*
1.	66.7	97.0	.41	.42	.43	68.2	95.5	.41	.50	.43
2.	72.7	100	.51	.69	.63	63.6	100	.30	.39	.40
3.	63.6	100	.26	.26	.27	77.3	95.5	.58	.53	.40
4.	72.7	100	.42	.49	.55	68.2	95.5	.39	.36	.23
5.	57.6	100	.22	.41	.44	63.6	100	.29	.48	.44
6.	51.5	100	.07	.17	.21	59.7	100	.27	.48	.45
7.	72.7	100	.48	.51	.57	72.7	100	.44	.45	.46
8.	60.6	100	.25	.29	.30	59.1	100	.23	.35	.32
9.	36.4	100	.24	.16	.14	59.1	100	.22	.30	.32
10.	72.7	100	.52	.62	.63	68.2	100	.37	.49	.43
11.	54.6	100	.12	.04	.06	59.1	95.5	.20	.21	.20
12.	66.7	100	.05	.05	.04	72.7	95.5	.27	.27	.15
13.	54.6	100	.06	.06	.08	59.1	100	.18	.20	.16
14.	72.7	100	.49	.55	.58	77.3	95.5	.54	.44	.39
15.	63.6	100	.29	.35	.36	50.0	95.5	.04	.04	.05
16.	60.6	100	.24	.41	.43	77.3	100	.43	.56	.54

NB. EA = exact agreement; AA = adjacent agreement; K = kappa; SR = Spearman Rank; ICC = intra class correlation.

Mean Cohen's kappa coefficient for exact agreement was .29 (*SD* = .17; range: .05-.52). For the staff member and the parent, mean percentage of exact agreement was 65.9 (*SD* = 8.13; range: 50-77.3%), and mean percentage of adjacent agreement was .98 (*SD* = 2.33; range: 95.5-100%). Mean Cohen's kappa coefficient for exact agreement was .32 (*SD* = .14; range: .04-.58). Then, Spearman rank order correlation coefficients were calculated for each item (see Table 3). Mean Spearman correlation was .34 (*SD* = 0.20; range .04-.69) and .38 (*SD* = 0.14; range: .04-.56) between staff members and between a staff member and a parent, respectively.

Finally, inter-rater agreement of item severity score was calculated using the intraclass correlation coefficient. Mean intraclass correlation was .35 (*SD* = 0.21; range: .04-.63) for staff members and .33 (*SD* = .14; range: .05-.54) for parents. For staff members, intraclass correlations were statistically significant at $p < .05$, except for Items 3, 6, 9, and 11-13. For parents, all items were significant at $p < .05$, except for Item 4, 8, 9, 11-13 and 15.

The intraclass correlation for total severity score between pairs of staff members was .58 ($p < .01$) and Spearman correlation for total severity score was .58 ($p < .01$). For the three factors, correlations were .67 ($p < .01$), .45 ($p < .01$), and .13 (ns.), respectively. Spearman correlations were .69 ($p < .01$), .49 ($p < .01$), and .10 (ns.), respectively. Between pairs of a staff member and a parent, intraclass correlation for total severity score was .55 ($p < .01$); Spearman correlation for total severity score was .64 ($p < .01$). Intraclass correlations for the three factors were .62 ($p < .05$), .28 (ns.), and .45 ($p < .01$), respectively, with Spearman correlations of .45 ($p < .05$), .78 ($p < .01$), and .32 (ns.).

4.3.5 Convergent validity

The convergent validity was calculated by correlating total scores on the BFRS-R and the Sameness Questionnaire (SQ). A statistically significant (Pearson) correlation was obtained between the total scores of the BFRS-R and the SQ, $r = .51$; $p < .01$. Also, a significant correlation was found between the SQ-score and Flexibility towards objects (Factor 1), $r = .37$; $p < .05$, and between the SC-score and Flexibility towards the environment (Factor 2), $r = .56$; $p < .01$.

4.4 Discussion

The present study was the first to investigate internal consistency, intra-rater and inter-rater agreement, and convergent validity of a new scale (i.e., the BFRS-R) for the assessment of behavioral flexibility in children with various types of developmental disabilities. The total scale has an excellent internal consistency and consistency for individual items ranged from poor to excellent. Factor analysis revealed three factors (i.e., Flexibility towards objects, Flexibility towards the environment, and Flexibility towards persons), with internal consistency ranging from modest to excellent.

Inter-rater and intra-rater reliability were assessed using exact and adjacent agreement. Adjacent agreement was excellent, whereas exact agreement was modest to good. Inter-rater reliability was good for the total scale. Finally, convergent validity was assessed using the Sameness Questionnaire of Prior and MacMillan (1973). Significant moderate correlations were obtained between the total Sameness Questionnaire and the total BFRS-R and between the SQ and Factors 1 and 2 of the BFRS-R.

The present data suggest that this scale has good potential for assessing situations related to behavioral flexibility in children with developmental disabilities. In clinical practice, the BFRSR might be used to identify the type of situations and extent to which individuals show a resistance to change or an insistence on sameness. The BFRS-R is a reliable tool that may be helpful in enabling clinicians to target interventions for scenarios that are problematic for the child. The BFRS-R can be completed independently by the parent or staff member and is easier to administer than more comprehensive yet elaborate tools measuring related aspects of behavioral flexibility (such as for example, the Autism Diagnostic Interview-Revised; Lord, Rutter, & Le Couteur, 1994). The results should interpreted with caution given the fact that the ratings were provided by parents and direct care staff working in two Dutch day care centers for children with developmental disabilities. It is not clear if the BFRS-R would produce similarly reliable ratings when used by teachers or clinicians in other settings and with other samples of children with developmental disabilities. Clearly, further research is necessary to extend these preliminary findings.

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Appendix A. The Behavioral Flexibility Rating Scale - Revised

Background

The Behavior Flexibility Rating Scale - Revised (BFRS-R) seeks to assess to what extent various SITUATIONS AND CHANGES cause problems for the person. The BFRS-R can be completed individuals who know the individual well enough to provide accurate information about the person's ability to be flexible and cope with change. Generally, anyone who has known and cared for the individual for at least 6 months could complete the BFRS-R.

Directions

Please rate the extent to which each of the following 16 situations is a problem for the individual by marking the best answer.

0 – means that the situation is not at all a problem for the person. He or she copes easily with the situation.

1 – means that the situation causes only mild problems and that these are only short-lived. The person might complain or fuss a little bit and for a short period of time (for 1 to 2 minutes). He/she might even tantrum mildly (e.g., stomp their feet, cry), but eventually the person accepts the situation and calms down and copes with the situation.

2 – means that the situation causes severe problems. The situation may lead to a major tantrum. The tantrum might include aggression, screaming, and/or self-injury. The person never accepts the situation and things have to be returned to how they were before or the person has to be removed from the situation to calm down.

To what extent is each of the following situations a problem for the person?		Severity of the Problem		
		No	*Mild*	*Severe*
1.	A commenly used object is misplaced and cannot be found	0	1	2
2.	A planned event is delayed or cancelled with little warning because of unforseen circumstances	0	1	2
3.	The person is required to move from their current location and go to another location	0	1	2
4.	An object in the environment has been moved or repositioned from its usual location or position	0	1	2
5.	The person wants something what is not available	0	1	2
6.	An object or some materials that the person was using breaks or malfunctions	0	1	2
7.	A usual routine is altered or changed, for example the parent takes a new route home from school	0	1	2
8.	An unexpected interaction occurs with another person, for example a stranger tries to talk to the person	0	1	2
9.	The person becomes momentarily separated from his/her family or group	0	1	2
10.	Materials break, causing a premature end to an activity	0	1	2
11.	Another person is doing something annoying, for example making noise	0	1	2
12.	Objects or materials are not returned to their proper place at the end of an activity	0	1	2
13.	A new object, item, or person has been added to the environment	0	1	2
14.	An activity is interrupted before the person was able to finish the task	0	1	2
15.	A new activity is introduced into the person's routine	0	1	2
16.	Another person tries to use favorite possessions of the person	0	1	2

Chapter 5

Behavioral Flexibility in Children with Autism Spectrum Disorder and Intellectual Disability

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Abstract

Children with autism spectrum disorder (ASD) have particular difficulty with behavioral flexibility, but the knowledge base on behavioral flexibility in children with a diagnosis of ASD plus intellectual disability (ID) compared to children with ID only is still scarce. The aim of the present study was to assess behavioral flexibility in 111 children (84 boys) with ASD (87 autistic disorder; 24 PDD-NOS) plus ID (IQ range: 10.59-72.67) and compare their scores to those of a control group consisting of 65 children with ID only (42 boys). Their age range was between 2:7 and 9:11 years/months. Behavior flexibility was measured using a Dutch version of the Behavioral Flexibility Rating Scale-Revised (Green et al., 2006; Peters-Scheffer et al., 2008). Results showed that behavioral flexibility in children with ASD plus ID was predicted by autism severity, developmental age, and initiating social interaction. A lack of behavioral flexibility seems to influence emotional and behavioral problems and maternal stress, but not adaptive behavior.

5.1 Introduction

Children with autism spectrum disorder (ASD) display impairments in social interaction and communication and show a restricted repertoire of activities and interests (American Psychiatric Association, 2000). Several studies associate these restricted and repetitive behaviors and interests with executive dysfunctioning and most clearly with the domain of cognitive flexibility (e.g., Lopez, Lincoln, Ozonoff, & Lai, 2005; South, Ozonoff, & McMahon, 2007). This is defined as the ability to adapt thoughts or actions in response to situational changes (Geurts, Corbett, & Solomon, 2009). In natural settings, deficits in flexibility are frequently reported in individuals with ASD (e.g., Gioia, Isquith, Kenworthy, & Barton, 2002), but laboratory studies using neuropsychological tests such as Wisconsin Card Sorting Test and the Trail Making Test or other experimental cognitive paradigms have yielded inconsistent findings (e.g., Corbett, Constantine, Hendren, Rocke, & Ozonoff, 2008; Hill & Bird, 2006; Lopez et al., 2005; South et al., 2007). Findings might be confounded by characteristics of the participants (e.g., intellectual functioning, verbal ability, age and co-occurring disorders), and task demands such as the explicitness of the task instruction and the amount of disengagement required to perform the switch (Geurts et al., 2009; Van Eylen et al., 2011). As stated by Geurts et al. (2009), based on face-validity, cognitive flexibility seems related to the insistence of sameness and behavioral rigidity (i.e., lack of behavioral flexibility) observed in individuals with ASD, but connecting results of the cognitive flexibility measures to behavioral flexibility in everyday situations has been complex.

Next to executive dysfunctioning, alternative explanations for the lack of behavioral flexibility in individuals with ASD have been proposed, including (a) a homeostatic mechanism to

reduce over-arousal, (b) an inability to cope with unpredictability, (c) obsessive-compulsive disorder, (d) a desire for self-stimulation, and (e) a lack of central coherence (Green et al., 2006; Turner, 1999). As these 'theories' are not entirely exclusive, it seems plausible that they complement each other in explaining the onset and the maintenance of behavioral flexibility in individuals with ASD (Turner, 1999). The lack of behavioral flexibility is one of the core features of ASD. However, our knowledge base on the nature and extent of behavioral flexibility in individuals with ASD is scarce, especially about situations in which children with ASD show a lack of behavioral flexibility. Ecologically valid measures are required to resolve the paradox between cognitive and behavioral flexibility (Geurts et al., 2009).

One of the few instruments available for assessing behavioral flexibility is the Behavior Flexibility Rating Scale (BFRS) and the Behavioral Flexibility Rating Scale-Revised (BFRS-R), which were developed by Green and her colleagues (Green et al., 2006; Green et al., 2007; Pituch et al., 2007) for the purpose of identifying specific situations in which individuals with developmental disabilities show an insistence on sameness. Green et al. (2006) administered the BFRS to 726 individuals with autistic disorder, Asperger syndrome and Down syndrome, and developed its factor structure (Pituch et al., 2007), while Didden et al. (2008) added a control group of individuals with non-specific Intellectual Disability (ID) and a control group consisting of individuals with Angelman syndrome. Results of these studies showed that individuals with autistic disorder and Asperger syndrome showed significantly more problems in behavioral flexibility than individuals with Down syndrome and Angelman syndrome. When diagnosis was controlled for, no significant relationship between behavioral flexibility and gender or age was found.

However, Green et al. (2006) and Didden et al. (2008) used the same participants with ASD and were not able to ensure the representativeness of the sample due to limitations with the data collection methods. As data in Green et al. (2006) were collected using an internet survey, diagnosis of ASD was established through parental report instead of more reliable standardized measures, such as the Autism Diagnostic Observation Schedule (ADOS; Lord, Rutter, DiLavore, & Risi, 2006) or the Autism Diagnostic Interview (Lord, Rutter, & Le Couteur, 1994). This makes verification of the diagnosis of ASD in the earlier studies impossible. Furthermore, in both studies no data were collected on child variables, such as cognitive functioning, adaptive behavior, and autism severity. Therefore, factors that might predict and/or which might influence behavioral flexibility were not investigated.

In light of these limitations, we aimed to improve the aforementioned studies by confirming the diagnosis of ASD and ID by using reliable and standardized measures and by including several child variables to determine which child factors might predict behavioral flexibility in children with ASD. The aim of the present study was to (a) assess behavioral flexibility in children with ASD (i.e., either autistic disorder or PDD-NOS) plus ID and compare them to children with ID only, (b) explore which factors predict and are influenced by behavioral flexibility in individuals with ASD plus ID, and (c) explore differences in behavioral flexibility scores between individuals with autistic disorder plus ID and those with PDD-NOS plus ID.

Turner (1999) divides repetitive and stereotyped behavior of individuals with ASD into higher level and lower level behavior. She suggests that lower level behavior such as manipulation of objects and stereotyped behavior are more frequent in individuals with lower IQ, while higher level behavior such as repetitive language, circumscribed interests, unusual attachments to objects, and the insistence on sameness are more common in individuals with higher IQ. However, in accordance with some other studies, Gabriels, Cuccaro, Hill, Ivers, and Goldson (2005) found a significantly higher prevalence of sameness behavior in children with ASD with low nonverbal IQ when compared to children with a higher non-verbal IQ. Nevertheless, since typically developing children display an insistence on sameness when they are between two and four years of age (Evans et al., 1997), we assumed that behaviors related to behavioral flexibility such as those measured with the BFRS-R require a certain level of development. Within our sample, which included children with a developmental age between 11 and 41 months (M = 28.70; SD = 7.50), we therefore expected that children with higher developmental age would experience more problems in behavioral flexibility than children with lower developmental age.

Furthermore, it is likely that children with more severe behavioral inflexibility might also experience more difficulties in learning, especially in natural learning environments. Flexibility appears to be a requirement for extracting relevant information from different stimuli (e.g., verbal vs. non-verbal, auditory vs. visual), persons and contexts. For example, Berger, Aerts, van Spaendonck, Cools, and Teunisse (2003) showed that in a group of 30 high functioning adults with ASD improvements in social competence are related to cognitive shifting ability. Consequently, we hypothesized that children with more severe behavioral inflexibility would have lower adaptive behavior scores.

Next, it was hypothesized that as the severity of autism increased, behavioral flexibility would decrease. As the three subtypes of social interaction and communication (i.e., aloof, passive, and active-but-odd; see Wing & Gould, 1979) may refer to distinct subgroups of children with ASD, they were included to further address heterogeneity (Beglinger & Smith, 2001). Children who were classified as active but odd were expected to have few behavioral flexibility issues, while those classified as aloof were expected to have the highest rates of behavioral inflexibility (Castelloe & Dawson, 1993; Wing & Gould, 1979).

A positive relationship between high behavioral flexibility, early social communication skills (i.e., joint attention, behavioral requests, and social interaction) and advanced language ability was expected. In particular, it was hypothesized that children who are responsive, attentive and have high receptive language skills may have a better understanding of their parents' communication and consequently able to anticipate unexpected changes in their environment. Furthermore, children with better expressive skills may express their needs and ask for clarification in ambiguous situations, which may decrease problems in behavioral flexibility.

Finally, we expected that behavioral inflexibility might be burdensome to both the child and the parent. It was hypothesized that children with higher scores on behavioral inflexibility would display more emotional and behavioral problems. Furthermore, mothers of children with problems regarding behavioral flexibility might be experiencing higher levels of maternal stress than mothers of children who are more flexible.

5.2 Method

5.2.1 Participants and Setting

Participants were 176 children. One hundred and eleven of them (84 boys) represented the experimental group and were diagnosed with ASD and ID (17.1% profound ID, 31.5% severe ID, 30.6% moderate ID, 18.9% mild ID; 1.8% borderline ID). Of the 111, 87 had received a diagnosis of autistic disorder and 24 were classified as having PDD-NOS. The remaining 65 children represented the control group.

Prior to their inclusion, all children in the experimental group had received a diagnosis of ASD and ID from a clinician who was independent of the study and in accordance with the DSM-IV (American Psychiatric Association, 2000) or ICD-10 criteria (World Health Organization, 1992). For all children, this diagnosis was confirmed by the ADOS (Lord et al., 2006), the Childhood Autism Rating Scale (CARS; Schopler, Reichler, & Rochen Renner, 2007), Mullen Scales of Early Learning (MSEL; Mullen, 1995) and the Vineland Adaptive Behavior Scales (VABS; Sparrow, Balla, & Cicchetti, 1984) as assessed by the first author. The Wing Subgroups Questionnaire (WSQ; Castelloe & Dawson, 1993) indicated that 79 children had the aloof subtype, while 20 children had the passive subtype and 12 the active-but-odd subtype. Characteristics of the participants of the experimental group are displayed in Table 1.

The remaining children represented the control group, which was comprised of 65 children (42 boys) with non-specific ID (n = 44), physical impairment (n = 12), Angelman syndrome (n = 6), and Down syndrome (n = 3). All were between 3 and 9 years of age (M = 5.02, SD = 1.99). Data on the participants of the control group were collected during a study conducted by Peters-Scheffer et al. (2008), which sought to determine the psychometric properties of the BFRS-R, but only children between 3 and 9 years from the earlier study were included in the current study. Although no formal measures of IQ were administered for the control group, all had ID and attended the same (pre)schools as the children with ASD plus ID in the experimental group. Therefore, the experimental group and the control group were considered to be comparable in terms of their cognitive functioning (i.e., level of ID, IQ, and adaptive functioning).

Table 1. Characteristics of the participants in the experimental group with ASD and ID and Pearson's correlation coefficients for the Behavioral Flexibility Rating Scale - Revised (BFRS-R) total and subscales, and all other measures for children with ASD plus ID (N = 111).

	Autistic disorder plus ID (*n*=87)		PDD-NOS ID (n=24)		Pearson's correlations			
	M (SD)	*Range*	*M (SD)*	*Range*	*T*	*O*	*E*	*p*
Chronological age (years; months)	5;7 (17.19)	3;7-9;0	5;11 (19.88)	2;7 - 9;11	.05	.01	.08	.10
Cognitive functioning								
Developmental age in months	28.70 (7.50)	11.25-40.50	21.53 (7.57)	4.75-43.25	41**	.46**	.26**	.17
IQ	45.77 (15.57)	13.55-71.05	32.85 (14.42)	10.59-72.67	.27**	.32**	.15	.08
Non Verbal IQ	48.71 (15.76)	16.87-76.85	37.92 (15.33)	11.18-75.58	.23*	.28**	.11	.04
Adaptive behavior in months								
Composite	22.96 (6.80)	13-45	18.84 (5.47)	11-43	.34*	.36*	.26**	.13
Communication	30.54 (10.28)	17-53	23.26 (9.28)	12-53	.29**	.31**	.22*	.15
Daily Living Skills	25.21 (8.65)	13-47	21.39 (7.98)	11-45	.39**	.40**	.30**	.16
Socialization	26.96 (8.05)	13-44	20.69 (5.26)	11 - 41	.24*	.26**	.19*	.14
Autism								
ADOS total	9.08 (156)	7-12	16.71 (2.83)	10-24	-.33**	-.32**	-.28**	-.22*
ADOS communication	3.42 (1.35)	2-7	6.37 (1.40)	2-10	-.20*	-.21*	-.13	-.17
ADOS social interaction	6.08 (1.95)	2-11	10.41 (2.33)	6-16	-.42**	-.38**	-.40**	-.28**
CARS	39.17 (6.92)	26-50	41.25 (5.43)	28-53	.30**	.27**	.27**	.19*
Early Social Communication Scales[3]								
Joint attention: initiating	14.06 (9.54)	0.37	5.77 (7.55)	0-35	.19	.19	.14	.16
Joint attention: responding	149.64 (54.20)	41.66-200	80.49 (64.32)	0-200	.41**	.43**	.30**	.27**
Behavioral requests: initiating	26.53 (8.28)	12-43	22.37 (6.60)	2-37	.14	.19	.06	.-08
Behavioral requests: responding	84.02 (23.69)	25-100	62.45 (33.46)	0-100	.37**	.40**	.24*	.15
Social interaction: initiating	3.06 (1.98)	0-7	2.41 (1.75)	0-8	.38**	.42**	.30**	.17
Social interaction: responding	9.12 (3.57)	4-16	6.35 (3.09)	0-18	.37**	.39**	.27**	.23*
Language								
Receptive language (PPVT)	28.17 (5.24)	21-43	24.17 (3.89)	21-39	.29**	.29**	.21**	.16
Receptive language (RDLS)[1]	26.92 (8.40)	14-41	17.65 (6.40)	1-43	.37**	.39**	.27**	.22**
Expressive languauge (WO)[2]	26.83 (9.12)	14-43	18.27 (7.34)	14.43	.31**	.32**	.25**	.20*

Table 1.: Continuation

	Autistic disorder plus ID (*n*=87)		PDD-NOS ID (n=24)		Pearson's correlations			
	M (SD)	*Range*	*M (SD)*	*Range*	*T*	*O*	*E*	*p*
Emotional and behavioral problems[2]								
Total	68.92 (30.96)	19-137	64.15 (23.62)	7-118	.64**	.61**	.57**	.34**
Internalizing	21.46 (11.10)	7-47	20.99 (8.93)	3-40	.64**	.57**	.58**	.40**
Externalizing	24.83 (11.61)	3-45	23.33 (10.61)	1-58	.48**	.51**	.39**	.20*
Maternal stress[2]					.33**	.35**	.24*	.13*

Note. [1] *n* = 110; [2] *n* = 108; [3] *n* = 95. T= Behavioral flexibility, total scale; O = Behavioral flexibility toward objects; E = Behavioral flexibility toward the environment; P = Behavioral flexibility toward persons; ADOS = Autism Diagnostic Observation Schedule; CARS = Childhood Autism Rating Scale; PPVT = Peabody Picture Vocabulary Test; RDLS = Reynell Developmental Language Scales; WO = vocabulary test of the Schlichting Test for Language Production; * $p < .05$. ** $p < 01$.

5.2.2 Instruments

Behavioral Flexibility

The Behavior Flexibility Rating Scale – Revised (BFRS-R; Green et al., 2007) is a scale for assessing behavioral flexibility in individuals with developmental disabilities. The BFRS-R is a revised version of the BFRS. In addition to several wording changes, the revision covered the exclusion of one item and the inclusion of two new items. Using a 3-point Likert-type scale, ranging from zero ('not a problem at all) to two ('the situation causes severe problems'), caregivers rate the severity of challenging behavior that are considered to be triggered by specific and unexpected events and changed routines that could be problematic to the individual. Thus higher scores on the total scale and subscales indicate greater behavioral inflexibility. Factor analysis revealed three factors: (a) flexibility toward objects, (b) flexibility toward the environment, and (c) flexibility toward persons. Internal consistency and intrarater and interrater reliability of the total scale were found to be good to excellent (Peters-Scheffer et al., 2008) and the validity was adequate (Green et al., 2008). For more information regarding the BFRS-R, the reader is referred to Green et al. (2007), Green et al. (2008), Ollington, Green, and Sigafoos (2010), and Peters-Scheffer et al. (2008).

Cognitive functioning

The Mullen Scales of Early Learning (Mullen, 1995) were used to assess the cognitive level of the children with ASD and those with ID. Developmental age was calculated as the average developmental age on the four subscales: fine motor, visual reception, receptive language, and expressive language. Since most children were typically too old and/or too low functioning to determine standardized scores, a ratio IQ was calculated using the following formula: developmental age divided by chronological age and multiplied by 100.

Adaptive behavior

Adaptive behavior levels were assessed using the survey form of the Vineland Adaptive Behavior Scales (Sparrow et al., 1984), a semi-structured interview conducted by a trained interviewer with parents. The VABS consists of a composite score and three subscales: Communication, Daily Living Skills, and Socialization. Age equivalents in months were used in the analyses.

Autism

The Autism Diagnostic Observation Schedule (Lord et al., 2006) is a semi-structured observation of children in a controlled setting, which is used to evaluate social and communicative functioning in individuals suspected of having an ASD. Depending on the language level of the child, one of four developmental modules of the ADOS is administered. A higher score indicates that a child displays more characteristics of autism. Autism severity was measured using the Childhood Autism Rating Scale (Schopler et al., 2007), a 15-item rating scale completed by an observer on a 4-point scale. Scores are summed to obtain a total score with higher scores indicating greater severity of autism. The Wing Subgroup Questionnaire (Castelloe & Dawson, 1993) is a questionnaire with 13 behavioral domains (e.g., communication, social approach, play, imitation, motor behavior, resistance to change) on which parents rate their child's behavior on a scale from 0 (never) to 6 (always) for each domain. A summary score is calculated for each subtype and the highest summary score is considered to indicate the child's subtype.

Early communication and language

The Early Social Communication Scales (ESCS; Mundy et al., 2003) is a videotaped semi-structured observational instrument. The scale measures how the child initiates and responds to tasks involving joint attention, as well how the child responds to behavioral requests and social interaction. Toys and activities are used to elicit social and communicative behavior in an ecologically valid context. Higher scores on the subscales indicate better performance. The first author administered the ESCS, and videotapes were scored by four raters, who were unaware of the exact aim of the study including the other scores of the participants. Interrater reliability was assessed using videotaped data from 28.7% of the children and intraclass correlation coefficients between the paired ratings of the 6 subscales ranged from .66 to .73, suggesting good reliability (Cicchetti, 1994).

Receptive language was measured by the comprehension scales of the Dutch version of the Reynell Developmental Language Scales (RDLS; Van Eldik, Van Der Meulen, Van Der Meulen, Schlichting, & Lutje Spelberg, 1995) and the Peabody Picture Vocabulary Test (PPVT; Dunn & Dunn, 1997). The RDLS consist of 87 items divided into 12 sections in which complexity of tasks increases (e.g., Where is the chair?, Place the doll on the chair). The PPVT measures receptive vocabulary as the child needs to indentify the picture named by the experimenter through pointing.

Expressive language was measured by the vocabulary test of the Schlichting Test for Language Production. This test measures expressive language as the child needs to name objects and pictures (Schlichting, van Eldik, Lutje Spelberg, Van Der Meulen, & Van Der Meulen, 1995).

Emotional and behavioral problems

The Child Behavior Checklist for ages 1.5 - 5 (CBCL; Achenbach & Rescorla, 2000) is a 99-item questionnaire to assess behavioral and emotional problems in children from 1.5 to 5 years. For each item, the respondent indicates whether it is not true (0), somewhat or sometimes true (1) or very true or often true (2) now or in the past two months. Thus, higher scores on the scales represent more emotional and behavioral problems. The CBCL consists of seven small band scales (i.e., aggressive behavior, anxious/depressed, attention problems, emotionally reactive, sleep problems, somatic complaints, and withdrawn), an internalizing, an externalizing, and a total problem scale.

Parental stress

Parental stress was measured by the Dutch version of the Parental Stress Index – short form (the Nijmeegse opvoedingsstress index – verkort, NOSI-K; De Brock et al., 1992). In general, mothers seem to experience more stress than fathers (Dąbrowska & Pisula, 2010) which can complicate between-subject comparison due to the gender effect of the parent. To avoid the confounding effect parent gender, only mothers - as primary caregivers - were asked to complete the NOSI-K. A higher score represents more maternal stress.

5.2.3 Procedure

The children were identified by approaching local (pre)schools for children with ID in The Netherlands. Schools distributed letters to the parents of children who met the following intake criteria: (a) chronological age between 2 and 10 years, (b) a documented diagnosis of ID and/or ASD as assessed by a psychiatrist or psychologist using psychometrically reliable and valid measures, and (c) children lived at home so that parents were able to provide information about their child. All parents gave their written consent and did not receive any honorarium for their participation.

Once participants were selected, the first author scheduled in-home interviews with the parents to administer the VABS and the CARS. A week before the parental interview parents completed the BFRS-R, the CBCL, the NOSI-K and the WSQ. The questionnaires were sent out by mail along with an information letter with contact information and instructions on how to complete the questionnaires. During the interview parents returned the completed questionnaires to the first author. If not returned during the interview the first author sent a reminder within four weeks.

In the same month during which the interview was held four assessments at the preschool or school of the child were scheduled to administer the MSEL, the ADOS, the ESCS, and the language tests. Tests were administered by the first author in a separate room at the (pre) school.

5.3 Results

5.3.1 Between-group analyses

Based on previous findings on the psychometric properties of the BFRS-R (Peters-Scheffer et al., 2008), a total mean and individual means for the three subscales were calculated for children with autistic disorder plus ID, children with PDD-NOS plus ID and children with ID only. Total and subscale means and mean item scores of the three groups are presented in Table 2.

To test for differences between children with autistic disorder plus ID, children with PDD-NOS plus ID and children with ID a univariate analysis of variance (ANOVA) was conducted with the BFRS-R total scale and a multivariate analysis of variance (MANOVA) was performed with scores on the subscales as dependent variables. There was a significant effect of diagnosis on the subscales (Λ = .82, $F(6, 340) = 5.77$; $p < .001$), but not on the BFRS-R total scale ($F(2, 172) = 1.17$; $p = .31$).

Separate univariate ANOVA's on the subscales did not reveal a significant effect of diagnosis on either the object sub-scale ($F(2,172) = 2.54$, $p = .08$) or the environment sub-scale ($F(2, 172) = 1.25$, $p = .29$). However, a significant effect of diagnosis was revealed for behavioral flexibility toward the persons sub-scale ($F(2, 172) = 11.21$; $p < .001$). The Games-Howell post hoc test revealed that behavioral flexibility toward persons was significantly higher in children with autistic disorder plus ID than in children with ID ($p < .001$). No differences in behavioral flexibility toward persons were found between children with PDD-NOS plus ID and children with ID ($p = .79$) and between children with autistic disorder plus ID and PDD-NOS plus ID ($p = .16$).

In sum, multivariate analyses indicated that diagnosis significantly affected behavioral flexibility. However, the precise nature of this relation is yet unclear. Therefore, further analyses were conducted in the group of children with ASD plus ID to explore which variables are associated with behavioral flexibility.

Table 2. Mean scores and standard deviations on the Behavioral Flexibility Rating Scale - Revised (BFRS-R) total, the subscales and the items for the total sample (N = 176).

Item	Children with autistic disorder and ID (*n* = 87)		Children with PDD-NOS and ID (*n* = 24)		Children with ID (*n* = 65)	
	M	*SD*	*M*	*SD*	*M*	*SD*
Chronological age in years	5.33	1.77	4.96	1.71	5.02	1.99
Behavioral flexibility (total)	9.59	6.23	11.65	7.35	9.52	5.46
Behavioral flexibility: objects	6.08	3.95	7.08	3.68	5.20	3.14
Behavioral flexibility: environment	2.00	1.84	2.65	2.52	2.34	1.83
Behavioral flexibility: persons	0.60	0.83	1.13	1.29	1.32	0.99
1. Item misplaced	0.51	0.63	0.74	0.69	0.52	0.59
2. Event postponed	0.69	0.75	0.87	0.82	0.51	0.56
3. Move from current location	0.57	0.68	0.70	0.56	0.48	0.56
4. Item deleted/ moved	0.36	0.57	0.43	0.59	0.38	0.52
5. Item unavailable	0.89	0.69	1.13	0.69	0.94	0.58
6. Item broken	0.85	0.86	0.83	0.78	0.66	0.57
7. Change in routine	0.68	0.67	0.83	0.72	0.45	0.52
8. Unexpected interaction	0.28	0.52	0.65	0.78	0.65	0.59
9. Separated from group or family	0.32	0.54	0.48	0.73	0.68	0.56
10. Activity interrupted due to broken item	0.91	0.79	0.91	0.79	0.66	0.59
11. Annoying behavior	0.75	0.80	0.75	0.80	0.77	0.61
12. Item is put in wrong place	0.28	0.56	0.28	0.56	0.26	0.51
13. New item added to the environment	0.22	0.42	0.22	0.42	0.42	0.53
14 Activity interrupted before finishing	0.96	0.71	0.92	0.77	0.72	0.57
15. New activity introduced	0.65	0.71	0.40	0.56	0.51	0.53
16. Other uses processions	1.04	0.71	0.98	0.79	0.92	0.51

5.3.2 Variables associated with behavioral flexibility

Besides chronological age, diagnosis and behavioral flexibility scores, no additional data were available for the children with ID. Therefore, only children with ASD plus ID were included in the analysis conducted to determine variables associated with behavioral flexibility. First, correlations were calculated between the total score, the subscales of the BFRS-R and variables related to cognitive functioning, adaptive behavior, ASD, early social communication skills, language, emotional and behavioral problems, and maternal stress. Pearson's correlation coefficients revealed significant associations between behavioral flexibility and cognitive functioning (i.e., developmental age, IQ, non-verbal IQ), adaptive behavior, autism severity, emotional and behavioral problems, parental stress and early social communication, and language (see Table 1).

5.3.3 Autism severity and subtype

Based on percentile scores on autism severity measured with the CARS, children were divided into four groups. Means and standard deviations are displayed in Table 3. A multivariate analysis of variance was conducted with autism severity as an independent variable and the subscale scores as dependent variables. There was a significant effect of autism severity on behavioral flexibility (Λ = .85; $F(9, 255.69) = 2.01$; p = .04). Separate univariate analyses of variance indicated significant effects of autism severity on the behavioral flexibility total score ($F(3, 107) = 4.12$; p = .01), the object sub-scale ($F(3, 107) = 3.09$; p = .03), the environment sub-scale ($F(3, 107) = 3.18$; p = .03) and the persons sub-scale ($F(3, 107) = 3.30$; p = .02). Planned contrasts (repeated) revealed no significant differences between severity groups (all ps > .05).

Table 3. Mean scores on the Behavioral Flexibility Rating Scale - Revised (BFRS-R) total and the subscales specified by autism severity for those children diagnosed with ASD plus ID (N = 111).

	Behavioral flexibility: total		Behavioral flexibility: objects		Behavioral flexibility: environment		Behavioral flexibility: persons	
Autism severity	*M*	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*
First quartile (30-35)	7.05	5.62	4.67	3.10	1.57	1.94	0.24	0.62
Second quartile (36-40)	7.92	4.84	5.15	3.03	1.42	1.55	0.58	0.70
Third quartile (41-43)	10.96	7.65	6.59	4.59	2.48	2.17	1.04	1.13
Fourth quartile (44-53)	12.05	6.17	7.35	3.87	2.73	2.02	0.81	1.02

Furthermore, as the social subtypes may refer to distinct subgroups, multivariate analyses were performed with the Wing subtype (i.e., aloof, active-but-odd, and passive) as an independent variable and the sub-scales scores as dependent variables. There were no significant differences in behavioral flexibility between subgroups (Λ = .94; $F(6, 212) = 1.15$; $p = .34$).

At first glance, the significant effect of autism severity on behavioral flexibility seems to contradict the results in which children with autistic disorder plus ID displayed fewer problems in behavioral flexibility than children with PDD-NOS plus ID. However, although groups were similar on most background variables, an independent *t*-test revealed that, on average, children with PDD-NOS plus ID ($M = 28.70$; $SD = 7.50$) had a significantly higher developmental age than children with autistic disorder plus ID ($M = 21.53$; $SD = 7.57$; $t(109) = 4.12$, $p < .001$). Therefore, the influence of developmental age (as assessed using the four subscales of the MSEL) on behavioral flexibility was determined next.

5.3.4 Developmental age

Based on developmental age, children with ASD and ID were divided in six subgroups. Children with a developmental age below 9 months ($n = 1$) and above 44 months ($n = 1$) were excluded from the analysis due to small sample sizes. Descriptives are displayed in Table 4. Results of a multivariate analysis on the sub-scales showed that behavioral flexibility is significantly affected by the developmental age of the child (Λ = .71; $F(15, 281.98) = 2.50$; $p < .01$).

Univariate ANOVA's on the outcome variables revealed a significant effect of developmental age on behavioral flexibility (total; $F(5, 104) = 5.19$; $p < .001$), the object sub-scale ($F(5, 104) = 6.60$, $p < .001$), and the environment sub-scale ($F(5, 104) = 2.45$; $p < .05$). However, the effect of developmental age on the persons sub-scale was not significant ($F(5, 104) = 1.11$; $p = .36$).

Planned contrasts (repeated) revealed significant differences between children with a development age between 15 to 20 months and 21 to 26 months on the total score of behavioral flexibility ($t(5) = 2.78$; $p < .01$) and the object sub-scale ($t(5) = 3.34$; $p < .001$). Other repeated contrasts between developmental age groups were not significant (all ps > .09). However, when applying a Bonferroni correction to correct for family-wise error ($p < .003$), only the contrast between the children with a developmental age between 15 to 20 months and 21 to 26 months on the object scale remained significant.

5.3.5 Language and early social communication

Fewer problems in behavioral flexibility were expected in responsive and attentive children who had good language skills. Therefore, a hierarchical regression analysis was performed with behavioral flexibility as the dependent variable. Receptive and expressive language and the sub-scales of the early social communication scales (i.e., initiating and responding to joint attention, initiating and responding to behavioral request and initiating and responding to social interaction) were the independent variables.

Table 4. Mean scores on the Behavioral Flexibility Rating Scale - Revised (BFRS-R) total and the subscales for all participants with ASD plus ID specified by developmental age (N = 110).

		Behavioral flexibility: total		Behavioral flexibility: objects		Behavioral flexibility: environment		Behavioral flexibility: persons	
Developmental age	*n*	*M*	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*
9 to 14 months	22	6.14	5.39	3.50	2.96	1.55	1.97	0.50	0.67
15 to 20 months	20	7.15	6.52	4.45	3.58	1.45	2.01	0.65	0.99
21 to 26 months	32	11.84	4.42	7.72	3.67	2.38	1.74	0.63	0.91
27 to 32 months	22	10.68	5.91	6.73	3.74	2.27	1.45	0.73	1.03
33 to 38 months	12	13.58	6.44	8.25	2.63	3.08	2.71	1.17	1.27
39 to 44 months	2	18.50	9.19	10.50	3.54	5.00	4.24	1.50	0.71

In the first and second step, the six subscales of the ESCS were entered in a stepwise manner. Only the subscales responding to joint attention and initiating social interaction contributed significantly to the model. In the third step, receptive and expressive language were entered, but these variables did not contribute significantly to the model ($F_{change}(2, 87) = 0.576$; $p = .56$). Table 5 displays the results at each step (i.e., the unstandardized regression coefficient [*B*] and standard error of the unstandardized regression coefficient [*SE B*] and the standardized regression coefficient [*β*]).

R^2 was significantly different from zero at the end of each step. All three models significantly improve the ability to predict behavioral flexibility, with the first model being the best (model 1 $F(1, 90) = 20.92$; $p < .001$; model 2 $F(2, 89) = 17.52$; $p < .001$; model 3 $F(4, 87) = 8.96$; $p < .001$). After step 3, 29.2% of the variance in behavioral flexibility was accounted for.

5.3.6 Overview of variables contributing to behavioral flexibility

To determine the relative contribution of the variables to behavioral flexibility, a hierarchical regression analysis with behavioral flexibility as the dependent variable and developmental age, severity of autism, initiation of social interaction and responding to joint attention as independent variables was performed. Table 6 displays the results at each step.

Table 5. Multiple regression to predict behavioral flexibility from joint attention, social interaction and receptive/expressive language for children with ASD plus ID (n = 104).

	Step 1			Step 2			Step 3		
	B	*SE B*	*B*	*B*	*SE B*	*β*	*B*	*SE B*	*B*
Constant	6.18	1.07		3.93	1.20		1.56	3.74	
Responding to joint attention	0.04	0.01	.44**	0.04	0.01	.37**	0.03	0.01	.28*
Initiating social interaction				1.14	0.34	.31**	1.12	0.34	.31**
Receptive language (PPVT)							0.07	0.10	.08
Expressive language							0.07	0.19	.05

*Note: R^2 = .19 for step 1; ΔR^2 = .09 for step 2; ΔR^2 = .01 for step 3; * p < .05, ** p < .01.*

R^2 was significantly different from zero at the end of each step. All three models significantly improved the ability to predict behavioral flexibility (model 1 $F(1, 93) = 25.00$; $p < .001$; model 2, $F(2, 92) = 23.49$; $p < .001$; model 3, $F(4, 90) = 15.23$; $p < .001$). Although a significant predictor when entered in combination with the other subscales of the ESCS, in combination with developmental age and severity of autism, responding to joint attention no longer contributed significantly to the model. After step 3, in which developmental age, severity of autism, responding to joint attention and initiating social interaction are included, 40.4% of the variance in behavioral flexibility was accounted for.

Table 6. Multiple regression to predict behavioral flexibility from developmental age, severity of ASD, joint attention and initiating social interaction for children with ASD plus ID (n = 104).

	Step 1			Step 2			Step 3		
	B	*SE B*	*B*	*B*	*SE B*	*β*	*B*	*SE B*	*B*
Constant	1.41	1.84		-16.23	4.54		-15.24	4.37	
Developmental age	0.38	0.08	.46**	0.42	0.07	.52**	0.24	0.10	.29*
Severity of ASD				0.40	0.10	.36**	0.39	0.09	.34**
Responding to joint attention							0.02	0.01	.21
Initiating social interaction							0.84	0.32	.23**

*Note: R^2 = .21 for step 1; ΔR^2 = .13 for step 2; ΔR^2 = .07 for step 3; * p < .05, ** p <.01.*

5.3.7 Adaptive behavior

As we expected that a lack of behavioral flexibility would have a negative effect on the development of children with ASD, a regression analysis between total behavioral flexibility score as an independent variable and adaptive behavior as a dependent variable was conducted. As adaptive behavior is associated with developmental age (Schatz & Hamdan-Allen, 1995), the average developmental age on the MSEL was entered in the first step. In the second step, the total score on the BFRS-R and the subscales were entered in a stepwise manner. However, as only developmental age contributed significantly to the model, total behavioral flexibility (t = 0.08; p = .93) and behavioral flexibility toward objects (t = -0.29; p = .77), toward the environment (t = 0.98; p = .33), and toward persons (t = -0.03; p = .97) were excluded from the analysis.

To assess the direct effect of behavioral flexibility on adaptive behavior, a hierarchical regression analysis with adaptive behavior as the independent variable was conducted. In the first step, behavioral flexibility and the subscales were entered stepwise, while developmental age was entered in the second step. Although behavioral flexibility toward objects significantly predicted adaptive behavior in the first step, it no longer contributed significantly to the model when developmental age was entered in the second step. Results of both analyses are displayed in Table 7.

Table 7. Multiple regression to predict adaptive behavior from developmental age and behavioral flexibility for children with ASD plus ID (N = 111).

Model 1				Model 2			
	B	*SE B*	*B*		*B*	*SE B*	*B*
Step 1:				Step 1:			.36*
Constant	5.65	0.99		Constant	16.30	1.01	
Developmental age	0.61	0.04	.82*	Behavioral flexibility: objects	0.56	0.14	
				Step 2:			
				Constant	5.68	1.00	
				Behavioral flexibility: objects	-0.03	0.10	-.02
				Developmental age	0.62	0.05	-.83*

*Note: For model 1: R^2 = .68. For model 2: R^2 = .13 for step 1; ΔR^2 = .55 for step 2 * p <.01.*

R^2 was significantly different from zero at the end of each step. Both models significantly improve the ability to predict adaptive behavior (model 1: $F(1,109) = 226.59$; $p < .001$; model 2: $F(2,108) = 112.39$; $p < .001$). That is, both models explained 68% of the variance. Hence, the associations between behavioral flexibility and adaptive behavior can be explained in terms of developmental age. There is a positive linear relation between developmental age and behavioral inflexibility and between developmental age and adaptive behavior, therefore, when developmental age is not controlled for, significant correlations were found between behavioral flexibility and adaptive behavior (see Table 1). However, when developmental age is controlled for, no significant correlation was found between behavioral flexibility and adaptive behavior ($r = .01$; $p = .93$).

5.3.8 Emotional and behavioral problems

To assess the effect of behavioral flexibility on emotional and behavioral problems, a stepwise regression was conducted with behavioral flexibility and the subscales as independent variables and the total score of the CBCL as a dependent variable. The total behavioral flexibility score contributed significantly to behavioral and emotional problems ($B = 2.50$; $SE\ B = 0.29$; $\beta = .64$; $p < .001$). This model significantly improved the ability to predict behavioral and emotional problems ($F(1,106) = 74.90$; $p < .001$) with 41.4 % of the variance in behavioral and emotional problems accounted for. As the subscales made no contribution to the model, they were excluded from the analysis (behavioral flexibility toward objects: $t = 0.01$; $p = .99$; behavioral flexibility toward environment: $t = 0.40$; $p = .69$; behavioral flexibility toward persons: $t = -0.68$; $p = .50$).

To determine which sub-scales of the CBCL were associated with behavioral flexibility, Pearson's correlations were calculated between the total scale of behavioral flexibility and the subscales of the CBCL. There were significant associations, with small effects between behavioral flexibility and sleep problems ($r = .22$; $p < .05$), attention problems ($r = .27$; $p < .01$) and withdrawn behavior ($r = .19$; $p < .05$), moderate effects between behavioral flexibility and externalizing behavior ($r = .48$; $p < .001$), anxiety ($r = .59$; $p < .001$), somatic complaints ($r = .42$; $p < .001$), and aggression ($r = .58$; $p < .001$). There were large effects between behavioral flexibility and CBCL total ($r = .64$; $p < .001$), internalizing ($r = .64$; $p < .001$) and the emotional reactive subscale ($r = .71$; $p < .001$).

5.3.9 Maternal stress

To assess the effect of behavioral flexibility on maternal stress, a stepwise multiple regression analysis was performed between the three sub-scales of the BFRS-R as independent variables and maternal stress as the dependent variable to determine which sub-scales contributed significantly to maternal stress.

Only behavioral flexibility toward objects contributed significantly to maternal stress, (*B* = 2.47; *SE B* = 0.65*; β* = .35; *p* < .001) with 12.2% of the variance in maternal stress accounted for. This model significantly improved the ability to predict maternal stress, ($F(1,102) = 14.21$; $p < .001$). As the behavioral flexibility toward the environment ($t = 0.11$; $p = .92$), and behavioral flexibility toward persons ($t = -0.36$; $p = .80$) did not contribute significantly to the model they were excluded from the analysis.

5.4 Discussion

This study was the first to explore which variables are associated with behavioral flexibility in children with ASD plus ID. In particular, behavioral flexibility scores were assessed in children with autistic disorder plus ID, PDD-NOS plus ID and ID only, using the Dutch version of the BFRS-R (Green et al., 2006; Green et al., 2007; Peters-Scheffer et al., 2008). Our main finding was that in children with ASD and ID an increase in developmental age and autism severity is associated with more problems in behavioral flexibility. The present study also displays a significant effect of diagnosis on behavioral flexibility. However, the precise nature of this relationship remains unclear as differences between groups might be explained by differences in developmental age between groups. The significant effect of developmental age on behavioral flexibility is in accordance with results found by Bartak and Rutter (1976), in which higher functioning children with ASD had significant more rituals and experienced more problems in adapting to new situations than did lower functioning children. However, the same study reported that lower functioning children displayed more resistance to environmental change than higher functioning children.

The effect of autism severity is in line with studies of Didden et al. (2008) and Green et al. (2006) who found more problems in behavioral flexibility reported in individuals with autistic disorder and Asperger Syndrome than in individuals with Angelman syndrome and Down syndrome. However, Didden et al. (2008) found no significant differences between individuals with non-specific ID and ASD on the total scale of the BFRS-R. The difference between the studies may be attributed to a lack of differentiation in the Didden et al. (2008) study between autistic disorder and PDD-NOS. They also included children, adolescents and adults in their sample. Furthermore, precise data on IQ and level of social functioning in the ID samples are lacking and direct comparison is therefore difficult. However, differences in chronological age and living setting as well as variation in other domains could explain differences in results between both studies.

Although results suggest that behavioral flexibility was predicted by severity of autism, no effect of social subtype (i.e., aloof, passive, and active-but-odd) on behavioral flexibility was found. However, since children with the aloof subtype had a significantly lower developmental age than children with the passive or active-but-odd subtype, the absence of a difference in behavioral flexibility between subtypes might be explained by differences in developmental age.

To assess how (a lack of) behavioral flexibility influences child and family functioning, data were collected on adaptive behavior, emotional and behavioral problems and maternal stress. Although behavioral flexibility did not predict adaptive functioning, results might indicate that a lack of behavioral flexibility predicts emotional and behavioral problems and increased levels of maternal stress. Evidence for an association between variables, however cannot be considered as evidence of causation. Therefore research addressing the question of whether changes in the causal variable (i.e., behavioral flexibility) precedes changes in the outcome (i.e., maternal stress and emotional and behavioral problems) is warranted. Furthermore, as behavioral problems are a strong predictor of maternal stress (see Peters-Scheffer, Didden, & Korzilius, 2012), scores on maternal stress can be affected by the presence of behavioral problems, the association between behavioral flexibility and maternal stress is unclear. Future research needs to address these issues using a longitudinal design.

Behavioral problems and maternal stress seem to affect the effectiveness of early behavioral intervention (Osborne, McHugh, Saunders, & Reed, 2008; Symes, Remington, Brown, & Hastings, 2005), currently considered as the treatment of choice for children with ASD (Eldevik et al., 2009; Peters-Scheffer, Didden, Korzilius, & Sturmey, 2011; Rogers & Vismara, 2008). Therefore, treatment that focuses on enhancing behavioral flexibility seems critical (Green et al. 2007; Ollington et al., 2010) and should commence early. Consequently, studies regarding variables associated with behavioral flexibility are warranted and useful in developing interventions which may enable children with ASD to enhance their capacities to understand and manage unpredictable and changing situations.

Some limitations of the study should be mentioned. A shortcoming is the lack of formal IQ measures in the control group, as is the heterogeneity of the control group which consists of children with various diagnoses. Furthermore, we measured behavioral flexibility using only one data source (i.e., parental report on the BFRS-R). Although subjective, parents can report information about several situations and about behaviors which might not be observed in a short assessment. However, in vivo assessment of behavioral flexibility in naturalistic settings and multiple informants (e.g., teachers) completing the BFRS-R would have strengthened the study (see for example: Green et al., 2008; Ollington et al., 2012). An additional limitation was the uneven sample sizes, including the small sample size for the group of children with PDD-NOS plus ID compared to those with autistic disorder plus ID, which may have influenced the findings.

Since demographic data on the children with ID only, were unavailable, we were unable to match the participants on demographic characteristics such as developmental age. Thus, differences between diagnostic groups may be influenced by differences in demographic characteristics. Furthermore, we were not able to assess the relationship between behavioral flexibility and other variables in children with ID only and, therefore, it is not possible to determine whether the relation between behavioral flexibility and developmental age and initiations of social interactions is specific for children with ASD or also representative for other diagnostic groups.

Clearly, further research is necessary to extend these preliminary findings on behavioral flexibility in individuals with ASD to enhance the comprehension of parents and professionals about the functioning and behavior of individuals with ASD. Moreover, knowledge about behavioral flexibility and associated factors may contribute to improvements in (early) intervention for individuals with ASD. For example, recent work using the BFRS-R as part of a play-based assessment has highlighted the need for developing interventions that involve problem solving and tolerance building as opposed to accommodating the child's lack of flexibility. This is particularly important as during daily routines and interactions, change is unavoidable (Green et al., 2008; Ollington et al., 2010).

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Chapter 6

Maternal Stress predicted by Characteristics of Children with Autism Spectrum Disorder and Intellectual Disability

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Abstract

To determine maternal stress and child variables predicting maternal stress, 104 mothers of children with autism spectrum disorder (ASD) and intellectual disability (ID) completed the Dutch version of the Parental Stress Index (PSI; De Brock, Vermulst, Gerris, & Abidin, 1992) every six months over a period of two years. The level of maternal stress remained stable over time. Child characteristics predicting maternal stress are behavioral inflexibility toward objects and initiating social interactions. However, these factors do not predict maternal stress when analyzed in combination with children's emotional and behavioral problems measured on the Child Behavior Checklist (CBCL; Achenbach & Rescorla, 2000). The subscales emotionally reactive behavior, withdrawn behavior and attention problems explain a third of the variance in maternal stress. This study revealed no relation between maternal stress and children's developmental age and IQ, receptive and expressive language, adaptive behavior, severity and subtype of ASD, behavioral flexibility toward the environment and persons, initiating and responding to joint attention, initiating and responding to behavioral requests, responding to social interactions and the other subscales of the CBCL. Findings are discussed in relation to the clinical and non-clinical norm groups of the PSI, the limitations of the study and clinical practice.

6.1 Introduction

Parenting a child with autism spectrum disorder (ASD) may face parents with unique challenges due to the nature of ASD, as children with ASD are significantly impaired in social interaction and communication and show restricted and stereotyped patterns of behaviors (APA, 2000). Additionally, intellectual disability (ID), anxiety, sleeping and eating disturbances, temper tantrums, self injury and aggressive behavior, social isolation and difficulties in self-care are frequently seen in children with ASD, causing considerable challenges to parents on a daily basis (e.g., Cotton & Richdale, 2010; Matson & Shoemaker, 2009; Rodrigue, Morgan, & Geffken, 1991; Schreck, Williams, & Smith, 2004; White, Oswald, Ollendick, & Scahill, 2009). Many parents experience the period in which their child obtains the diagnosis of ASD as additionally stressful, especially when there is a lack of clarity about the diagnosis, the diagnosis is set comparatively late and when there is a delay between the first concerns and the final diagnosis (Brogan & Knussen, 2003; Howlin & Moore,1997; Moh & Magiati, 2012). Concerns about the future of the child, his or her level of cognitive and communicative impairment, physical health and needs and abilities to get accepted in the community, to function independently and to obtain help may even further elevate stress (Baxter, Cummins, & Yiolitis, 2000; Koegel et al., 1992; Konstantareas & Homatidis, 1989; Ogston, Mckintosh, & Myers, 2011).

Furthermore, early intervention of a young child with ASD may be very intensive, and selecting, coordinating and advocating treatment can be burdensome (Johnson & Hastings, 2002; Peters-Scheffer, Didden, Korzilius, & Sturmey, 2011; Trudgeon & Carr, 2007). Finally, families are faced with financial issues associated with having a child with ASD due to their child's challenging behavior, therapy costs and lifestyle changes that may cause heightened stress levels (Sharpe & Baker, 2007).

However, while it is known that parenting a child with ASD may cause elevated levels of stress, not all parents with a child with ASD report heightened stress levels (Ornstein Davis & Carter, 2008). Stress in parents of children with ASD seems related to parent characteristics as gender, age and coping style (Dabrowska & Pisula, 2010; Dunn, Burbine, Bowers, & Tantleff-Dunn, 2001; Hastings & Johnson, 2001; Herring et al., 2006) and (perceived) levels of social and professional support (Bromley, Hare, Davison, & Emerson, 2004; Dabrowska & Pisula, 2010; Dunn et al., 2001; Hastings & Johnson, 2001). In addition, interrelationships between mothers, fathers and other family members influence parental stress. For example, Hastings (2003) found that child behavioral problems and father's mental health (i.e., anxiety and depression) were associated with mother's stress.

Also, child variables may be linked to increased levels in parental stress. Most studies have focused on the severity of the child's disability and behavioral problems and found that the latter may be a more prominent stress or for parents than the severity of the disability itself (Bromley et al., 2004; Hastings, 2002; Hastings et al., 2005; Herring et al., 2006; Lecavalier, Leone, & Wiltz, 2006). However, studies on the relationship between maternal stress and other variables related to diagnosis are inconclusive. For example, Mori, Ujiie, Smith, and Howlin (2009) reported no relation between parental stress and IQ and results about the relation between maternal stress and impaired adaptive behavior are mixed (Beck, Hastings, Daley, & Stevenson, 2004; Lecavalier et al., 2006; Tomanik, Harris, & Hawkins, 2004). On the other hand, several studies report that higher parental stress is associated with higher autism symptom scores (Hastings & Johnson, 2001; Konstantareas & Homatidis, 1989; Konstantareas & Papageorgiou, 2006). Other child factors associated with higher parental stress are amongst others lower social skills (Baker-Ericzen, Brookman-Frazee, & Stahmer,2005), less responsiveness to interactions and social relatedness (Ornstein Davis & Carter, 2008; Kasari & Sigman, 1997), temperament (Konstantareas & Papageorgiou, 2006) and repetitive and self-injurious behavior (Konstantareas & Homatidis,1989).

As stated in Lecavalier et al. (2006) some of the outcomes of instruments used in above studies may be influenced by the child's level of functioning or the presence of behavioral problems and as a result obscure the associations between particular child characteristics and parental stress (e.g., the CARS, which is used to measure autism severity contains items regarding hyperactivity and anxiety). Furthermore, not all studies have used reliable measures for child characteristics of ASD, behavior and developmental age, but relied on reports of the mothers. As longitudinal studies addressing maternal stress are scarce, this study investigates if and how child characteristics influence maternal stress using a longitudinal design.

As data was collected on several variables (i.e., cognitive functioning, adaptive behavior, autism subtype and severity, behavioral flexibility, communication and behavioral problems), an attempt was made to provide a comprehensive view of child characteristics and their relative contribution to maternal stress.

We aimed at improving above studies in several ways. To confirm the diagnosis of ID and assess developmental age and IQ the Mullen Scales of Early Learning (MSEL; Mullen, 1995) and the Vineland Adaptive Behavior Scales (VABS; Sparrow, Balla, & Cicchetti, 1984) were administered in a relatively large sample of children (n = 104). As children with lower developmental age and adaptive behavior are more dependent upon their parents to meet their needs, we expect higher maternal stress in mothers of children with lower developmental age and lower levels of adaptive behavior.

Subsequently, the diagnosis of ASD was confirmed by administering the Autism Diagnostic Observation Schedule (ADOS; Lord, Rutter, DiLavore, & Risi, 2006) and the Childhood Autism Rating Scale (CARS; Schopler, Reichler, & Rochen Renner, 2007). To further address heterogeneity of ASD, subtypes of social interaction and communication were included in the analyses as these three subtypes (i.e., aloof, passive, and active-but-odd; see, Wing & Gould,1979) may correspond to distinct subgroups of children with ASD (Beglinger & Smith, 2001). Castelloe and Dawson(1993) suggested that the aloof and active-but-odd subtypes fall at two ends of the continuum with the most autistic children in the aloof group and the least autistic in the active-but-odd group. In concurrence with the literature, we hypothesize that as the severity of autism increases, maternal stress increases and that higher maternal stress is reported by mothers of children with the aloof subtype than in the active-but-odd group with the passive subtype in the middle.

As poor communication skills are related to increased levels of stress (Baxter et al., 2000), data were collected on early social communication skills (i.e., joint attention, behavioral requests and social initiations) and receptive and expressive language. We expect that mothers of children with less communication skills experience more maternal stress than mothers of children who have better communication skills.

Next to impairments in communication and social interaction, the insistence on sameness is one of the core features of ASD and there is accumulating evidence of an increased risk of problems with behavioral flexibility in individuals with ASD (Green et al., 2006; Didden et al., 2008; Peters-Scheffer, Didden, Sigafoos, Green, & Korzilius 2013). We hypothesize that behavioral inflexibility may be burdensome to the mothers and that mothers of children with more problems regarding behavioral flexibility experience more maternal stress than mothers of children who are more flexible.

Considering the interplay between maternal stress and behavioral problems, data were collected on emotional and behavioral problems as well. We hypothesize that mothers of children with more behavioral problems experience more stress than mothers of children with fewer behavioral problems. Finally, we expect that behavioral problems predict parental stress over time (Lecavalier et al., 2006). A longitudinal design was used and all variables were measured over a period of two years.

One hundred and four mothers completed measures on stress, and their child's behavioral problems, behavioral flexibility, severity of autism and adaptive behavior. In addition, data on cognitive functioning, (non-verbal) communication and language were collected in the children. Due to the absence of a control group, outcomes of the children with ASD and ID were compared to those of the clinical and non-clinical norm groups of the Parental Stress Index (PSI; De Brock, Vermulst, Gerris, & Abidin, 1992). Next, predictors of maternal stress were explored.

6.2 Method

6.2.1 Participants and setting

Participants were 104 mothers of children with ASD and ID enrolled in longitudinal study on the development of children with ASD and ID in the Netherlands. Over a period of two years, data were collected on maternal stress, and a range of child variables related to cognitive functioning, adaptive behavior, autism severity and subtype, early social communication, language, behavioral flexibility and emotional and behavioral problems.

All children (78 males) had received a clinical diagnosis of ASD (82 autism; 22 Pervasive Developmental Disorders-Not Otherwise Specified [PDD-NOS]) and ID (17 profound ID; 33 severe ID; 33 moderate ID; 19 mild ID; 2 borderline ID) from a child psychiatrist or a multidisciplinary diagnostic clinic independent of the study. In all children the diagnosis of ASD was confirmed by the Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2006) and the Childhood Autism Rating Scale (CARS; Schopler et al., 2007) and the diagnoses of ID was confirmed by the Mullen Scales of Early Learning (MSEL; Mullen, 1995) and the Vineland Adaptive Behavior Scales (VABS; Sparrow et al., 1984) administered by the first author at baseline. All children attended a preschool or school for children with ID. Information about the demographic characteristics is presented in Table 1.

6.2.2 Variables and measures

All measures were selected for their good psychometric properties and their applicability to children with ASD and/ or ID.

Parental stress

Parental stress was measured by the Dutch version of the Parental Stress Index, short form: the Nijmeegse Opvoedingsstress Index (PSI; De Brock et al., 1992). Using a six-point Likert-type scale, caregivers rated 25 items regarding the upbringing of their child. In the manual of the PSI a non-clinical and a clinical norm group are described and due to the absence of a control group outcomes of the present study are compared to these norm groups.

In general, mothers seem to experience more stress than fathers (Dabrowska & Pisula, 2010), complicating between-subject comparison due to the gender effect of the parent. To avoid the confounding effect of the gender of the parent, only mothers - as primary caregivers - were asked to complete the PSI.

Table 1. Characteristics of the participants and Pearson's correlations with maternal stress at baseline.

Child variables	Descriptives			Maternal stress	
	n	*M*	*SD*	*r*	*p*
Cognitive functioning					
Developmental age in months	104	23.13	7.85	.09	.39
IQ	104	35.71	15.20	-.02	.87
Non-verbal IQ	104	40.41	15.76	-.03	.77
Adaptive behavior in months					
Composite	104	19.70	5.50	.12	.22
Communication	104	24.85	9.64	.02	.85
Daily living skills	104	22.24	7.92	.16	.11
Socialization	104	22.15	6.18	.05	.65
Behavioral flexibility					
Behavioral flexibility: total	102	10.18	6.48	.34	<.001
Behavioral flexibility: objects	102	6.37	3.81	.37	<.001
Behavioral flexibility: environment	102	2.19	2.02	.23	.02
Behavioral flexibility: persons	102	0.70	0.96	.15	.15
Emotional and behavioral problems					
CBCL Total	102	66.22	25.47	.55	<.001
Internalizing	102	21.46	9.46	.51	<.001
Externalizing	102	24.05	10.84	.49	<.001
Autism					
ADOS total	104	15.13	4.09	.01	.93
ADOS communication	104	5.79	1.85	-.04	.70
ADOS social interaction	104	9.50	2.88	.02	.84
CARS	104	40.71	5.88	.18	.06
Early Social Communication Scales					
Initiating joint attention	88	7.16	8.52	-.07	.52
Responding to joint attention	88	94.15	66.49	-.04	.69
Initiating behavioral requests	88	23.30	6.83	-.02	.47
Responding behavioral requests	88	67.69	31.41	.07	.87
Initiating social initiations	88	2.52	1.79	.24	.03
Responding to social initiations	88	6.80	3.28	-.00	.97
Language					
Receptive language	104	25.11	4.53	.07	.46
Expressive language	101	20.13	8.39	-.07	.50

Developmental level

The Mullen Scales of Early Learning (MSEL; Mullen, 1995) is a standardized measure to assess the developmental level of children from birth to 68 months of age. A developmental age was calculated averaging age equivalent scores on the visual reception, fine motor, receptive language and expressive language scales. Since most children were typically too old and/or too low functioning to determine standardized scores, a ratio IQ was calculated using the following formula: developmental age divided by chronological age and multiplied by 100.

Autism

The Autism Diagnostic Observation Schedule (ADOS; Lord et al., 2006) is a semi-structured observation to assess social and communicative functioning in individuals suspected of having an ASD. The Childhood Autism Rating Scale (CARS; Schopler et al., 2007) was administered as a measure of symptom severity. The Wing Subgroups Questionnaire (WSQ; Castelloe & Dawson, 1993) is a questionnaire with 13 behavioral domains (e.g., communication, social approach, play, imitation, motor behavior, resistance to change) on which parents rate their child's behavior. A summary score is calculated for each subtype (i.e., aloof, passive and active-but-odd) and the highest summary score is considered to indicate the subtype.

Adaptive behavior

The Dutch version of the Vineland Adaptive Behavior Scale-survey form (VABS; Sparrow et al., 1984) was used to measure adaptive behavior across three domains: Socialization, Communication and Daily Living Skills. Based on the subscales a composite score was derived.

Emotional and behavioral problems

The Child Behavior Checklist (CBCL; Achenbach & Rescorla, 2000) is a well-known and widely used questionnaire with 100 items on various problem behaviors grouped into seven syndrome scales: emotional reactive, anxiety, somatic complaints, withdrawn, sleep problems, aggressive behavior and attention deficits. In addition, scores on internalizing, externalizing and total scales were calculated. Based on the developmental and chronological age of the participants the CBCL 1.5-5 years was used.

Behavioral flexibility

The Behavioral Flexibility Rating Scale-revised (BFRS-R; Green et al., 2006; 2007) is a scale for assessing behavioral flexibility in individuals with developmental disabilities. Using a three-point Likert-type scale, ranging from 0 ('not a problem at all') to 2 ('the situation causes severe problems'), caregivers rated the severity of challenging behavior as a result to specific and unexpected events and changed routines that could be problematic to the individual. The BFRS-R comprises three factors: (1) flexibility toward objects, (2) flexibility toward the environment, and (3) flexibility toward persons (Peters- Scheffer et al., 2008).

Early communication

The Early Social Communication Scales (ESCS; Mundy et al., 2003) are designed to assess nonverbal communication behavior (e.g., joint attention, behavioral requests and social interactions) in young children during a videotaped semi-structured observation. The first author administered the ESCS and videotapes were scored by four raters, unaware of the exact aim of the study and other scores of the participants. Inter rater reliability was assessed using videotaped data from 29% of the children and intra class correlation coefficients between the paired ratings of the six subscales ranged from .66 to.73 suggesting good reliability (Cicchetti, 1994).

Language

Receptive language was measured by the Peabody Picture Vocabulary Test (PPVT; Dunn & Dunn, 1997), while expressive language was measured by the vocabulary test of the Schlichting Test for language production (Schlichting, van Eldik, Lutje Spelberg, van der Meulen, & van der Meulen, 1995).

6.2.3 *Procedure*

The children were identified by local preschools and schools for children with ID in The Netherlands. The (pre)schools distributed letters to the parents of children who met the following inclusion criteria: (1) chronological age from 2 to 9 years,(2) a documented diagnosis of ID and ASD (i.e., autism or PDD-NOS) set by a psychiatrist or a psychologist supported by psychometrically reliable and valid measures, (3) children lived at home at the start of study, (4) absence of medical conditions which could interfere with data collection (e.g., severe epilepsy, visual impairment) and (5) written consent of the parents.

Once participants were selected, the first author scheduled an in-home interview with the parents to administer the VABS and the CARS. Six, 12, 18 and 24 months later the questionnaires were administered to the parents during an interview by phone. A week before the interview, the CBCL, BFRS-r, WSQ and the PSI were sent out by mail to the parents along with an information letter and a prepaid and addressed return envelope. The information letter provided contact details and the first time instructions about completing the questionnaires. If the parents had not returned the questionnaires within 4-6 weeks, the researcher sent a reminder. Families did not receive any honoraria for their participation.

In the same month as the first interview was held the first author assessed the MSEL, ADOS, ESCS and the language tests to the child at the preschool or school and the same tests, except for the ADOS, were administered after 12 and 24 months. The ADOS was only administered after 24 months.

6.3 Results

6.3.1 Maternal stress

Scores of maternal stress over two years are displayed in Table 2. Scores remained relatively stable over time and ANOVA repeated measures indicated no significant differences between the five assessments over time (F(3.15, 327.78) = 1.37; p = .25). One sample t-tests were used to compare the means to the norms of the clinical group of the PSI and no significant differences were found between the mean of the clinical norm group (M = 85.9; SD = 24.3) and the five assessments in the ASD and ID group (M = 80.65-85.00; SD = 25.97-29.16), all ps > 05. However, means of the five assessments of the ASD and ID group were considerably higher than the mean reported of the mothers of the non-clinical norm group of the PSI (M = 54.4; SD = 19.3), all ps < .001. Between 36 and 45% of the mothers in the ASD and ID group scored at or above the 95th percentile of the stress scores reported by the non-clinical norm group. Hence, mothers of children with ASD and ID experience more stress than mothers of the PSI norm group consisting of typically developing children, but no differences were found between the maternal stress experienced by mothers of the ASD and ID group and the clinical norm group of the PSI.

6.3.2 Child variables associated with maternal stress

Pearson correlations were calculated to explore which variables are associated with maternal stress. These analyses showed that there were associations with small to moderate effects between maternal stress and the initiation of social initiations, a moderate effect on behavioral flexibility and large effects on emotional and behavioral problems. Further, a trend was found between maternal stress and autism severity as measured by the CARS. No associations were found between maternal stress and children's level of cognitive functioning, adaptive behavior, language, responding to and initiating behavioral requests and joint attention. The results of these analyses are shown in Table 1. As associations between maternal stress and the other variables after 6, 12, 18 and 24 months were not substantial different from the associations of the baseline, only the baseline results are reported in Table 1.

Developmental age and adaptive behavior

Although correlations revealed no significant associations between maternal stress and developmental age or IQ, a one- way ANOVA was performed with maternal stress as dependent variable and six categories of developmental age as independent variable (i.e., 9-14, 15-20, 21-26, 27-32, 33-38 and 39-44 months). There was no significant difference on maternal stress between the categories of developmental age (F(5,97) =1.37; p = .24), nor on the categories of severity of ID (i.e., borderline, mild, moderate, severe, profound; F(4,99) = 0.73; p = .57). Furthermore, there was no significant difference in maternal stress between the categories of adaptive behavior (i.e., 9-14, 15-20, 21-26, 27-32, 33-38 and 39-44 months; F(5,98) = 0.56, p = .73).

Table 2. Maternal stress during baseline and after 6, 12, 18 and 24 months. Percentages of scores representing respectively very low (VL), low (L), below average (BL), average (A), above average (AA), high (H) and very high (VH) compared to the norms of the clinical group of the PSI are displayed.

	n	*M*	*SD*	*Range*	*VL*	*L*	*BA*	*A*	*AA*	*H*	*VH*
Baseline	104	81.64	26.73	26-144	16.3	5.8	9.6	40.4	15.4	6.7	5.8
After 6 months	96	85.00	29.16	25-150	11.5	9.4	14.6	27.1	16.7	11.5	9.4
After 12 months	95	80.65	25.97	25-142	12.8	9.6	12.8	38.3	14.9	7.4	4.3
After 18 months	75	83.09	27.75	28-142	10.7	6.7	20.0	32.0	14.7	5.3	10.7
After 24 months	79	84.99	28.09	27-143	10.3	6.4	14.1	38.5	10.3	7.7	12.8

Autism severity and subtype

As children with ASD are a heterogeneous group, several analyses were conducted to explore within-group differences related to ASD. First, an independent *t*-test was conducted to explore differences in maternal stress between mothers of children with PDD-NOS (n = 22; M = 81.36; SD = 24.06) and those of children with autism (n = 82; M = 81.72; SD = 27.54). On average, mothers of children with PDD-NOS experienced comparable levels of stress to mothers of children with autism, $t(102) = -.06$; $p = .96$. Based on percentile scores on the CARS, participants were divided into four sub groups and a one-way ANOVA was conducted with autism severity as independent variable and maternal stress as dependent variable. Results did not reveal a significant effect of autism severity on maternal stress ($F(3,100) = 1.56$; $p = .20$).

As the subtypes may refer to distinct subgroups, a one-way ANOVA was performed with Wing's subtypes (i.e., aloof, active-but-odd and passive) as independent and maternal stress as dependent variable. There were no significant differences in maternal stress between subgroups, ($F(2,101) = 0.61$; $p = .55$).

Emotional and behavioral problems

To assess whether emotional and behavioral problems predict maternal stress a simple regression analysis between emotional and behavioral problems as independent variable and maternal stress as dependent variable was performed. Emotional and behavioral problems accounted for 30.1% of the variance in maternal stress ($B = .57$; $SE\ B = .09$; $\beta = .55$; $p < .001$, $F(1,100) = 43.06$, $p < .001$).

Subsequently, a regression analysis was conducted to determine which subscales contributed significantly to maternal stress. A stepwise method was used whereby maternal stress was entered as the dependent variable and the raw scores of the subscales emotionally reactive, anxiety/depressed, somatic complaints, withdrawn, sleep problems, aggressive behavior and attention problems as independent variables. Table 3 displays the unstandardized regression coefficient *(B)* the standard error of the unstandardized regression coefficient *(SE B)* and the standardized regression coefficient (β) at each step.

R^2 was significantly different from zero at the end of each step. All three models significantly improved the ability to predict maternal stress, with the first model the best, (model 1: $F(1,100) = 32.65$; $p < .001$; model 2: $F(2,99) = 21.81$; $p < .001$; model 3: $F(3,98) = 16.95$; $p < .001$). As the predictors sleep problems ($t = -0.38$; $p = .71$), anxiety/ depressed ($t = -0.61$; $p = .54$), somatic complaints ($t = -0.12$; $p = .90$) and aggressive behavior ($t = 1.27$; $p = .21$) did not contribute significantly to the models (results reported for model 3), they were excluded from the analyses. After step 3, in which emotionally reactive, withdrawn and attention problems were included, 34.2% of the variance in maternal stress was accounted for.

Table 3. Multiple regression to predict maternal stress from emotional and behavioral problems for children with ASD and ID (n = 102).

	Step 1			Step 2			Step 3		
	B	*SE B*	*β*	*B*	*SE B*	*β*	*B*	*SE B*	*β*
Constant	58.76	4.54		46.11	6.16		39.50	6.67	
Emotionally reactive	3.40	0.59	.50**	2.66	0.63	.39**	2.18	0.65	.32**
Withdrawn				2.25	0.77	.27**	2.09	0.76	.25**
Attention problems							1.89	0.82	.21*

*Note: R2 = .25 for step 1; ΔR2 = .06 for step 2; ΔR2 = .04 for step 3; * p < 05. ***

Behavioral flexibility

As behavioral flexibility correlated significantly with maternal stress, a stepwise multiple regression analysis with the subscales of behavioral flexibility as independent variables and maternal stress as dependent variable was conducted to determine which subscales contributed significantly to maternal stress. Only behavioral flexibility toward objects contributed significantly to maternal stress ($B = 2.56$; SE B = 0.65; $\beta = .37$; $p < .001$) with 13.6% of the variance in maternal stress accounted for. This model significantly predicted maternal stress ($F(1,100) = 15.75$; $p < .001$). As behavioral flexibility toward the environment ($t = -0.31$; $p = .76$), and behavioral flexibility toward persons ($t = -0.24$; $p = .81$) did not contributed significantly to the model, these were excluded from the analysis.

Early social communication and language

As children's poor communication skills may increase maternal stress, a regression analysis was conducted including the domains of early social communication (i.e., initiating and responding to joint attention, behavioral requests and social interactions), receptive and expressive language and maternal stress.

Only initiating social interaction significantly predicted maternal stress *(B* = 3.55; *SE B* = 1.62; *β* = .24; *p* = .03) with 5.5% of the variance accounted for (*F*(1,83) = 4.84; *p* = .03). As initiating (*t* = 1.03, *p* = .31) and responding to joint attention (*t* = 0.62; *p* = .54), initiating (*t* = 0.15; *p* = .88) and responding to behavioral requests (*t* = 0.34; *p* = 74), responding to social interaction (*t* = 0.82; *p* = .42), receptive (*t* = 0.25; *p* = .81) and expressive language (*t* = 1.22; *p* = .23) did not contributed significantly to the model they were excluded from the analysis.

Table 4. Multiple regression to predict maternal stress from emotional and behavioral problems (i.e., emotionally reactive behavior, withdrawn behavior and attention problems, behavioral flexibility toward objects and initiating social interaction for children with ASD and ID (n = 88).

	Step 1			Step 2					Step 3
	B	*SE B*	*β*	*B*	*SE B*	*β*	*B*	*SE B*	*β*
Constant	38.16	7.31	.32**	36.99	7.63	.27*	35.75	7.77	.30*
Emotionally reactive	2.19	0.73	.29**	1.89	0.91	.30**	2.05	0.92	.30**
Withdrawn	2.52	0.86	.15	2.58	0.87	.14	2.57	0.87	.12
Attention problems	1.39	0.97		1.33	0.97	.07	1.12	1.00	.02
Behavioral flexibility toward objects				0.47	0.84		0.15	0.91	.09
Initiating social interaction							1.40	1.55	

*Note: R^2 = .35 for step 1; ΔR^2 = .00 for step 2; ΔR^2 = .01 for step 3; *p < 05, **p < 01.*

Relative contribution of child characteristics to maternal stress

To assess for relative contributions of the child's characteristics to maternal stress a hierarchical regression was employed. As emotional and behavioral problems see ma consistent predictor of maternal stress, the sub scales emotionally reactive, with drawn and attention problems were entered in the first step and behavioral flexibility toward objects in the second step. In the third step, initiating social interaction was entered. Table 4 displays the relevant statistics. R^2 was significantly different from zero at the end of each step, with the first model the best in predicting maternal stress (model 1: *F*(3,82) = 14.70; *p* < .001; model 2: *F*(4,81) = 11.01; *p* < .001; model 3: *F*(5,80) = 8.95; *p* < .001). *F*change was not significant between model 1 and 2 (*F*(1,81) = 0.31; *p* = .58) and between model 2 and 3 (*F*(1,80) = 0.81; *p* = .37). The first model, which included emotionally reactive behavior, withdrawn behavior and attention problems, accounted for 35% of the variance in maternal stress.

6.3.3 Maternal stress and child's emotional and behavioral problems over time

As the child's emotional and behavioral problems seem to be the most important predictor for maternal stress, the stability of emotional and behavioral problems and maternal stress over time was examined using Mplus. Across-lagged model that specified within-time correlations between maternal stress and behavioral problems (e.g., maternal stress at baseline with behavioral problems at baseline), stability effects for each variable (e.g., maternal stress at 12 months predicted by maternal stress at baseline) and cross-legged effects (e.g., behavioral problems at baseline predict stress at 12 months). The overall fit of the model was evaluated by the CFI (Bentler, 1990), TLI (Tucker & Lewis, 1973), SRMR (Hu & Bentler, 1999) and RMSEA (Steiger & Lind, 1980) indexes. Values of the TLI and CFI > 0.95 and the RMSEA < 0.05 are considered excellent fit, while a SRMR < .08 is generally considered a good fit (Hu & Bentler,1999). The proposed model is displayed in Fig. 1 and consistent with the data, $\chi^2(4) = 7.19$; relative $\chi^2 = 1.80$; *TLI* = .95;CFI = 99; *SRMR* = 03. Yet, the RMSEA of 0.11 indicates poor fit. However, the RMSEA can be misleading when degrees of freedom are small and the sample size is not large. Significant within-time correlations and standardized parameter estimates were found between maternal stress and emotional and behavioral problems and stability effects for maternal stress and emotional and behavioral problems were also significant. However, no significant relations were found for the cross-legged effects. Table 5 displays the estimates, standard errors and the standardized coefficients for the model.

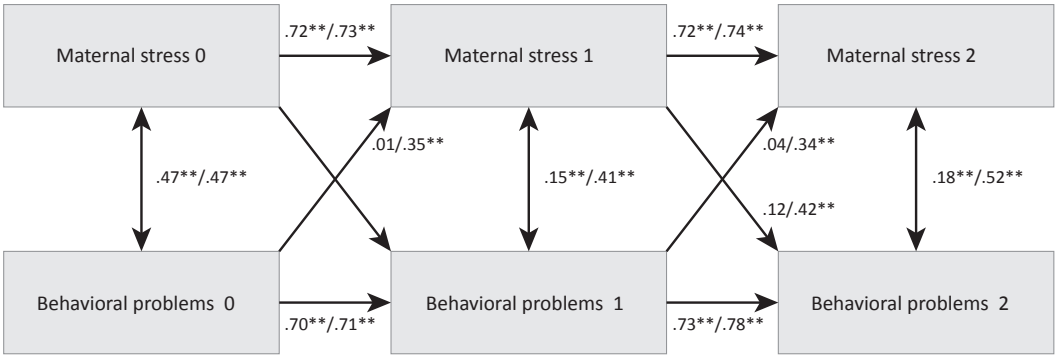

*Figure 1. Model on maternal stress and behavioral problems with standardized parameter estimates (StdXY) and correlations (italicized) at baseline (0), after 12 months (1) and after 24 months (2); * p <.05; ** p <.01.*

Table 5. Estimates, standard errors (SE) and standardized coefficients (StdXY) of the model on maternal stress and behavioral problems at baseline (0), after 12 months (1), and after 24 months (2).

	Estimates	SE	Est./SE	StdXY
Maternal stress 1 on maternal stress 0	0.72	0.09	8.05	0.72
Maternal stress 1 on behavioral problems 0	0.01	0.11	0.06	0.01
Maternal stress 2 on maternal stress 1	0.82	0.10	7.95	0.72
Maternal stress 2 on behavioral problems 1	0.06	0.12	0.49	0.04
Behavioral problems 1 on behavioral problems 0	0.59	0.08	7.59	0.70
Behavioral problems 1 on maternal stress 0	0.03	0.08	0.32	0.03
Behavioral problems 2 on behavioral problems 1	0.90	0.09	10.15	0.73
Behavioral problems 2 on maternal stress 1	0.13	0.09	1.40	0.12
Maternal stress 0 with behavioral problems 0	270.15	85.01	3.18	0.47
Maternal stress 1 with behavioral problems 1	71.43	30.03	2.38	0.15
Maternal stress 2 with behavioral problems 2	125.56	34.55	3.63	0.18

6.4 Discussion

This study investigates maternal stress in 104 mothers of children with ASD and ID who were between 2 and 9 years old. Data were collected over a period of two years. Level of maternal stress was compared to that of two norm groups. Besides, child characteristics predicting maternal stress were explored. When compared to the norms of the Dutch version of the PSI, mothers of children with ASD and ID experience more stress than mothers of typically developing children, but no differences in stress were found between mothers of children with ASD and ID and the clinical norm group of the PSI. The latter finding seems deviant from findings of other studies in which parents of children with ASD present greater levels of stress than those of children with other disabilities (e.g., Dabrowska & Pisula, 2010; Eisenhower, Baker, & Blacher, 2005; Estes et al., 2009; Griffith, Hasting, Nash, & Hill, 2010).

While most studies used control groups of children with mixed etiology ID/ developmental disabilities (e.g., Estes et al., 2009; Griffith et al., 2010), Down syndrome (e.g., Dabrowska & Pisula, 2010; Eisenhower et al., 2005; Griffith et al., 2010), or typically developing children (Eisenhower et al., 2005), this study compared the scores of the children with ASD and ID to the non-clinical and clinical norm group of the PSI. According to the PSI manual, the clinical group of the PSI is comprised of families recruited through professionals of mental health care institutions in two regions of the Netherlands, but additional background information on child or family characteristics (e.g., SES, income, IQ, diagnosis) is lacking (De Brock et al., 1992). As a substantial subset of clients of the mental health care institutions has a diagnosis of ASD, this may cloud comparisons and explain why no significant differences were found between the experimental group and the clinical norm group of the PSI.

Most families in the clinical norm group were waiting for treatment. In accordance with a study of Schieve, Blumberg, Rice, Visser, and Boyle (2007) in which parents with special services needs indicated enhanced stress levels compared to parents without special care needs, parents of the clinical group may have experienced increased stress levels in this period before treatment onset. This may also explain why no significant differences were found between the clinical norm group and the children with ASD and ID in our study.

Although, stability effects for maternal stress and emotional and behavioral problems overtime and within-time associations between maternal stress and behavioral problems were found, maternal stress did not predict emotional and behavioral problems one year later, nor did emotional and behavioral problems predict maternal stress one year later. Either this suggest the absence of a longitudinal relation between both variables, or problems with the power. As the sample size of the present study was relatively small and the interval between measures relatively long, further research should investigate the longitudinal relation between maternal stress and emotional and behavioral problems using a larger sample size and smaller intervals between assessments.

No significant effect for developmental age, severity of ID or adaptive behavior on maternal stress was found. Furthermore, diagnosis (i.e., PDD-NOS vs. autism), subtype (i.e., aloof, passive, active-but-odd) or severity of ASD did not influence maternal stress. However, emotional and behavioral problems accounted for a third of the maternal stress, mostly caused by emotionally reactive behavior, with drawn behavior and attention problems. This study clearly indicates that behavioral problems are more associated with maternal stress than any other child characteristic and that both variables remain relatively stable over a period of two years. The latter results are in accordance with other studies that consistently found that behavioral problems of children with ASD predict maternal stress (e.g., Bromley et al., 2004; Estes et al., 2009; Hastings, 2003; Hastings et al., 2005).

The absence of an effect of developmental age, adaptive behavior and autism severity on maternal stress, however, was in contrast to our expectations and might be explained by a lack of variation in scores within our sample. As all children had ASD and ID, the prognosis for most children was poor (Billstedt, Gillberg, & Gillberg, 2005) and since daily living skills were severely affected in all children, all participants required greater assistance during most basic activities during the day than typically developing children. In contrast to our study some studies did find a relation between maternal stress and cognitive functioning (e.g., Konstantareas & Homatidis, 1989; Konstantareas & Papageorgiou, 2006), adaptive behavior (e.g., Tomanik et al., 2004) and autism severity (e.g., Hastings & Johnson, 2001; Konstantareas & Homatidis, 1989; Konstantareas & Papageorgiou, 2006). However, as already stated by others (e.g., Lecavalier et al., 2006) these relations might be clouded by other variables as behavioral problems. For example, the CARS, which is used to measure autism severity contains items regarding hyperactivity and anxiety.

As children with ASD and ID require intensive care and treatment, parents must contribute substantially to the development of their children and are frequently and actively involved in

their child's therapy. Early intervention based on applied behavior analysis (EIBI) is currently recognized the treatment of choice for children with ASD and results in increased cognitive, social and communication skills and reductions in challenging behavior (Eldevik et al., 2009; Peters-Scheffer et al., 2011; Rogers & Vismara, 2008). Moreover, although EIBI seems to decrease parental stress in the long term (Birnbrauer & Leach, 1993), an important concern of EIBI programs is parental and child stress, especially because parental stress is related to the outcome of EIBI programs (Osborne, McHugh, Saunders, & Reed, 2007). Research implies that parental stress associated with caring for a child with ASD is open to psycho educational treatment (Bristol, Gallagher, & Holt, 1993) and it seems important that parents reduce their stress levels before commencing EIBI since parents who experience stress are less able to contribute to their child's treatment. As indicated by the present study, emotional and behavioral problems contribute significantly more to parental stress than ID or (severity of) ASD, and therefore need to be identified and addressed with priority in EIBI programs.

The current study has some limitations in the sample and method that should be considered in interpreting its results. Parents of the children with ASD were selected through public (pre)schools from most regions in the Netherlands and seem to comprise are representative sample of parents of children with ASD and ID in the Netherlands. However, as involvement in an extensive longitudinal study is time-consuming, parents with the highest levels of stress may decline participation and caution is needed when generalizing these results to the population of children with ASD. As parents had restricted time available, we were limited in our data collection and were therefore notable to collect additional background information on parental and family functioning and data on both parents. As fathers are typically less involved in the daily care of children (Bristol, Gallagher, & Schopler, 1988), we decided to collect data in the mothers. However, it is uncertain if effects found in mothers hold for both parents. As a child functions in a family, family functioning may directly and indirectly through the child's behavior and functioning affect maternal stress (Harris, 1994).

Maternal stress and emotional and behavioral problems were measured using only one data source, i.e. maternal report on the PSI and the CBCL and thereby possible generating common method variance. Although subjective, parents can report information about several situations and behavior, which might not be observed in a short assessment by a clinician. However, in vivo assessment of parental stress and behavioral problems in the home or school setting and multiple informants completing multiple questionnaires would strengthen the study.

Results of this study mark the importance of addressing emotional and behavioral problems in children with ASD and ID as these problems cause more parental stress than children's diagnosis, developmental delay or gender (see also Herring et al., 2006) and parental stress has a negative impact on treatment outcome (Osborne et al.,2007). Therefore, emotionally reactive behavior, withdrawn behavior and attention problems are crucial intervention targets in (early) intervention in children with ASD and ID and optimizing those skills may reduce parental stress and directly and indirectly improve children's functioning.

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Part II
Effectiveness of Early Behavioral Treatment

Chapter 7

A Meta-Analytic Study on the Effectiveness of Comprehensive ABA-based Early Intervention Programs for Children with Autism Spectrum Disorders

Research in Autism Spectrum Disorders, 2011, 5, 60-69

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Peter Sturmey

Abstract

Excitement and controversy have surrounded the effectiveness of Early Intensive Behavioral Intervention (EIBI) for young children with autism. The purpose of this meta-analysis was to investigate the effectiveness of EIBI based on applied behavior analysis in young children with Autism Spectrum Disorders (ASD). There were 11 studies with 344 children with ASD. Quality of studies was assessed using the Downs and Black Checklist. Experimental groups who received EIBI outperformed the control groups on IQ, non-verbal IQ, expressive and receptive language and adaptive behavior. Differences between the experimental and control groups were 4.96–15.21 points on standardized tests. These results strongly support the effectiveness of EIBI.

7.1 Introduction

Autism Spectrum Disorder (ASD) is characterized by severe and sustained impairment in communication and social interaction and restricted patterns of ritualistic and stereotyped behaviors manifested prior to age 3 years (APA, 1994). In approximately 26–40% of young children with ASD intellectual disability (ID) is also present (Baird et al., 2000; Chakrabarti & Fombonne, 2001). A range of behavior problems are also common, including self-injury, anxiety, compulsions, withdrawal, uncooperative behavior, aggression, and destruction of property (Gadow, DeVincent, Pomeroy, & Azizian, 2004; Lecavalier, 2006; McClintock, Hall, & Oliver, 2003). There are many intervention approaches including applied behavior analysis (ABA), diets and vitamins, floor time, holding, medication, options, Picture Exchange Communication System, sensory integration, speech and music therapy, special education and visual schedules (Green et al., 2006; Hess, Morrier, Heflin, & Ivey, 2008); however, there is little empirical evidence for the effectiveness of many of these approaches and available evidence shows mixed results (Foxx, 2008; Howlin, 1997; Schechtman, 2007; Smith, 1999).

Building on research from the 1960s, Early Intensive Behavioral Intervention (EIBI) is the most often studied type of intervention for children with ASD (Matson & Smith, 2008). It is based on principles of operant learning and focuses on remediation of deficient language, imitation, pre-academics, self-help and social interaction skills (Sturmey & Fitzer, 2007) which are broken down into discrete components and taught on a one-to-one basis in school and/or at home, typically using discrete trial teaching (with subsequent planned generalization), reinforcement, backward chaining, shaping, extinction, prompting and prompt fading (Duker, Didden, & Sigafoos, 2004). Parental participation is considered essential to achieve generalization and maintenance. EIBI is effective when it is both intensive (i.e.,approximately 40 h per week) and extensive –minimally 2 years (Lovaas, 2003; Matson & Smith, 2008).

Studies have reported mixed outcomes (Eikeseth, 2009). Several descriptive reviews have concluded that, although EIBI generally has meaningful benefits for young children with ASD, there were large individual differences in treatment response and most children continued to require specialized services (Eikeseth, 2009; Howlin, Magiati, & Charman, 2009; Matson & Smith, 2008; Rogers & Vismara, 2008; Shea, 2004; Smith, 1999).Two meta-analyses (Eldevik et al., 2009; Reichow & Wolery, 2009) found an average medium to large effect size for IQ change despite using different effect sizes (standardized mean change effect size versus a standardized mean difference effect size) and differences in study selection criteria. Eldevik et al. (2009) also found a medium effect size on change of the adaptive behavior composite. Several studies have also reported supplementary measures of adaptive behavior; however, meta-analyses have not systematically analyzed full scale, verbal and performance IQ and measures of adaptive behavior. Thus, it is unclear if the effects of EIBI are robust across all these measures. Therefore, this meta-analysis synthesized the outcomes of comprehensive EIBI programs in which data were collected in group designs using full scale, verbal and performance IQs and measures of adaptive behavior.

7.2 Method

7.2.1 Search methods and selection of studies

There were three search strategies. First, computerized literature searches of MedLine, Psychinfo and Eric were conducted using the keywords ''behavioral treatment'' or ''behavioral intervention'' in combination with ''autism'', ''autism spectrum disorder'' or ''pervasive developmental disorder''. Second, a manual search of the following journals was performed: American Journal on Mental Retardation/American Journal on Intellectual and Developmental Disabilities, Autism, Behavioral Interventions, Behavior Modification, Focus on Autism and Other Developmental Disabilities, Journal of Applied Behavior Analysis, Journal of Autism and Developmental Disorders, Journal of Intellectual Disability Research, Intellectual and Developmental Disabilities/Mental Retardation, Research in Autism Spectrum Disorders and Research in Developmental Disabilities. Third, recent publications on EIBI were inspected to confirm the manual and computer searches. Each article on EIBI retrieved through the manual or computerized search was checked on relevant studies. All EIBI studies in young children with ASD were selected and reviewed if: (1) interventions addressed all three core deficits in autism using ABA; (2) only studies with a pre-test post-test control group were included; (3) all participants had a diagnosis of Autism Spectrum Disorder, including DSM-III, DSM-III-R DSM-IV or ICD diagnosed Autistic Disorder (AD) and Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS); (4) children were aged 10 years or younger at treatment onset; (5) studies contained quantitative outcome data including means and standard deviations on standardized measures of IQ, language and adaptive behavior; and (6) the study was published in English in a peer-reviewed journal between 1980 and 2009. Figure 1 gives an overview of the selection and exclusion process.

Eleven studies met inclusion criteria (see Table 1). Ten were retrieved by computer search and a manual search and reference tracking resulted in one additional study. Each study sample could contribute only one data point to the meta analysis; therefore, since Eikeseth, Smith, Jahr, and Eldevik (2002, 2007) used the same participants, these two studies were treated as one study. Only Smith, Groen, and Wynn (2000) was a fully randomized control trial. Other studies used a pre-test post-test control group design, which was not fully randomized. A second reviewer examined the first 50 articles of the MedLine database. Agreement between the reviewer and the first author was 100%. Study quality was assessed by two independent reviewers using Downs and Black's (1998) checklist.

7.2.2 Data extraction

Outcome variables were IQ and adaptive behavior. All means and standard deviations were obtained directly from published papers when available. When the study did not provide these data (Smith et al., 2000: Non-verbal IQ, Expressive and Receptive Language), the standard scores were calculated using the following formula: outcome in months/ chronological age in months x 100. When studies did not report means and standard deviations of pre- and post-tests, the study was excluded. For each study, mean differences and standard deviations between baseline and treatment were calculated. When a study had two control groups, a weighted mean and standard deviation was calculated, since the similarity between both control groups and the experimental group and the control groups of the other studies made it problematic to select one control group over the other. The meta-analysis was conducted using meta-analysis with interactive explanations (MIX) (Bax, Yu, Ikeda, Tsuruta, & Moons, 2006, 2008).

7.3 Results

7.3.1 Study characteristics

Participants' average age ranged from 33.56 (Magiati, Charman, & Howlin, 2007) to 65.68 months (Eikeseth et al., 2002). Reported gender was 65.70% male, 10.47% female; 23.84% was not reported. All had an Autism Spectrum Disorder (47.09% autistic disorder; 12.79% PDD- NOS; 40.12% not specified) and average IQ ranged from 27.52 (Smith, Eikeseth, Klevstrand, & Lovaas, 1997) to 76.53 (Magiati et al., 2007). Experimental groups received on average 12.5 (Eldevik, Eikeseth, Jahr, & Smith, 2006) to 38.6 h (Sallows & Graupner, 2005) of EIBI for 10 months to more than 2 years. Control groups consisted of less intensive EIBI (<10 h per week; Smith et al., 1997), 12.5–29.08 h per week eclectic treatment (Eikeseth et al., 2002; Eldevik et al., 2006; Howard, Sparkman, Cohen, Green, & Stanislaw, 2005), parent-directed ABA (Sallows & Graupner, 2005; Smith et al., 2000) or treatment as usual (e.g., public early intervention, nursery provision, Portage, school based intervention; Howard et al., 2005; Magiati et al., 2007; Reed, Osborne, & Corness, 2007; Remington et al., 2007; Sheinkopf & Siegel, 1998. Howard et al. (2005) and Reed et al. (2007) had two control groups. Table 1 shows the demographic characteristics.

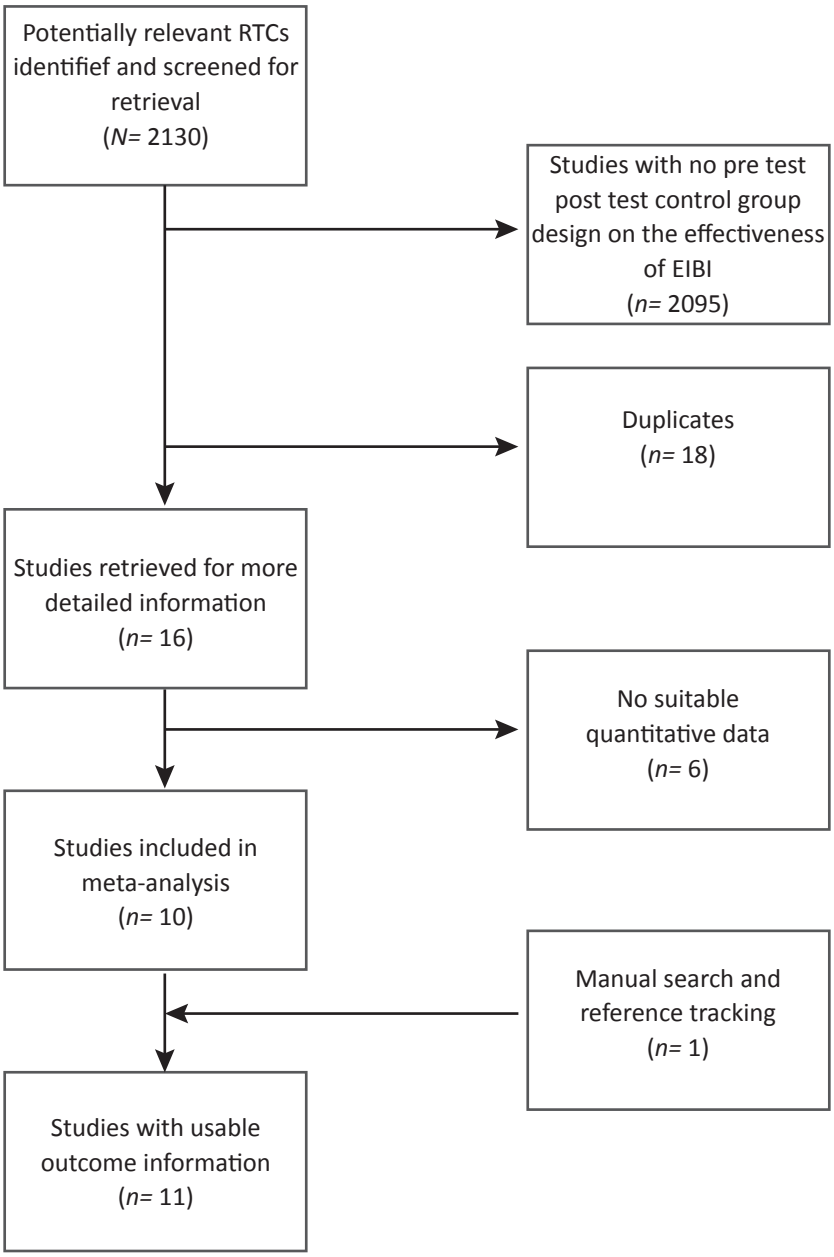


Figure 1. Study identification, screening and selection

Table 1. Characteristics of included studies. N.B. EG = Experimental group; CG = Control Group; CA = Chronological Age in months at onset of treatment; NR. = not reported; AD = Diagnosis of Autistic Disorder by start of the treatment; PDD = Diagnosis of Pervasive Developmental Disorder-Not Otherwise Specified by start of the treatment; ASD = A diagnosis of Autism Spectrum Disorder; this include Autistic Disorder (AD) and Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS); Treatment hours are hours reported in the first year of treatment.

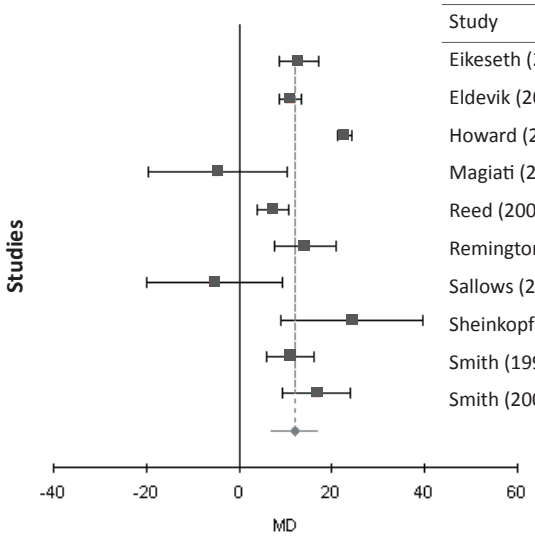
Study	*Sample EG*	*Sample CG*	*Intervention EG*	*Intervention CG*	*Outcome*
Eikeseth et al., (2002; 2007)	N: 13 (8 boys) CA: 66.31 (11.31) IQ: 61.92 (11.31) ASD: 13	N: 12 (11 boys) CA: 65.00 (10.95) IQ: 65.17 (14.97) ASD: 13	28.00 h per week 1-1 beavhioral treatment based on Lovaas using a discrete trial format	29.08 h eclectic treatment per week	The EG made significant larger gains than the CG on IQ, receptive and expressive language and adaptive behavior.
Eldevik et al., (2006)	N: 13 (10 boys) CA: 53.0 (9.5) IQ: 41.0 (15.2) AD: 13	N: 15 (14 boys) CA: 49 (16.9) IQ: 47.2 (14.7) AD: 15	12.5 h. 1–1 behavioral treatment based on Lovaas using a discrete trial format; for 20.3 months	12 h eclectic 1;1 treatment for 21.4 months	The EG displayed significant more change than the CG on intellectual functioning, language and the communication domain of the VABS; no significant differences on other measurements
Howard et al., (2005).	N: 29 (25 boys) CA: 30.86 (5.16) IQ: 58.54 (18.15) AD: 24 PDD-nos: 5	CG1 N: 16 (16 boys) CA: 34.56 (6.53) IQ: 59.88 (14.85) AD: 9 PDD-nos: 7 CG2 N: 16 (13 boys) CA: 37.44 (5.68) IQ: 53.69 (13.50) AD: 12 PDD-nos: 4	25 – 40 h per week 1–1 behavioral treatment based on ABA using a discrete trial format	CG1: 15 h per week public early intervention (eclectic treatment in small groups) CG2: 30 h per week 1:1 or 1:2 eclectic treatment	At follow-up the EG outperformed on intelligence, language and adaptive behavior.
Magiati et al., (2007)	N: 28 (27 boys) CA: 38.0 (7.2) IQ: 83.0 (27.9) AD: 19 PDD: 9	N: 16 (12 boys) CA: 42.5 (7.8) IQ: 65.2 (26.9) AD: 13 PDD: 3	32.4 h 1–1 behavioral treatment based on Lovaas using a discrete trial format; ≥2 years	25.6 h nursery provision using several developmental and behavioral approaches; ≥2 years	No significant differences in cognitive ability, language, play or severity of ASD. Large individual differences in IQ and language level.

Study	*Sample EG*	*Sample CG*	*Intervention EG*	*Intervention CG*	*Outcome*
Remington et al., (2007)	N: 23 (boys: n.r.) CA: 35.7 (4.0) IQ: 61.43 (16.43) ASD: 23	N: 21 (boys: n.r.) CA: 38.4 (4.4) IQ: 62.33 (16.64) ASD: 21	25.6 h 1–1 behavioral treatment based on ABA using a discrete trial format; ≥2 years	Standard provision for young children with autism	The EG outperformed the CG on intelligence, language, daily living skills and positive social behavior.
Reed et al., (2007)	N: 12 (11 boys) CA: 40 IQ: 55.6 (13.8) ASD: 12	CG1: N: 20 (18 boys) CA: 43 IQ: 51.9 (20.1) ASD: 20 CG2: N: 16 (n.r. boys) CA: 38 IQ: 53.3 (16.1) ASD: 16:	30.4 h per week 1–1 generic ABA programs (Lovaas, CABAS and Verbal behavior)	CG1: 12.7 h per special nursery placement in small classes CG2: 8.5 h per week portage, a home-based parent administered teaching program.	EG made greater intellectual and educational gains than children in CG1 and CG 2. CG1 made larger gains than CG2.
Sallows and Graupner, (2005)	N: 13 (11 boys) CA: 35.00 (4.86) IQ: 50.85 (10.57) AD: 13	N: 10 (8 boys) CA: 37.10 (5.36) IQ: 52.10 (8.98) AD: 18	38.60 h per week Clinic directed 1–1 behavioral treatment based on Lovaas; ≥2 years	31.67 h per week parent-directed 1–1 behavioral treatment based on Lovaas; ≥2 years (6 h supervision per month).	After 4 years treatment no group differences existed. The IQ increased from 51 to 76 and 11 children had an IQ over 85.
Sheinkopf and Siegel, (1998)	N: 11 (n.r. boys) CA: 33.8 (6.2) IQ: 62.8 (27.4) AD: 10 PDD-nos: 1	N: 11 (n.r. boys) CA: 35.2 (5.5) IQ: 61.7 (20.2) AD: 10 PDD-nos: 1	19.45 h per week 1–1 home-based behavioral treatment based on Lovaas for 15.36 months	10.70 h school based intervention and 0.44 h other one-to-one therapies.	Children in the EG had higher post treatment IQ scores and post treatment measurements displayed a modest group differences on autism severity
Smith et al., (1997)	N: 11 (11 boys) CA: 36 (6.90) IQ: 28 (4.90) ASD: 11	N: 10 (8 boys) CA: 38 (5.40) IQ: 27 (5.40) ASD: 10	≥30 h 1–1 behavioral treatment based on Lovaas using a discrete trial format; ≥2 years	≤10 h 1–1 behavioral treatment based on Lovaas using a discrete trial format; ≥2 years	In follow-up the EG had higher IQ scores than the CG. 10/11 children in the EG used spoken words versus 2/11 in the CG.
Smith et al., (2000)	N: 15 (12 boys) CA: 36.07 (6.00) IQ: 50.53 (11.18) AD: 7 PDD: 8	N: 13 (11 boys) CA: 35.77 (5.37) IQ: 50.69 (13.88) AD: 7 PDD: 6	24.52 h 1–1 behavioral treatment based on Lovaas using a discrete trial format;≥2 years	3–9 months 5 h per week parent training in 1–1 ABA and 1 h supervision per week.	The EG outperformed the CG at follow-up on IQ, visual spatial skills, language and academic skills and had less restrictive school placements. No differences in behavior problems and adaptive functioning.

7.3.2 Child outcomes

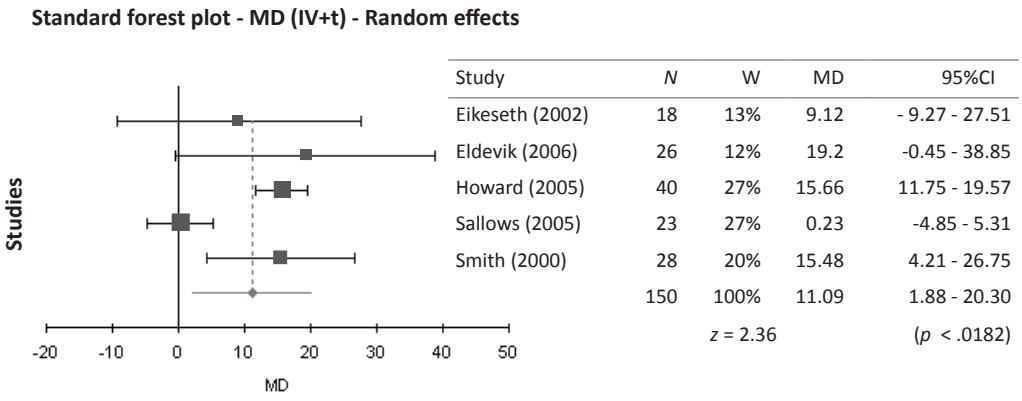
The EIBI group outperformed the control group on all dependent variables. Full scale and non-verbal IQ improved in the EIBI group 11.98 and 11.09 points more than in the control groups, respectively. In receptive and expressive language, the average increases were 13.94 and 15.21 points more, respectively. The EIBI groups surpassed the control groups on composite adaptive behavior, communication, daily living skills and socialization subscales the experimental groups surpassed the control groups by 5.92, 10.44, 5.48, and 4.96 points, respectively. Consistent with the results based on mean differences, Cohen's *d* indicates moderate (adaptive behavior: daily living skills subscale) to large effect sizes (IQ, non-verbal IQ, adaptive behavior, receptive and expressive language). Figs. 2-9 summarize the means, confidential intervals and standard deviations for each study and totals on each dependent variable. Table 2 displays Cohen's *d* for each study on each dependent variable.

The mean quality score (Downs & Black, 1998) was 24.65 out of 32 (*SD* = 1.29; range: 23–27). Intraclass correlation (average measures, two-way random effects model using an absolute agreement definition) between the two reviewers was 0.70 (p = 0.04; 95% *CI*: -0.15 to 0.93). As described in Bax et al. (2009) publication bias and statistical heterogeneity were attested with funnel plots, adjusted rank correlations, Galbraith plots and Tau-squared measures.



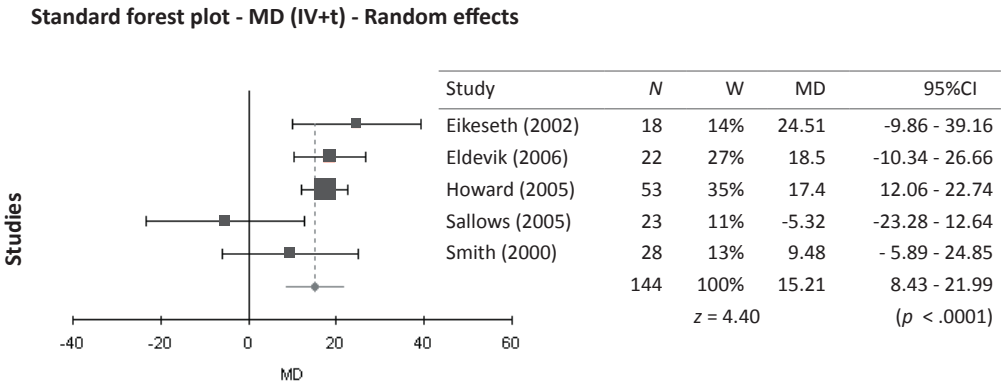
Study	*N*	W	MD	95%CI
Eikeseth (2002)	25	12%	12.83	8.67 - 16.99
Eldevik (2006)	16	12%	11.1	8.69 - 13.51
Howard (2005)	58	13%	22.65	21.07 - 24.23
Magiati (2007)	43	6%	-4.7	-19.74 - 10.34
Reed (2007)	48	12%	7.32	3.84 - 10.79
Remington (2007)	44	11%	14.24	7.74 - 20.74
Sallows (2005)	23	6%	-5.27	-19.86 - 9.32
Sheinkopf (1998)	20	6%	24.3	9.09 - 39.51
Smith (1997)	21	11%	11	5.82 - 16.18
Smith (2000)	28	10%	16.71	9.45 - 23.97
	336	100%	11.98	6.73 - 17.23
		z = 4.47		(*p* < .0001)

Figure 2. Result of EIBI on IQ in individuals with ASD. MD = Mean Difference; IV = Inverse Variance; t = Estimate of the between study variance where the Weight given to each study is calculated by the inverse sum of the within study and between study variance estimates; z = z-score. The z-statistic determines the size of the effect of EIBI when all studies are combined.



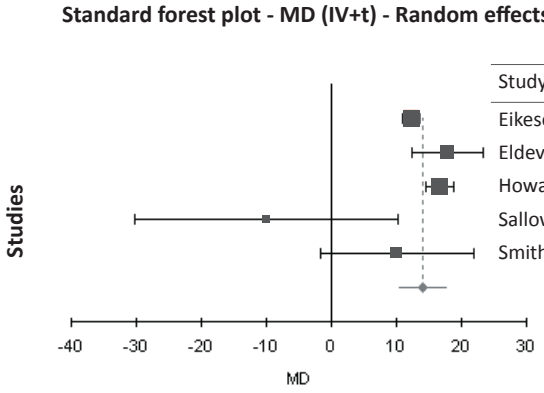
Study	*N*	W	MD	95%CI
Eikeseth (2002)	18	13%	9.12	- 9.27 - 27.51
Eldevik (2006)	26	12%	19.2	-0.45 - 38.85
Howard (2005)	40	27%	15.66	11.75 - 19.57
Sallows (2005)	23	27%	0.23	-4.85 - 5.31
Smith (2000)	28	20%	15.48	4.21 - 26.75
	150	100%	11.09	1.88 - 20.30
		z = 2.36		(*p* < .0182)

Figure 3. Result of EIBI on Non-verbal IQ in individuals with ASD. MD = Mean Difference; IV = Inverse Variance; t = Estimate of the between study variance where the Weight (W) given to each study is calculated by the inverse sum of the within study and between study variance estimates; Z = z-score. The Z-statistic determines the size of the effect of EIBI when all studies are combined.



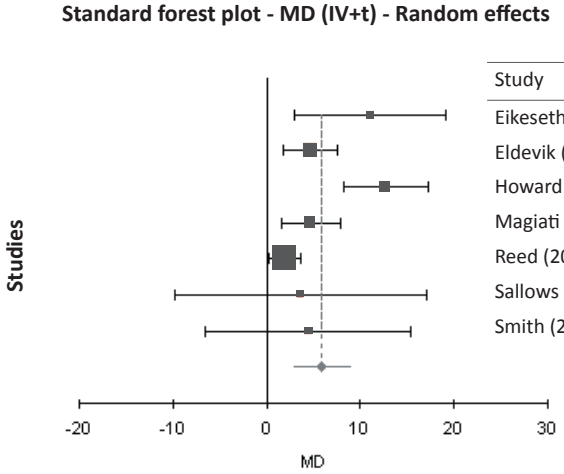
Study	*N*	W	MD	95%CI
Eikeseth (2002)	18	14%	24.51	-9.86 - 39.16
Eldevik (2006)	22	27%	18.5	-10.34 - 26.66
Howard (2005)	53	35%	17.4	12.06 - 22.74
Sallows (2005)	23	11%	-5.32	-23.28 - 12.64
Smith (2000)	28	13%	9.48	- 5.89 - 24.85
	144	100%	15.21	8.43 - 21.99
		z = 4.40		(*p* < .0001)

Figure 4. Result of EIBI on Expressive Language in individuals with ASD. MD = Mean Difference; IV = Inverse Variance; t = Estimate of the between study variance where the Weight (W) given to each study is calculated by the inverse sum of the within study and between study variance estimates; z = z-score. The z-statistic determines the size of the effect of EIBI when all studies are combined.



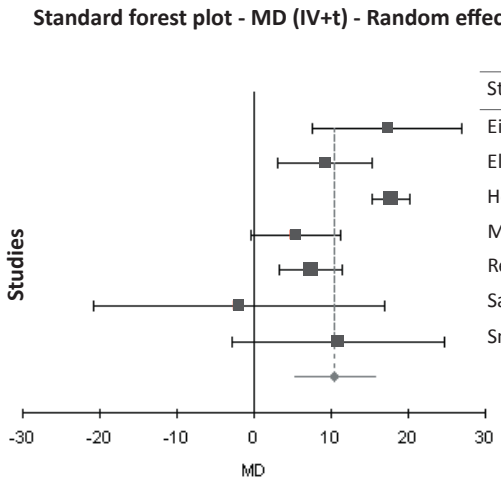
Study	*N*	W	MD	95%CI
Eikeseth (2002)	18	35%	12.27	10.96 - 13.58
Eldevik (2006)	22	21%	17.8	12.31 - 23.29
Howard (2005)	53	33%	16.57	14.42 - 18.72
Sallows (2005)	23	3%	-10	-30.31 - 10.31
Smith (2000)	28	8%	10.09	-1.76 - 21.94
	144	100%	13.94	10.14 - 17.75
		$z = 7.19$		($p < .0001$)

Figure 5. Result of EIBI on Receptive Language in individuals with ASD. MD = Mean Difference; IV = Inverse Variance; t = Estimate of the between study variance where the Weight (W) given to each study is calculated by the inverse sum of the within study and between study variance estimates; z = z-score. The z -statistic determines the size of the effect of EIBI when all studies are combined.



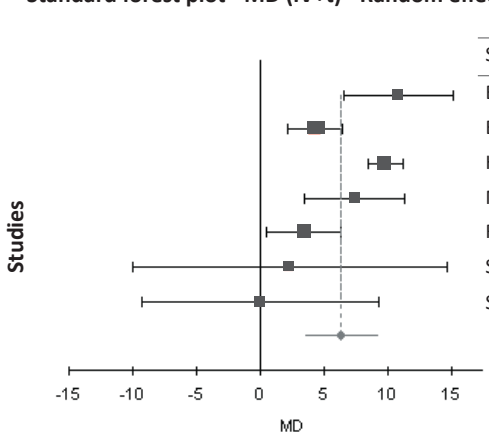
Study	*N*	W	MD	95%CI
Eikeseth (2002)	25	35%	11.06	2.93 - 19.19
Eldevik (2006)	26	21%	4.7	1.80 - 7.60
Howard (2005)	54	33%	12.73	8.20 - 17.26
Magiati (2007)	40	20%	4.7	1.54 - 7.86
Reed (2007)	48	23%	1.91	0.20 -3.62
Sallows (2005)	23	5%	3.66	-9.83 - 17.15
Smith (2000)	28	6%	4.42	-6.59 - 15.43
	244	100%	5.92	2.72 - 9.13
		$z = 3.622$		($p < .0003$)

Figure 6. Result of EIBI on Adaptive Behavior in individuals with ASD. MD = Mean Difference; IV = Inverse Variance; t = Estimate of the between study variance where the Weight (W) given to each study is calculated by the invers sum of the within study and between study variance estimates; z = z-score. The z- statistic determines the size of the effect of EIBI when all studies are combined.



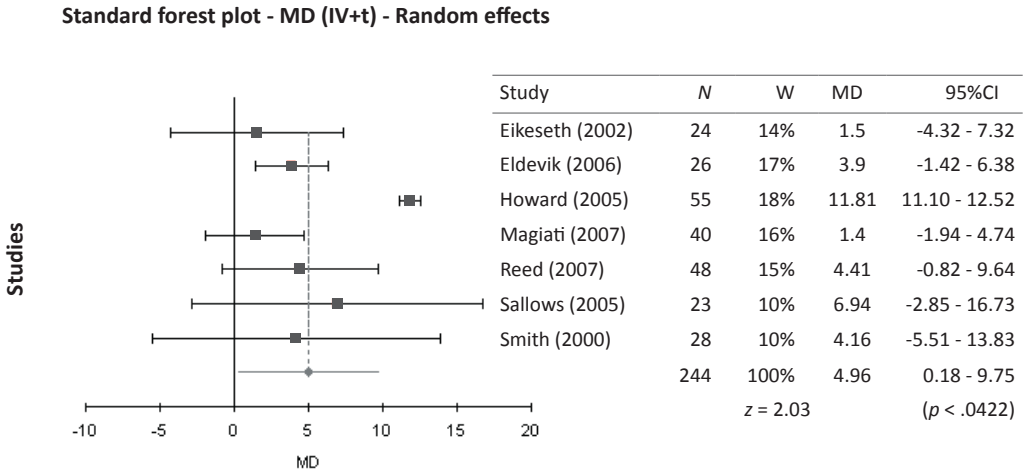
Study	*N*	W	MD	95%CI
Eikeseth (2002)	24	13%	17.29	7.61 - 26.97
Eldevik (2006)	26	17%	9.2	3.05 - 15.35
Howard (2005)	56	20%	17.75	15.21 - 20.29
Magiati (2007)	40	17%	5.4	-0.34 - 11.14
Reed (2007)	48	19%	7.36	3.19 - 11.53
Sallows (2005)	23	6%	-1.97	-20.84 - 16.90
Smith (2000)	28	9%	10.9	-2.91 - 24.71
	245	100%	10.44	5.03 - 15.85
		$z = 3.79$		($p < .0002$)

Figure 7. Result of EIBI on Adaptive Behavior: communication subscale in individuals with ASD. MD = Mean Difference; IV = Inverse Variance; t = Estimate of the between study variance where the Weight (W) given to each study is calculated by the inverse sum of the within study and between study variance estimates; z = z-score. The z-statistic determines the size of the effect of EIBI when all studies are combined.



Study	*N*	W	MD	95%CI
Eikeseth (2002)	24	5%	3.73	-5.38 - 12.84
Eldevik (2006)	26	6%	4.3	-4.08 - 12.68
Howard (2005)	55	22%	8.82	4.92 - 12.72
Magiati (2007)	40	22%	7.4	4.45 - 11.35
Reed (2007)	48	37%	3.75	1.06 - 6.44
Sallows (2005)	23	3%	2.31	-10.04 - 14.66
Smith (2000)	28	5%	0.02	-9.27 - 9.31
	244	100%	5.49	3.36 - 7.61
		$z = 5.07$		($p < .0001$)

Figure 8. Result of EIBI on Adaptive Behavior: daily living skills subscale in individuals with ASD. MD = Mean Difference; IV = Inverse Variance; t = Estimate of the between study variance where the Weight (W) given to each study is calculated by the inverse sum of the within study and between study variance estimates; z = z-score. The z-statistic determines the size of the effect of EIBI when all studies are combined.



Study	*N*	W	MD	95%CI
Eikeseth (2002)	24	14%	1.5	-4.32 - 7.32
Eldevik (2006)	26	17%	3.9	-1.42 - 6.38
Howard (2005)	55	18%	11.81	11.10 - 12.52
Magiati (2007)	40	16%	1.4	-1.94 - 4.74
Reed (2007)	48	15%	4.41	-0.82 - 9.64
Sallows (2005)	23	10%	6.94	-2.85 - 16.73
Smith (2000)	28	10%	4.16	-5.51 - 13.83
	244	100%	4.96	0.18 - 9.75
		z = 2.03		(*p* < .0422)

Figure 9. Result of EIBI on Adaptive Behavior: socialization subscale in individuals with ASD. MD = Mean Difference; IV = Inverse Variance; t = Estimate of the between study variance where the Weight (W) given to each study is calculated by the inverse sum of the within study and between study variance estimates; z = z-score. The z-statistic determines the size of the effect of EIBI when all studies are combined.

Funnel plots indicated some publication bias; however, this was not confirmed by adjusted rank correlations which indicated that publication bias was absent (all *p*'s > 0.22). Galbraith plots showed there was statistical heterogeneity. IQ and the communication and daily living skills domains of the Vineland Adaptive Behavior Scale (VABS) had diverse variances. Tau-squared measures of heterogeneity showed rather high values for full scale IQ, expressive language and VABS communication domain (π^2: 56.30, 27.23 and 35.98, respectively; with theoretical range from 0 to 100) and low values for non-verbal IQ, the receptive language and the composite score, the daily living skills domain and the socialization domain of the VABS (π^2: 0.70, 0.75, 0.14, 1.30 and 0.75, respectively). Thus, the meta-analysis for EIBI contains statistically heterogeneous studies.

Table 2. Cohen's d for each study on each dependent variable. NV-IQ = non-verbal IQ; EL = expressive language; RL = receptive language; ABC = adaptive behavior composite; Com = adaptive behavior: communication subscale; DLS = adaptive behavior: daily living skills; Soc = adaptive behavior: socialization subscale.

	Cohen's *d* for each study on each dependent variable							
Studies	*IQ*	*NV-IQ*	*EL*	*RL*	*ABC*	*Com*	*DLS*	*Soc*
Eikeseth et al. (2002)	2.36	0.39	1.73	9.50	1.09	1.51	0.35	0.19
Eldevik et al. (2006)	3.55	1.11	1.90	2.71	1.25	1.15	0.39	1.21
Howard et al. (2005)	7.24	2.29	1.76	4.20	1.62	3.66	1.10	8.12
Magiati et al. (2007)	-0.19				1.06	0.57	1.66	0.29
Reed et al. (2007)	2.06				0.72	1.91	0.91	0.97
Remington et al. (2007)	1.30							
Sallows and Graupner (2005)	-0.28	0.03	-0.23	-0.38	0.20	-0.08	0.14	0.52
Sheinkopf and Siegel (1998)	1.40							
Smith et al. (1997)	1.75							
Smith et al. (2000)	1.63	1.03	0.45	0.62	0.28	0.56	0.15	0.31
Total	2.00	0.98	1.10	2.91	0.91	1.32	0.68	1.49

7.4 Discussion

Children with ASD participating in EIBI generally outperformed children receiving other treatments or treatment as usual on both IQ and adaptive behavior measures. This confirms findings from other studies on EIBI (Eikeseth, 2009; Howlin et al., 2009) and two other recent meta-analyses (Eldevik et al., 2009; Reichow &Wolery, 2009). The average differences of 11.09 to 15.21 standardized points in scores between the experimental and control groups on IQ, non-verbal IQ and receptive and expressive language and the large effect sizes may be considered clinically significant (Hojat & Gang, 2004).

Consistent with Eldevik et al. (2009), this study found smaller differences on adaptive behavior between the experimental and the control group (4.96–10.44 points) suggesting that future applied work might focus more intensively to improve child

adaptive behavior. This might include a greater quantity of teaching and/or improved quality of teaching of skills in these domains.

Results varied considerable between studies and participants. Differences may be attributable to treatment intensity (Lovaas, 1987), EIBI quality, intensity of supervision (Eikeseth, Hayward, Gale, Gitlesen, & Eldevik, 2009), participant characteristics, and the control group's treatment, if any. Further research should determine which child characteristics, beside baseline IQ and age at start of treatment, are related to treatment outcome (Harris & Handleman, 2000).

Children who do not make dramatic responses are often readily identifiable within a few weeks or months of EIBI (Sallows & Graupner, 2005).

Future research might evaluate what strategies should be adopted to further improve outcomes for these children who may need even more intense EIBI or perhaps technically very precise teaching and a very high degree of treatment integrity to accelerate development. Perhaps these children enter EIBI with key deficits that are not readily remediable with routine EIBI. These might include acquisition of prerequisite skills such as sitting and attending, joint attention skills, and acquisition of praise as a secondary reinforcer or perhaps some of these children have interfering challenging behavior that routine EIBI does not address effectively in the first few months of intervention. An alternate explanation might be that the quality of the teaching that these children receive might be poor and staff and parents working with these children require more careful training and supervision than other staff.

Since a meta-analysis is only based on published studies, publication bias is a threat to validity (Torgerson, 2006). Funnel plots and rank correlation tests of expressive language suggested some evidence for publication bias. More studies with positive than non-significant or negative results are published (Torgerson, 2006); however, another explanation might be that behavioral treatment is indeed effective. Galbraith plots showed that there was indication of statistical heterogeneity. This could be explained in terms of differences in characteristics of the treatment (e.g., setting, amount of supervision provided, intensity) and the participants (age at treatment onset, IQ at treatment onset, diagnosis). This seems typical for the field and for autistic children.

As Reichow and Wolery (2009) and Eldevik et al. (2009) stated, results need to be interpreted cautiously, since studies in this area contain several methodological limitations including small sample sizes, non-randomized assignments to groups, non-uniform assessments protocols, use of quasi-experimental designs, lack of equivalent groups, lack of adequate fidelity measures, unknown characteristics of comparison conditions, and selection bias (Boyd, 1998; Eikeseth, 2009; Gresham & MacMillan, 1997; Levy, Kim, & Olive, 2006; Mundy, 1993; Schopler, Short, & Mesibov, 1989; Wheeler, Baggett, Fox, & Blevins, 2006); only one study (Smith et al., 2000) was a fully randomized control trial. Despite these potential limitations, this meta analysis demonstrated that EIBI has a moderate to large effect in young children with autism on full scale and non-verbal IQ and adaptive behavior.

Acknowledgments

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Chapter 8

Low Intensity Behavioral Treatment Supplementing Preschool Services for Young Children with Autism Spectrum Disorders and Severe to Mild Intellectual Disability

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Abstract

This study evaluated the effectiveness of low intensity behavioral treatment (on average 6.5 hours per week) supplementing preschool services in 3–6-year-old children with autism spectrum disorder and severe to mild intellectual disability. Treatment was implemented in preschools (i.e., daycare centers) and a discrete trial teaching approach was used. Twelve children in the treatment group were compared to 22 children receiving regular intervention. At pre-treatment, both groups did not differ on chronological age, developmental age, diagnosis and level of adaptive skills. Eight months into treatment, children receiving behavioral treatment displayed significantly higher developmental ages and made more gains in adaptive skills than children from the control group. No significant differences between groups were found on autistic symptom severity and emotional and behavioral problems.

8.1 Introduction

Young children with autism spectrum disorders (ASD) and intellectual disabilities (ID) show restricted, repetitive behaviors and deficits in social reciprocity and communication (O'Brien & Pearson, 2004). They show internalizing and externalizing behavior problems (Harley, Sikora, & McCoy, 2008) and display more impairment in adaptive behavior than children with ID only, mostly caused by lower functioning on communication and socialization (Carpentieri & Morgan, 1996). Results of relatively many studies have shown that with intensive behavioral treatment, based on applied behavior analysis (ABA), substantial gains can be made in cognitive, language, academic and adaptive skills (see e.g., Eldevik et al., 2009; Eikeseth, 2009; Green, 1996; Howlin, Magiati, & Charman, 2009; Matson & Smith, 2008; Reichow & Wolery, 2009; Rogers & Vismara, 2008; Schreibman, 2000). For example, in a study by Lovaas (1987) behavioral treatment was given to 19 children with ASD during 40 hours per week using a discrete trial format. This group was compared to two control groups: one receiving 10 hours or less behavioral treatment and one group not receiving behavioral treatment. Although these groups were comparable on several variables at pre-treatment, after 2 years the experimental group outperformed both control groups on educational placement and IQ. Results were maintained at follow-up (McEachin, Smith, & Lovaas, 1993). Lovaas' approach (1987) has been replicated in older children (Eikeseth, Smith, Jahr & Eldevik, 2002; Eikeseth, Smith, Jahr, & Eldevik, 2007), children with ID (Smith, Eikeseth, Klevstrand, & Lovaas, 1997), and in other settings, such as home (Cohen, Amerine-Dickens & Smith, 2006; Sheinkopf & Siegel, 1998) and school (Eikeseth et al., 2002). Results of these studies show that while fewer children achieved average levels of functioning than in the Lovaas study (1987), a subset of children obtained an improvement in IQ. In general, better outcomes are reported for children who start treatment early and in children with a higher IQ (e.g., Harris & Handleman, 2000). Since the study published by Lovaas (1987) several empirical studies were conducted with a variety

of treatment hours per week. More intensive treatment (up to approximately 40 hours per week) is associated with the best results (e.g., Eikeseth et al., 2002; Eldevik, Eikeseth, Jahr, & Smith, 2006; Sallows & Graupner, 2005; Smith et al., 1997).

Despite its effectiveness, intensive behavioral treatment is difficult to implement in clinical practice and parents reported several barriers implementing such intensive treatment (Johnson & Hastings, 2001). Primary concerns are the availability of professionals (e.g., recruiting and maintaining a suitable team) and obtaining funding of the treatment from the education departments. Also, concerns about the school, child and impact on the family were reported. Therefore, children with ASD may not have the opportunity to start intensive treatment and less intensive treatment could be an accessible option. Studies on the effects of lower intensity treatment (in terms of hours of one-to-one treatment) are therefore warranted. Until present, only one study has evaluated the effects of low intensity behavioral treatment by comparing 13 children (mean IQ of 41) receiving behavioral treatment to 15 children (mean IQ of 47) receiving the same amount of eclectic treatment (Eldevik et al., 2006). The behavioral treatment consisted of one-to-one-training and implementation of behavioral intervention in the classroom for an average of 12.5 hours per week. Parents generalized the learned skills in their children to other settings, like home. The eclectic group received the same amount of treatment, which was based on a combination of alternative communication and/or total communication, sensory motor treatment, TEACCH, and other approaches. After 2 years, the behavioral treatment group had made a significantly larger progress on intellectual functioning, language comprehension, expressive language and the Communication scale of the VABS than the eclectic treatment group. Also, the behavioral treatment group had significantly lower scores on several pathology items assessed with Lovaas' measure of pathology (see Lovaas, 1987), including peer play, toy play, toilet training, affectionate behavior and general pathological symptoms. No significant effects of treatment on non-verbal intelligence, the adaptive behavior composite, daily living skills and socialization were found. Although gains were smaller than in other studies, this study suggested that even low intensity behavioral treatment can be effective in young children with ASD and ID.

Presently, our knowledge base of the effects of low intensity treatment is scarce (see e.g., Eldevik et al., 2006). Other studies, therefore, should be conducted in this area. This study evaluated the effects of low intensity one-to-one behavioral treatment in young children with ASD and ID by providing less than ten hours of one-to-one behavioral treatment per week for a period of 8 months. Although behavioral treatment mutates and evolves over time and between program differences exist (Love, Carr, Almason, & Petursdottir, 2009), treatment procedures are based on operant conditioning (e.g. stimulus control, task analysis, reinforcement, prompting, generalization, discrete-trial instruction, incidental teaching). A comprehensive skill based hierarchically organized and developmentally sequenced curriculum is used, which is individualized to each child' strengths and deficits. First, treatment is provided in a one-to-one setting at home, but over time gradual and systematic transitions to small-group and large-group formats and to other environments (as preschool, kindergarten, school) are

made when children developed the skills required to learn in those settings. Generally, the training is directed by individuals with advanced training in ABA and experience with young children with autism and parents are active as co therapists of their children. Most programs provided 20–30 hours of structured treatment per week and additional informal instruction and practice during the children's other waking hours for 2 or more years. Treatment is generally started at 3 or 4 years of age (Green, Brennan, & Fein, 2002). In this study the children received 5–10 hours structured one-to-one training at the preschool. Next to the one-to-one training, the children attended a preschool for children with ID. In this preschool, teachers were instructed and stimulated to use the principles of behavioral treatment to teach new skills and generalize skills learned in the training in a daily context. As usual in behavioral treatment, parents were instructed and stimulated to generalize the skills their child had learned to as many settings in the community as possible and some parents acted as co therapist for their children. The intervention period in this study is relatively short in comparison to other studies. To prevent children changing (pre)schools during the intervention period, a intervention period of 8 months was chosen to avoid a co founding influences of different schools during the intervention period. Aim of this study was to assess the effects of low intensity one-to-one behavioral treatment with an environment that informally uses behavioral principles, on developmental age and adaptive skills in 12 children with ASD and ID. Additionally, symptom severity and emotional and behavioral problems were assessed, as they could interfere with treatment and change as a result of the treatment.

8.2 Method

8.2.1 Participants and setting

All participants attended a preschool setting for children with ID and met the following criteria: (1) a diagnosis of Autistic Disorder (AD) or Pervasive Developmental Disorder Not Otherwise Specified (PDD-NOS) and ID based on DSM-IV criteria established by a licensed and independent psychologist or psychiatrist; level of IQ was assessed by standard intelligence tests (e.g., Wechsler Preschool and Primary Scale of Intelligence-Revised, SON-2.5–7, Bayley Scales of Infant Development), (2) a chronological age below 7 years and (3) absence of medical conditions (e.g., visual impairment; uncontrolled epilepsy) that could interfere with treatment. The treatment group consisted of 12 children who started behavioral treatment in a preschool. Parents gave their written consent in all cases. Twenty-two children comprised the control group and these children visited preschools in which no one-to-one behavioral treatment was given.

Table 1 shows chronological age, developmental age, estimated ratio of the mental developmental index, adaptive behavior composite and the total score of the Scale of Pervasive Developmental Disorder in Mentally Retarded Persons (PDD-MRS; Kraijer, 1999).

Results from *t*-tests showed no statistically significant differences between groups on chronological age, $t(32) = 0.16$, $p = .88$, developmental age, $t(32) = 1.01$, $p = .32$, symptom severity of ASD, $t(32) = 0.88$, $p = .39$, and level of adaptive skills, $t(32) = 0.89$, $p = .38$.

Table 1. Demographic characteristics of the participants at pre-treatment.

Variabele	Treatment group			Control group		
	M	*SD*	*Range*	*M*	*SD*	*Range*
Chronological age in months	53.50	5.52	42-62	52.95	11.14	38-75
Developmental age in months	25.92	7.57	17-40	23.32	6.33	15-36
Mental Developmental Index/ IQ	47.00	10.33	31-64	45.73	15.99	21-77
VABS-composite in months	20.83	6.69	13-35	19.18	4.14	11-30
Raw score PDD-MRS	11.58	4.42	1-18	12.91	3.79	3-18

Note: PDD-MRS = Pervasive Developmental Disorder in Mentally Retarded Persons; VABS = Vineland Adaptive Behavior Scales.

8.2.2 Design

A non-randomized pretest posttest control group design was used to assess the effects of low intensity behavioral treatment. Before intake measures were collected, the third author assigned children based on therapist' availability to the treatment group or the control group. Parents and teachers were not informed about the exact aim and design of the study and were told the study was about the development of young children with ASD and ID. They were unaware of whether their child was in the experimental or in the control condition.

8.2.3 Treatment

All participants attended 13 preschools for children with ID for an average of 28.38 hours per week (*SD* = 3.83; range: 16-36 h). A class consisted of 6-10 children with comparable disabilities and levels of functioning, and two or three teachers. Classes typically started at 9 a.m. and ended at 3 p.m. on weekdays. The treatment consisted of elements of TEACCH to structure and visualize the environment (such as visual timetables, routines, and workstations), incidental teaching, structured play and activities in a group setting. Individual physiotherapy, speech therapy, music therapy or play therapy with a maximum of 1 hour per week was provided to each child. Treatment was supervised by an experienced psychologist or special educator. In addition to the group program, the treatment group received 5-10 hours (*M*= 6.29; *SD* = 1.31) one-to-one treatment per week, based on the work of Lovaas (2003) using a one-to-one discrete trial format in which no aversive stimuli were used.

The treatment was provided at the preschool and began with five basic treatment targets: attention skills, responding to simple requests, non-verbal imitation, matching and communication (e.g., pointing, PECS). Depending on the child's developmental level and parental requests other targets were added. Additional programs addressed self-help skills, labeling objects and abstract concepts, identifying actions, play skills, social interaction and emotions. Teachers of the children in the treatment group also informally used ABA principles in the group context (e.g., prompting, discrete trials, extinction and reinforcement) to teach new skills and generalize mastered skills to the daily context. As in the group setting staff primarily focused on managing daily activities (e.g. eating, toileting, free play, leisure activities) and only limited worked on weekly treatment targets, these hours spend in the group setting were not counted as ABA treatment hours.

Treatment was provided by trainers and teachers of the preschool, who were supervised by a special educator with 5 years of extensive experience in applying ABA in young children with autism. Before the start of the behavioral treatment all parents, trainers and teachers received a workshop on how to apply techniques of applied behavior analysis (e.g., discrete trials, reinforcement and prompting). In the workshop, programs were demonstrated and role played. Furthermore, supervision was given through monthly staff meetings, in which all teachers received feedback on video fragments of their training sessions. Monthly, treatment sessions were observed and the trainer was provided with feedback during and after treatment using verbal instruction and modeling. A subset of parents (25%) and class room teachers (66%) trained the children for 1 hour per week. In additional monthly meetings individually organized for each child parents and all teachers were informed of the skills the child had learned in training. Parents and class room teachers were instructed to generalize the skills by implementing these skills in every day routines.

As was the case in the Lovaas (1987) study, no formal measures were taken to assess procedural integrity, but through treatment protocols, the use of manuals (Leaf & McEachin, 1999; Lovaas, 2003), meetings, video-recordings during treatment, data-sheets with trial-by-trial data and an individual learning plan with detailed descriptions of each program, and supervision by a special educator, fidelity was facilitated in the treatment group (Eikeseth, 2001). Through reports of parents, teachers and psychologists, the absence of one-to-one behavioral treatment in the control group was confirmed.

8.2.4 Instruments and data-collection

At pre-treatment and after 8 months of treatment all children were assessed with the Dutch version of the Bayley Scales of Infant Development (Second Edition) to determine developmental age and an estimated ratio of the mental developmental index (van der Meulen, Ruiter, Lutje Spelberg, & Smrkovsky, 2002). The children with a developmental age of 30 months or more were additional assessed with the SON-2.5–7 (Snijders, Tellegen, Winkel, & Laros, 1996). Eight children (four of the treatment group) completed this non-verbal intelligence test.

After 8 months the children were assessed either with the SON-2.5–7 or the BSID-II-nl depending on the test used in pre-treatment.

The BSID-II-nl measures mental development up to 42 months. An estimated ratio score (mental age/chronological age) was used for all children who scored below the norms or were older than 42 months. The SON-2.5–7 determines non-verbal intelligence in children between 2.5 and 7.0 years old. Both tests have been validated and are commonly used for children with ID and autism (Snijders et al., 1996; van der Meulen et al., 2002).

At pre- and post-treatment and at two (i.e., period 2), four (i.e., period 3) and six (i.e., period 4) months of treatment the Dutch survey version of the Vineland Adaptive Behavioral Scales (de Bildt & Kraijer, 2003), the Dutch translation of the Child Behavior Checklist (CBCL; Achenbach & Rescorla, 2000) and the Dutch version of the PDD-MRS (Kraijer, 1999) were completed by interviewing the parents of all children. To simplify interpretation of the results only the pre- and posttreatment scores are reported. The Vineland Adaptive Behavior Scales (Sparrow, Balla, & Cicchetti, 1984) were used to measure adaptive behavior and provide an adaptive behavior composite score and scores in three domains: Communication, daily living skills and socialization. Reliability and validity of the Dutch survey version are high in individuals with ID (de Bildt, Kraijer, Sytema, & Minderaa, 2005). Since standard scores were not available for all children and only range from 1 to 10, age equivalents are reported and analyzed. The behavioral problem scale of the CBCL is a parent-completed questionnaire consisting of 100 items assessing emotional and behavioral problems and reliability and validity are adequate. American norms were used to calculate standard scores (Achenbach & Rescorla, 2000). The PDD-MRS is a 12-item questionnaire that can be used in the screening for ASD in children and adults with ID (Kraijer, 1999). It is based on the three aspects of ASD: Communication, Social behavior and Stereotyped behavior. The PDD-MRS is commonly used to screen for ASD. Reliability and validation of the PDD-MRS are good (Kraijer and de Bildt, 2005).

8.2.5 Statistical analysis

The statistical analysis focused on the differences between the treatment group and the control group over time. First, *t*tests were conducted to confirm resemblance on several variables between both groups at pre-treatment. Second, Multivariate and Univariate GLM repeated measures were used to assess group differences over time.

8.3 Results

8.3.1 Developmental age and mental developmental index (MDI)

At pre-treatment, *t*-tests did not reveal any significant differences between the treatment group and the control group, on developmental age, $t(19.49) = 1.01$, $p = .32$, and the estimated ratio score of the MDI, $t(31) = 0.28$, $p = .78$.

However, after 8 months of treatment, the treatment group outperformed the control group (see Table 2). Univariate GLM repeated measures (Greenhouse-Geisser) revealed significant interaction effects on developmental age, $F(1,32) = 23.37$, $p < .01$, and the estimated ratio score of the MDI, $F(1,32) = 26.96$, $p < .01$.

8.3.2 Adaptive skills

At pre-treatment no significant differences were found on the Vineland-composite, $t(16) = 0.78$, $p = .45$, and the domains communication, $t(19) = 0.47$, $p = .65$, daily living skills, $t(16) = 1.59$, $p = .13$, and socialization, $t(32) = 1.78$, $p = .08$. Since the subscales of the VABS were correlated, $r = .42$ to $r = .86$, a multivariate GLM repeated measures was used to analyze the differences on the subscales. A significant interaction effect, $\lambda = 0.71$, $F(12,21) = 4.33$, $p < .01$, indicated an improvement in both groups on all scales, but the progress in the treatment group was significantly larger. Univariate GLM repeated measures (Greenhouse-Geisser) also revealed significant interaction effects on the domains communication, $F(1,32) = 6.48$, $p = .02$, daily living skills, $F(1,32) = 13.17$, $p < .01$, and socialization, $F(1,32) = 44.86$, $p < .01$, representing a significantly larger progress in the treatment group (see Table 2).

An univariate GLM repeated measures analysis revealed a significant interaction effect on the VABS composite score at treatment over time, $F(1,32) = 15.68$, $p < .01$. As expected, both the treatment group and the control group made gains in adaptive skills. However, in the treatment group these gains were significantly larger (see Table 2).

8.3.3 Symptom severity of autism

No significant differences on symptom severity of autism appeared between both groups at pre-treatment, $t(20) = 0.88$, $p = .39$, and post-treatment, $t(27) = 0.84$, $p = .41$. GLM repeated measures shows a significant effect of time, $F(1,32) = 6.22$, $p = .02$, indicating a decrease of symptom severity over time in both groups.

8.3.4 Emotional and behavioral problems

Using *t*-tests, significant differences between both groups appeared on the total T-score, $t(21) = 2.37$, $p < .05$, and on the internalizing scale, $t(25) = 3.33$, $p < .01$, of the CBCL at pre-treatment. Differences on external scales were not significant, $t(18) = 1.32$, $p = .20$. GLM repeated measures indicated no significant effect over time and treatment for the total T-score, $F(1,32) = 0.40$, $p = .53$, the internalizing scale, $F(1,32) = 0.33$, $p = .57$, and the external scale, $F(1,32) < 0.01$, $p = .96$.

Table 2. Developmental age in months, estimated mental developmental index/IQ and F values for the treatment group versus the control group on pre- and post-treatment measures.

Variable	Treatment group				Control group				Statistics	
	Pre-treatment		Post-treatment		Pre-treatment		Post-treatment			
	M	*SD*	*M*	*SD*	*M*	*SD*	*M*	*SD*	*F*	*p*
Developmental age in months	25.92	7.57	34.83	10.89	23.32	6.33	25.73	8.26	$F(1, 32) = 23.37$	<.01
Mental developmental index/ IQ	47.00	10.33	55.83	14.94	45.73	15.99	43.73	16.74	$F(1, 32) = 26.96$	<.01
VABS-composite in months	20.83	6.69	31.75	10.96	19.18	4.14	22.05	7.47	$F(1, 32) = 15.68$	<.01
VABS-communication in months	26.92	12.12	39.42	15.39	25.00	10.00	29.95	13.39	$F(1, 32) = 6.48$	.02
VABS-daily living in months	23.83	7.28	33.25	9.04	20.14	4.68	23.23	7.70	$F(1, 32) = 13.17$	<.01
VABS-socialization	20.75	4.54	34.08	8.14	24.64	8.18	25.14	7.21	$F(1, 32) = 44.86$	<.01
CBCL-total	60.00	8.37	58.25	8.02	66.91	7.70	63.23	7.98	$F(1, 32) = 0.40$	.53
CBCL- internalizing	60.58	5.58	59.08	7.74	67.55	6.27	64.41	8.45	$F(1, 32) = 0.33$	.57
CBCL-externalizing	58.92	10.82	54.33	8.52	63.59	7.89	58.86	6.26	$F(1, 32) < 0.01$	.96
PDD-MRS raw score	11.58	4.42	10.25	3.14	12.91	3.79	11.27	3.84	$F(1, 32) = 0.06$	.82

8.4 Discussion

In this study, 12 children with ASD and ID receiving on average 6.5 hours additional behavioral treatment per week were compared to 22 children receiving treatment as usual. During 8 months all children made significant progress on developmental age and adaptive skills. However, the improvement on developmental age and adaptive skills was significantly larger in the children who received behavioral treatment supplementing preschool services. No significant changes were found on severity of ASD and on emotional and behavioral problems. The present study is the first study in this area that was conducted in The Netherlands.

A higher IQ and more intensive treatment are associated with better treatment outcome (e.g., Harris & Handleman, 2000; Lovaas, 1987). Nevertheless, this study and the study of Eldevik et al. (2006) found good results on intellectual functioning and the Communication domain of adaptive behavior. Only this study found significant differences on all adaptive domains. By contrast, Eldevik et al. (2006) found a positive effect of behavioral treatment on several measures of pathology. Differences in outcome between this study and Eldevik et al. (2006) can be explained in terms of treatment and methodology. While the children of Eldevik et al. (2006) received 12.5 hours behavioral treatment, children in the present study received only 6.5 hours of one-to-one treatment. However, additionally to the one-to-one treatment the children in the present study received a behavioral approach in the group setting. Combining the one-to-one training and the informal behavioral approach in the group setting children in the present study received on average 28 hours of behavioral treatment, considerably more than in Eldevik et al. (2006).The supervision in this study is limited to 1 staff meeting of 2 hours per month per location (1–5 children), supervision during treatment (on average 1 hour per month per child) and 1 meeting with parents and staff of 1–2 hours per month for each child. While the children in this study received on average 1.5 hours of supervision per week, normally supervision during an EIBI program contains up to 10 hours per week (Eikeseth, Hayward, Gale, Gitlesen, & Eldevik, 2009; Hayward, Gale, & Eikeseth, 2009). Since intensity of supervision is related to treatment outcome, this could explain differences in outcome between the studies, as in the Eldevik et al. (2006) 1–4 hours consultation per week and 2 hours supervision per child per week were provided.

The results of this study indicate that the largest group differences on adaptive skills are found from 2 to 6 months into treatment. Possibly, the effect of low intensity behavioral treatment diminishes over time. It remains to be investigated if this could explain the more modest results on adaptive behavior reported by Eldevik et al. (2006) after 2 years of treatment. In the present study the control group received treatment as usual, while the treatment group received an additional treatment. By contrast, the control group in Eldevik et al. (2006) received an eclectic treatment. Skills learned in the eclectic treatment as well as the expectations of the parents of the eclectic treatment could account for smaller group differences. This study found no treatment effect on symptom severity of ASD. However, it is unclear whether this can be attributed to the treatment or to the sensitivity of the PDD-MRS, used to

assess symptom severity of ASD. As this measure is a screening tool, the PDD-MRS may not be sensitive enough to measure treatment effects (Kraijer and de Bildt, 2005). In further research other instruments (e.g., CARS; Schopler, Reichler, & Rochen Renner, 2007) should be used to assess the treatment effect on symptom severity of ASD.

By contrast to this study and Eldevik et al. (2006) the Lovaas's study (1987) in which one of the control groups received less than 10 hours behavioral treatment per week for 2–3 years no significant differences were found on IQ and school placement at all in comparison to another control group that did not receive behavioral treatment at all. As Eldevik et al. (2006) have stated, comparing the low intensity group of Lovaas' study to more recent research is complicated due to missing data on the exact training hours, advances in the treatment methodology and other methodological issues.

Anecdotally, parents and teachers of children in the experimental group reported as soon as after a few months into treatment that their child was more attentive to other people and events, more calm and concentrated. Some parents reported the most important improvement in the area of communication. Because the child understood the parents better and was better able to express his or her needs, challenging behavior was prevented, so parents could engage in every day situations (e.g., go to a store or visit family) with their child. Parents and teachers evaluated the learned ABA techniques as useful in coping with challenging behavior and teaching new skills to their child and used them in every day life.

This study has several limitations and strengths. Although typical for this kind of studies, the sample size is relatively small, the children are not randomly assigned to the treatment and control groups and the authors conducted part of the assessments (which is also mentioned by Eldevik et al., 2006). Although studies with random assignment to groups are superior, they are difficult to realize in clinical practice in terms of cost and ethics (Kasari, 2002; Matson & Smith, 2008). Selection on therapist availability is a common and a generally accepted alternative (see e.g., Smith et al., 1997).

Although more research is necessary in this area, preliminary results of this study suggest that children with ASD and ID may make significant gains with low intensity behavioral treatment. While treatment results are promising, the questions remains in which degree differences are clinically significant. Clinical significance is usually defined as returning to normal functioning (Jacobson, Roberts, Berns, & McGlinchey, 1999) and in this study treatment did not enable any of the participants to perform in the average range of functioning. Although the differences between the treatment group and the control group of this study are substantially smaller than in other EIBI studies (e.g. Lovaas, 1987; Sallows & Graupner, 2005), the participants were intellectually more impaired and the intervention period was relatively short. Further research over a longer period of time including measures of social validation is necessary to conclude if low intensity behavioral treatment supplementing preschool based services is an effective option if intensive behavioral treatment is not possible.

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Chapter 9

Effectiveness of Low Intensity Behavioral Treatment for Children with Autism Spectrum Disorder and Intellectual Disability

Pending minor revisions accepted for publication

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Abstract

To determine the effectiveness of low intensity behavioral treatment (LIBT) supplementing regular treatment in young children with autism spectrum disorder (ASD) and intellectual disability (ID) standardized tests of cognition, adaptive behavior, interpersonal relations, play, language, characteristics of autism, emotional and behavioral problems, behavioral flexibility, early social communication, and maternal stress were administered in a treatment group (*n* = 20), receiving 4-10 hours LIBT per week and a control group (*n* = 20) receiving treatment as usual. At baseline, no differences were found between groups (mean chronological age: 5;3 years; mean developmental age: 1;11 years) on several key variables, but after two years of intervention the treatment group outperformed the control group on IQ, developmental age, adaptive behavior, interpersonal relations, play and receptive language, and less autistic symptoms were seen in treatment group. Following intervention, no differences between groups were found on expressive language, behavioral flexibility and maternal stress. Progress in developmental age, adaptive behavior, interpersonal relations, play and receptive and expressive language was clinically and reliably significant for the majority of the LIBT group.

9.1 Introduction

Autism Spectrum Disorder (ASD) is a disorder associated with severe impairments in communication and social interaction and stereotyped and restricted patterns of behavior, with an onset in early childhood (American Psychiatric Association, 2000). Many individuals with ASD have intellectual disability (ID) and a majority may never mature to independence in adult life (Mordre et al., 2012; Yeargin-Allsopp et al., 2003). Behavioral inflexibility, anxiety, sleeping and eating disturbances, temper tantrums, aggression and self-injurious behaviors are common in children with ASD (e.g., Hartly, Sikora, & McCoy, 2008; McClintock, Hall, & Oliver, 2003).

Although a wide range of educational and therapy options for ASD are available, early intensive behavioral intervention (EIBI) currently enjoys the strongest research validation for effectiveness in ASD and evidence is accumulating that EIBI successfully addresses the core symptoms of ASD and increases the likelihood of being included in mainstream settings later on (Eikeseth, 2009; Matson & Smith, 2008; Peters-Scheffer, Didden, Korzilius, & Sturmey, 2011; Reichow & Wolery, 2009; Smith, Groen, & Wynn, 2000).

Although EIBI programs comprise slightly different approaches, they share some essential features. All are based on learning principles derived from Applied Behavior Analysis (ABA) and common reinforcement-based procedures include prompting, stimulus generalization, fading, modeling, task analysis, error correction, functional analysis, and shaping procedures. Usually, a discrete trial format is used to foster systematic teaching of distinct behaviors and to ensure repetition and structured presentations of tasks.

The curriculum is comprehensive, developmentally sequenced and individualized and addresses pre-academic skills (e.g., attending, imitation, matching), language, daily living skills, social skills and behavioral problems (e.g., stereotypy, self-injury). Ideally, EIBI programs are implemented for 30-40 hours per week for two years or more and instruction is primarily delivered in a one to one format gradually extending to (small) groups. Parent involvement is crucial to facilitate generalization and maintenance of new skills (Eikeseth, 2009; Green, Brennan, & Fein, 2002; Hayward, Gale, & Eikeseth, 2009; Howlin, Magiati, & Charman, 2009; Peters-Scheffer et al., 2011).

EIBI yields significant improvements in children with ASD, especially when intervention is implemented intensively (i.e., 40 hrs per week; Eldevik, Hastings, Hughes, Jahr, Eikeseth, & Cross, 2010; Granpeesheh, Dixon, Tarbox, Kaplan, & Wilke, 2009; Lovaas, 1987; Reed, Osborne, & Corness, 2007; Reichow & Wolery, 2009) and lasts at least two years (Reichow & Wolery, 2009). For example, Reed, Osborne, and Corness (2007) studied the effectiveness of home-based early behavioral intervention for children between 2;6 to 4;0 years old and found that over a period of 9-10 months high-intensity behavioral approaches (on average 30 hours per week) produced greater gains in intellectual and educational functioning than low-intensity programs (mean 12 hours per week). Granpeesheh et al. (2009) evaluated the outcome of 245 children receiving EIBI and found a significant increase in new skill acquisition with increased treatment hours. However, when analyzed in age groups, differences in the relationship between treatment hours and treatment outcome were found. For children between 2-7 years of age an association between the number of treatment hours per week and the number of mastered behavioral objectives was found, while children between 7-12 years did not show a significant relationship between treatment hours and the number of behavioral objectives mastered. In this age group participants mastered approximately 17 behavioral objectives per months regardless if the child was receiving high or low levels of treatment hours. In addition, two meta-analytic studies addressed the intensity of treatment (Makrygianni & Reed, 2010; Virués-Ortega, 2010). Both studies found that children's gains on intellectual and adaptive behavior benefited from intervention intensity. However, progress on language skills benefited more from intervention duration than from intervention intensity (Virués-Ortega, 2010).

Next to treatment intensity and duration, intensity and quality of supervision seems to be an important element to successful EIBI (Eikeseth, Hayward, Gale, Gitlesen, & Eldevik, 2009). Better outcome is reported for children with lower chronological age, a higher IQ, more adaptive behavior and less severe autism at the start of the treatment (Ben-Itzchak & Zachor, 2007; Eldevik et al., 2010; Granpeesheh et al., 2009; Harris & Handleman, 2000; Remington et al., 2007; Zachor & Ben-Itzchak, 2011).

Despite its effectiveness, several constraints in the implementation of EIBI in clinical settings are expressed. In clinical settings a majority of children starts treatment later and a substantial subset (16%) has a lower IQ than recommended in research-based studies such as those conducted by Lovaas (1987) and Sallows and Graupner (2005; Mudford, Martin, Eikeseth, & Bibby, 2001).

Also, parents report barriers in implementing EIBI, including problems in recruiting and maintaining a suitable and qualified team and a lack of financial and physical resources, rules and regulations (e.g., laws and requirements related to autism, education, insurance coverage and individual education plans). These barriers in implementation lead to modified, and generally less favorable, programs in which children receive fewer hours of intervention and program supervision is infrequent and of lesser quality than recommended (Grindle, Kovshoff, Hastings, & Remington, 2009; Johnson & Hastings, 2002; Trudgeon & Carr, 2007).

Hence, in clinical practice the implementation of intensive EIBI is difficult. However, low intensity behavioral treatment (LIBT) may be provided as circumstances may limit the amount of treatment that a child can obtain. For example, in LIBT the costs may be lower, less professionals may be involved and less stress may be put on the family and child. Next to LIBT, additional treatment may be provided in a group setting, leaving time to play and socialize with peers (Eldevik, Eikeseth, Jahr, & Smith, 2006). Therefore, the effectiveness of LIBT and the conditions in which significant progress is found must be examined.

Since the literature base on LIBT is scarce (see for an example: Eldevik et al., 2006) and no literature on comprehensive treatment in the Netherlands existed, a study was conducted by Peters-Scheffer, Didden, Mulders, and Korzilius (2010) in which twelve children received on average 6.5 hours of behavioral treatment supplementing preschool services in the community. Eight months into treatment, the children who received behavioral treatment displayed significantly higher developmental ages and made more gains in adaptive skills than children who received regular treatment at a preschool. Regular treatment consisted of elements of TEACCH to structure and visualize the environment, incidental teaching, structured play, activities in a group setting, and maximally one hour individual therapy per week. No significant differences were found on autistic symptom severity and emotional and behavioral problems. Shortcomings of the study were that the sample size was relatively small, the assessment battery rather restricted, the intervention period relatively short and no measures on treatment fidelity were collected. Therefore, the present study is conducted to extend the literature on the effectiveness of LIBT in 40 children with ASD and ID. Children in both groups attend preschools or schools for children with ID and the children in the treatment group receive additional LIBT between four to ten hours per week. Over a period of two years standardized data are collected on developmental age, adaptive behavior, interpersonal relations, play, language development, autism severity, early social communication skills, maternal stress, behavioral flexibility, and emotional and behavioral problems. Also, data of the behavioral treatment program and on treatment fidelity are collected.

Aim of the study is to address the effect of adding in up to ten hours of one-to-one delivered LIBT into community-based (pre)school programs for children with ID on the outcomes of children with ASD and ID. In the treatment model used, treatment is provided by therapists and (pre)school teachers. In addition, teachers and parents are also trained in behavioral techniques and made responsible for generalizing the learned skills to the daily environment of the child.

9.2 Method

9.2.1 Participants and Setting

Forty children between 3;1 and 7;10 years (*M* = 62.52 months; *SD* = 16.96) participated. All children had previously been diagnosed with a DSM-IV or ICD-10 diagnosis of intellectual disability (ID) and autism or Pervasive Developmental Disorder, Not Otherwise Specified (PDD-NOS) by a clinician who was independent of the study. For all children, the diagnosis of ASD was confirmed by the Autism Diagnostic Observation Schedule (ADOS; Lord, Rutter, DiLavore, & Risi, 2006), and the Childhood Autism Rating Scale (CARS: Schopler, Reichler, & Rochen Renner, 2007), while the diagnosis of ID was confirmed by the Mullen Scales of Early Learning (MSEL; Mullen, 1995) and the Vineland Adaptive Behavior Scales (VABS; Sparrow, Balla, & Cicchetti, 1984). All instruments were assessed by the first author. No additional major medical diagnoses, such as severe epilepsy or visual problems, were reported in the sample.

The 20 children (18 male; 18 autism, 2 PDD-NOS) of the behavioral treatment group were recruited through letters distributed by Driestroom, an ABA center located in the eastern part of the Netherlands. Parents of 27 children gave their written consent and 20 were included in the study. Exclusion was based on cancellation of the treatment within 3 months after onset due to lack of funding (*n* = 2) or collaboration of the school the child attended (*n* = 1). One child was excluded, because he did not meet the criteria for ASD on the ADOS and CARS and three others were excluded as follow-up data after 24 months could not be obtained since parents moved abroad (*n* = 1) or could not be contacted (*n* = 2). The Wing Subgroups Questionnaire (WSQ; Castelloe & Dawson, 1993) indicated that 19 children had the aloof subtype, while one child had the passive subtype.

Participants in the control group were drawn from an ongoing longitudinal study (on the development of children with ASD and ID; e.g., Peters-Scheffer, Didden, & Korzilius, 2012; Peters-Scheffer, Didden, Green, Sigafoos, & Korzilius, 2013) in 100 children with ASD and ID by a reviewer independent of the study and received the community services that their family selected. The reviewer, blind of the exact aim of the study and the follow-up data of the children, selected 20 children to comprise the control group (18 male; 19 autism; 1 PDD-NOS). Participants in the control group were followed over the same period of time as children of the treatment group (i.e., 2007-2011). Pairs of children were individually matched on the following variables: pre-treatment chronological age (within 6 months; mean difference = -1.25; *SD* = 4.03), diagnosis (i.e., autism versus PDD-NOS) and IQ ratio (mean difference = 2.70; *SD* = 5.58) as measured by the mean age equivalence score/chronological age on the visual reception, fine motor, receptive language and expressive languages subscales of the MSEL (Mullen, 1995). Parents of the children in the control group were not actively seeking behavioral intervention and at intake the children were enrolled in preschools and public schools for children with ID. Fourteen children had the aloof subtype, four the passive subtype and two the active-but-odd subtype.

At treatment onset independent *t*-tests did not reveal significant differences between groups on chronological age ($t(38) = 0.14$; $p = .89$), developmental age ($t(38) = 0.04$; $p = .97$), ratio IQ ($t(38) = 0.09$; $p = .93$), adaptive behavior ($t(37) = 0.86$; $p = .39$), emotional and behavioral problems ($t(34) = .11$; $p = .91$), receptive language ($t(38) = 0.24$; $p = .81$), expressive language ($t(38) = 0.33$; $p = .74$), behavioral flexibility ($t(34) = 0.19$; $p = .85$), parental stress ($t(32) = 1.15$; $p = .26$), severity score of the ADOS ($t(38) = 1.63$; $p = .11$), and the CARS ($t(36) = 1.76$; $p = .08$). Table 1 shows the descriptives of the treatment and the control group.

9.2.2 Dependent measures

Most instruments were standardized instruments designed to assess a broad range of skills in both typically and atypically developing children and were assessed in a distraction free environment at the children's (pre)school. Additionally, at baseline, 12, and 24 months into treatment the VABS and the CARS were completed by the first author during a parent interview and a week before the interview was scheduled the parents completed the Child Behavior Checklist (CBCL; Achenbach & Rescorla, 2000), the Behavioral Flexibility Rating Scale - revised (BFRS-r; Green et al., 2007; Peters-Scheffer et al., 2008) and the Dutch-translated and revised version of the Parenting Stress Index (PSI; De Brock, Vermulst, Gerris, & Abidin, 1992).

Developmental age

The Mullen Scales of Early Learning (MSEL; Mullen, 1995) is a standardized developmental test for children from birth to 68 months of age and contains five subscales: gross motor, fine motor, visual reception, expressive language, and receptive language. The latter four scales were administered. The MSEL yields a single standard score (the Early Learning Composite). However, as the chronological age of some children was outside the standard age range for which the test was developed and some raw scores were below the lowest scores for which standard score are provided not all standardized scores could be calculated. In these cases, it is common practice for researchers and clinicians to rely on age-equivalents or ratio IQs (Bishop, Cuthrie, Coffing, & Lord, 2011). Therefore, developmental ages and ratio IQ were used in the analysis. Ratio IQ was calculated by the mean age equivalence score/chronological age on the visual reception, fine motor, receptive language and expressive languages subscales of the MSEL. Although not validated with the Dutch population, good internal, test-retest, and interrater reliability, as well as good convergent validity with the BSID are reported for the MSEL (Mullen, 1995). Using age equivalent scores (ratio IQs) of the MSEL, Bishop et al. (2011) found high agreement between the MSEL and standardized norm scores of the Differential Ability Scales.

Table 1. Means (SD) of the treatment group and the control group at baseline, after 12 months and after 24 months reported for cognitive functioning, adaptive behavior, social emotional development, early social communication, language, autism, emotional and behavioral problems, behavioral flexibility, and maternal stress.

Measure	Baseline		12-month assessment		24-month assessment	
	Treatment	Control	Treatment	Control	Treatment	Control
Cognitive functioning						
Developmental age	23.34 (7.32)	23.43 (6.34)	33.76 (8.57)	27.58 (8.87)	39.70 (11.99)	32.44 (11.55)
Ratio IQ	40.66 (20.07)	40.14 (18.27)	48.17 (19.31)	39.26 (18.85)	48.12 (19.71)	39.42 (19.89)
Visual reception	26.30 (8.47)	26.95 (5.46)	36.70 (9.19)	31.65 (8.16)	44.50 (14.39)	36.10 (11.99)
Fine motor	27.50 (6.20)	27.65 (6.43)	37.05 (9.68)	30.00 (8.37)	44.45 (14.66)	34.65 (10.37)
Receptive language	19.75 (9.26)	20.15 (8.57)	32.85 (11.88)	25.35 (11.28)	36.55 (11.63)	30.80 (13.27)
Expressive language	19.80 (8.32)	18.95 (9.12)	28.45 (10.72)	23.30 (11.43)	33.30 (12.02)	28.20 (14.03)
Adaptive Behavior [b]						
Composite	18.35 (3.41)	19.82 (4.71)	29.82 (9.89)	22.94 (5.39)	37.35 (13.05)	26.71 (9.84)
Communication	23.94 (7.64)	24.35 (9.80)	36.53(13.78)	28.59 (10.40)	43.71 (17.68)	32.35 (14.56)
Daily living skills	20.82 (6.12)	23.00 (9.26)	32.53 (9.31)	26.65 (9.72)	39.29 (11.13)	29.71 (12.15)
Socialization	19.76 (3.36)	22.88 (5.79)	32.47 (9.63)	26.47 (6.57)	39.35 (10.58)	29.71 (9.99)
Social emotional development [c]						
Interpersonal relationships	14.44 (5.19)	16.94 (6.50)	23.88 (6.47)	20.75 (5.74)	29.25 (9.60)	22.31 (6.59)
Play and leisure time	15.38 (5.82)	18.75 (5.87)	28.44 (8.02)	22.81 (5.34)	36.19 (12.97)	25.31 (7.58)
Early social communication						
Initiating Joint attention	7.43 (6.02)	7.64 (9.52)	7.21 (6.70)	5.71 (5.51)	11.50 (7.62)	11.21 (7.75)
Responding to Joint attention	96.60 (62.68)	118.80 (58.92)	101.81 (68.50)	121.73 (79.74)	84.70 (73.19)	95.31 (83.88)
Initiating requests	24.64 (4.77)	25.71 (4.50)	26.29 (7.49)	24.71 (5.73)	26.36 (5.21)	26.86 (4.75)
Responding to requests	69.16 (35.05)	70.07 (22.53)	84.56 (19.38)	81.31 (20.39)	88.21 (17.60)	89.33 (15.90)
Initiating social interaction	3.21 (1.48)	2.07 (1.49)	3.07 (1.54)	3.57(1.74)	3.79 (2.36)	3.29 (2.02)
Responding to social interaction	7.50 (2.74)	7.00 (2.91)	9.14 (3.63)	7.36 (3.23)	9.79 (3.98)	9.07 (3.45)
Receptive language	25.00 (4.48)	24.70 (3.21)	30.35 (7.87)	27.10 (5.35)	34.30 (10.54)	29.30 (7.42)
Expressive language	18.35 (6.72)	17.65 (6.64)	27.45 (9.89)	21.80 (9.83)	34.15 (14.54)	30.80 (15.12)
Autism; total score ADOS	17.00 (3.28)	15.45 (2.72)			12.05 (5.41)	15.15 (4.26)
Autism; total score CARS [a]	43.84 (4.30)	40.79 (6.20)	37.21 (3.95)	40.11 (5.65)	34.89 (3.62)	39.95 (4.62)
Emotional/ behavioral problems [d]	67.00 (26.38)	68.29 (33.47)	63.50 (21.31)	60.43 (26.92)	52.86 (23.52)	65.21 (32.62)
Behavioral flexibility [d]	10.00 (6.96)	11.29 (6.64)	9.86 (6.60)	10.36 (6.37)	9.14 (4.59)	11.14 (6.49)
Maternal stress [e]	78.38 (28.75)	95.08 (30.31)	80.85 (32.68)	83.00 (26.78)	71.38 (30.76)	87.08 (31.43)

Note. N = 40; except for [a] n =38, [b] n = 34; [c] n = 32; [d] n = 28; [e] n = 26.

Adaptive behavior

The Vineland Adaptive Behavior Scales (VABS; Sparrow et al., 1984; Dutch version by De Bildt & Kraijer, 2003) is a semi-structured parent interview that assesses adaptive behavior and, yields standard scores and developmental ages on three domains: communication, daily living skills, and socialization. Based on these scores the VABS also provides a composite score. Age equivalents in months were used in the analyses. In children and adolescents with ID, good construct validity and reliability of the Dutch version is reported (de Bildt, Kraijer, Sytema, & Minderaa, 2005).

Social-emotional development

The Vineland Social-Emotional Early Childhood Scales (SEEC) is a semi-structured interview administered to the parents to measure social-emotional functioning for children from zero to six years and provides developmental ages on the three domains: interpersonal relationships, play and, leisure, and coping skills (Sparrow, Balla, & Cicchetti, 1998). Age equivalents in months were used in the analyses. Although not validated with the Dutch population, internal consistency, test-retest, concurrent validity are adequate in an American sample (Sparrow et al., 1998).

Subtype, symptoms and severity of ASD

The Autism Diagnostic Observation Schedule (ADOS) is a semi-structured standardized interactive schedule that measures autism symptoms in communication, social interaction, play and stereotyped behavior. The ADOS consists of four developmentally sequenced modules of which only one is administered depending on the child's expressive language (Lord et al., 2006). The Childhood Autism Rating Scale (CARS; Schopler et al., 2007) is a 15-item behavioral rating scale developed to distinguish children with ASD from developmentally disabled children without ASD. The CARS was completed by the first author based on the assessments and parental report. A total score classifies children's behavior as having no autism (15-29.5), mild to moderate autism (30-36.5) and severe autism (37-60). The CARS is not validated with the Dutch population, but psychometric studies on the CARS yield good interrater and test retest reliability, high internal consistency and high convergent validity. The Wing Subgroups Questionnaire (WSQ; Castelloe & Dawson, 1993) is a questionnaire with 13 domains (e.g., communication, social approach, play, imitation, motor skills, resistance to change) that classifies children with ASD into one of three subtypes (i.e., aloof, passive, active-but-odd). For each domain, parents rate their child's behavior on a scale from 0 to 6 and a summary score is calculated for each subtype. The highest summary score is considered to indicate the child's subtype. Except to obtain wants or needs, children with the aloof subtypes rarely approach others spontaneously and they seem to reject social or physical contact. Although children with the passive subtype are also characterized by a lack of spontaneous social approaches to others, they engage in social interactions as long as the other person structures the interaction.

Children in the active-but-odd subgroup approach other socially, but in a peculiar, naive or one-sided manner (Castelloe & Dawson, 1993). O'Brien (1996) found moderate to good internal consistency and good inter rater reliability for the WSQ. However, the WSQ is not validated for the Dutch population.

Early social communication and language

The Early Social Communication Scales (ESCS; Mundy, Delgado, Block, Venezia, Hogan, & Seibert, 2003) is a videotaped semi-structured observational instrument to measure how the child initiates and responds to tasks involving joint attention, behavioral requests and social interaction. A set of toys and activities are used to elicit social and communicative behavior in an ecologically valid context (Mundy et al., 2003). The first author administered the ESCS following the manual and videotapes were scored by five raters, who were unaware of group status (e.g., treatment and control group), other scores of the participants and the exact aim of the study. Interrater reliability was assessed using videotaped data from 20% of the children at each time point and on the 6 subscales the average intraclass correlation between the paired rating was .62, suggesting good reliability (Cicchetti, 1994).

Receptive language was measured by the Peabody Picture Vocabulary Test (PPVT; Dunn & Dunn, 1997), while expressive language was measured by the vocabulary test of the Schlichting Test for Language Production (Schlichting, van Eldik, Lutje Spelberg, van der Meulen, & van der Meulen, 1995). On both language tests, age equivalents in months were used in the analyses. Both tests are validated in the Dutch population and have sufficient to good psychometric properties.

Emotional and behavioral problems

The Dutch version of the Child Behavior Checklist for the ages 1.5 to 5 years (CBCL; Achenbach & Rescorla, 2000) was completed by one of the parents to assess the parent's report on the child's behavioral and emotional problems of the last 3 months. Ratings on a 3-point scale to the 99 items on emotional and behavioral problems result in scores for seven behavioral categories: emotionally reactive, anxiety/depressed, somatic complaints, withdrawn, sleep problems, attention problems, and aggressive behavior. Sum scores were calculated on internalizing, externalizing and total behavior and higher scores represent more emotional and behavioral problems. Internal consistency, test-retest reliability, discriminative power, and convergent validity of the Dutch sample were adequate (Koot, Van Den Oord, Verhulst, & Boomsma, 1997).

Behavioral Flexibility

The Dutch version of the revised Behavior Flexibility Rating Scale (BFRS-R; Green et al., 2007) was used to assess severity of behavioral flexibility. One of the parents completed a three point Likert scale on 16 items referring to specific and unexpected events and changed routines that may prove problematic for children with ASD and related developmental disabilities.

The scale has a total scale and three subscales: flexibility towards objects, flexibility towards the environment and flexibility towards persons and psychometric proportions (i.e., internal consistency and intra-rater and inter-rater reliability) of the Dutch version are good to excellent (Peters-Scheffer et al., 2008). Higher scores on the BFRS-r represent more difficulties in behavioral flexibility.

Maternal stress

ASD has substantial impact on family functioning and parental stress and high levels of parenting stress may counteract the effectiveness of intervention (Osborne, McHugh, Saunders, & Reed, 2008) As parents for fill a crucial part in funding and facilitating the program and arrange the maintenance and generalization of new skills (Johnson & Hastings, 2002; Trudgeon & Carr, 2007), maternal stress was measured. Mothers completed the short version of the Dutch-translated and revised version of the Parenting Stress Index: the Nijmeegse Ouderlijke Stress Index – verkort (PSI; De Brock et al., 1992). Twenty-five parenting stress-related statements were scored on a 6-point Likert scale comprising a total stress score. Higher scores represented greater parenting stress. The NOSI-K has shown good psychometric properties (De Brock et al., 1992).

Interrater agreement

At baseline, 1 year and 2 years after treatment onset the children were evaluated by the first author who was independent of the treatment. For 57 assessments, data on inter rater reliability on the MSEL was obtained. A second rater, who was independent of the study and unaware of the group status, the exact aim of the study and the first examiner's ratings, used video recordings to score assessments of the MSEL. Intraclass correlation coefficient between the two raters was .99 (single measures). Also, interrater reliability was calculated on 24% of the assessments of the total scales of the VABS, 18% of the CARS, 29% of the CBCL, 31% of the PSI and 29% of the BFRS-R. Intra class correlations were .99, .95, .97, .99, and .99, respectively, suggesting excellent reliability. Finally, during the base-line or the second follow-up in 50% of the assessments a second VABS was completed using a (pre)school teacher of the child as informant (n = 32). The intraclass correlation coefficient between the parent and teacher ratings of .96, indicates excellent reliability, which may suggest that parental report was minimally biased (Cicchetti, 1994).

9.2.3 Intervention

Both behavioral and eclectic treatment took place in public preschools or schools for children with ID. As is usual in the Netherlands, preschools and schools were open from 8.30 am – 3.30 pm, Monday through Friday. Classes were composed from six to twelve children with comparable disabilities and developmental level, as well as two to three teachers. During individual treatment sessions the child worked alone with his or her therapist in a separate room.

When not in these sessions the child was mainstreamed with his or her classmates. Incidentally, a child of the behavioral treatment group was shadowed by the therapist. However, this always occurred within reported treatment hours.

The behavioral treatment group

The behavioral treatment group was provided with one-to-one intervention for 4 to 10 hours per week over a period of 2 years. The treatment procedure and curriculum were based on those initially described by Lovaas (Leaf & McEachin, 1999; Lovaas, 2003). Procedures based on the principles of ABA (e.g., systematic reinforcement, stimulus control, discrete trial teaching, task analysis, chaining, generalization, functional behavior assessment) were used and treatment progressed gradually and systematically from relatively simple tasks to more complex skills (Leaf & McEachin, 1999; Hayward, Gale, & Eikeseth, 2009; Lovaas, 2003). No aversives were employed. Treatment was implemented in the child's natural environment (e.g., home, community and (pre)school) to increase the likelihood of the child using the skills acquired (Lovaas, 1993).

Each child was provided with an Individual Education Plan (IEP) based on ongoing analysis of current strengths and needs and a logbook, in which information about the programs was provided and progress was reported. Every program description comprised information on the name of the program, target behavior, instructions procedures (e.g., prompting, shaping), used materials, expected or appropriate responses of the child, criteria for mastering the skill and the next program to be taught. After each treatment session a detailed record in the logbook was completed by the therapist, providing information about the treatment (therapist, date, duration), trial-by-trial data, target behavior, progress of the child and next stage to be taught. When necessary, data on challenging behavior (e.g., aggressive, stereotypic, self-injurious behavior) were collected.

A program consultant and three to seven therapists were assigned to every child and provided behavioral treatment for 48 weeks per year. Children received on average 4.98 hours one-to-one treatment per week (*SD* = 1.45; range: 1.32-7.11)[1]. Main responsibilities of the program consultant were supervision to the therapists, and meetings with the team, parents, teachers, school staff and other professionals involved with the child. The program consultant also completed several clinical administrative tasks related to the case, such as programming, task analysis and functional assessment and conducted several one-to-one sessions with the child. Although no Board Certified Behavior Analysts were involved, all program consultants had an MsC in Special Education or Psychology and took several courses (at university level) in ASD, ID, Functional Analysis, and ABA. In addition, an experienced clinical MSc in special education with extensive experience with ABA, ASD and ID was available for consultation.

[1] Reported treatment hours are hours actually received by the child and taking in account absence of the child due to personal circumstances as illness and vacations. On request of the parents and the preschool the hours of one child were kept deliberately low.

She assisted the consultants in setting up the programs and solved complex situations regarding challenging behavior of the children and coaching of therapists. On average, program consultants spent 4.39 hours per week on each child.

Therapists were employed part-time while they were enrolled in university (mostly special education or psychology) or worked at the (pre)school as teacher or staff member. The therapists received a comprehensive training in ABA and worked under close supervision of the program consultants. During two training sessions of three hours verbal and written information about ABA, ASD, ID and teaching procedures was provided and under supervision of the behavioral consultant, therapist analyzed video fragments of training sessions. Furthermore, procedures based on the principles of ABA were modeled and practiced during role-play with feedback. After the training, therapists worked alongside experienced therapists.

Therapists, teachers, staff members and parents participated in semi-monthly meetings in which the child's progress, current en new programs were discussed and (additional) ABA principles were taught. Based on video recordings of the treatment, feedback on teaching procedures and progress was provided. In these meetings, parents and teachers received specific guidelines to generalize and maintain new learned skills and were trained and stimulated to use ABA strategies during daily activities.

Table 2 displays the critical features of each participant's curriculum as recommended by Lechargo and Carr (2008). Due to a lack of cooperation of schools or difficulties in obtaining funding, nine children received one year behavioral treatment instead of two years. Although one child switched to another ABA provider after one year, all remaining children received approximately two year behavioral intervention.

The control group

Information on the treatment of the control group was provided by the children's parents and during a short interview of the teacher or staff member, responsible for the daily care of the child. Children in the control group received standard care for children with ASD and ID in the Netherlands and attended a (pre)school for children with ID. In most (pre)schools the environment and tasks were highly structured as outlined by the TEACCH methodology (Schopler, Mesibov, & Baker, 1982). Intervention comprised a mixture of interventions, including elements from the TEACCH-program, Picture Exchange Communication System (PECS; Bondy & Frost, 2002), individualized speech therapy and sensory integration therapy. Each child's program comprised individualized goals and objectives in language, play, social, emotional, sensory-motor, pre-academic and daily living domains. Intervention was based on the child's characteristics and staff's expertise and preferences. A clinical psychologist or special educator (MSc) provided information and advice to the parents and the professional team.

Treatment integrity and procedural fidelity

Several formal and informal efforts were made to ensure treatment integrity in the treatment group. Each child had his or her own individual education plan in which criteria, procedures and task analyses of each target behavior were described in detail. In addition, treatment was described in a manual and in general programs.

Table 2. Overview of programs of the individual children in the LIBT group.

	1	2	3	4	5	6	7	8	9	10	11	12	13	14	15	16	17	18
Compliance, attention, and non-verbal communication	x	x	x	x	x	x	x	x	x	x	x	x	x	x	x	x	x	x
Imitation	x	x	x	x	x	x	x	x	x	x	x	x	x	x	x	x	x	x
Matching	x	x	x	x	x	x	x	x	x			x	x	x	x	x	x	
Categorization	x	x	x	x						x	x	x	x	x		x	x	
PECS	x		x	x	x	x	x		x	x	x	x		x	x	x	x	x
Fine motor skills	x			x	x	x			x		x	x	x		x	x		
Gross motor skills	x			x	x				x									
Receptive language	x	x	x	x	x	x	x	x	x	x	x	x	x	x	x	x	x	x
Expressive language	x	x	x	x		x				x	x		x	x	x	x	x	
General knowledge	x			x						x	x		x			x	x	
Memory										x	x		x					
Play	x	x		x	x	x		x	x		x	x	x	x	x	x	x	
Adaptive behavior		x	x	x	x	x			x	x	x	x	x	x		x	x	
Emotions: labeling and imitation	x	x			x						x						x	
Age level (pre) academic skills	x	x	x	x	x	x		x	x	x	x	x	x	x	x	x	x	
Social Interaction/ communication		x	x	x		x				x	x		x	x		x	x	

Note. Due to computer problems, the individual education programs of two children were not available.

All therapists were trained in how to employ the intervention techniques central to the study. Furthermore, procedures and target behavior were explained and practiced during two weekly to monthly meetings. Therapists videotaped their sessions and were observed by the program consultant. Through analysis of the videotapes and feedback during treatment sessions the implementation of the treatment was frequently supervised to ensure that the treatment was being implemented in a reliable manner and carried out as intended.

Procedural fidelity was measured by three independent reviewers using an observation schedule, developed by the first author, three programs consultants and a special educator (see for more information Peters-Scheffer, Didden, Korzilius, & Sturmey, submitted). Using event recording, the observer scored the correct or incorrect implementation of the components of a Discrete Trial (e.g., discriminative stimulus, prompt, response time, feedback, error correction, positive practice trial and the intertrial interval), omission or commission of these components and when applicable procedural errors (e.g., incorrect stimulus, multiple stimuli, incorrect prompt [location, level], absence of contingent feedback). For each video fragment, the percentage of correct implementation was calculated for all trials and each component. Three percent of the treatment sessions were scored and for each trainer at least one video fragment was included (range: 1-11). The three reviewers concluded that in all sessions treatment was based on ABA and that procedural fidelity was 90.3%.

9.3 Results

9.3.1 Statistical significance

Developmental age and IQ

To test for differences between the treatment and the control group on developmental age and IQ two repeated measure designs with developmental age or IQ as within-subject factor and treatment (i.e., treatment as usual and LIBT) as between-group factor were conducted. As for both developmental age and IQ, Mauchly's test indicated that the assumption of sphericity was violated ($\chi^2(2) = .51$; $p < .001$ for developmental age; $\chi^2(2) = .74$; $p < .01$ for IQ), degrees of freedom were corrected using Greenhouse-Geisser estimates of sphericity ($\varepsilon = .67$ for developmental age; $\varepsilon = .79$ for IQ).

Although developmental age increased over time in both treatment and the control group, ($F(1.35, 51.10) = 113.60$; $p < .001$), the increase was significantly larger in the treatment group, ($F(1.35, 51.10) = 11.04$; $p = .001$). In both groups assessments between baseline and the first follow-up ($F(1, 19) = 113.06$; $p < .001$ for the treatment group; $F(1, 19) = 18.08$; $p < .001$ for the control group) and between the first and second follow-up ($F(1, 19) = 33.57$; $p < .001$ for the treatment group; $F(1, 19) = 18.00$; $p < .001$ for the control group) were significant. For both groups, MANOVAs and ANOVAs with repeated measures on the subscales of the MSEL showed significant effects for developmental age over time. Repeated contrasts displayed that only the progress of the control group on the fine motor scale between the baseline-assessment and the first follow-up was not significant ($F(1, 19) = 3.72$; $p = .07$).

In addition, there was a significant effect of treatment on IQ (ε = .79; F(1.59, 60.36) = 13.70; p < .001). ANOVAs with repeated measures conducted for the separate groups showed a significant effect of time in the treatment group (F(2, 38) = 20.35; p <.001) with significant increases in IQ between baseline assessment (F(1, 19) = 33.85; p < .001) and the IQ remaining relatively stable between follow-up assessments after 12 and 24 months (F(1, 19) = .002; p = .97). However, no effect of time in the control group was found, (F(1.53, 29.16) = 0.29; p = .69), indicating no differences in IQ between assessments. Descriptives are displayed in Table 1.

Adaptive behavior

First, a repeated measures design with adaptive behavior over time as within factor and treatment as group factor (Greenhouse-Geisser: ε = .70) was used to test for differences between the treatment and the control group on the total scale of the VABS. As expected, both groups made significant gains in adaptive skills (F(1.40, 44.70) = 59.47; p < .001), but in the treatment group the gains were significantly larger (F(1.40, 44.70) = 13.58; p < .001).

Since the subscales of the VABS (i.e., communication, daily living skills, and socialization) were correlated (r = .28 to r = .85) a multivariate variance analysis with repeated measures was used to analyze the differences on the subscales. Results indicated an improvement over time in both groups on all sub-scales, (Λ = .23; F(6, 27) = 15.32; p < .001); however, the progress of the treatment group was significantly larger (Λ = .56; F(6,27) = 3.52; p = .01). ANOVAs with repeated measures (Greenhouse-Geisser) revealed significant interaction effects in all domains (communication: ε = .75; F(1.49, 47.74) = 7.48; p < .004), daily living skills, ε = .74; F(1.48, 47.47) = 15.57; p < .001), and socialization, ε = .85; F(1.70, 54.38) = 16.40; p < .001), representing a significantly larger progress in the treatment group (see Table 1). Repeated contrasts conducted for both groups indicated significant progress between the baseline and the first follow-up assessment and between the first and the second follow-up on all domains.

Social emotional development

Since the subscales of the Vineland-SEEC (i.e., interpersonal relations and play skills), used to measure social emotional development, were correlated (r = .70 to r = .87) a MANOVA with repeated measures was used to analyze the differences on the subscales. A significant effect over time was found (Λ = .23; F(4, 27)= 22.69; p < .001). However, the progress of the treatment group was significantly larger, (Λ = .52; F(4, 27) = 6.15; p = .001) than the progress of the control group.

Separate ANOVAs with repeated measures (Greenhouse-Geisser) also revealed significant interaction effects on interpersonal skills (ε = .64; F(1.29, 38.55) = 10.23; p = .001) and play skills (ε = .78; F(1.57, 46.96) = 17.17; p < .001) and in both groups repeated contrasts indicated significant progress over time in all periods on all subscales.

Language and early social communication

A repeated measures design with receptive language over time as within factor and treatment as group factor revealed a significant interaction effect on receptive language (F(2,76) = 3.35; p = .04), representing a significantly larger progress in the treatment group (see Table 1). Repeated contrasts conducted for both groups indicated significant progress between the baseline and the first follow- up assessment and between the first and the second follow-up.

Although expressive language improved over time in both groups (ε = .70; F(1.40, 53.18) = 51.21; p < .001), no significant interaction effect on expressive language (F(1.40, 53.18) = 1.50; p = .23) was found. Repeated contrasts showed significant changes between the baseline and the first follow-up assessment and between the first and the second follow-up for both groups.

Next, a MANOVA with repeated measures was conducted with the subscales of the ESCS (i.e., initiating and responding to joint attention, interaction and responding to behavioral requests and initiating and responding to social interaction) as within factors. Although no significant effect between both groups appeared (Λ = .75; F(6, 21) = 1.16; p = .36), there was a significant effect of time, (Λ = .29; F(12, 15) = 3.08; p = .02). Separate univariate variance analyses with repeated measures on the scales revealed a significant effect of time on initiating joint attention (F(2, 52) = 8.08; p = .001), responding to behavioral requests (Greenhouse-Geisser: ε = .80; F(1.61, 41.75) = 10.05; p = .001) and responding to social interaction (F(2, 52) = 4.77; p = .01), but non-significant effects on responding to joint attention (F(2, 52) = 1.17; p = .32), initiating behavioral requests (F(2, 52) = 0.65; p = .53) and initiating social interaction *(*F(2, 52) = 2.51; p = .09).

Subtype, symptoms and severity of ASD

First, an ANOVA with repeated measures was conducted on autism severity as measured with the CARS. A significant interaction effect between time and treatment was revealed, (F(2, 72) = 18.20; p < .001). ANOVAs with repeated measures conducted for both groups showed a significant effect of time for the treatment group (ε = .77; F(1, 54, 27.65) = 55.77; p < .001), but not for the control group, (F(2, 36) = 0.35; p = .71). For the treatment group, repeated contrasts indicated significant decreases in symptom severity between the baseline and the first assessment (F(1, 18 = 48.45; p < .001) and between both follow-up assessments (F(1, 18 = 15.02; p = .001).

Next, an ANOVA with repeated measures was conducted on the total scale of the ADOS and a MANOVA with repeated measures on the subscales communication and social interaction, yielding significant interaction effect on the total scale (F(1, 38) = 12.54; p = .001) and the subscales, (Λ = .75; F(2, 37) = 6.20; p < .01). ANOVAs with repeated measures conducted for the separate groups show that the decrease over time holds for both the communication (F(1, 19) = 11.34; p < .01), and the social interaction scale (F(1, 19) = 20.23; p < .001) in the treatment group, but not in the control group (communication: F(1, 19) = .07; p = .80; social interaction: F(1, 19) = 0.17; p = .69).

Emotional and behavioral problems

First, a repeated measures design was used to test for differences between the treatment and the control group on the total scale of the CBCL. As expected, emotional en behavioral problems changed significantly over time ($F(2, 52) = 3.67$; $p = .03$). There was also a marginally significant interaction effect of treatment over time ($F(2, 52) = 3.03$; $p = .06$), indicating a larger decrease in emotional and behavioral problems in the treatment group.

Separate ANOVAs with repeated measures for both groups revealed a significant effect over time for the treatment group ($F(2, 26) = 4.49$; $p = .02$), but not for the control group ($F(2, 26) = 1.78$; $p = .19$). In the treatment group, repeated contrasts showed significant changes between the first and the second follow-up ($F(1, 13) = 7.50$; $p = .02$), but not between the baseline and the first follow-up assessment ($F(1, 13) = 0.41$; $p = .53$).

This marginally significant effect was not confirmed with a MANOVA with repeated measures on the subscales of the CBCL, which showed no effect of time ($\Lambda = .85$; $F(4, 23) = 1.05$; $p = .15$) nor of treatment ($\Lambda = .84$; $F(4, 23) = 1.09$; $p = .16$) on emotional and behavioral problems.

Behavioral flexibility

First, a repeated measures design with the total scale over time as within factor and a multivariate repeated measure design with the subscales of the BFRS-r over time as within factors were used to test for differences between the treatment and the control group on behavioral flexibility. No significant changes in behavioral flexibility over time were found on the total scale (Greenhouse-Geisser; $\varepsilon = .81$; $F(1.62, 42.20) = 0.56$; $p = .66$), nor on the subscales ($\Lambda = .85$; $F(6, 21) = .61$; $p = .72$).

Maternal stress

Reported stress levels are comparable to scores of the Dutch clinical norm group of the PSI (De Brock et al., 1992) and to stress scores of mothers of children with ASD and ID reported in another study (Peters-Scheffer et al., 2012). Scores are considerable higher than the mean maternal stress scores of the non-clinical norm-group of the PSI. To test for differences in stress between mothers in the treatment and the control group, a repeated measures design with maternal stress over time as within factor and treatment as between group factor was used. There was no significant effect of time ($F(2, 48) = 1.51$; $p = .23$) and treatment on maternal stress, ($F(1, 24) = 1.16$; $p = .29$).

9.3.2 Effect sizes

Effect sizes were calculated for each outcome measure by dividing the difference in change score of the experimental en the control group by the original standard deviation of the sample (Cohen, 1988). Large effect sizes were found for change scores on developmental age (Cohen's $d = 1.09$), adaptive behavior (Composite: Cohen's $d = 1.74$; Communication:

Cohen's *d* = 1.41; Daily Living Skills: Cohen's *d* = 1.62; Socialization: Cohen's *d* = 2.61), autism severity (ADOS: Cohen's *d* = 1.51; CARS: Cohen's *d* = 1.50), play (Cohen's *d* = 2.42), interpersonal relations (Cohen's *d* = 1.57), and receptive language (Cohen's *d* = 1.22). Small effects were found for ratio IQ (Cohen's *d* = 0.40), maternal stress (Cohen's *d* = 0.33), and expressive language (Cohen's *d* = 0.40).

Next, the effect sizes were compared with the average effect sizes calculated in a meta-analytic study conducted by Peters-Scheffer et al., (2010) on EIBI and the effect sizes of a study investigating LIBT (Eldevik et al., 2006). The large effect sizes found on receptive language and the composite score and the communication and socialization domains of the VABS are consistent with other studies on EIBI (receptive language: Cohen's *d* = 2.91; composite: Cohen's *d* = 0.91; communication: Cohen's *d* = 1.32; socialization: Cohen's *d* = 1.49) and LIBT (receptive language: Cohen's *d* = 2.71; composite: Cohen's *d* = 1.25; communication: Cohen's *d* = 1.15; socialization: Cohen's *d* = 1.21).

However, effect sizes on (ratio) IQ, expressive language and the VABS domain Daily Living Skills are inconsistent between studies. While Peters-Scheffer et al. (2010) and Eldevik et al. (2006) found large effect sizes on (ratio) IQ (Peters-Scheffer et al.: Cohen's *d* = 2.00; Eldevik et al.: Cohen's *d* = 3.55) and expressive language (Peters-Scheffer et al.: Cohen's *d* = 1.10; Eldevik et al.: Cohen's *d* = 1.90), this study found small effect sizes. In contrast, in this study large effect were found on the daily living skills domain of the VABS, while Peters-Scheffer et al. found a moderate effect (Cohen's *d* = 0.68) and Eldevik et al. a small effect (Cohen's *d* = 0.39) .

9.3.3 Clinical significance

Reliable and clinical change

In addition to the group effects, individual change was analyzed by calculating individual reliable change and clinical significance. First, reliable change was determined by calculating the Reliable Change Index (RCI; Jacobson & Truax, 1991) with the following formula, in which x_1 and x_2 are the pre- and post treatment scores of the participant, s_1 the standard deviation for the pre-treatment score and r_{xx} the internal consistency or test-retest reliability of the measure used:

$$RCI = \frac{X_2 - X_1}{\sqrt{2(s_1\sqrt{1 - r_{xx}})}}$$

Table 3 displays the standard error of difference. Clinical significance is usually defined as returning to normal functioning (Jacobson, Roberts, Berns, & McGlinchey, 1999). Given the severity of ID and ASD in the present study's sample this criterion seems to be too stringent, and as a result clinical significant change (CSC) was calculated using the following formula, in which M_1 is the average pretreatment score and s_1 the standard error of the pretreatment score: $CSC = M_1 \pm (2 \times s_1)$.

The minimum score for obtaining clinical significance for each variable is reported in Table 3, as well as an overview of the number of children obtaining reliable and clinical significant improvement, reliable but no clinical significant change, clinical but no reliable significant change and no reliable and clinical significant change. More children of the treatment group obtained clinical and reliable significance on developmental age, adaptive behavior, interpersonal relationships, play and leisure time, receptive language, expressive language, autism severity on the ADOS and the CARS, and responding to social interaction than in the control group. However, in the control group more children obtained clinical and reliable significance on the CBCL, and maternal stress. The frequency of children obtaining clinical and reliable significance was equal between groups on ratio IQ, behavioral flexibility, initiating and responding to joint attention, initiating and responding to behavioral requests and initiating social interaction.

Table 3. Frequencies of children in the treatment and the control group obtaining (1) clinical and reliable significance, (2) reliable but no clinical significance, (3) clinical, but no reliable significant change, and (4) no reliable and clinical significant change.

Variable	Reliable Change	Clinical Significant Change	Treatment group				Control group			
	SE_{diff}	CL	1	2	3	4	1	2	3	4
Developmental age	2.87	> 36.89	0	3	2	15	0	0	1	19
Ratio IQ	8.04	> 78.29	10	10	0	0	6	6	0	8
Adaptive Behavior [b]	1.44	> 26.70	13	4	0	0	5	6	1	5
Interpersonal relationships [c]	3.19	> 26.75	11	5	0	0	3	4	1	8
Play and leisure time [c]	3.38	> 28.81	11	5	0	0	0	7	3	6
Initiating JA [c]	4.18	> 21.60	1	2	1	12	1	3	1	11
Responding to JA [c]	31.12	> 221.64	0	3	0	13	0	2	0	14
Initiating requests [c]	16.51	> 34.14	0	0	3	13	0	0	1	15
Responding to requests [c]	28.98	> 127.14	0	1	0	15	0	0	0	16
Initiating SI	0.67	> 5.71	3	6	0	7	3	3	0	10
Responding to SI [c]	1.46	> 12.79	3	7	0	6	1	5	2	8
Receptive language	1.20	> 32.56	10	7	0	3	5	5	0	10
Expressive language	3.36	> 31.20	11	3	0	6	8	6	0	6
ASD Total (ADOS)	1.13	< 10.07	7	7	1	5	3	3	1	13
ASD Symptom severity (CARS) [a]	1.84	< 31.49	3	14	0	2	1	4	0	14
Emotional/ behavioral problems [d]	8.55	< 12.86	0	6	1	8	1	3	1	10
Behavioral flexibility [d]	2.89	<-2.31	0	2	0	13	0	1	0	14
Maternal stress [c]	8.89	< 29.80	0	5	1	7	1	3	3	7

Note. N = 40; except for [a] n =38, [b] n = 34; [c] n = 32; [d] n = 30; [c] n = 26; SEdiff = Standard error of differences (i.e., indicating the spread of change scores if no effect appeared); CL = Clinical level (i.e., minimum score for obtaining clinical significance): JA = joint attention; SI = social initiations.

Post-treatment diagnoses of ASD and ID

Furthermore, data were collected on post-treatment diagnosis and school placement. Forty-five percent of the treatment group (eight autism; one PDD-NOS) versus 80% of the children of the control group (15 autism; one PDD-NOS) had similar pretreatment and post treatment diagnoses. In the control group, 20% of the children were classified with autism at pre-treatment, but received a post treatment diagnosis of PDD-NOS versus 45% of the children in the treatment group. Also, 10% of the children of the treatment group, at pretreatment classified with PDD-NOS and autism, were classified as not autistic (versus none of the children in the control group).

Based on their IQ scores, 45% children of the treatment group (one 'none'; one mild ID; three moderate ID, four severe ID) versus 95% children of the control group (one none, two mild, five moderate, ten severe; one profound) had similar levels of ID at pre treatment and post treatment. However, in the treatment group 55% of the children reduced their level of ID (three from profound to severe; five from severe to moderate; two from moderate to mild; one from borderline to none) versus only 5% in the control group (one from profound to severe).

School placement

Since school placement can be seen as a real life measure of cognitive and social functioning (Kazdin, 1993), information on school placement was obtained. At baseline, all children attended a preschool or school for children with ID. After two years, one child (5%) of the treatment group was placed in regular education, one child (5%) attended a school for children with language delays, nine children (45%) a preschool for children with ID and nine children (45%) a school for children with ID, while in the control group nine children (45%) visited a preschool for children with ID, one child (5%) a preschool for children with developmental delays and ten children a school or children with ID (50%).

9.3.4 Predictors of progress

To determine whether child variables are associated with the progress during the behavioral treatment, Pearson's correlations were calculated between the child variables at pre treatment (i.e., chronological age, developmental age, IQ, adaptive behavior, autism severity, language, early social communication, emotional and behavioral problems, behavioral flexibility) and the progress children made between the baseline and second follow-up (i.e., after 2 years) on developmental age, IQ, adaptive behavior, expressive and receptive language, autism severity (on both the ADOS and the CARS), emotional and behavioral problems, behavioral flexibility and maternal stress.

Results are displayed in Table 4 and suggest that hours of treatment per week and developmental age, IQ, adaptive behavior, play skills, and receptive language at pretreatment are related to the progress of children with ASD and ID following two years of LIBT.

Table 4. Pearson correlations between (pre) treatment variables and progress on developmental age, IQ, adaptive behavior, language, autism, emotional and behavioral problems, behavioral flexibility and maternal stress of the treatment group.

Variables	Progress between baseline and the second follow-up									
	DA	IQ	AB	RL	EL	AU-C	AU-A	EBP	Flex	Stress
Hours treatment per week	.62**	.34	.24	.35	.47*	.28	-.56*	-.24	.34	-.03
Chronological age	-.08	.25	-.13	-.09	-.19	-.03	.37	-.18	-.43	-.80**
Developmental age	.61**	.25	.57*	.36	.69**	.13	-.46*	-.05	-.13	-.17
IQ	.49*	-.02	.42	.28	.64**	.15	-58**	.02	.13	.29
Adaptive Behavior	.55*	.29	.57*	.39	.48*	.16	-.40	-.01	-.22	-.51
Interpersonal relationships	.31	.18	.44	.29	.49*	.27	-.18	-.13	-.05	.59*
Play and leisure time	.54*	.36	.77**	.32	.55*	.06	-.45	-.03	-.17	-.27
Initiating Joint attention	.16	-.04	.27	-.14	.17	-.25	-.09	-.16	-.58*	.34
Responding to Joint attention	.29	.04	.33	-.12	.18	.31	-.38	-.04	.11	-.22
Initiating behavioral Requests	.07	-.06	.11	.00	-.04	-.20	-.07	.25	.13	.23
Responding to requests	.38	.20	.22	.22	.35	-.48*	-.49*	-.03	.36	-.52
Initiating social interaction	-.23	-.16	.07	-.25	-.12	-.10	-.03	.37	-.59*	-.17
Responding soc. interaction	.52*	.28	.35	.19	.18	-.03	-.42	-.01	-.16	.05
Receptive language	.57**	.38	.44	.59**	.45*	.08	-38	-.23	-.22	-.20
Expressive language	.58**	.18	.50*	-.05	.33	-.11	-.40	.33	-.06	.24
ASD Total (ADOS)	-.07	-.14	-.06	.26	.18	.44	-.11	-.04	.68*	-.26
ASD severity (CARS)	-.26	-.07	-.41	.00	-.20	-.24	.14	.11	-.39	.43
Emotional/ beh. problems	.07	.21	-.11	-.19	-.14	-.25	.09	.17	-.68*	-.14
Behavioral flexibility	.07	.03	.27	-.11	-.09	-.25	-.12	.32	.59*	-.31
Maternal stress	.24	.21	.02	.25	.10	-.28	.11	.04	-.67*	-.23

*Note. * p < .05;** p < .01; DA = progress on developmental age in months , IQ = progress on IQ, AB = progress on adaptive behavior in months, RL = progress on receptive language in months, EL = progress on expressive language in months, AU-C = difference in severity of ASD on the CARS, AU-A = difference in severity of ASD on the ADOS, EBP = difference in emotional and behavioral problems, flex = difference in behavioral flexibility, stress = difference in maternal stress.*

9.4 Discussion

Children receiving LIBT for 4 to 10 hours per week were compared at pre treatment, 1 year and 2 years into treatment to a matched control group of children receiving standard (pre) school interventions. Children exposed to the LIBT were higher in IQ at the time of follow-up than children who received regular treatment (a mean difference of about 9 points). Also, the children in the LIBT group outperformed the children in the standard care group on developmental age, adaptive behavior, interpersonal skills, play and receptive language. At follow-up, fewer characteristics of autism and behavioral problems were reported in the treatment group. However, no significant differences were found on expressive language, behavioral flexibility and maternal stress between groups. As compared to the progress between baseline and the first follow-up, less progress between the first and the second follow-up was seen, which may be the result of the nine children who terminated treatment after one year. However, some other studies also reported a lower learning rate in the second year of treatment as compared to the first year (e.g., Eikeseth, Klintwall, Jahr, & Karlsson, 2012). These lower learning rates reported in the second year of treatment may perhaps indicate a ceiling effect.

In the treatment group, 50% of the children showed reliable and clinical progress on developmental age, 50% on receptive language and 77% on adaptive behavior versus 30%, 25% and 39% of the children in the control group. Approximately a third of the children in the treatment group showed a reliable and clinical decrease in autism symptomology on the ADOS versus 15% of the children in the control group. These results are consistent with previous studies on LIBT in which the treatment group outperformed the control group on developmental age (Eldevik et al., 2006; Peters-Scheffer et al., 2010), adaptive behavior (Peters-Scheffer et al.) and receptive language (Eldevik et al.). In contrast to Eldevik et al., no significant differences between groups were found on expressive language. However, children in our experimental group made comparable progress on expressive language as the children in the study of Eldevik et al. Hence, the absence of a difference between groups, might be explained by the progress shown by our control group. Their expressive language scores progressed with approximately seven standardized points, while in the study of Eldevik et al. standardized scores of the control group decreased.

The differences between groups reported in the present study are substantially smaller than reported in studies in which treatment was implemented more intensively and in which more supervision was provided. This is to be expected as these variables predict a favorable treatment outcome (e.g., Eikeseth et al., 2009; Granpeesheh et al., 2009; Lovaas, 1987; Sallows & Graupner, 2005). Alternatively, the more modest results could be due to child characteristics such as chronological age and IQ at treatment onset. Regardless of the treatment program, the prognosis for children with an higher IQ is better (Dietz, Swinkels, Buitelaar, Van Daalen, & Van Engeland, 2007; Eaves & Ho, 2004) and although Eikeseth et al.'s studies (2002; 2007) show that older children may benefit from EIBI programs, a lower chronological age at intake seems to be associated with better outcome (Granpeesheh et al., 2009).

However, many children with ASD do not have an opportunity to participate in an EIBI program before 4 years of age. As research in community settings (Mudford et al., 2001) shows that children are entering EIBI programs with lower IQ and higher chronological age than recommended, research in this older and more impaired population is needed.

As is common in applied research, the present study has several methodological limitations. First, no fully randomized controlled trial design was used and assignment to treatment group was based on availability of LIBT at the (pre)school the child visited. Based on the data files, a reviewer blind to follow-up scores, identified an IQ-, severity of ASD-, CA-matched child for each child receiving EIBI. This resulted in two groups who were very similar on key dependent measures before treatment began and were followed over the same period of time. In this way it was possible to form a group of children who met participation criteria and received other services than EIBI, providing an opportunity to collect data in real life circumstances.

ASD has a substantial impact on family function and parental stress. However, as in most studies in the area of ASD, this study primarily focused on child outcome and no data - except on maternal stress - were collected on family well-being and demographics (e.g., availability of respite care, (in)formal social support, family coping strategies, depression). Collecting such data might give a more complete picture of the results as parental stress is related to child outcome (Osborne et al., 2008) and parents for fill a crucial part in funding and facilitating the program and arrange the maintenance and generalization of new skills (Johnson & Hastings, 2002; Trudgeon & Carr, 2007). Unfortunately, no data were collected on the recommendations about maintenance and generalization given to parents and staff during LIBT, nor on the time and the quality parents and staff worked on maintaining and generalizing skills. As there might be a relationship between how much time parents and staff spend working on maintenance and generalization and overall treatment outcome for their child, future studies should address this issue.

Although estimated long-term savings of EIBI seem to overweigh costs of the program (Chasson, Harris, & Neely, 2007; Jacobson, Mulick, & Green, 1998; Peters-Scheffer, Didden, Korzilius, & Matson, 2012), obtaining funding for the program was highly challenging for many parents and consequently nine children had to terminated the program before they completed the 24 months. Hence, the recruiting and retention of participants proved to be difficult and the sample size of the study remained therefore relatively small.

Future research should focus on how the individual LIBT should be supplemented to provide a full time program. For example, by developing complementing group programs for children with ASD and by designing guidelines for parents and staff members to learn and generalize skills to children with ASD in the daily situation. As suggested by Sheinkopf and Siegel (1998), intense involvement of parents, teachers and staff members in LIBT could compensate for less intensive treatment, as parents use their teaching techniques learned while providing treatment to their child and extend the therapy outside the formal treatment sessions. More research is needed to determine the extent to which parents and teacher use their teaching skills to teach children skills in a more informal setting.

In sum, results of this study show that when practical reasons (e.g., costs, availability of professionals) or concerns regarding the impact of EIBI on the child and family limit the intensity of the treatment, LIBT supplemented with (pre)school services can be an effective alternative. Despite the fact that the children in the treatment group were exposed to fewer treatment hours, had a lower IQ and a higher chronological age at intake than the children in most previous studies (Peters-Scheffer et al., 2011; Virués-Ortega, 2010), they showed statistically and clinically significant gains on developmental age, adaptive behavior, interpersonal relation, play and receptive language when compared to children receiving treatment as usual in the Netherlands.

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Part III
Health Care Conditions Required for Early Behavioral Treatment

Chapter 10

Cost Comparison of Early Intensive Behavioral Intervention and Treatment as Usual for Children with Autism Spectrum Disorder in the Netherlands

Research in Developmental Disabilities, 2012, 33, 1763-1772

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Abstract

Early Intensive Behavioral Intervention (EIBI) may result in improved cognitive, adaptive and social functioning and reductions in autism severity and behavioral problems in children with Autism Spectrum Disorder (ASD). For a subset of children, normal functioning may be the result. However, due to the intensity (20-40 hrs per week for 2-3 years with a low child staff ratio) implementation costs are high and can be controversial. Estimated costs for education, (supported) work and (sheltered) living for individuals with ASD in the Netherlands are applied in a cost-offset model. A compelling argument for the provision of EIBI is long term savings which are approximately € 1,103,067 from age 3 to 65 years per individual with ASD. Extending these costs to the whole Dutch ASD population, cost savings of € 109.2 to € 182 billion have been estimated, excluding costs associated with inflation.

10.1 Introduction

Autism Spectrum Disorder (ASD) is one of the most common developmental disorders. The disorder is characterized by impairments in communication and social interaction, by repetitive behaviors and by limited areas of interest (American Psychiatric Association, 2000; Baron-Cohen, Scott, Allison, Williams, Bolton, Matthews, & Brayne, 2009). In 50 to 80% of the individuals with ASD an intellectual disability (ID) is also present (Goldberg Edelson, 2006). Although individuals with ASD present great variability in severity and clinical picture, their prognosis without treatment is generally poor. ASD is a chronic disability and due to the unique social and communicative difficulties the majority of individuals involved requires professional care throughout their lives (Billstedt, Gillberg, & Gillberg, 2005; Mordre, Groholt, Knudsen, Sponheim, Mykletun, & Myhre, 2012). As more people are being diagnosed with ASD and require specialized services (Wing & Potter, 2002; Yeargin-Allsop, Rice, Karapurkar, Doernberg, Boyle, & Murphy, 2003), the costs of public health and social welfare programs are increasing. Järbrink and Knapp (2001) estimated the lifetime costs (including costs such as family expenses, medication and daycare) to care for an individual with ASD in Britain more than € 2.5 million, excluding the costs associated with typical child rearing.

Currently, early intervention based on applied behavior analysis (EIBI) is considered the treatment of choice for children with ASD (Eikeseth, 2009). Although EIBI programs vary slightly in their approach, all programs are characterized by the following essential features: (1) systematic use of behavior analytic principles, (2) treatment is comprehensive, (3) systematic development from simple to more complex skills with a transfer to natural settings, (4) functional assessment of individual behavior and individualization of goals and instructional procedures, (5) the use of scientific methods to evaluate the effects of treatment, (6) early age of treatment onset, (7) a low child staff ratio, and (8) involvement and training of parents and significant others.

Usually, treatment is implemented intensively (i.e., 20 – 40 hours per week) for a long period of time (i.e., two years or more; Eikeseth, 2009; Green, Brennan, & Fein, 2002; Leaf & McEachin, 1999; Lovaas, 2003), although less intensive EIBI has also shown positive outcomes (e.g., Eldevik, Eikeseth, Jahr, & Smith, 2006; Peters-Scheffer, Didden, Mulders, & Korzilius, 2010).

Four of the five meta-analyses included in an overview of Reichow (2012) concluded that EIBI is an effective intervention strategy for many children with ASD and results in increased cognitive, social and communication skills and reductions in challenging behavior. However, great variability in outcome within and between studies is seen, with some children making rapid and remarkable progress, while other children's gains are limited (Eikeseth, 2009; Peters-Scheffer, Didden, Korzilius, & Sturmey, 2011; Reichow & Wolery, 2009). These differences in outcome are influenced by child and family factors (e.g., age of treatment onset, co-morbid conditions and pre-treatment IQ, autism symptom severity and language) and treatment characteristics (e.g., treatment intensity, treatment duration, treatment quality, and intensity and quality of supervision; Allen & Warzak, 2000; Ben-Itzack & Zachor, 2007; Davis, Smith, & Donahoe, 2002; Eikeseth, Hayward, Gale, Gitlesen, & Eldevik, 2009; Eldevik et al., 2006; Granpeesheh, Dixon, Tarbox, Kaplan, & Wilke, 2009; Lovaas, 1987; Peters-Scheffer et al., 2010; Smith, Eikeseth, Klevstrand, & Lovaas, 1997; Smith, Groen, & Wynn, 2000; Smith, Klevstrand, & Lovaas, 1995).

Due to treatment intensity and duration, EIBI is an expensive treatment (albeit highly effective), but only a few studies have been conducted regarding the financial costs and benefits of EIBI to children with ASD. Based on the outcome of the studies by Lovaas (1987) and McEachin, Smith, and Lovaas (1993), Jacobson, Mulick, and Green (1998) estimated the costs and benefits of EIBI for children with ASD in Pennsylvania. In their model, in which EIBI outcome ranged from regular education without support (47%) and less intensive special education (42%) to intensive special education (11%), cost savings ranged from $ 187,000 to $ 203,000 per child for ages 3 to 22 years and from $ 656,000 to $ 1,082,000 per child for ages 3-55 years. Authors concluded that the estimated savings outweigh the differences in initial treatment costs for EIBI. Translating outcome into a cost dichotomization (i.e., successfully or unsuccessfully mainstreamed in regular education), Chasson, Harris, and Neely (2007) estimated that with a success rate of 72%, $ 208,500 per child would be saved by the state of Texas across 18 years of education with EIBI. Motiwala, Gupta, and Hon (2006) used more conservative efficacy rates and therefore estimated savings lower than Jacobson et al. (1998) and Chasson et al. (2007) between 34.479 to 53.720 Canadian Dollars per individual.

Cost-effectiveness studies by Jacobson et al. (1998) and Chasson et al. (2007) have estimated costs exclusively based on best outcome studies (i.e., Lovaas, 1987; McEachin et al., 1993; Sallows & Graupner, 2005) and although Motiwala et al. (2006) used more conservative efficacy rates, none of the studies included less favorable outcomes studies published after Lovaas (1987).

In these studies children received fewer hours than recommended (e.g., Anderson, Avery, DiPietro, Edwards, & Christian, 1987), program supervision was infrequent or of less quality (e.g., Bibby, Eikeseth, Martin, Mudford, & Reeves, 2002) or children had a higher pretreatment chronological age (e.g., Eikeseth, Smith, Jahr, & Eldevik, 2002; 2007) or lower IQ (e.g., Smith et al., 1997) than in the Lovaas study (1987). It is unlikely that in community-based programs children obtain the same outcome as in the Lovaas study (Mudford, Martin, Eikeseth, & Bibby, 2001). Therefore, by including studies with less favorable outcome our study may provide a more realistic estimate of the outcome of community based EIBI and the potential costs and savings of EIBI in the Netherlands.

The present study was designed to provide a cost-offset analysis of EIBI relative to treatment as usual for children with ASD in the Netherlands. An estimate of the effects of EIBI on financial costs was investigated. To date only one study regarding the effectiveness of EIBI has been published in the Netherlands (Peters-Scheffer et al., 2010). Therefore, potential cost savings including different outcomes of EIBI were based on a number of international studies and presented per child and extended to the ASD population in the Netherlands.

10.2 Cost-offset analysis

To analyze the cost and benefits of EIBI in the Netherlands, efficacy rates of EIBI and treatment as usual based on meta-analytic studies were determined first. Next, costs were identified for individuals with ASD from age 3 to 65 years including costs for education, work and living and total costs were calculated for individuals with ASD who received EIBI or treatment as usual including different outcomes (i.e., normal functioning, reduced dependency, or dependency). Lastly, avoided costs for the Netherlands through the provision of EIBI were calculated per child and for the Dutch ASD population.

10.2.1 Efficacy rates

As still relatively few children in the Netherlands receive EIBI and pre-treatment and/or post treatment data of most children is lacking, efficacy rates of both the EIBI group and the treatment as usual group were based on published literature. Following Jacobson et al. (1998), children were categorized into three groups according to their level of functioning. The first group was comprised of children who achieve normal functioning, participate in regular education with little or no support and who are vocationally productive adult workers. The second group consisted of children who participate in less intensive special education and evince reduced dependency throughout their lives, while the third group requires continuing specialized and intensive educational and adult services.

Behavioral intervention

Given the controversy (e.g., Schopler, Short, & Mesibov, 1989) regarding the reported efficacy of the Lovaas study (1987) and several replications (e.g., Sallows & Graupner, 2005), the efficacy figures used in our study are based on the results reported in six meta-analytic studies regarding EIBI that were recently published (i.e., Eldevik, Hastings, Hughes, Jahr, Eikeseth, & Cross, 2009; Makrygianni & Reed, 2010; Peters-Scheffer et al., 2011; Reichow & Wolery, 2009; Spreckley & Boyd, 2009; Virués-Ortega, 2010). Common measures reported in the included studies are IQ, adaptive behavior, language and school placement. Although subject to parental advocacy and school policy, school placements seem to be the best real world efficacy measurement of academic and social competence (Kazdin, 1993) and are therefore used in our analysis.

Characteristics of the studies included in the meta-analyses are displayed in Table 1. As the study of Matos and Mustaca (2005) was in Spanish, it was excluded from the analysis. Also, the control groups of the studies of Ben-Itzack, Lahat, Burgin, and Zachor (2008) and Harris, Handleman, Gordon, Kristoff, and Fuentes (1991) were excluded, as they were not comprised of children with ASD. Sixteen of the studies reported school placements. However, the follow-up studies by Lovaas (1987; i.e., McEachin et al., 1993) and Eikeseth et al. (2002; i.e., Eikeseth et al., 2007) used the same participants as the original studies and were therefore combined with the original papers. The actual treatment hours for the children in the low intensity group of Lovaas (1987) and Smith et al. (2000) are not reported. As these children received minimal EIBI treatment (i.e., less than 10 hours per week), school placements of these groups of children were excluded from the analyses. Therefore, 14 studies were included in the analysis. There were 292 children with a mean chronological age of 41.45 months (30.2-66.3) and a mean pretreatment IQ of 60.17 (50.5-83.0). On average, children received 32.54 hours of treatment per week (20-40) for 27.01 months (12-36). After treatment, 29% were placed in regular treatment, 34% were placed in less intensive special education and 37% were placed in special education.

Treatment as usual

Six studies also report school placements of children in a control group who had a mean chronological age of 42.88 months (33.2-65.0) and mean pretreatment IQ of 62.73 (59.4-65.2). After eclectic treatment or treatment as usual, 11% of the children were placed in regular treatment, 8% were placed in less intensive special education and 81% were placed in special education. These rates are roughly in line with studies on outcome of adolescents and adults with autism (e.g., Levy & Perry, 2011).

Table 1. Characteristics of the studies included in the meta-analysis (i.e., Eldevik, Hastings, Hughes, Jahr, Eikeseth, & Cross, 2009; Makrygianni & Reed, 2010; Peters-Scheffer, Didden, Korzilius & Sturmey, 2011; Reichow & Wolery, 2009; Spreckley & Boyd, 2009; Virués-Ortega, 2010).

	EIBI group								Control group								
	Participants			Treatment		Outcome			Participants				Treatment		Outcome		
Study	*N*	*CA*	*IQ*	*Hrs*	*Du*	*1*	*2*	*3*	*N*	*CA*	*IQ*	*Type*	*Hrs*	*Du*	*1*	*2*	*3*
Anan et al. (2008)	72	44	52	15	3												
Anderson et al. (1987)	14	43	57	15-25	12-24	0	31	69									
Baker-Ericzen et al. (2007)	158	49	75		3												
Ben-Itzchak et al. (2008)	44	27	71	45	12												
Ben-Itzchak and Zachor (2007)	25	27	51	>35	12												
Bibby et al. (2002)	66	45	45	30	32	5	53	42									
Birnbauer & Leach (1993)	9	38		19	22				5	33	45			22			
Boyd and Corley (2001)	22	41	62	30-40	23	0	41	59									
Cohen et al. (2006)	21	30		35-40	36	48	33	19	21	33	59	E		36	5	0	95
Eikeseth et al. (2002; 2007)	13	66	62	28	31	38	0	62	12	65	65	E	29	33	8	0	92
Eldevik et al. (2006)	13	53	41	13	20				15	49	47	E	12	21			
Harris and Handleman (2000)	27	49	59	35-45		41	0	59									
Harris et al. (1991)	9	50	68	35-45	12	0	89	11									
Howard et al. (2005)	29	31	59	25-40	14				16	37	54	E	25-30	13			
									16	35	60	E	15	15	5	48	48

	EIBI group								Control group								
	Participants			Treatment		Outcome			Participants				Treatment		Outcome		
Study	*N*	*CA*	*IQ*	*Hrs*	*Du*	*1*	*2*	*3*	*N*	*CA*	*IQ*	*Type*	*Hrs*	*Du*	*1*	*2*	*3*
Lovaas (1987) and	19	35	53	40	24+	47	42	11	21	<42	>40	U		24+			
McEachin et al. (1993)	19	41	46	<10	24+	0	42	58	16	43	65	E	27	26	0	0	100
Magiati et al. (2007)	28	38	83	33	24	0	82	18	20	43	51	E	13	9			
Reed et al. (2007a)	12	40	56	30	9				16	38	53	P	9	9			
Reed et al. (2007b)	14	43	57	30	9-10												
	13	41	49	13	9-10		0										
Remington et al. (2007)	23	36	61	26	24	74	43	26	21	38	62	U		24	48	0	52
Sallows and Graupner (2005)	13	35	51	38	48	48		9									
	10	37	52	31	48		20	50									
Sheinkopf and Siegel (1998)	11	34	63	27	16	30			11	35	62	U	11		0	0	100
Smith et al. (1997)	11	36	28	30	24												
	10	38	27	<10	24		13	60									
Smith et al. (2000)	15	36	51	25	33	27	21	79									
	13	36	51		24	0	25	25									
Weiss (1999)	20	42		40	24	50											

Note. CA= average age in months; Hrs= average number of hours per week of treatment; Du= average number of months of treatment; 1= percentage of children placed in regular education with no support, minimal support (e.g., part-time support with shadow tutor, fading the shadow tutor) or unknown support; 2= percentage of children with regular school placement with full-time individual support or part-time EIBI, less intensive special education (e.g., for children with communication impairments or mild ID), mixture of special education and regular education placement, private school with small classes; 3= percentage children receiving one-to-one-treatment and special education (e.g., autism specific schools, generic special needs schools, self-contained classes); U= treatment as usual; E= Eclectic treatment; P= Portage program. Since Sallows and Graupner (2005) report educational placement for the parent-directed and clinic- directed group together, placements are reported for the two groups together.

However, the rates noted above are more pessimistic than rates provided by the Dutch Association for Autism (Nederlandse Vereniging voor Autisme, 2008). They reported that 36% of the adults lived independently (18% with a partner) and 10% lived independently but with support. Twenty-nine percent had a paid job (25% with sufficient income to provide in their living), 35% worked voluntary (e.g., voluntary job, traineeship, sheltered work with support), 13% worked in a sheltered environment, and 13% participated in structured daytime activities. About 20% of the adults had no structured day care or (supported) work.[1] Half of the adults received a security income from the Dutch Government (so-called WAJONG) as major source of income and the majority (74%) indicated they needed professional support in conducting their work with 55% actually receiving this support. Therefore, emulating Motiwala et al. (2006), also the most positive figures (Freeman, 1997) were included to estimate costs for children who receive standard care in the Netherland. In Freeman's study, 25% of the participants attained normal functioning, while 25% evinced semi-independent living, and 50% were very dependent at adulthood.

10.2.2 Costs

Costs were calculated from age 3 to 65 years for individuals with ASD who received EIBI or treatment as usual including different outcomes (e.g., normal functioning, reduced dependency, or dependency). ASD can be reliably diagnosed between two and three years of age (Kleinman et al., 2008) and costs after 65 years of age are difficult to estimate due to health costs, retirement and pension. Also, some researchers assume a higher mortality rate among individuals with ASD (Mouridsen, Brønnum-Hansen, Rich, & Isager, 2008; Picket, Xiu, Tuchman, Dawson, & Lajonchere, 2011). Estimated costs in Euros are displayed in Table 2.

Education

In the Netherlands, typically developing children receive, on average, 8 years of primary education, four to six years secondary education, and four years of intermediate or higher vocational education or university. Attending school is compulsory from the age of five, but most children start primary school when they are 4 years old (student staff ratio 14.6 to 1) and graduate between 20 to 22 years of age (Minne, Webbink, & van der Wiel, 2009; OCW, 2008).

Approximately 5% of the children in primary school attend special education (Centraal Bureau voor de Statistiek, 2009), divided into less intensive special education (student staff ratio 5.9 to 1; Minne et al., 2009) and intensive special education (student staff ratio 3.5 to 1; Minne et al., 2009).

[1] Some of the participants mentioned multiple jobs.

Table 2. Indication of costs of an individual with ASD with various outcomes between 3 to 65 years of age in the Netherlands.

Costs	*Treatment as usual*	*EIBI*
Normal range of functioning		
EIBI (weekly 32.54 hrs, 27.01 months)		€100,000
8 years primary education	€32,000	€32,000
5 years secondary education (average)	€42,500	€42,500
4 years college or university (average)	€40,000	€40,000
Total from 3-65 years	**€114,500**	**€214,500**
Reduced dependency in adulthood		
EIBI (weekly 32.54 hrs, 27.01 months)		€100,000
2 year preschool	€58,000	
8 years less intensive primary education	€71,200	€71,200
4 years secondary school	€44,000	€44,000
2 years intermediate vocational education	€20,000	€20,000
15 years suppl. aid to dependent children	€12,687	€12,687
47 years of security income	€583,452	€583,452
Living and working with support	€1,882,075	€1,882,075
Total from 3-65 years	**€2,671,414**	**€2,713,414**
Dependency in adulthood		
EIBI (weekly 32.54 hrs, 27.01 months)		€100,000
2 years preschool for children with ID	€58,000	
14 years primary/ secondary school for children with ID	€257,600	€257,600
15 years suppl. aid to dependent children	€12,687	€12,687
47 years of security income	€583,452	€ 583,452
Living and working with intensive support and intensive care	€3,354,317	€3,354,317
Total from 3-65 years	**€4,266,056**	**€4,308,056**

Less intensive special education is attended by students with learning difficulties, while intensive special education serves children with visual or hearing impairments, children with severe communication impairments (including hearing problems), children with ID and children with psychiatric disorders. Some children with ASD are enrolled in special classes, but other children with ASD are mainstreamed with a mix of children with other psychiatric disorders and/or developmental disabilities such as Down syndrome, learning disabilities and ID. Children with ASD who have average or higher intellectual and linguistic ability have increasingly joined regular education in the Netherlands. Sometimes these children received additional support provided by special education staff financed by the Ministry of Education, Culture and Science (Student-linked budget; LGF). Costs for this alternative method are equivalent to the cost for special education.

After primary special education, the majority children with learning difficulties or disabilities visit secondary special education (59%; Dienst Uitvoering Onderwijs, 2010). Most of the remaining children (7%) receive secondary education with a strong emphasis on practical skills (LWOO and PRO; Dienst Uitvoering Onderwijs, 2010).

In these approaches additional support is provided. After completing PRO, the majority of the students starts working (supported, regular or in combination with education BBL; Heijnens, 2009), while 90% of the children completing LWOO continues their education at an intermediate vocational education for three or four years (Oosterling, Brouwer, & Nijman, 2010). Therefore, two years of intermediate vocational education were included in the calculations.

All education costs were derived directly from websites and reports of the Dutch Government (OCW, 2008) or from studies conducted for the Dutch Government (i.e., Centraal Bureau voor de Statistiek, 2009; Oosterling et al., 2010). In 2007, annual costs for primary education per child were € 4,000 in regular education, € 8,900 in special education and € 18,400 in intensive special education (Minne et al., 2009). In 2008, annual costs per child were € 8,500 for regular secondary education, with € 11,000 for LWOO and PRO and € 18,400 for special secondary education. Annual cost for intermediate vocational education, higher vocational education and university (excluding costs for research and development) are € 10,000, € 9,200 and € 9,200 (Centraal Bureau voor de Statistiek, 2009; Oosterling et al., 2010) and were set at € 10,000 for the purpose of this analysis.

To our knowledge, no exact numbers of children with ASD receiving educational services in the Netherlands exist. However, the Dutch Association for Autism (Nederlanse Vereniging voor Autisme, 2008) conducted a study in 2,275 individuals with ASD (i.e., 43% PDD-NOS; 28% autism; 26% Asperger and 3% multiple complex developmental disorder) and found that approximately 39% of the children with ASD in primary school visited regular education. The remaining 23% received less intensive special education, while the other 35% received intensive special education. During secondary school, 53% of the children with ASD received special education. Approximately half of the children received additional support, while an additional 20% needed extra support. Almost 6% of the children with ASD had no educational services.

Child costs

No specific autism related costs were assumed for normal-functioning children, but parents of (semi-) dependent children between 3 and 18 years old receive a compensation in the costs of raising a child with a disability, which is € 845.80 per year (Sociale Verzekeringsbank, 2011). In addition, in the Netherlands, (parents of) individuals with ASD can apply for a client-linked budget to fund additional therapies (e.g., speech therapy, physiotherapy), as well as daytime activities and care. Although children with ASD use a variety of effective and ineffective therapies (e.g., Green et al., 2006; Thomas, Morrissey, & McLaurin, 2007), no exact numbers exist of the amount and characteristics of children with ASD that receive such additional therapies and (specialized) services. Therefore, therapy costs were omitted from the analysis since we assumed that costs were balanced across all children and offer little to the analysis.

Adult costs

Besides education, no specific (autism related) costs were assumed for normal-functioning adults after 18 years of age, as these adults provide their own income and pay taxes. In the Netherlands, individuals with developmental disabilities with an onset before 17 years of age are eligible for security income (WAJONG), which is 75% of the minimal wage. In 2011, the minimal monthly wage varied between € 653 and € 1435.20 depending on the chronological age of the individual (Uitvoeringsinstituut Werknemersverzekeringen, 2011).

Costs for adult living and day programs or supported work were obtained from the Dutch government (Zorgzwaartepakketten Sector GZ; Bureau HHM, 2010). Based on client characteristics (e.g., level of ID, care, motor functioning, problem behavior, nursery needs, psychosocial functioning and adaptive behavior), clients were categorized into one of the eight intensity levels of care and support. Each intensity level corresponds to a daily budget to provide for living expenses (including care and support) and for a structured day program, consisting of adapted and supported work or an alternative program to replace schooling or working with an emphasis on the maintenance or development of cognitive and adaptive skills and the regulation of behavior.

Those with reduced independence were assumed to be categorized into the second intensity level, in which individuals were able to function relatively independently in a sheltered environment. Staff needs to supervise and provide minimal help with adaptive skills and psychosocial functioning. Some support is needed with reading, writing, calculations, daily routines, decision-making and problem solving. In general, clients present no behavioral and psychiatric problems. According to Bureau HHM (2010) daily costs were € 109.71.

Needs of the dependent group seem in accordance with the fifth intensity level of care in which clients receive intensive comprehensive support and care. Individuals in this group were only able to function in society and engage in social relationships with support and sometimes staff needed to regulate behavioral problems. Care, support and supervision regarding communication, psychosocial functioning and daily living skills were provided 24 hours per day. Daily costs were estimated at € 195.53 (Bureau HHM, 2010).

EIBI

Program costs fall into five general categories (i.e., personnel, capital assets, transportation, materials and supplies and miscellaneous; Escobar, Barnett & Goetze, 1994), which were used to estimate EIBI program costs. Next to a home-based model of EIBI, also center-based EIBI was provided in the Netherlands and estimated costs were assumed to be representative for both. Although EIBI programs vary slightly in intensity, structure and supervision, most programs provide 20 to 40 hours of intervention, which is implemented by 5 to 7 therapists generally for 2 to 6 years with the average child requiring 3 years of treatment.

Intensity and duration of the program and intensity and quality of supervision are related to outcome (Granspeheeh et al., 2009; Reichow & Wolery, 2009) and have a significant impact on costs (Escobar et al., 1994). The intensity of the treatment was estimated by calculating the average intensity (*M* = 32.54 hours) and duration (*M* = 27.01 months) of the included studies to determine efficacy (see 2.1.2). EIBI programs are supervised by a competent clinician with knowledge of and experience in implementing advanced learning principles in different types of learners. Average amount of supervision was estimated at 5 hours per month (Eikeseth et al., 2009) with monthly costs set at € 500. In sum, for the total duration of the program children received on average of 3.809 hours of EIBI, while therapists and parents received on average of 135 hours of consultation. Employment wages are based on the average cost per hour for staff as described in the collective bargaining agreement 2009-2011 (Vereniging Gehandicaptenzorg Nederland, 2010) and set at € 13.84 per hour. Professional time was estimated at € 66,217 for the total duration of the program. Additional annual costs for the program (e.g., travel time, expanses, materials) were estimated at € 15,000 per child. Therefore, total program costs sum up to approximately € 99,967 and to calculate cost-offset, EIBI was set at € 100,000 for the total program.

10.2.3 Cost-offset analysis

As seen in Table 3 costs can be broken down into different child outcome and for children who receive treatment as usual and EIBI. Although large differences are visible in the outcome percentages between children receiving EIBI and treatment as usual as reported in the meta-analytic studies (i.e., 29% normal functioning, 34% reduced dependency, and 37% dependent vs. 11% normal functioning, 8% reduced dependency, and 81% dependent), the percentages of children attaining normal outcome are comparable between the EIBI group and the Freeman study (i.e., 25% vs. 29%). However, after EIBI relatively more children obtained reduced dependency compared to children who received treatment as usual in the Freeman study (i.e., 34% vs. 25%).

Per child savings

Next, we broke down the costs into differential child outcomes and compared the costs to those of children who received treatment as usual. As no expectations can be made per child, analyses were conducted as a function of the percentage (i.e., 11% normal functioning, 8% reduced dependency, and 81% dependent) resulting in an average estimate of costs of € 3,681,813 per individual. Results were also compared to Freeman (1997; average estimated costs: € 2,829,507 per individual; i.e., 25% normal functioning, 25% reduced dependency, and 50% dependent). Table 3 shows the gains and losses for each outcome of EIBI with children who received treatment as usual as baseline.

Table 3. Differential child outcomes of EIBI compared to the cost to those of children who received treatment as usual.

	Total costs 3-65 years with EIBI	Projected costs savings (Control groups)	Projected costs savings (Freeman, 1997)
Independent (29%)	214,500	3,467,313	2,615,007
Reduced dependent (34%)	2,713,414	968,399	116,093
Dependent (37%)	4,308,056	-626,243	-1,478,549
Average per child	2,578,746	1,103,067	250,761

Avoided costs for the Netherlands

Next, the costs and benefits were extended to all individuals with ASD living in the Netherlands. In 2010, the population of the Netherlands consisted approximately of 16.500.000 individuals, of which 3.928.334 were younger than 20 years old and each year approximately 182.000 children are born (Centraal Bureau voor de Statistiek, 2011). The number of individuals with ASD in the Netherlands is unknown (Gezondsheidsraad, 2009). According to estimates published in the international scientific literature, the prevalence of ASD is currently 60 to 100 per 10.000 (Baron-Cohen et al., 2009; Fernell & Gillberg, 2010). Hence, approximately 99.000 to 165.000 individuals with ASD live in the Netherlands, of which 23.570 to 39.283 are between 0 and 20 years of age. Each year approximately 1.092 to 1.820 children with ASD are born. When average child savings (€ 1,103,067) were applied to the prevalence rates, savings are estimated at € 1,204,549,164 to € 2,007,581,940 per birth year cohort and € 25,999,289,190 to € 43,331,780,961 when all children under 20 years had received or will receive EIBI. Approximately € 109,203,633,000 to € 182,006,055,000 can be avoided by society when all individuals with ASD received EIBI. Using the more conservative child savings based on Freeman (1997; € 250,761), savings are estimated at € 273,831,012 to € 456,385,020 per birth year cohort and € 5,910,436,770 to € 9,850,644,363 when all children under 20 years had received or will receive EIBI. In total, roughly € 24,825,339,000 to € 41,375,565,000 can be avoided by society when all individuals with ASD received or will receive EIBI.

10.3 Discussion

The current study provides a cost comparison of EIBI relative to treatment as usual for children with ASD in the Netherlands. Based on efficacy rates published in meta-analytic studies on EIBI (Eldevik et al., 2009; Makrygianni & Reed, 2010; Peters-Scheffer et al., 2011; Reichow & Wolery, 2009; Spreckley & Boyd, 2009; Virués-Ortega, 2010) estimated costs avoided for society by the provision of EIBI on a large scale are approximately € 1,103,067 per child and, extended through the school-aged population (i.e., children with ASD between 0 and 20 years), € 26 to € 43.3 billion. As concluded earlier by Jacobson et al. (1998), estimated savings seem to outweigh the costs of EIBI, which are approximately € 100,000 per program.

As with other cost-benefits studies (e.g., Chasson et al., 2007; Jacobson et al., 1998), our analysis is based on several assumptions and therefore provides an indication of future costs and savings, excluding costs associated with inflation. In addition, changes in treatment, development of new treatments, changes in public health services and funding may influence the costs and benefits of interventions including EIBI. In this analysis we assumed that children receive their diagnosis before the age of three and consequently initiate EIBI at three years of age. However, many children receive their diagnosis at a later age (Nederlandse Vereniging voor Autisme, 2008) and consequently start treatment at higher chronological age, which may result in different (i.e., less favorable) outcomes and cost savings.

In this analysis, costs regarding education, security income and (supported) living and working were included. However, costs associated with having a child with ASD are not limited to these costs. For example, children with ASD may require specialized childcare and extracurricular activities. Often parents must reduce their work hours and family activities are limited as a result of raising a child with ASD (Sharpe & Baker, 2007). Järbrink, Fombonne, and Knapp (2003) found that parents of children with ASD had weekly out-of-pocket costs of €76 to € 116, excluding expenses for education, early intervention, health services, medication and income losses (on average € 268 per week).

Since we used meta-analytic studies to estimate effectiveness, effectiveness and cost savings of EIBI were more conservative than reported in other studies, in which percentages of successfully mainstreaming children between 47% and 72% were used to calculate the costs and benefits (Chasson et al., 2007; Jacobson et al., 1998). Contrary to Jacobson et al. and Chasson et al. who assumed that most children with ASD would remain in special education throughout childhood, the effectiveness of treatment as usual in this study was based on the results of the control groups included in the meta-analytic studies in which 11% of the participants obtained normal functioning, 8% reduced-dependency, and 81% were dependent in adulthood. In addition, results were compared to Freeman (1997), who suggested that 25% of the individuals with ASD attained normal functioning, 25% reduced dependency, while 50% were very dependent in adulthood. This prognosis seems more positive than reported in other studies (e.g., Billstedt et al., 2005; Mordre et al., 2012) and estimated savings in this analysis (€ 250,761 per child) would likely underestimate the costs that would be saved.

Several studies exploring the predictors of successful EIBI have been published and found that amongst others treatment intensity, treatment duration, intensity and quality of supervision, and pretreatment chronological age, IQ and autism severity are related to treatment outcome (e.g., Ben-Itzack & Zachor, 2007; Eikeseth et al., 2009; Granpeesheh et al., 2009; Lovaas, 1987). Currently, estimates of the individual contributions of these predictors to the efficacy of EIBI and consequently the costs savings are imprecise and cannot be integrated in cost-offset analyses yet. However, as research on these predictors is mounting, future cost-offset studies should include these predictors to provide a more accurate estimate of cost savings.

Since few children in the Netherlands are engaged in EIBI programs, efficacy data had to be based on the existing literature. It is yet unknown to which extent these data can be generalized to the Dutch situation. Although research seems to confirm our assumptions (Nederlandse Vereniging voor Autisme, 2008), it is uncertain to which extent school placement scores predict later social and economic functioning (e.g., employment in the future, independent living). In addition, school placement does not account for within-group differences. Although EIBI in several children may not result in changes in school placement, adaptive behaviors (e.g., toilet training, independency in eating, dressing) are learned reducing their care needs and dependency.

As research on EIBI is expanding, current treatment programs may be improved and better decisions may be made about whether a child should receive EIBI. As not all children may respond positively to EIBI, further research should explore effective treatment options. This strategy may result in greater savings as additional costs due to ineffective EIBI could be avoided and more children could engage in effective treatment and consequently be mainstreamed into regular education. However, even after treatment, a substantial subset of children retains impairments related to ASD (e.g., impairments in social interaction and communication and persistent patterns of restricted and stereotyped behavior). Future research should determine how teachers, professionals and parents address the needs of these children.

While researchers have shown that in a substantial subset of children with ASD, EIBI can result in lasting improvements in IQ scores and adaptive behavior (Reichow, 2012), many children with ASD are still receiving controversial and unsupported treatments (Green et al., 2006). However, to make EIBI generally available to children with ASD in the Netherlands, several difficulties in implementing EIBI need to be eliminated. First, appropriate funding is required for identifying children with ASD at an early age and implementing the EIBI program. In addition, professionals and parents need to be educated about the costs and benefits of early interventions including EIBI and therapists and consultants need to be properly trained in applying EIBI as the quality of treatment is related to treatment outcome. These changes in policy may improve the quality of life of children with ASD and result in substantial cost savings to society.

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Chapter 11

Therapist Characteristics Predict Discrete Trial Teaching Procedural Fidelity

Pending minor revisions accepted for publication

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Abstract

Early intensive behavioral intervention is generally effective for children with autism spectrum disorder but is associated with variability in treatment outcome and quality of treatment delivery may contribute to this. This study examined the relationship between therapist personality, attitude towards individuals with a disability and perceived relationship between therapist and child on procedural fidelity. Discrete Trial Teaching was provided at a pre-school for children with intellectual Disabilities. Seventy DTT sessions between 22 therapists and 35 children were videotaped and analyzed. Data on therapist's attitude towards individuals with a disability, therapist's personality traits, and perceived relationship between therapist and child were also collected. Procedural fidelity was high and significantly related to therapist's attitude towards individuals with a disability, therapist's openness to experience, and perceived relationship between therapist and child. Therapists with high procedural fidelity tended to have a more positive attitude towards individuals with disabilities on the cognitive dimension, a more negative attitude towards individuals with disabilities on the affect dimension, lower levels of openness to experience, and perceived the relationship between themselves and the child as less positive.

11.1 Introduction

Early intervention based on applied behavior analysis (EIBI) is currently recognized the treatment of choice for children with autism spectrum disorder (ASD; Eldevik, Hastings, Hughes, Jahr, Eikeseth, & Cross, 2009; Peters-Scheffer, Didden, Korzilius, & Sturmey, 2011; Rogers & Vismara, 2008). A frequent approach used in EIBI is Discrete Trial Teaching (DTT), which is particularly important in the early stages of treatment and mostly combined with more naturalistic approaches (e.g., incidental teaching) to foster generalization (Lovaas, 2003; Smith, 2001).

Although EIBI is generally effective for children with ASD, there is great variability in outcomes (Eikeseth, 2009; Peters-Scheffer et al., 2011; Reichow & Wolery, 2009). Results of studies investigating predictors of outcome are ambivalent, but a subset of studies shows that outcome is associated with child and family factors, including age of treatment onset (e.g., under the age of 4; Granpeesheh, Dixon, Tarbox, Kaplan, & Wilke, 2009), pre-treatment IQ, autism symptom severity and speech acquisition early in intervention (Ben-Itzack & Zachor, 2007; Smith, Groen, & Wynn, 2000), co-morbid conditions (Smith, Eikeseth, Klevstrand, & Lovaas, 1997; Smith, Klevstrand, & Lovaas, 1995) and parental stress (Osborne, McHugh, Saunders, & Reed, 2008) and treatment features, such as treatment intensity and duration (Eldevik, Eikeseth, Jahr, & Smith, 2006; Granpeesheh et al., 2009; Lovaas, 1987; Peters-Scheffer, Didden, Mulders, & Korzilius, 2010) and intensity and quality of supervision (Eikeseth, Hayward, Gale, Gitlesen, & Eldevik, 2009).

Even small changes in treatment procedures can influence child outcome (Kelly, Green, & Sigman, 1998) suggesting that the quality of treatment delivery can predict treatment outcome (Allen & Warzak, 2000; Bibby, Eikeseth, Martin, Mudford, & Reeves, 2001). Treatment quality is dependent on therapists reproducing intervention during daily sessions (procedural fidelity) and the extent to which treatment is implemented as designed (treatment integrity). While treatment integrity centers on the role of consultant to generate a treatment program that meets the child's needs, procedure fidelity focuses on the accurate use of particular treatment procedures and the skills and judgements of the therapist shown during treatment (Symes, Remington, Brown, & Hastings, 2005).

During EIBI research, treatment in general is implemented with good treatment integrity and procedural fidelity (Sallows & Graupner, 2005; Smith, Buch, & Gamby, 2000) but in community-based programs procedural fidelity and treatment integrity is likely lower (Love, Carr, Almason, & Petursdottir, 2009; Mudford, Martin, Eikeseth, & Bibby, 2001). Since children learn better when therapists are appropriately trained (Dib & Sturmey, 2007; Sarakoff & Sturmey, 2008) many researchers have improved treatment quality by training paraprofessionals, professionals and parents to implement EIBI correctly (Crockett, Flemming, Doepke, & Stevens, 2007, Dib & Sturmey, 2007; Leblanc, Ricciardi, & Luisselli, 2005; Sarakoff & Sturmey, 2004; 2008; Thomson, Martin, Arnal, Fazzio, & Yu; 2009); however, there is little research to determine the relationship between therapist individual differences and their performance during EIBI. An exception is Symes et al. (2005) in which 19 therapists who implemented DTT were interviewed about factors they considered to enhance or reduce correct treatment delivery. According to them, both training in behavioral analysis theory and techniques and therapist's patience and child factors, such as likeability, motivation, compliance, intellectual competence and behavior problems all related to child outcomes. Symes et al. (2005), however, did not observe procedural fidelity. Therefore, this study investigated the relationship between therapist characteristics and therapist-child relationship and treatment integrity using objective measures of treatment integrity. Given the characteristics of EIBI and the significant learning deficits and challenging behavior displayed by children with ASD and ID, therapists might need specific skills or personality traits to provide adequate treatment. "Personality" may seem an unusual variable for behaviorists to study, yet, Skinner (1953, pp. 284) did indeed discuss personality as a "functionally unified set of responses" (p. 285). The observations made in this study can be usefully interpreted in this light.

Among researchers, there seems to be consensus that there are five replicable, broad dimensions which represent personality at the broadest levels of abstraction: (1) extraversion, (2) agreeableness, (3) conscientiousness, (4) neuroticism (emotional stability), and (5) openness to experiences. In short, extraversion refers to an energetic approach to the social and material world and involves traits like assertiveness, positive emotionality, activity and sociability. Conscientiousness involves prescribed impulse control that facilitates task- and goal directed behavior like following norms and rules, planning, organizing and prioritizing tasks, thinking before acting and postponing gratification.

Agreeableness describes a prosocial and communal orientation towards others and comprises traits such as trust, tender-mindedeness, modesty and altruism. Openness to experiences refers to the complexity, originality, width and depth of the person's mental and practical life, while neuroticism describes negative emotionality like feeling nervous, sad, anxious, and tense. This classification facilitates the possibility to study specified personality domain instead of observing all behaviors that comprise an individual's personality (John & Srivastava, 1999).

Over different jobs and settings conscientiousness predicted overall job performance, while the other dimensions predicted job performance for job categories or specific tasks (Barrick & Mount, 1991). As EIBI needs to be delivered accurately and precisely, we hypothesized that therapists with higher scores in conscientiousness and lower scores in openness to experiences, will show higher levels of procedural fidelity. We also expected that therapists with higher scores in agreeableness show higher procedural fidelity and that increased scores of extraversion will be related to lower levels of procedural fidelity. The dimension neuroticism was examined on an exploratory basis. Therapists in Symes et al. (2005) suggested that competent, likeable, motivated and compliant children are associated with more effective service delivery and that effective treatment delivery was hindered by the therapists' negative reaction to child behavior (Symes et al., 2005). Therefore, we expect higher procedural fidelity in therapist-child dyads with a better therapist-child relationship than in dyads in which the relationship is of lesser quality. Furthermore, we expect that therapists with a negative attitude towards individuals with a disability have lower procedural fidelity scores.

11.2 Method

11.2.1 Participants and Setting

At the time of the study, 34 paraprofessional line therapists worked at the ASD treatment program, of which 22 therapists participated. All were female and provided DTT to one or more children with ASD and ID in an educational setting for young children and ID (M = 3.82; SD = 2.67; range: 1-11). All had worked for at least 2 months in this setting. On average, therapists had 22.77 months experience in providing DTT (SD = 25.36; range: 2-103) for 6.32 hours per week (SD = 4.10; range: 2-20). Therapists were aged 20-58 years (M = 28.82; SD = 9.98). Thirty-five children participated and all had autism or PDD-NOS and mild to severe ID with a mean ratio IQ of 40. A clinician independent of the study determined diagnoses based mainly on the Autism Diagnostic Observation Schedule (ADOS; Lord, Rutter, DiLavore, & Risi, 2006), the ADI-r (Lord, Rutter, & Couteur, 1994), the Mullen Scales of Early Learning (MSEL; Mullen, 1995), the Bayley Scales of Infant Development Second Edition (Van der Meulen, Ruiter, Spelberg, & Smrkovsky, 2002) and/or the Vineland Adaptive Behavior Scales (VABS; Sparrow, Balla, & Cicchetti, 1984). Children's ages ranged from 3;5-10;1 years with a developmental age between 1;5-3;4 years. They were chosen for the program based on availability.

The study took place at an ASD treatment program, which provided EIBI services in the eastern part of the Netherlands to children between 2 to 10 years of age classified with ASD and ID. The treatment format was ABA with an emphasis on DTT combined with incidental teaching methods. Each child's team included the program consultant, a senior therapist and several therapists delivering the DTT program. Senior therapists had a master's degree and at least 1 years experience in delivering ABA at the project. The consultant held a master's degree in special education and had eight years of experience in treatment of children with ASD with or without ID using ABA treatment procedures.

11.2.2 Procedure and Measurements

Three master special education students collected data on procedural integrity. They selected video fragments at random from a container which had all the video clips of each dyad (n = 37) and, when unavailable, they randomly filmed a treatment session (n = 33) and scored them using an instrument (see below) developed for the purpose of this study.

For each trainer-child dyad one session was scored (average duration 7.6 minutes (range 5 to 10 minutes; minimally 10 trials) and the number of sessions scored per therapist ranged from 1 to 11 (M = 3.05; SD = 2.52). Although DTT is one method of teaching used during EIBI, only DTT sessions conducted at the table were scored as the structured character of these trials facilitates the measurement of procedural fidelity.

11.2.3 Personal and job related questionnaire

Therapists were asked to provide personal and job-related information including their age, level of education, experience in delivering DTT and prior acquaintance with an individual with a disability (ASD and/or ID).

11.2.4 Attitudes towards individuals with a disability

The Multidimensional Attitudes Scale towards individuals with disabilities (MAS; Findler, Vilchinsky, & Werner, 2007) was used to assess therapist's attitude based on three dimensions: affect, cognition and behavior. After reading a vignette describing an interaction between Michelle and an individual in a wheelchair, participants marked on a 5-point Likert-type scale 47 items to which degree they believed the item accurately reflected how Michelle would feel, think or act in that situation. The MAS yields a total score (range: 47-235) and scores on the three dimensions. A high score on the MAS represents a negative attitude towards individuals with a disability.

11.2.5 Child-therapist-relationship

For each therapist-child dyad, therapists completed the Dutch version of the Student-Teacher-Relationship Scale (STRS; Cornelissen & Verschueren, 2002) to measure perceived relationship between therapist and child. The STRS is a questionnaire comprised of 28 items with a 5-point Likert-type scale (range: 28-140) and consists of three subscales: closeness (positive aspects of the relationship between the therapist and the child), conflict (negative aspects of the relationship between the therapist and the child such as child challenging behavior) and dependency (negative aspects of the relationship related to possessive behavior). The STRS has good psychometric properties for individuals with ID (Roeden, Maaskant, Koomen, Candel, & Curfs, 2012). Twenty-two teachers and 35 children were included, who formed 70 therapist-child dyads. Therapists completed the STRS for one to eleven children (M = 3.05; SD = 2.52), while one to seven therapists completed the STRS for each child (M = 2.29; SD = 1.55). In the analysis each dyad was used once.

11.2.6 Personality factors

The Dutch version of the NEO-Five Factor Inventory (NEO-FFI; Hoekstra, Ormel, & De Fruyt, 1996) was used to measure neuroticism, extraversion, openness to experiences, agreeableness, and conscientiousness. For each scale, therapists rated twelve items on a 5-point Likert-type response format. Raw factor scale scores were transformed into stanine scale scores using Dutch norms (M = 5; SD = 2).

11.2.7 Procedural fidelity

Dependent variable was therapist's correct use of the components of DTT during five to ten minutes of one DTT session (minimally ten trials). An observation instrument was developed to measure correct use of DTT based on treatment manuals (Leaf & McEachin, 1999; Lovaas, 2003), interviews with senior therapists, the training package of the ABA provider and individual education plans of the children, and included the following components: Discriminative stimulus, prompt, response time, feedback, error correction, positive practice trial and inter trial interval (see for a description Appendix A). Using event recording, the observer scored the correct or incorrect implementation of the component, omission or commission of the component and when applicable procedural errors occurred (e.g., incorrect discriminative stimulus, multiple stimuli, incorrect prompt [location, level], and absence of contingent feedback).

The percentage of correct implementation was calculated for each trial using the following formula: Number of correct components per trial divided by the total number of components of the trial (i.e., correct and incorrect implemented components, with exclusion of irrelevant components [e.g., positive practice trial after a correct response of the child]) and multiplied by 100.

Total procedural fidelity for each session was computed as the average percentage of correct implementation across trials. In addition, for all fragments the correct implementation of each component of DTT was calculated by dividing the frequency of the correct implementations of the component over all trials by the total number of trials and multiplied by 100.

Prior to scoring, the three observers and first author practiced scoring using videotaped sessions until inter-observer agreement (IOA) of at least 90% was achieved, which was obtained in two sessions (in total: 1.5 hours). An agreement was defined as observers scoring an item on the checklist identically. A disagreement was defined as the observers scoring an item on the checklist differently (including omission and commissions). On five treatment sessions the three observers obtained a mean IOA of 95% (range: 91-100%). The remaining videotapes were scored by one of the three observers and 20% of the sessions was scored by a second observer. The mean IOA was 97% (range: 96-100%).

11.3 Results

11.3.1 Procedural fidelity

The average procedural fidelity for each fragment ranged between 74.29 to 100% (*M* = 90.38, *SD* = 5.86) and average scores for individual therapists ranged from 82.93 to 98.61% (*M* = 91.50, *SD* = 4.57). Procedural fidelity and procedural fidelity for the components were > 90% for all components except for the component feedback (67.82%; See Table 1).

Table 1. Average procedural fidelity (%) for each therapist and fragment.

	Procedural fidelity					
	Average per therapist (*N* = 22)			Average per fragment (*N* = 70)		
Component	*M*	*SD*	*Range*	*M*	*SD*	*Range*
Total	91.50	4.57	82.93-98.61	90.38	5.86	74.29-100.00
Stimulus	92.23	9.19	66.67-100.00	91.87	11.70	40.74-100.00
Prompt	93.58	9.39	68.18-100.00	91.94	14.75	36.36-100.00
Response time	99.43	1.29	95.56-100.00	99.02	0.04	77.78-100.00
Feedback	67.82	13.90	47.32-90.91	64.19	21.41	22.22-100.00
Error correction	97.44	3.91	85.78-100.00	96.53	9.09	40.00-100.00
Positive practice trial	93.86	5.86	83.33-100.00	93.15	10.89	40.00-100.00
Inter-trial interval	96.64	5.32	76.92-100.00	96.57	0.08	50.00-100.00

11.3.2 Predictors of Procedural fidelity

Therapist's characteristics are presented in Table 2. Since stanine scores of one to three are below average, four to six average, and seven to nine above average, therapists had average personality scores on the NEO-FFI as compared to the Dutch norms.

First, a hierarchal multiple regression analysis was conducted to assess the contribution of personality traits and attitude towards individuals with a disability to procedural fidelity. Attitudes and traits are both seen as relatively stable, but in general personality traits are more resistant to transformation than attitudes as the latter might change as new information or experiences become available (Ajzen, 2005). Therefore, in a stepwise manner, personality traits neuroticism, extraversion, openness to experiences, agreeableness, and conscientiousness were entered in the first step and scores on the cognition, behavior, and affect scales of the MAS in the second step (see Table 3). R^2 was significantly different from zero at the end of each step and the three models significantly improved the ability to predict procedural fidelity (model 1: $F(1,19) = 4.90$, $p = .04$; model 2: $F(2,18) = 6.65$, $p < .01$; model 3: $F(3,17) = 7.58$, $p < .01$). After step 3, in which openness to experiences and the affect and cognition scale of the MAS were included, 57% of the variance of the procedural fidelity was accounted for; however, as only openness to experience, and the affect and cognition scales of the MAS contributed significantly to the model, in step 3 neuroticism ($t = -0.50$; $p = .62$), extraversion ($t = 0.90$; p = .38), agreeableness ($t = -0.79$; $p = .44$), conscientiousness ($t = 1.46$; $p = .16$), and the behavior scale of the MAS ($t = -0.17$; $p = .87$) were excluded from the analysis. To assess the relationship between procedural fidelity and the therapist-child relationship, Pearson correlations were computed using data from 70 therapist-child dyads.

Table 2. Means, standard deviations and ranges of therapist characteristics (N = 22).

Variable	*Mean*	*SD*	*Range*
Personality (stanine)			
Neuroticism	4.77	1.19	2-7
Extraversion	6.05	1.70	2-9
Openness to experiences	6.32	1.84	4-9
Agreeableness	6.36	0.95	4-8
Conscientiousness	5.36	1.65	2-8
Attitudes			
Cognition	53.86	10.48	31-78
Behavior	31.52	6.49	22-50
Affect	28.29	5.02	18-37
Relationship therapist-child			
Total	99.56	10.48	75-122
Closeness	34.31	5.40	23-46
Dependency	9.56	2.71	5-17
Conflict	26.84	9.16	14-51

Table 3. Multiple regression analysis to predict procedural fidelity from openness to experience and the affect and cogniton scale of the MAS (N = 22).

	B	*SE B*	*B*
Step 1:			
Constant	97.90	3.17	
Openess to experience	-1.08	0.49	-.45*
Step 2:			
Constant	107.01	4.44	
Openess to experience	-0.79	0.44	-.33
Affect	-0.20	0.08	-.49*
Step 3:			
Constant	103.73	4.17	
Openess to experience	-0.84	0.39	-.35*
Affect	-0.33	0.09	-.79**
Cognition	0.34	0.14	.50*

*Note: * p < .05, ** p <.01.*

Based on Cohen's (1992) criteria, associations with a medium effect size were found between procedural fidelity and the total scale (r = -.28; p = .02) and between procedural fidelity and the closeness scale of the STRS (r = -.39, p <.01). Hence, therapists who experienced a positive relationship with the child displayed lower procedural fidelity than therapists who experienced the relationship as less positive. No significant relationships were found between procedural fidelity and the dependency scale (r = .06, p = .63) and the conflict scale (r = .09, p = 48) of the STRS.

11.4 Discussion

Until recently, relatively little attention has been given to the impact of individual therapist characteristics on the procedural fidelity and the outcome of treatment for children with ASD and/or ID. This study found that therapist's personality trait openness to experiences was associated with procedural fidelity. Therapists who displayed more openness to experiences showed lower procedural fidelity: this is in line with our expectations as these individuals experience more difficulties in conforming to rules and schedules (Hoekstra et al., 2007).
Also, therapists who experienced a positive relationship with the child displayed lower procedural fidelity than therapists who experienced the relationship as less positive. Finally, therapists with negative emotions towards persons with a disability displayed lower procedural fidelity as opposed to individuals with positive emotions towards individuals with a disability; however, higher procedural fidelity was seen in therapists with less negative cognitions towards individuals with ID. These observations suggest that therapist characteristics do indeed predict their performance during DTT.

Skinner's analysis of personality as an explanatory fiction may yield some interesting implications. For example, for those therapists who experience a positive relationship with the child engaging in interactions with children is often highly reinforcing. Therefore, it might be beneficial for them to engage in other activities where sociable interaction is desirable at times other than DTT, thereby reducing the reinforcing value of engaging in sociable interactions during DTT that may inhibit learning. Alternately, or in addition to the previously mentioned strategy, therapists whose characteristics interfere with certain essential components of DTT may require more training and supervision than other therapists to ensure that they teach the children correctly and effectively.

While it may be possible to speculate that personality tests might be useful in selecting therapists, the magnitude of this relationship is very modest and hence not likely to be helpful in predicting the performance of individual therapists reliably. Future studies might investigate whether the ability in ABA principles on questionnaires with a vignette and/ or role play might predict future therapist performance. Also, a work sample before and during training and therapist's responsiveness to training and supervision might predict future therapist behavior.

Overall procedural integrity was relatively high; however, these data may be deceptive. Integrity data on the feedback component of DTT were low and this component includes reinforcement of correct responses and error correction, which are essential components of DTT that may lead to child progress. Therefore, next to overall procedural fidelity, fidelity scores on subscales should be reviewed to obtain a full picture of therapist performance. Further, even where treatment integrity appears relatively high, the ranges were very wide which might hide problems with individual therapists. Only when these data are validated with data on child response acquisition and reduction in maladaptive behavior or other evidence of learning (Dib & Sturmey, 2007; Sarakoff & Sturmey 2008) can we be sure that these measures of staff performance is valid. Although we do expect that there is a clinical difference between therapist with low and high scores on procedural fidelity, future studies should further explore this by including measures of child behavior in addition to staff behavior and explore data on overall procedural fidelity and the components of DTT.

The present study has several limitations. First, limited data were collected on child variables. Future studies might investigate to which extent child characteristics influence therapist procedural fidelity. Children's challenging behavior, compliance, motivation and learning rate might influence how demanding and reinforcing treatment delivery to a therapist is.
Also, child factors such as likeability, motivation, compliance, intelligency and behavior problems might be related to the relationship between the therapist and child and indirectly influence procedural fidelity. Therefore, future research should adress the gap between child characteristics and procedural fidelity. Second, the present sample of therapists were drawn from one service setting where there may have been relatively good staff training and support, which might have attenuated the relationship between therapist individual differences and treatment integrity.

This may limit the generality of these results. Likewise this study only studies staff implementation of only one intervention method. Thus, future research should also determine to what extent these findings can be generalized to other less structured teaching methods such as Pivotal Response Training (Koegel, Koegel, Harrower & Carter, 1999). Finally, the small sample size may have enlarged the chance of making a Type II error. In addition, the number of predictors in the regression analyses was relatively large in relation to sample size and therapists and children were included in multiple therapist-child dyads. Therefore, these are tentative results that need to be corroborated in larger samples in future research.

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Appendix A. Components of a Discrete Trial (e.g., Duker, Didden, & Sigafoos, 2004; Leaf & McEachin, 1999; Lovaas, 2003; Smith, 2001)

Component of a discrete trial	Operational definitions
Discriminative stimulus	After obtaining the attention of the child (e.g., by calling the name of the child), a brief, clear instruction (e.g., "Do this") or question (e.g., "What is it?") as described in the child's Individual Education Plan (IEP), was provided once by the therapist to evoke a response from the child.
Prompt	A stimulus (e.g., physical guidance, verbal model) added immediately after the discriminative stimulus (within two seconds) as described in the IEP of the child, to increase the probability that the child responds correctly to the discriminative stimulus.
Response time	The time between the discriminative stimulus and the response of the child (in our program: five to seven seconds).
Feedback	Immediately after a correct response of the child, the therapist provides the child with praise in combination with behavior specific feedback ("Good job, you pointed correctly to the ball"). Feedback was intermediately combined with a tangible reinforcer. Reinforcement and reinforcement schedules were described in the IEP of the child. After an error or positive practice trial, a behavior specific comment at a neutral tone is made (e.g. "that is pointing to the ball").
Error correction	If the child responds incorrectly to the stimulus presented by the therapist, the therapist intervenes directly and finishes the trial providing the child with full assistance so that the child performs the target behavior and a forthcoming error is prevented. When the child shows no response within seven seconds after the stimulus, the therapist finishes the trial providing the child with full assistance so that the child performs the target behavior.
Positive practice trial	With full assistance (i.e., physical guidance or a model) and at a neural tone the whole trial is repeated to practice the target behavior to provide the child with an extra opportunity to display the appropriate behavior. A correct positive practice trial consists of 5 steps: 1) the therapist gets the attention of the child (e.g., by calling the child name); 2) the therapist provides the child with the correct materials in the manner described in the IEP of the child; 3) the trainer provides the child with the correct instruction as describe in the IEP of the child; 4) the therapist provides the child with full assistance to perform the target behavior; and 5) the therapist provides behavior specific feedback at a neutral tone (e.g., "that is pointing to the ball").
Inter trial interval	A brief pause of one to five seconds between the consequence and the discriminative stimulus of the next trial.

Chapter 12

Conclusions and General Discussion

This thesis focuses on the development and treatment of young children with ASD and ID. Results are presented of studies on: (1) the behavioral characteristics of young children with ASD and ID, (2) the effectiveness of early behavioral treatment, and (c) the health care conditions required for effective early behavioral treatment. In the 12th and final chapter, general results and limitations will be discussed, suggestions will be given for future research and implications for policy and clinical practice are discussed.

12.1 Behavioral characteristics of children with ASD and ID

In Part I of the thesis, five studies are presented regarding the behavioral characteristics of children with ASD and ID. To illustrate how behavioral characteristics may affect early development in children with ASD and ID, the understanding of other's intentions and a lack of behavioral flexibility were chosen as two examples associated with the three core deficits of ASD (i.e., deficits in communication and social interaction, and repetitive and restricted patterns of behavior and interests). In addition, the influence of behavioral characteristics of children with ASD and ID on maternal stress was investigated.

12.1.1 Understanding of intentions of others

To determine whether children with ASD and ID see others as intentional beings, who initiate and react to goal-directed actions, the understanding of other's intentions and associated variables in children with ASD and ID were explored in the studies described in the Chapter 2 and Chapter 3. The findings of these studies suggest that children with ASD and ID have difficulty in understanding intentions of others as manifested during imitation and non-verbal communication (gazing and pointing). The understanding of other's intentions is predicted by the child's developmental age and may result in difficulties in acquiring early social communication skills and language. Compared to typically developing children or children with other developmental disabilities, children with ASD and ID require a higher developmental age to succeed in tasks related to the understanding of other's intentions (e.g., Behne, Carpenter, & Tomasello, 2005; Bellegamba & Tomasello, 1999; Bellegamba, Camaioni, & Colonnesi, 2006; John & Mervis, 2010; Meltzoff, 1995). This higher developmental age required to succeed in such tasks suggests a delayed development of intention understanding.

Findings are in line with the social motivation model and may partly explain why children with ASD and ID experience difficulties in (social) learning in the natural environment. It underlines the importance of functional reinforcement in learning as it states that social consequences of behavior in the natural environment are sometimes insufficient for children with ASD and ID to engage in social learning and interaction. Therefore, children with ASD and ID may become less proficient in processing social information necessary for social learning (Dawson, Webb, & McPartland, 2005).

Several studies show that behavioral strategies can be used to teach children with ASD and ID joint attention skills including pointing and gaze following (see for a review: White et al., 2011) and imitation skills (e.g., Cardon & Wilcox, 2011; Ganz, Bourgeois, Flores, & Campos, 2008). Instead of teaching these behaviors to children with ASD and ID, future studies may use behavioral strategies to increase the reinforcing value of social stimuli by systematically pairing tangible with social reinforcers to enhance social orienting in children with ASD and ID. It should be investigated if the increase in social orienting has a collateral effect on social learning behaviors that were not targeted during treatment (e.g., imitation, joint attention) and development.

Furthermore, results reported in Chapter 2 and 3 imply that parents and professionals need to realize that young children with ASD and ID are not always directing their attention to or are capable of understanding the intentions of their caregivers' actions. When using observational learning, parents or professionals should focus the child's attention to the social stimuli and use tangible reinforcement to learn and strengthen behavior. With the help of professionals, parents may consider to establish the reinforcing value of social stimuli by using stimulus-stimulus paring. Using imitation, parents and professionals must initially model the whole act instead of only a part of the act or state the goal of the action. If necessary, additional prompts can be provided to help the child to succeed in the task. Prompt fading procedures, in which decreasing assistance to the child is provided, can be used to prevent prompt dependency (see Duker, Didden, & Sigafoos, 2004).

12.1.2 Behavioral flexibility

As the knowledge base on the nature and extent of behavioral flexibility in individuals with ASD and ID is scarce as compared to the extensive literature published on other impairments related to ASD (Healy & Leader, 2011), we focused on behavioral flexibility as measured by the Behavioral Flexibility Rating Scale in Chapter 4 and 5 (BFRS-r; Green et al., 2006; 2007; Pituch et al., 2007). The study described in Chapter 4 suggests that the BFRS-r has excellent internal consistency, good inter-rater and intra-rater reliability and evidence of validity based on convergence with the Sameness Questionnaire (Prior & MacMillan, 1973). In the study described in Chapter 5, the BFRS-r was used to assess behavioral flexibility in children with ASD (i.e., autism and PDD-NOS) plus ID and compared scores to those with ID only. Although the precise nature of the effect of diagnosis on behavioral flexibility remains unclear, our main finding was that in children with ASD and ID a higher developmental age and more severe autism is associated with more problems in behavioral flexibility. Further, behavioral flexibility seems to influence emotional and behavioral problems and maternal stress, but not adaptive behavior.

Despite that individuals with ASD show clear flexibility deficits during everyday behavior, results on neuropsychological measures, with exception of the Wisconsin Card Sorting Task, are inconsistent (Geurts, Corbett, & Solomon, 2009).

For example, Teunisse and colleagues (2012) found no significant correlations between neurological tests and parental-based ratings of behavioral flexibility as measured on the BFRS-r and the Behavior Rating Inventory Executive Functioning. Future studies should shed more light on this disparity between cognitive and behavioral flexibility by comparing behavioral flexibility scores on parent's ratings and neurological tests measuring flexibility in a broader ASD sample including preschool children with ASD and ID.

Next to social-communication skills, flexibility is required for learning in the social environment. Restricted interests and a fixation on familiar and predictable objects and routines may prevent the child from engaging in social interaction, exploring new objects and environments and acquiring new knowledge and skills. In addition, difficulties in behavioral flexibility are associated with heightened levels of parental stress. Interventions aiming at reducing the insistence of sameness and increasing behavioral flexibility in individuals with ASD and ID are warranted. Therefore, future studies should evaluate if and how behavioral strategies can be used to teach children with ASD and ID to cope with unpredictable changing environments for example by teaching the child socially appropriate behavior as alternative to challenging behavior (see Green et al., 2007).

12.1.3 Maternal Stress

Children with ASD and ID require more or less intensive care and treatment, which has a substantial impact on family functioning and parental stress. The study described in Chapter 6 shows that emotional and behavioral problems contribute more to parental stress than the child's diagnosis or developmental delay. This is in line with other studies (e.g., Herring et al., 2006).

Parental stress is associated with the outcome of behavioral treatment programs. For example, Osborne, McHugh, Saunders, and Reed (2008) showed that early behavioral treatment produced fewer gains when parents reported high levels of parental stress, especially when intervention was more intensive. Therefore, before commencing intervention parents should reduce their stress levels. Emotional and behavioral problems of the children need to be addressed with priority in behavioral treatment programs.

Parental distress might be seen as a contraindication to (intensive) behavioral treatment, which requires daily involvement of the parents in their child's treatment. However, next to a negative effect on parents, involvement in their child's treatment may also result in positive effects such as less depression and stress, greater satisfaction and a more positive attitude towards their child (Baker, 1996). Also, the increased skills of their child and reduced behavioral problems might add to lower stress levels. Consequently, parental stress is included as an outcome measure in one of the studies in Part II, in which the effectiveness of behavioral treatment is investigated. In contrast to other studies that found that behavioural treatment resulted in lower levels of parental stress (Birnbauer & Leach, 1993), our study found no effect of behavioural treatment on parental stress.

However, given the interrelationship between parental stress and behavioral treatment found in many other studies, parental stress should be considered as a key factor in determining the effectiveness of early interventions for children with ASD and ID.

12.2 Effectiveness of early behavioral treatment

In Part II the effectiveness of early behavioral treatment on the development and behavior of children with ASD and ID was addressed. In a meta-analysis, we synthesized the results on early intensive behavioral intervention (EIBI), while in two longitudinal intervention studies the effects of low intensity behavioral treatment (LIBT) on the development and behavior of children with ASD and ID were investigated.

12.2.1 Effectiveness of early behavioral Intervention

Based on 11 studies with 344 participants, the meta-analysis described in Chapter 7 strongly supports the conclusion that EIBI in children with ASD and with and without ID is effective. Experimental groups who received EIBI outperformed the control groups on IQ, non-verbal IQ, expressive and receptive language and adaptive behavior with differences of 5 to 15 point on standardized tests. With our meta-analysis, five other meta-analyses appeared (Eldevik et al., 2009; Makrygianni & Reed, 2010; Reichow & Wolery, 2009; Spreckley & Boyd, 2009; Virués-Ortega, 2010). Like our meta-analysis, they concluded that EIBI was an effective intervention for many children with ASD. Misinterpretation of the study conducted by Sallows and Graupner (2005) might explain why one meta-analysis found that EIBI was not better than treatment as usual (Reichow, 2012). Hence, although not effective for all children, EIBI has ample empirical support and should be strongly considered when selecting a treatment for children with ASD (Reichow, 2012).

Large variability in outcome is seen between participants and studies, which is amongst others related to differences in treatment intensity (Granspeesheh, Dixon, Tarbox, Kaplan, & Wilke, 2009). Relatively few studies have investigated the minimal intensity of behavioral treatment that is needed to achieve significant gains (see for an exception: Eldevik, Eikeseth, Jahr, & Smith, 2006). Therefore, we conducted two longitudinal studies (duration: 8 and 24 months) with a pre-test post test control group design to assess the effectiveness of 4-10 hours per week LIBT supplementing (pre)school services. Results showed that children with ASD and ID aged between 3-10 years made significantly more progress on developmental age, receptive language and adaptive skills as compared to children receiving treatment as usual at their (pre)school. Also, after two years children exposed to LIBT showed fewer characteristics of ASD and fewer behavioral problems. No differences between groups were found on expressive language, behavioral flexibility and maternal stress.

Our results are generally in line with results reported by Eldevik et al. (2006) who also found that children with ASD and ID receiving LIBT (12.5 hrs per week) outperformed children receiving eclectic treatment. As expected, the differences between the treatment group and control group in LIBT studies were smaller than reported in many EIBI studies. Next to differences in the intensity of treatment and supervision, this may be explained by the lower IQ and higher chronological age at treatment onset which are also related to treatment outcome (e.g., Granpeesheh et al., 2009).

Albeit effective, most studies investigating the effectiveness of behavioral treatment fail to fulfill basic research criteria such as random assignment to groups, non-uniform assessment protocols, the lack of adequate fidelity measures and unknown characteristics of comparison conditions (Reichow, 2012). Only one randomized control trial on EIBI was published (i.e., Smith, Groen, & Wynn, 2000). As it is unethical to deny children with ASD effective treatment, studies are very labor intensive and costs are high, conducting studies of this type is challenging and methodological limitations need to be considered in this light (Kasari, 2002; Matson & Smith, 2008; Smith, Eikeseth, Klevstrand, & Lovaas, 1997).

Our intervention studies have several limitations and strengths in design. As randomized control trials were difficult to realize in clinical practice in terms of costs and ethics, we used pre-test post test control group designs. In the field, this is a common and a generally accepted alternative (Kasari, 2002; Matson & Smith, 2008; Smith et al., 1997). Parents and staff were actively involved in the treatment of their children, so no attempt could be made to blind the participants to the intervention they had received. Although assignment to groups was not randomized, at intake groups were not significantly different on several measures.

An uniform assessment protocol was used. The experimental and the control group were followed over the same period of time (i.e., 2005-2007 and 2007-2011) and the same lengths of follow up were used. As different measures of the same construct may result in considerably different outcomes, the same measures were used across time and groups. Analyses in Chapter 9 show that, although the researcher was aware of group assignment, assessments in both the experimental as the control groups were reliable.

In contrast to most studies, which only used measures of IQ, communication and adaptive behaviour to determine treatment outcome, we used broad range of outcome measures, which were valid, reliable and all vital to establish the effect of treatment on child development (Matson, 2007). However, as in other studies only short term effects were reported (i.e., follow-up after two years).

Although we studied a center-based program implemented in the community, treatment fidelity was facilitated by the use of treatment protocols and manuals (e.g., Leaf & McEachin, 1999; Lovaas, 2003), meetings, video-recordings during treatment, data-sheets with trial-by-trial data and an individual learning plan with detailed descriptions of each program, and supervision by a special educator. Compliance to treatment was further assessed by registration of treatment hours. In addition, in the study described in Chapter 9 measures of treatment fidelity were included.

In our studies, children started treatment later and had a lower IQ than in most other studies implemented in a research setting (e.g., university) (Lovaas, 1987; Sallows & Graupner, 2005). As this is in accordance with other programs implemented in the community (see Mudford, Martin, Eikeseth, & Bibby, 2001), our sample seems representative for children served in clinical practice. In addition, treatment was implemented by clinicians employed at Stichting De Driestroom, a service provider for individuals with ID in the eastern part of the Netherlands and regular funding was used. Therefore, the studies described in Chapter 8 and 9 provide evidence for real-world effectiveness of LIBT in the Netherlands and contribute to reducing the gap between intervention implemented in research settings and within the community (Jacobson & Mulick, 2000).

Although there have been a number of methodological concerns about our and other behavioral treatment studies, results suggest that behavioral treatment is effective in increasing cognitive and adaptive functioning in children with ASD and ID and may result in decreased autism severity and behavioral problems. However, still controversies exist regarding which children respond best and to what extent. Studies with improved research methodology are still warranted (Matson & Smith, 2008).

12.3 Health care conditions

Part III of this thesis addresses the health care conditions that are required to provide behavioral treatment of high quality to children with ASD and ID in the community. Several studies report that major barriers to implement behavioral treatment are associated with the quality of programs implemented in the community (e.g., obtaining and maintaining a well-trained and suitable team) and the high costs of the program related to the difficulties in obtaining funding (Johnson & Hastings, 2002; Trudgeon & Carr, 2007). Therefore, we conducted a cost-offset study regarding the costs and benefits of EIBI. Next to this, we investigated to what extent therapists' characteristics contribute to the quality of the treatment.

12.3.1 Cost and Benefits of EIBI

Life-time costs associated with ASD are high and approximately three million Euro for an individual with ASD and ID (Järbrink & Knapp, 2001). However, with appropriate treatment, individuals with ASD and ID may become less dependent of professional care, reducing life-time costs.

The study described in Chapter 10 sought if long-term benefits of EIBI may overweigh the initial costs associated with the program. Costs between 3-65 years of age were estimated for individuals with ASD with different outcomes (i.e., dependency, reduced-dependency and independency). Based on behavioral treatment studies, percentages of outcome after treatment were estimated.

After EIBI, 29% of the individuals with ASD were placed in regular education, 34% in less intensive special education and 37% in special education versus 11%, 8% and 81% respectively after treatment as usual or eclectic treatment. This resulted in estimated long-term savings, which are approximately € 1,100,000 from age 3-65 years per individual with ASD. Extending these costs to the whole Dutch ASD population, cost savings of € 109.2 to € 182 billion have been estimated, excluding costs associated with inflation.

Results of our study provide a compelling argument for the provision of EIBI and are in line with other cost-offset studies indicating that the long-term savings of behavioral treatment seem to overweigh costs of the programs (Chasson, Harris, & Neely, 2007; Jacobson, Mulick, & Green, 1998; Motiwala, Gupto, & Hon, 2006). However, future studies should integrate estimates of the individual contributions to the outcome (e.g., intensity of treatment, child characteristics) to provide a more precise estimate of the costs and benefits of behavioral treatment.

12.3.2 Quality of LIBT

Next to the costs of the program, quality of programs implemented in the community is a concern of many parents and professionals. The study described in Chapter 11 found that the therapist's attitude towards individuals with a disability, therapist's personality traits agreeableness and openness to experience, and the perceived relationship between the therapist and the child contributed significantly to procedural fidelity of the treatment delivered. This finding is of importance as several studies indicate that procedural fidelity affects the efficacy of learning of children during behavioral treatment (e.g., Downs, Conley Downs, & Rau, 2007; Groskreutz, Groskreutz, & Higbee, 2011; Grow et al., 2009; Sarakoff & Sturmey, 2008). Given the modest relationships between variables, these variables may not predict individual therapist performance, but can be used in adapting treatment, training and supervision procedures. For example, more training and supervision can be provided to those therapists of whom personality traits interfere with certain essential components of DTT to ensure treatment of high quality.

Quality of treatment provided in the community needs to be reliably assessed and monitored so that parents can make informed decisions about the enrolment of their child in LIBT or EIBI. Guidelines regarding the quality of treatment and objective instruments to measure treatment quality need to be developed. An example for evaluating the quality of discrete trial teaching is the Discrete Trial Teaching Evaluation Form (Fazzio, Arnal, & Martin, 2007). It should be investigated if there is a relationship between performance on such measure and overall quality of the programs and long term child outcome. In addition, instruments to measure the quality of other components of behavioral treatment still need to be developed. For example, instruments to measure the quality of behavioral techniques used during classroom routines.

12.4 Future directions and reflections on early behavioral treatment

This thesis extends the limited knowledge base on LIBT and provides promising results of a center-based behavioral approach for children with ASD and ID that is implemented in the community. However, several questions remain to be answered. For example, future research should address long term (maintenance) effects over 5, 10 and 20 years and to which extent additional treatment is necessary to maintain treatment gains (Matson & Smith, 2008). Also, more needs to be learned about the side-effects of behavioral treatment on the child and family. LIBT is advised when professionals and parents are concerned that more intensive treatment is too stressful for the child. Therefore, future studies should investigate the influence of treatment intensity on the well being of the child and the family and explore factors such as chronological age and IQ that might moderate this relation.

Outcome between and within behavioral treatment studies is variable. Future studies should result in adapted approaches and guidelines regarding the intensity, duration, content and quality of treatment necessary to achieve optimal outcome for different types of children. Treatment characteristics and pre-treatment child variables across a broad range of measures need to be analyzed to identify the treatment components with the greatest effect on development and to differentiate children who successfully respond to behavioral treatment from non-responders.

After behavioral treatment, many children remain dependent from professional care and future studies should investigate how parents and professionals should address the needs of these children. Given their continued dependency from professional care, some might argue to which degree differences between the treatment and the control group are clinically significant. However, even little progress on standardized measures such as IQ could represent valuable improvements in quality of life for children with ASD and ID and their families. For example, if children acquire basic communication and adaptive behavior skills, this may prevent or reduce challenging behavior (Sigafoos, Arthur, & O'Reilly, 2003; Wacker, Berg, & Harding, 2002).

The intense involvement of parents and professionals in LIBT may partly compensate for the provision of less intensive one-to-one treatment. In our studies, parents and professionals were instructed in behavioral techniques and the (pre)school teachers were responsible for delivering part of the treatment themselves. When trained, parents are able to assist in generalization and equipped to use the many opportunities they have to practice skills throughout their natural interaction with child. It is likely that this knowledge changes the general interaction with their child and consequently has a collateral effect on the child's development as the child is now instructed during all waking hours. As suggested by Sheinkopf and Siegel (1998) generalized training skills outside the treatment setting informally extent treatment, which has implications for an analysis of treatment intensity. However, data supporting this hypothesis are lacking and future research is warranted.

Future research should also focus on how the individual one-to-one treatment should be supplemented to provide a full-time treatment program to children with ASD and ID. This treatment program should be implemented in a communicative rich and stimulating environment in which learning principles could be used to provoke social initiatives of the child and facilitate learning during social interactions accounting for the impairments seen in social attention and processing (see also Chapter 2 and 3). Teaching and reinforcing behavior in the daily context promotes generalization and the likelihood of the child displaying the behavior in the daily context over time.

As in other countries where behavioral treatment programs for children with ASD are just establishing, competent therapists and supervisors are scarce and no evidence-based training programs in applying behavioral treatment for parents and therapists exist. This may result in poorly trained staff providing treatment with reduced quality. Consequently, in future studies evidence-based training programs for parents and professionals should be developed to learn them how to conduct LIBT of high quality, how to generalize learned behavior, teach new behavior in the daily situation and how to manage challenging behavior using functional analysis (Dillenburger, Keenan, Gallagher, & McElhinney, 2004).

Given the heightened level of stress seen in parents of children with ASD and ID, training should be relatively short and practical and should consist of some theory underlying behavioral treatment and the techniques used with an emphasis on practical exercise such as analyzing videos and role-play. As many parents do not have the time to engage in intensive individual treatment sessions with their child, parent training should focus on how to use learning principles in teaching adaptive behavior and reducing challenging behavior during normal routines with the child. Coaching of parents in the home should be an important component of parent training, as coaching on the job with performance feedback is necessary to maintain long-term results (LeBlanc, Ricciardi, & Luiselli, 2005).

Most studies on early behavioral intervention have studied the effectiveness in preschool children with ASD with developmental delays. Further research should address the needs and outcomes of children with only ASD or ID. For example, Eldevik, Jahr, Eikeseth, Hastings, and Hughes (2010) showed that behavioral treatment is effective in improving adaptive behavior in children with ID only. Future studies should investigate how based on behavioral phenotypes, behavioral treatment programs can be tailored to accelerate learning in children with distinctive profiles (e.g., children with Down syndrome).

A few studies (Chapter 8; 9; Eikeseth, Smith, Jahr, & Eldevik, 2002; 2007) provide some evidence for the effectiveness of behavioral treatment in relatively older children, but future studies should investigated to which extend behavioral treatment is effective in adolescents and adults with ASD and/or ID, which skills these programs should target and how these programs can be implemented in treatment facilities in the community.

Based on videotapes in some cases ASD can be differentiated from ID when children are only one year of age (Osterling, Dawson, & Munson, 2002). Future research should investigate if behavioral treatment is applicable in children younger than 2-3 years and with which

intensity, as overstressing the child should be prevented. Although research indicates that a lower chronological age at treatment onset, is associated with better outcome, no studies address at which age, with which intensity and with which precursors children should commence treatment (see also Matson & Smith, 2008; Shattuck & Grosse, 2007).

12.5 Implications for policy and clinical practice

To make behavioral treatment of high quality widely available to children with ASD and ID in the Netherlands, some changes in policy need to be made. To begin with, challenges related to the funding of the treatment need to be resolved. However, even though the results of four cost-offset studies performed by independent groups provide ample evidence that behavioral treatment may result in long-term savings, obtaining funding for behavioral treatment remains a major hurdle for many families to take. Often families initially fund treatment themselves, while further funding from the authority is sought.

In conducting the studies in Part II it appeared challenging to persuade financiers to fund our treatment and nine children had to discontinue their treatment in the second year. Financiers considered the regular treatment provided at the (pre)school as sufficient and refrained from investing in additional one-to-one treatment. However, as the studies in Chapter 8, 9 and 10 show that (pre)school services supplemented with behavioral treatment are more (cost) effective than (pre)school services only, additional one-to-one treatment should be funded to improve the functioning of children with ASD and ID.

Furthermore, as quality of treatment is related to outcome, quality needs to be reliably assessed and monitored so that parents can make informed decisions about the enrolment of their child in behavioral treatment and financiers can decide which programs to fund. Due to constraints associated with behavioral treatment provided in community settings (e.g., lack of funding, shortage of qualified therapists and specialized supervision, and children not fitting the profile of the program [Johnson & Hastings, 2002; Love, Carr, Almason, & Petursdottir, 2009]), it is unlikely that programs implemented in the community achieve and maintain levels of quality as evinced in programs implemented in research settings. However, quality of programs can be improved by developing evidence-based training programs for parents and professionals in applying behavioral treatment.

In a similar vein, ASD needs to be identified at a younger age. Since in the Netherlands children receive their diagnosis around their fifth birthday on average (Nederlandse Vereniging voor Autisme, 2008), many children start treatment later than recommended in most other studies. This has negative consequences for the outcome. Systematic screening of children with ASD with instruments such as the Early Screening of Autistic Traits questionnaire (ESAT; Swinkels et al., 2006), the Modified Checklist for Autism in Toddlers (M-CHAT; Robins, 2008) and the Baby and Infant Screen for Children with Autism Traits (BISCUIT; Matson, Boisjoli, & Wilkins, 2007) may facilitate early identification and diagnosis. Future studies should investigate and compare psychometric properties and applicability to the Dutch situation.

Finally, as low expectations regarding the educational achievements of children with ASD and ID and philosophies about their treatment may partly explain the low number of children enrolled in behavioral treatment in the Netherlands, parents and professionals need to be educated about evidence-based early intervention including behavioral treatment. This may facilitate parents when they have to decide upon the most appropriate treatment for their children (Kerr, Mulhern, & McDowell, 2000). Systematic and early information about intervention programs, may result in children enrolling in intervention programs at an early age. Future research exploring (combinations of) treatments implemented in the community should aid parents and professionals in this decision process.

As a whole, this thesis shows that children with ASD and ID are severely impaired in several areas of functioning and that this may negatively affect their development. The extent to which these impairments impact the child's prognosis depends on child factors such as IQ as well as on environmental factors such as the provision of appropriate and effective intervention. The meta-analysis and the two interventions studies included in Part II of this thesis provide additional evidence for the effectiveness of early behavioral treatment for children with ASD and ID. To ensure that in the Netherlands more children with ASD and ID can participate in early behavioral treatment of high quality, earlier and better screening of ASD is needed as are education programs to train parents, therapists, teachers and other professionals in the learning principles and techniques used in behavioral treatment. This may result in improved quality of life of children with ASD and ID and their family and long-term cost savings to the society.

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Summary

In this thesis, results are presented of studies on (1) the behavioral characteristics of children with autism spectrum disorder (ASD) and intellectual disability (ID), (2) the effectiveness of early behavioral treatment, and (3) health care conditions required for effective early behavioral treatment for children with ASD and ID. Designs and results of the studies conducted in this thesis are summarized below.

Part I: Behavioral Characteristics of Children with ASD and ID

In Part 1, five studies were conducted addressing the behavioral characteristics of children with ASD and ID. Aim of the studies described in Chapter 2 and Chapter 3 was to assess the ability of children with ASD and ID to infer other's intentions. In the study described in Chapter 2, 100 children aged 2-10 years were exposed to a hiding game, in which they had to locate a tangible reward that the experimenter hid in one of the two boxes on the table. In the warm-up condition, the children saw the experimenter hid the reward, while in the experimental trials the experimenter indicated the location of the reward by gazing or pointing to the container. Only 56 children performed above change (3 or 4 out of 4 trials correct) in the warm-up condition and were included in the analyses. Despite large individual differences, on average children performed above chance in the point condition, while the children performed under change in the gaze condition. Using a binomial test (7 out of 8 trials correct, $p = .03$), only 14% of the children performed above chance in both conditions.

In Chapter 3, a modified version of the behavioral re-entactment task was used to assess the understanding of others' intentions during an imitation task. In the first condition, the adult performed the target behavior, while in the second condition an unsuccessful attempt to perform the target behavior was demonstrated. Children with ASD and ID aged 2-10 years displayed significantly more target behavior and less off-task behavior in the first condition than in the second condition.

Results of both studies suggest that children with ASD and ID experience difficulties in understanding other's intentions and that better understanding is associated with a higher developmental age and better performance on tests measuring early social communication and language.

Next to impairments in social interaction and communication, children with ASD have particular difficulty with behavioral flexibility. For the purpose of measuring situations in which individuals with developmental disabilities display a lack of behavioral flexibility, Green and colleagues developed the Behavioral Flexibility Rating Scale (BFRS) and the BFRS-r. The study described in Chapter 4 assessed psychometric properties of the BFRS-r as completed by 76 direct care staff members and 56 parents for 70 children with developmental disabilities. Factor analysis revealed three factors (i.e., Flexibility towards objects, Flexibility towards the environment, and Flexibility towards persons) and results of several analyses indicated an excellent internal consistency and good intra-rater and inter-rater reliability of the total scale.

These data suggest that the BFRS-R may provide a reliable rating of behavioral flexibility when used by direct-care staff and parents of children with developmental disabilities.

In Chapter 5, this scale was used to assess behavioral flexibility in 111 children with ASD and ID (87 autism; 24 PDD-NOS; IQ range 10.59-72.67) and results were compared to those of 65 children with ID only. Their age ranged between 2:7 and 9:11 years. Results showed that behavioral flexibility in children with ASD plus ID was predicted by autism severity, developmental age, and initiating social interaction. Behavioral flexibility seems to influence emotional and behavioral problems and maternal stress, but not adaptive behavior.

As compared to mothers with typically developing children, maternal stress in mothers of children with ASD and ID is heightened. Since this has a negative impact on children's treatment outcome, aim of Chapter 6 was to investigate child variables predicting maternal stress. Hundred four mothers of children with ASD and ID completed the Dutch version of the Parental Stress Index every six months over a period of two years and maternal stress remained stable over time. No relationship was found between maternal stress and children's developmental age and IQ, receptive and expressive language, adaptive behavior, severity and subtype of ASD, behavioral flexibility, and early social communication (i.e., joint attention, behavioral requests, social interaction). However, children's emotional and behavioral problems did predict maternal stress; emotionally reactive behavior, withdrawn behavior and attention problems explained a third of the variance in maternal stress.

Part II: Effectiveness of Early Behavioral Treatment

In Part II, the effectiveness of early intensive behavioral treatment was determined. First in Chapter 7, a meta-analysis was conducted to investigate the effectiveness of early intensive behavioral treatment in young children with ASD. There were 11 studies with 344 children with ASD. Quality of studies was assessed using the Downs and Black Checklist. Experimental groups who received early intensive behavioral treatment outperformed the control groups on IQ, non-verbal IQ, expressive and receptive language and adaptive behavior. Differences between the experimental and control groups were 4.96-15.21 points on standardized tests. These results strongly support the effectiveness of early intensive behavioral treatment.

Next, two studies with a pre-test post test control group design were conducted to evaluate the effectiveness of low intensity behavioral treatment (4-10 hrs per week) supplementing (pre)school services in children with ASD and ID as compared to treatment as usual. Treatment was implemented in preschools (i.e., daycare centers) and schools for children with ID and a discrete trial teaching approach was used.

In the study described in Chapter 8, twelve children in the treatment group were compared to 22 children receiving regular intervention. All children had ASD and ID and were between 3-6 years old. At pre-treatment, both groups did not differ on chronological age, developmental age, diagnosis and level of adaptive behavior. Eight months into treatment, children receiving behavioral treatment displayed significantly higher developmental ages

and made more gains in adaptive behavior than children from the control group. No significant differences between groups were found on autistic symptom severity and emotional and behavioral problems.

Since the sample size of the first study was relatively small, the assessment battery rather restricted, the intervention period relatively short and no measures on treatment fidelity were collected, a second study was conducted in which 40 children with ASD and ID (mean chronological age: 5;3 years; mean developmental age: 1;11 years) participated. Over a period of 2 years, standardized data were collected on developmental age, adaptive behavior, interpersonal relations, play, language development, autism severity, early social communication skills, maternal stress, behavioral flexibility, and emotional and behavioral problems. Also, data of the behavioral treatment program and on treatment fidelity were collected. These results are described in Chapter 9 and replicate the results found in the first study. Children in the treatment group outperformed the control group on IQ, developmental age, adaptive behavior, interpersonal relations, play and receptive language, and less autistic symptoms were seen in treatment group. Following intervention, no differences between groups were found on expressive language, behavioral flexibility and maternal stress. Progress in developmental age, adaptive behavior, interpersonal relations, play and receptive and expressive language was clinically and reliably significant for the majority of the treatment group.

Part III: Health Care Conditions Required for Early Behavioral Treatment

The two major barriers in implementing early behavioral treatment in the community are related to the funding and quality of behavioral programs. Due to the intensity of many programs (20-40 hrs per week for 3 years with a low child staff ratio), implementation costs are high and can be controversial. However, in children with ASD these programs may result in improved cognitive, adaptive and social functioning and reductions in autism severity and behavioral problems. Consequently, these children may require less professional care, reducing their lifetime costs. In chapter 10, estimated costs for education, (supported) work and (sheltered) living for individuals with ASD in the Netherlands are applied in a cost-offset model. The provision of behavioral treatment may result in long-term savings, which are approximately € 1,103,067 from age 3-65 years per individual with ASD. Extending these costs to the whole Dutch ASD population, cost savings of € 109.2 to € 182 billion have been estimated, excluding costs associated with inflation. Hence, the estimated costs of early behavioral treatment (estimated at € 100,000) seem to overweigh long-term savings.

Behavioral treatment is associated with variability in treatment outcome and quality of treatment delivery may contribute to this. The study described in Chapter 11 investigates the relation between therapist characteristics and the quality of the treatment provided during Discrete Trial Teaching (DTT), an important component of early behavioral treatment. DTT

was provided at a pre-school for children with ID and 70 sessions between 22 therapists and 35 children with ASD and ID were videotaped and analyzed. Data on therapist's attitude towards individuals with a disability, therapist's personality traits, and perceived relationship between therapist and child were also collected. Procedural fidelity was high and significantly related to therapist's attitude towards individuals with a disability, therapist's openness to experience, and perceived relationship between therapist and child. Therapists with high procedural fidelity tended to have a more positive attitude towards individuals with disabilities on the cognitive dimension, a more negative attitude towards individuals with disabilities on the affect dimension, lower levels of openness to experience, and perceived the relationship between themselves and the child as less positive.

In sum, this thesis shows that children with ASD and ID display behavioral characteristics that may negatively impact their development. Without appropriate treatment, the prognosis of children with ASD and ID is poor. However, early behavioral treatment may accelerate the development of children with ASD and ID and may results in reduced autism severity and behavioral problems. In addition, behavioral treatment may result in long-term savings. However, behavioral treatment needs to be of high quality, which seems associated with therapist's characteristics.

Samenvatting

[Summary in Dutch]

In dit proefschrift worden resultaten gepresenteerd met betrekking tot (1) de gedragskenmerken van kinderen met een autisme spectrum stoornis (ASS) en een verstandelijke beperking (VB), (2) de effectiviteit van vroegtijdige gedragstherapie en (3) de voorwaarden voor het aanbieden van vroegtijdige gedragstherapie aan kinderen met ASS en een VB. De onderzoeksopzet en de resultaten van de studies uitgevoerd in dit proefschrift worden in dit hoofdstuk samengevat.

Deel I: Gedragskenmerken van kinderen met ASS en een VB

In deel I worden vijf studies beschreven waarin enkele gedragskenmerken van kinderen met ASS en een VB onderzocht worden. Doel van de studies beschreven in hoofdstuk 2 en 3 is om vast te stellen in hoeverre kinderen met ASS en een VB in staat zijn om de intenties van anderen te interpreteren. Aan de studie omschreven in hoofdstuk 2 namen 100 kinderen met ASS en een VB tussen de 2 en 10 jaar oud deel om te onderzoeken of zij tijdens een experimentele taak in staat waren om de communicatieve intenties van de onderzoeker af te leiden. Tijdens deze taak dienden de kinderen een tastbaar voorwerp (een stukje koek, snoep of speelgoed) te vinden dat de onderzoeker in een van twee boxen op tafel verstopte. Tijdens vier trials in de oefenconditie verstopte de onderzoeker het voorwerp in het zicht van het kind. Na de oefenconditie volgden acht experimentele trials, waarin de onderzoeker het voorwerp buiten het zicht van het kind verstopte en het kind het voorwerp diende te vinden aan de hand van de non-verbale hulp geboden door de onderzoeker (wijzen in de eerste conditie; blikrichting in tweede conditie). Slechts 56 kinderen presteerden boven kansniveau in de oefenconditie (drie of vier van de vier trials correct) en werden in de analyses geïncludeerd. Ondanks grote individuele verschillen, presteerden de kinderen gemiddeld boven kans in de eerste conditie (wijzen), terwijl zij onder kans presteerden in de tweede conditie (blikrichting). Gebruikmakend van een binomiale test (7 van de 8 trials correct, p =.03), presteerde slechts 14% van de kinderen boven kans in beide condities.

In hoofdstuk 3 werd een aangepaste versie van Meltzoff's *behavioral re-enactment* taak gebruikt, waarbij de onderzoeker in de experimentele conditie gedrag laat zien dat niet overeenstemt met het doel van de taak. In deze conditie doet de onderzoeker binnen 20 seconden drie (niet-succesvolle) pogingen om het kind het doelgedrag te laten zien. Voor het kind wordt het uiteindelijke doel van de taak niet zichtbaar. In de controle conditie doet de onderzoeker in ongeveer 20 seconden drie maal het doelgedrag behorend bij de taak voor. Kinderen met ASS en een VB laten meer doelgedrag zien in de controle conditie dan in de experimentele conditie.

Aan beide studies namen geen controlegroepen deel, waardoor het prematuur is om conclusies te trekken over het begrip van intenties door kinderen met ASS en een VB. Toch lijken beide studies te suggereren dat kinderen met ASS en een VB moeilijkheden ervaren op deze taken. Een beter begrip van intenties lijkt gerelateerd aan een hogere ontwikkelingsleeftijd en betere prestaties op testen die vroege sociale communicatieve vaardigheden en taal meten.

Naast beperkingen in de sociale interactie en communicatie, ervaren kinderen met ASS problemen met betrekking tot de gedragsflexibiliteit. Om situaties vast te stellen waarin personen met een ontwikkelingsstoornis gedragsinflexibiliteit laten zien, ontwikkelde Green en collega's de Behavioral Flexibility Rating Scale (BFRS) en een gereviseerde versie hiervan (BFRS-r). De studie beschreven in hoofdstuk 4 stelt de psychometrische kenmerken van de BFRS-r vast aan de hand van scores van 76 groepsleiders en 56 ouders die de BFRS-r invulden voor 70 kinderen met een ontwikkelingsstoornis.

Factoranalyse onthulde drie factoren (flexibiliteit ten aanzien van objecten, flexibiliteit ten aanzien van de omgeving en flexibiliteit ten aanzien van personen). Resultaten van diverse analyses laten een excellente interne consistentie, een goede intra- en interbeoordelaarsbetrouwbaarheid van de totale schaal en convergente validiteit met de Sameness Questionnaire zien. Deze data suggereren dat de BFRS-r een betrouwbare indicatie van gedragsflexibiliteit geeft wanneer deze wordt ingevuld door groepsleiders en ouders van kinderen met ontwikkelingsstoornissen.

In de studie omschreven in hoofdstuk 5 wordt gedragsflexibiliteit met de BFRS-r gemeten bij 111 kinderen (84 jongens) met ASS (87 autisme; 24 PDD-NOS) en een VB (IQ range 10.59-72.67). Resultaten worden vergeleken met die van een controlegroep bestaande uit 65 kinderen met een VB (42 jongens). De chronologische leeftijd lag tussen de 2;7 en 9;11 jaar/maanden. Resultaten laten zien dat gedragsflexibiliteit in kinderen met ASS en een VB voorspeld wordt door de ernst van ASS, de ontwikkelingsleeftijd en het initiëren van sociale interactie. Gedragsflexibiliteit lijkt van invloed te zijn op gedragsproblemen en opvoedingsstress, maar niet op adaptief gedrag.

Verschillende studies laten zien dat de opvoedingsbelasting van moeders met een kind met ASS en een VB hoger is dan opvoedingsbelasting van moeders met kinderen die zich normaal ontwikkelen. Aangezien een hoge opvoedingsbelasting van ouders een negatieve invloed heeft op de behandeluitkomsten van hun kinderen, werd in de studie omschreven in hoofdstuk 6 onderzocht welke kindvariabelen van invloed zijn op de opvoedingsstress van moeders. Honderdvier moeders met een kind met ASS en een VB vulden over een periode van twee jaar halfjaarlijks de verkorte Parental Stress Index in. Gedurende deze periode bleef de opvoedingsstress stabiel. Er werd geen relatie gevonden tussen opvoedingsstress ervaren door de moeders en de ontwikkelingsleeftijd en het IQ, de receptieve en expressieve taal, adaptief gedrag, ernst en subtype van ASS, gedragsflexibiliteit en vroege sociale communicatieve vaardigheden (joint attention, gedragsverzoeken en sociale interacties) van het kind. Echter, de gedragsproblemen van het kind voorspelden de opvoedingsstress van de moeders. Emotioneel-reactief gedrag, teruggetrokken gedrag en aandachtsproblemen voorspelden ongeveer een derde van de variantie in opvoedingsstress.

Deel II: Effectiviteit van vroegtijdige gedragstherapie

In deel II zijn drie studies uitgevoerd om de effectiviteit van vroegtijdige gedragstherapie te onderzoeken. Ten eerste is in hoofdstuk 7 een meta-analyse uitgevoerd om de effectiviteit van vroegtijdige intensieve gedragstherapie bij jonge kinderen met ASS nader vast te stellen. Vervolgens zijn in hoofdstuk 8 en 9 de resultaten beschreven van twee longitudinale studies naar de effectiviteit van laag intensieve gedragstherapie.

In de meta-analyse werden elf studies geïncludeerd waaraan 344 kinderen met ASS deelnamen. Bij de start van de behandeling waren de kinderen tussen 33.56 en 65.68 maanden oud en hadden zij een IQ tussen 27.52 en 76.53. De experimentele groepen ontvingen gemiddeld 12.5 uur tot 38.6 uur vroegtijdige gedragstherapie gedurende tien maanden tot meer dan twee jaar. Controle groepen ontvingen minder intensieve gedragstherapie (minder dan 10 uur per week), 13-29 uur per week eclectische behandeling, oudergestuurde gedragstherapie of reguliere behandeling. Kwaliteit van de studies werd gemeten met de Downs and Black Checklist. Kinderen uit de experimentele groep lieten meer vooruitgang op IQ, nonverbaal IQ, adaptief gedrag en expressieve en receptieve taal zien dan de controle groepen. Verschillen tussen de experimentele en controle groepen lagen tussen de 5-15 punten op gestandaardiseerde testen. Deze resultaten onderbouwen de effectiviteit van vroegtijdige intensieve gedragstherapie.

Aangezien er nauwelijks onderzoek is uitgevoerd naar de effectiviteit van laag intensieve gedragstherapie, zijn er twee studies met een pre-test-post-test controlegroep design uitgevoerd om de effectiviteit van laag intensieve gedragstherapie (4-10 uur per week) als aanvulling op het reguliere programma op het kinderdagcentrum of de school te bepalen. Behandeling werd uitgevoerd op kinderdagcentra en scholen, waarbij het accent lag op Discrete Trial Teaching (DTT).

In de studie omschreven in hoofdstuk 8 werden 12 kinderen uit de behandelgroep vergeleken met 22 kinderen die de reguliere behandeling op een kinderdagcentrum ontvingen. In de voormeting verschilden beide groepen niet met betrekking tot kalenderleeftijd, ontwikkelingsleeftijd, diagnose en niveau van adaptief gedrag. Na acht maanden behandeling hadden de kinderen in de behandelgroep gemiddeld een significant hogere ontwikkelingsleeftijd en lieten zij een grotere vooruitgang zien op adaptief gedrag dan de kinderen uit de controle groep. Geen significante verschillen werden gevonden met betrekking tot gedragsproblemen en ernst van autisme.

Gezien de relatief kleine steekproefgrootte, de beperkte testbatterij, de relatief korte interventieperiode en het gebrek aan data met betrekking tot de integriteit van behandeling, is er een tweede studie uitgevoerd waaraan 40 kinderen met ASS en een VB (gemiddelde kalenderleeftijd: 5;3 jaar; gemiddelde ontwikkelingsleeftijd: 1;11 jaar) deelnamen. Gedurende een periode van twee jaar werden gestandaardiseerde data verzameld op het gebied van ontwikkelingsleeftijd, adaptief gedrag, interpersoonlijke relaties, spel, taalontwikkeling, ernst van autisme, vroege sociaal communicatieve vaardigheden, opvoedingsstress, gedragsflexibiliteit en emotionele problemen en gedragsproblemen.

Ook werden gegevens met betrekking tot het behandelprogramma en de procedurele betrouwbaarheid verzameld.

De resultaten zijn beschreven in hoofdstuk 9 en repliceren de resultaten gevonden in de eerste studie. Naast dat kinderen uit de behandelgroep meer vooruitgang laten zien op IQ, ontwikkelingsleeftijd, adaptief gedrag, interpersoonlijke relaties, spel en receptieve taal, is er bij hen een grotere afname in autistische symptomen dan bij de kinderen in de controlegroep. In de behandelgroep werd eveneens een afname in gedragsproblematiek zichtbaar. Na twee jaar behandeling werden echter geen verschillen gevonden tussen de groepen op expressieve taal, gedragsflexibiliteit en opvoedingsstress. De vooruitgang op ontwikkelingsleeftijd, adaptief gedrag, interpersoonlijke relaties, spel, receptieve taal en expressieve taal was voor het merendeel van de kinderen in de behandelgroep klinisch en statistisch significant.

Deel III: Voorwaarden voor vroegtijdige gedragstherapie

De twee belangrijke barrières voor de implementatie van effectieve vroegtijdige gedragstherapie zijn de financiering en de kwaliteit van de programma's. Door de intensiteit van veel programma's (20-40 uur per week gedurende 3 jaar met een laag kind-staf ratio) zijn de kosten van deze programma's hoog en controversieel. Deze programma's kunnen echter leiden tot een verbeterd cognitief, adaptief en sociale functioneren en een afname in autistische kenmerken en gedragsproblemen bij kinderen met ASS. Dit kan er in resulteren dat personen met ASS op de lange termijn minder behandeling en professionele zorg nodig hebben, hetgeen de levenslange kosten geassocieerd met ASS kan verminderen.

In hoofdstuk 10 worden de geschatte kosten voor onderwijs, (begeleid) werken en (begeleid) wonen voor personen met ASS in Nederland toegepast in een kosten-batenmodel. Hieruit blijkt dat het aanbieden van vroegtijdige gedragstherapie (gemiddeld 33 uren per week gedurende twee jaar) kan resulteren in een lange termijnbesparing van ongeveer € 1.103.067 per persoon met ASS van 3 tot 65 jaar. Wanneer deze kosten vertaald worden naar de Nederlandse ASS populatie kunnen kostenbesparingen oplopen van € 109.2 tot € 182 miljard, exclusief de kosten gerelateerd aan inflatie. Kortom, de lange termijn besparingen lijken de kosten gerelateerd aan vroegtijdige gedragstherapie (geschat op € 100.000) te overschaduwen.

Hoewel vroegtijdige gedragstherapie over het algemeen effectief is, verschillen de behandeluitkomsten binnen en tussen studies. Omdat kwaliteit van behandeling hieraan mogelijk bijdraagt, wordt in de studie omschreven in hoofdstuk 11 de relatie tussen kenmerken van de trainer en de kwaliteit van de behandeling tijdens Discrete Trial Teaching (DTT) onderzocht. DTT is een belangrijke component van vroegtijdige gedragstherapie. DTT werd aangeboden op een kinderdagcentrum voor kinderen met een VB en 70 sessies uitgevoerd door 22 trainers bij 35 kinderen werden gefilmd en geanalyseerd. Er werden eveneens data verzameld met betrekking tot de persoonlijkheid van de trainer, de attitude ten opzichte van personen met een beperking en de relatie tussen de trainer en het kind. De procedurele betrouwbaarheid was hoog en significant gerelateerd aan de attitude van de trainer ten opzichte van personen met een beperking, de openheid voor ervaringen en de relatie tussen de trainer en het kind.

Trainers met een hoge procedurele betrouwbaarheid hebben op de cognitieve component een positievere attitude ten opzichte van personen met een beperking, op de affectieve component een negatievere attitude ten opzichte van personen met een beperking, staan minder open voor ervaringen en ervaren de relatie tussen de trainer en het kind als minder positief.

Tot slot

Samenvattend laat dit proefschrift zien dat kinderen met ASS en een VB gedragskenmerken laten zien die hun ontwikkeling negatief kunnen beïnvloeden. Zonder adequate behandeling is de prognose van kinderen met ASS en een VB slecht. Vroegtijdige gedragstherapie kan leiden tot een versnelling van de ontwikkeling en een afname van autistische kenmerken en gedragsproblemen. Daarnaast leidt de toepassing van vroegtijdige gedragstherapie mogelijkerwijs tot besparingen op de lange termijn. Voorwaarde hiervoor is dat er behandeling van hoge kwaliteit wordt aangeboden, hetgeen onder andere samen lijkt te hangen met kenmerken van de trainers.

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Nienke Peters-Scheffer

Curriculum Vitae
List of Publications

Curriculum Vitae

Nienke Peters-Scheffer is geboren op 15 augustus 1982 te Warnsveld. Tijdens de HBO Propedeuse Toegepaste Psychologie wordt haar interesse voor de studie Pedagogische Wetenschappen en Onderwijskunde gewekt. Na het behalen van haar propedeuse aan de Radboud Universiteit Nijmegen besluit zij haar studie een jaar te onderbreken om in weeshuis Hogar Alalay in La Paz (Bolivia) met straatkinderen te werken. In februari 2006 rondt Nienke haar studie Pedagogische Wetenschappen af binnen de afstudeerrichting Leren en Ontwikkeling met de specialisatie Verstandelijke Handicaps. Tijdens haar studie werkt Nienke in Logeerhuis Doetinchem van Stichting Zozijn.

Vanaf juni 2005 is Nienke binnen Driestroom, een instelling voor ondersteuning en zorg aan mensen met een verstandelijke beperking, werkzaam als onderzoeks- en gedragskundig medewerker bij een vroegtijdig interventieproject gericht op kinderen met een autisme spectrum stoornis en een verstandelijke beperking. Binnen dit project behandelt zij kinderen volgens gedragstherapeutische principes en heeft zij geholpen deze interventie verder vorm te geven. Daarnaast zet zij een studie naar de effectiviteit van dit project op.

In maart 2007 wordt Nienke bij de Radboud Universiteit Nijmegen aangesteld om deze studie vervolg te geven in een promotieonderzoek, hetgeen geleid heeft tot onderhavig proefschrift. Tijdens haar promotieonderzoek werkt zij eveneens als docent bij de vakgroep Orthopedagogiek en begeleidt zij werkgroepen, diverse Masterstages en Masterscripties.

Sinds april 2012 is Nienke aangesteld als universitair docent bij het Behavioral Science Institute en de vakgroep Orthopedagogiek van de Radboud Universiteit Nijmegen. Daarnaast werkt zij als senior onderzoeker en gedragskundige bij Driestroom. Haar onderzoekswerkzaamheden richten zich momenteel op de screening van jonge kinderen met een verstandelijke beperking op een autisme spectrum stoornis, het aanleren van vaardigheden bij kinderen en jongeren met een verstandelijke beperking en de ontwikkeling van scholingsprogramma's voor ouders en groepsleiding van mensen met een verstandelijke beperking.

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