

Effects of Intensive Behavioural Treatment and a focused Imitation
Intervention for young Children with Autism Spectrum Disorder

One plus One means More

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Abstract

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The general aim of this thesis was to investigate the effectiveness of interventions offered to children with autism attending the Habilitation services. Another aim was to investigate the efficacy of a new intervention within the setting of Swedish Habilitation services, namely Imitation Responding.

In Study I the aim was to investigate if habilitation services, after early screening, showed a positive outcome. The interventions were *Intensive Behavior Treatment (IBT)*, and *Eclectic Interventions* encompassing a selection of methods based on TEACCH or communication methods such as ComeAlong.

In Study II the aim was to explore if an intervention based on imitation, *Imitation Responding (IR)*, could function as a complement to a comprehensive *IL/IBT*, which is treatment as usual at the Habilitation services.

In Study III the aim was to examine if generalized joint attention had been developed in the group of children with autism that received *IR* followed by *IL/IBT* and the group of children with autism that received *IL/IBT* only.

Study I investigated the impact of intelligence on outcome of interventions from Habilitation service in a naturalistic study. The actual comprehensive program is Intensive Learning for young children with autism, an IBT intervention (acronym: IL/IBT) offered as regular or modified version. The Modified IL/IBT had only parents as trainers and low weekly intensity. The Regular IL/IBT had both parents and preschools staff as trainers and with planned training of 25 hrs/weekly. The third group encompassed Eclectic interventions. Child variables at initial assessment (IQ in particular) accounted for most of the outcome variance regardless of intervention group. Although the mean adaptive composite score did not change significantly between start of intervention and follow-up, the variance increased significantly. This underscores the need for regular monitoring of the child's functioning and developmental gain/loss during the intervention period and the necessity to consider a change or modification of the program in use.

Study II comprised of 40 children with Autism Spectrum Disorder (ASD), after receiving diagnosis they were randomized to either IL/IBT, which is standard treatment in child habilitation service, or the new treatment Imitation Responding (IR). IR is a focused program directed to increase the child's ability to respond to other's communication and to learn from others by utilizing a Being Imitated strategy, where the therapist imitates the child. All children got their first intervention during 12 weeks.

The IR group, mean age 41.6 months at start of intervention received treatment on average 2.2 hours/week while the IL/IBT group, mean age 40.1 months at start of intervention, received 14.4 hours/week. Before and after treatment all children were assessed with The Psycho Educational Profile - revised edition (PEP-R), and Vineland Behavior Adaptive Scales, second edition (VABS- II). Seven measures from the PEP-R and VABS-II, covering language, play, interpersonal relations, and visuo-motor imitation, were used as outcome measures. Results showed a significant increase in 6 out of 7 subscales for both intervention groups but no significant difference was noted in any of the subscales when comparing the groups. Only the IR group had a significant gain on the measure of interpersonal relations. We interpret the findings as support for IR to be used as a complementary intervention strategy alongside comprehensive programs such as IL/IBT.

Study III examined if the intervention IR or if IL/IBT could have an effect on the development of IJA in children with autism. One group received IR for three months followed by IL/IBT for 12 months, whereas the second group received only IL/IBT for the whole 15 months period. Two measures of IJA were used: A gaze shift score and a sum score of pointing and showing.

IJA did not change during the first three months of treatment, nor were any significant differences between the interventions groups noted. However, at the end of the 15-month-long intervention period the gaze shift score had increased significantly for the IR group followed by IL/IBT training in contrast to the IL/IBT only group. No significant change was noted for pointing and showing. Our results indicate that IR with the Being Imitated strategy might be useful if implemented early as a “start-up” – intervention, which is followed by an IL/IBT treatment.

Keywords: Autism, Interventions, Preschool Children, Imitation, IBT

Svensk sammanfattning

Syftet med avhandlingen var att utvärdera interventionsmetoder för små barn med autism och detta undersöks i tre studier.

I den första studien har syftet varit att utvärdera om vanligt förekommande interventionsmetoder inom habilitering minskar färdighetsbrister hos barn med autismspektrumstörningar, ASD (Autism Spectrum Disorders). Barnen rekryterades i samband med undersökning på BVC då de var cirka 2,5 år. De uppvisade då en annorlunda utveckling av kommunikativ förmåga, språklig förmåga och social utveckling. Vid en senare utredning bedömdes de uppfylla diagnoskriterier för autism. I samband med detta har barnen remitterats till Habiliteringen där de erbjuds behandling och stöd av skilda slag.

Huvudsakligen erhöll barnen två typer av behandling, antingen Intensivinlärning för små barn med autism, en IBT metod (IL/IBT) eller eklektiska metoder. Sedan början av 2000-talet har mångsidiga program som bygger på Tillämpad Beteende Analys (TBA) använts. I Västra Götalands-regionen har IL/IBT funnits sedan 2010. Programmet har utvecklats inom habiliteringen och är anpassat till svenska förhållanden.

IL/IBT har erbjudits i två varianter, Regulerad (föräldrar och förskola med handledning 2 ggr/månad där föräldrar och förskola har målet att träna barnet 20 – 25 timmar/vecka) och Modifierad (enbart föräldrar med handledning 1 gång/månad där föräldrar tränar barnet ca 10 timmar/vecka). Det andra alternativet omfattar eklektiska interventioner och har vanligtvis getts som TEACCH, kommunikationsprogrammet ”KomIgång”, eller stöd för bostadsanpassning och hjälp till föräldrarna att hantera beteendeproblem. Behandlingarna har utförts av multidisciplinära team.

I samband med diagnosticering och vid uppföljning två år senare, har barnen testats av ett oberoende team och på en klinik med detta uppdrag skilt från behandlingsteam.

Resultaten av studie I, visade att effekten av behandling varierade inom samtliga behandlingsgrupper. Det förekom barn med signifikant förbättrad adaptiv förmåga och generell funktionsnivå men också barn med låg eller utebliven förbättring av funktioner. Effekten av samtliga gruppers interventioner var associerade med intellektuell förmåga vid start.

Syftet med studie II har varit att utvärdera effekten av IR och IL/IBT i tolv veckor, direkt efter att barnet erhållit diagnos. Barnen randomiserades till behandling med antingen IR eller IL/IBT efter genomförd mätning av deras kognitiva, språkliga och sociala utveckling. Sju delskalor ur PEP-R och VABS-II, omfattande språk och sociala förmågor användes för att undersöka om interventionen hade någon effekt.

Metoden IR erbjöds till 21 barn, deras genomsnittsålder var vid start 41,6 månader. IL/IBT omfattade 19 barn som vid start var 40,1 månader.

IR bygger på en imitationsstrategi där den vuxne imiterar barnets beteenden, gester och vokala uttryck, i kontrast till IBT som erbjuder barn imitation och övningar där

barnet förväntas agera efter den vuxnas önskan eller begäran. IL/IBT är lämpat för barn som har förmåga att vilja och kunna ta delvis ögonkontakt och att vara motiverade för att vara tillsammans med och göra som andra. Barnen som fick IR rapporterades ha en genomsnittlig träning av 2,2 timmar per vecka. Interventionen gavs i förskolemiljö av en förskollärare eller barnskötare. Barnen i IL/IBT -gruppen hade en genomsnittlig träningstid av 14,4 timmar/vecka (hem och skola). En jämförelse mellan de två behandlingsgrupperna visade ingen signifikant skillnad mellan effekten av metoderna vid den korta uppföljningen. Båda behandlingsgrupperna har ett likartat resultat med en signifikant förändring för 6 av 7 delskalor. IR visade sig ha effekt på barnets förmåga att uttrycka sig och förstå tal, imitera, leka med andra i nivå med IL/IBT under en kort behandlingsperiod. Slutsatsen är att IR är ett användbart behandlingsalternativ till IBT under en 12-veckors period och som en första behandling.

Studie III utvärderade om IR, Being Imitated strategin, kunde utveckla barnens förmåga till att ta initiativ (ögonkontakt) med den vuxne för att få den vuxne att uppmärksamma det som barnet är intresserat av (Initierad Joint Attention) (IJA). Behandlingsgrupperna utgjordes av samma barn som i studie II d.v.s. de erhöll IR eller IL/IBT under 12 veckor men därefter erbjöds alla barn IL/IBT under tolv månader dvs. samtliga barn erhöll totalt 15 månaders behandling. Fördelningen av behandling var 13 barn som fått både IR och IL och 9 barn som endast fått IL/IBT. Bortfallet av barn berodde bl.a. på tekniska problem eller på att några barn vägrade medverka.

För att mäta barnets utveckling avseende delad uppmärksamhet (joint attention) användes Early Social Communication Scales (ESCS) som bland annat mäter två aspekter av delad uppmärksamhet: Dels förmågan att initiera uppmärksamhet (eng. initiation joint attention, IJA) med andra och dels förmågan att svara på andras initiativ till uppmärksamhet (eng. responding to joint attention, RJA). I avhandlingen studeras enbart IJA, d.v.s. att initiera ögonkontakt under en aktivitet eller för att visa något eller att använda gester som att peka eller visa något av intresse. Under de första 12 veckorna observerades ingen signifikant förändring av dessa initierande beteenden till delad uppmärksamhet hos någon av behandlingsgrupperna.

Vid uppföljande mätning 12 månader efter inledande behandling visade resultaten på en signifikant ökad förmåga att initiera och växla ögonkontakt hos de barn som fått IR initialt och därefter IL/IBT till skillnad mot barnen som enbart fått IL/IBT. När det gällde hur barnen använde pek/visa-beteende konstaterades ingen signifikant förändring varken mellan behandlingsgrupperna eller mellan för- och eftermätning. Resultatet visar att IR, Being Imitated strategin, är användbar som en inledande intervention för att utveckla initiering av delad uppmärksamhet.

Avhandlingens studier visar att de interventioner som vanligen ges i ordinarie habiliteringsverksamhet och deras effekt är avhängigt barnets kognitiva nivå vid start av

behandling. Detta talar för att utvärdering av kognitiv nivå måste ske och vara en del av varje behandlingsinsats för varje enskilt barn.

Den kortsiktiga behandlingen (tre månader) visar att IR med Being Imitated strategin och IL/IBT har jämförbara effekter på barnens språkförståelse och talutveckling, samt på de sociala färdigheterna imitation, lek och relationer till andra, trots att den totala träningsmängden för IR är mycket mindre än IL/IBT.

Vid långtidsuppföljning 12 månader efter avslutad behandling med IR och IL/IBT visade det sig att de barn som deltagit i både IR och i IL/IBT hade en signifikant säkerställd utveckling av initierad delad uppmärksamhet till skillnad mot barnen som enbart fått IL/IBT. Därför bör Interventionen IR erbjudas till barn med ASD under en inledande kort tidsperiod och följas av en längre tids behandling med IL/IBT.

List of abbreviations

- ABA Applied Behavior Analyses
- ADHD Attention Deficit Hyperactive Disorders
- ANOVA Analysis of Variance
- ASD Autism Spectrum Disorders
- CA Chronological age
- CDC Centers of Disease Control
- C-GAS Children's Global assessment Scale
- CI Confidence Interval
- CNC Child Neuropsychiatric Clinic
- DCD Developmental coordination disorders
- DSM Diagnostic and Statistical Manual of Mental Disorders
- DTT Discrete Trail Training
- EIBI Early Intensive Behavior Intervention or IBT
- EP Epilepsy
- ES Effect Size
- ESCS Early Social Communication Scales
- ESDM Early Start Denver Model
- ESSENCE Early Symptomatic Syndrome Eliciting Neurodevelopmental Clinical Examinations
- IBT Intensive Behavior Treatment
- IDD Intellectual Developmental Disorder
- IJA Initiating Joint Attention
- IL Intensive Learning for young children with autism
- IR Imitation Responding
- JA Joint Attention
- NET Natural Environment Training
- PECS the Picture Exchange System

PEP Psycho Educational Profile
RCT Randomized Control Trail
RIT Reciprocal Imitation Training
RJA Responding Joint Attention
T Time
TBA Tillämpad Beteende Analys (eng. ABA)
TEACCH Treatment and Education of Autistic and Related Communication-
Handicapped Children
VABS Vineland Adaptive Behavior Scales
WMA World Medical Association
WPPSI Wechsler preschool and Primary Scale of Intelligence
ZPD Zone of Proximal Development

Preface

This thesis is based on the following three articles:

I. Spjut Jansson, B., Miniscalco, C., Westerlund, J., Kantzer, A-K., Fernell, E., & Gillberg, C. (2016). Children who screen positive for autism at 2.5 years and receive early intervention: prospective naturalistic two-year outcome study. *Neuropsychiatric Disease and Treatment*, *12*, 2255-2263. doi:10.2147/NDT.S108899

II. Spjut Janson, B., Heimann, M., & Tjus, T. Comparing one brief and one comprehensive early intervention program for children with an autism spectrum disorder. (Submitted manuscript).

III. Spjut Janson, B., Koch, F-S., Tjus, T., & Heimann, M. Being Imitated improves joint attention in young children with ASD receiving IBT. (Manus in preparation).

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Birgitta Spjut Janson
Gothenburg, April, 2017

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Introduction

Children with autism spectrum disorders (ASD) are characterized by social communication deficits, rigid behaviour and learning difficulties. The nature of these children's learning difficulties can be understood as the effect of impaired abilities in communication, language, social inter-relatedness, motor coordination, attention and regulation of activity. Research and clinical experience (Volkmar, Rogers, Paul, Pelphrey, & 2014) have led to more and varied interventions, with the aim to increase the children's abilities to develop skills that support adaptation to the environment.

This thesis begins with a presentation of autism as a syndrome, the change of prevalence over decades, and the common co-morbidity with other neurodevelopmental disorders. The second part presents the ecological model formulated by Bronfenbrenner (1979), which outlines the perspectives of and relations to various levels of society. This will be followed by perspectives of learning, Piaget's individual developmental perspective, Skinner's behavioral theories of learning and, finally, the social theories of learning of Vygotsky and Bandura.

The third part of the thesis focuses on children's developmental processes on the basis of the theory of intersubjectivity, which encompasses the perception of social stimuli as well as the emergence of social understanding as evidenced by imitation and joint attention. The presentation contains both typical development and selected findings related to autistic behaviour. The fourth part includes a general presentation of a new intervention method, Imitating Responding, as well as more established programs for children with autism Intensive Behaviour Therapy (IBT) and the method Intensive Learning and some eclectic methods. The fifth part comprises ideas of evaluation in different studies. The last section comprises the three empirical studies conducted plus an integrative discussion.

Autism Spectrum Disorders according to DSM-5

The American Psychiatric Association (APA) (Washington, DC, 1994) publishes *Diagnostic and Statistical Manual of Mental Disorders* (DSM). In their fourth edition (DSM-IV, 1994) they used the umbrella term Pervasive Development Disorders for the central features of abnormal or impaired development in social interaction and communication, and restricted repertoire of activity and interest. The diagnostic terminology was Autistic Disorder, Asperger's Disorder, Retts Syndrome and Childhood Disintegrative Disorder. This manual has been used in the studies. In 2013, DSM-5 folded all subcategories into one umbrella diagnosis of Autism Spectrum Disorder (ASD). Today "autism" stands for a heterogeneous group of neurodevelopmental conditions defined by behavioural symptoms. ASD involves a broad spectrum of deficits in social, verbal and non-verbal skills, according to DSM-5 (APA, 2013). The disorder is marked by qualitative impairment in communication as well as social impairments and the appearance of restricted or routinized behaviours and interests (APA, 2013). The symptoms begin to appear before three years of age, but researchers and

clinicians still debate whether ASD can be reliably diagnosed even earlier on (e.g. Eikeseth, 2008; Webb, Jones, Kelly, & Dawson, 2014). Nevertheless, both international (NICE, 2013) and Swedish (SBU, 2013) guidelines stress the importance of extended treatment programs for young children with autism and of starting treatment as soon as problems are identified.

Prevalence of Autism

The prevalence of autism has increased throughout its history. To begin with, autism was considered an extremely rare disorder, but in one decade starting from 1994 the number of 6- 21-year-old individuals receiving services for autism in the US increased from 22,664 to 193,637 Centers of Disease Control (CDC, 2012). CDC reports an estimate of one child identified with ASD in every 68 children. Similar changes have been reported in Sweden, and for the last ten years an increasing number of young children below four years of age have received an ASD diagnosis (Hedvall et al., 2013). In Gothenburg, for example, 48 children were given an ASD diagnosis before the age of three in 2010, compared to only two children five years earlier (Nygren et al., 2012). The prevalence of 0.95% Confidence Interval (CI) at 95%: 0.82 - 1.08 for the autism symptom phenotype was reported by Lundström, Reichenberg, Ankarsäter, Lichtenstein and Gillberg (2015), who state that its prevalence among children in earlier times may have been underestimated, since it was reported to be 0.7% as early as the 1980s. This confirms autism as being one of the most common childhood development disabilities (Volkmar et al., 2014). ASD is almost three to four times more frequent among boys than girls in population cohorts and 5-14:1 in clinical settings (Gillberg, Cederlund, Lamberg, & Zeijlon, 2006; Kopp, Beckung, & Gillberg, 2010), and girls who receive an early diagnosis are more likely to have additional intellectual or behavioural problems (Dworzynski, Ronald, Bolton, & Happé, 2012).

Some factors that could impact the number of children being diagnosed with ASD are new diagnostic criteria, findings that girls are fulfilling criteria for ASD who were previously unidentified, and treatment guidelines that recommends early screening. One such factor is the change of criteria in DSM-V (APA, 2013) where Asperger's diagnosis has been included in the overall spectrum diagnosis of autism. Also the fact reported by Fernell, Eriksson and Gillberg (2013) that in recent studies girls and women have been "wrongly" diagnosed with other neurodevelopmental disorders than ASD (Fernell et al. 2013) is likely to have affected the recent change in prevalence.

Today's increased prevalence could also be related to the increasing evidence that early interventions have positive effects (e.g. Reichow, Barton, Boyd, & Hume, 2013) and guidelines recommending that interventions start as early as possible (CDC, 2017). This has resulted in outlined programs of early screening and highlighted symptoms of toddlers and young children.

Aetiologies of Autism Spectrum Disorder

ASD is diagnosed with criteria that rely on behavioural symptoms and the degree of impairment is most varied. Even if the criteria for ASD are defined by categories of symptoms, the symptoms within each category can vary between individuals. Variation also holds for aetiology and pathology states (Coleman & Gillberg, 2012). The clinical presentation of ASD varies according to severities within these symptom categories but is also to a considerable degree dependent on the associated neurodevelopmental disorders.

Family and twin studies show that ASDs are highly heritable neurodevelopmental disorders (Werling & Geschwind, 2015; Coleman & Gillberg, 2012). Despite the convincing genetic basis for ASD, a definite cause remains unknown in most individual cases. In the last 40 years, very significant progresses have been made in the understanding of genetics of ASD, but there are many aspects of the genetics that remain largely unknown (Bourgeron, 2016).

Several hundred candidate or risk genes have now been implicated in ASD (Maynard & Manzini, 2017). In about 20% of the cases with ASD, a specific underlying diagnosis can be expected (Coleman & Gillberg, 2012, p. 173). There are several genetic/chromosomal syndromes associated with ASD. In about 1% or more of individuals with ASD specific diseases such as Fragile X, Tuberous sclerosis and Rett syndrome, can be identified (Coleman & Gillberg, 2012, p. 172). Copy number variants refer to structural, submicroscopic variations, deletions and duplications of submicroscopic DNA segments, changing the chromosomal architecture and gene expression. There are certain microdeletion/microduplication syndromes associated with ASD.

There are also many other prenatal risk factors in ASD, intrauterine infections, autoimmune disorders and intrauterine toxic exposures affecting the fetal brain (Coleman & Gillberg, 2012).

In some children with ASD a perinatal aetiology can be identified, e.g. in children born extremely preterm (gestational age before 27-28 weeks) with risks related to the immature brain. An accelerated brain volume of young toddlers has also been reported by Courchesne (2002).

Environmental factors cannot explain many cases on their own, but in interaction with genes they may be associated with an increased risk for autism. Such environmental factors may be present during pregnancy, for example infections, drugs, toxic substances and exposure to stress (Beverdorf, et al., 2005).

Comorbidities & functional deficits

Autism is often combined with other neuropsychiatric and neurodevelopmental conditions such as intellectual developmental disorder, language disorder, developmental coordination disorder (DCD), tics, attention-deficit/hyperactivity disorder (ADHD), and epilepsy (EP) (Coleman & Gillberg, 2012).

When the first symptoms emerge early in life these conditions are also referred to as ESSENCE (Early Symptomatic Syndrome Eliciting Neurodevelopmental Clinical Examinations; Gillberg, 2010). Children who receive an early diagnosis often show, in contrast to typical children, a development characterized by an uneven cognitive profile illustrating differences between perceptual and language skills (Hagberg, Miniscalco & Gillberg, 2010). Among children diagnosed with ASD at a young age, the rate of associated intellectual and developmental disabilities is about 50% (Hedvall, et al., 2013). Problems with imitation (e.g. Gopnik, Capps, & Meltzoff, 2000; Vivanti & Rogers, 2011) and joint attention (Carpenter, Pennington & Rogers, 2002) are two of the earliest disabilities detected in children with autism. Several studies have found a strong correlation between imitation ability and joint attention in autistic children, even when controlling for developmental level (Escalona, Field, Nadel, & Lundy, 2002; Rogers, Hepburn, Stackhouse, & Wehner, 2003). Disturbances in the development of both verbal and non-linguistic social communication behaviours are also common (Charman & Campbell, 1997; Stone, Ousley, Yoder, Hogan, & Hepburn, 1997). Furthermore, language skills in children with ASD at ages 2-4 are the best predictors of adaptive skills at age 5 and of outcome at adulthood (Lord, Risi, & Pickles, 2004), which pinpoints the importance of early diagnosis and intervention in children with ASD.

Theories of learning

Every child needs support to learn and achieve skills. However children with ASD may require more individual support of both their social development and their academic learning than typical children. ASD greatly impacts their ability to interact with others and learn from others to give them information of activities and interactions. To more fully understand how intervention based on learning could promote positive development this dissertation will describe different perspectives of learning. This presentation includes the ecological model formulated by Bronfenbrenner (1979), which outlines the perspectives of and relations to various levels of society. This will be followed by perspectives of learning, Piaget's cognitive developmental perspective: Skinner's behavioral theories of learning and, finally, the social theories of learning of Vygotsky and Bandura.

Ecological theories of learning

Bronfenbrenner's (1979) ecological theory of human development has often been presented as a framework for identifying the ecological levels that are highly integrated and significant for research on children with disabilities and their families. In this thesis, relations within the context of child, parents, family, preschool, and habilitation service are in focus. Bronfenbrenner's theory comprises five distinct and nested concentric systems: Micro, Meso, Exo, Macro and Chrono. These different systems exert multidirectional influences between the child and the environment. The microsystem includes the child's immediate environment, that is, the family and the many ways the family exercises its influence, as in

parenting style, health, and nutrition, parents' responsibility for carrying out interventions from habilitation services, demographic and socioeconomic status, child care, health service, peers and neighborhood. The mesosystem includes connections between two or more systems, for example social, health care and preschool systems, as well as school organizations. It might also include connections with larger structures of the community with only an indirect relation to the child and the family. The exosystem contains regional policies and decisions that have an impact on community social services, health care and habilitation services, with an indirect impact on the child. These policies and decisions could be legislations for access to special education schools, interventions or programs for specific diagnoses provided by the Health care and Habilitation services. The macrosystem comprises aspects of layers including cultural characteristics, national political decisions influencing factors such as the physical environment and the integration of children with special needs into preschools, as well as systems that provide integration to enable people with disabilities to participate on equal terms in society. The fifth, the chronosystem relates to a child's experience of environmental events and to transitions that occur throughout life.

The demands made by the microsystem on parents to young children with autism, or children at a low developmental level, represent both typical and multiple roles connected to their children or their children's diagnosis. A tremendous strain and perhaps the largest transformation of parents' role is their need to receive help and support in their first and often early concern for their child's problem. Another role comprises the evaluation of their own appropriate support with a view to developing sustainable teamwork with professionals.

Simultaneously with struggling with conflicting feelings of sadness, anger and disappointment, parents must deal with their new insight into their child. Remington et al. (2007) found that fathers' level of depression increased when acquiring further knowledge of the child's need of support. Norlin and Broberg (2013) concluded that mothers of children with disabilities were more depressed than mothers of typical children. Parents are forced to shoulder an extensive collaborative role as partners between home, preschool, habilitation services and other social welfare instances within the mesosystem (Margalit, Al-Yagon, & Kleitman, 2006). In order to supply their child with optimized education, health and support, many parents find that their role has turned them into "advocates" when taking part of the individualized plans. This role requires parents who are informed and educated about autism, as well as in educational laws, available services and how to negotiate on behalf of their child (Grindle, Kovshoff, Hastings, & Remington, 2009). Parents also have a central role in finding treatment and in cooperating with supervisors as co-therapists during periods of intervention, or being educated in specialized knowledge about their children's dysfunction. Their role in everyday life also involves mastering of strategies or tasks that enable them to support their child to acquire new skills. On a general level, both parents and children need long-term involvement and intensive interactions with supportive teams (Volkmar et al., 2014). The efficacy of interventions depends on clinicians' understanding

of parental variables like stress, well-being and affect (Grindle et al., 2009). Interventions for young children should be family-centered, prioritizing the children and their families as well as making use of the children's natural environment, like their home or community settings (Volkmar et al., 2014).

Social theories of Learning

To create a base for understanding parts of early development and learning in typical children and the intervention strategies investigated in this thesis the theoretical work of Bandura, Vygotsky Piaget and Skinner, is presented. Theories of children's learning have to some extent created contradictory perceptions of child competencies: the socio-cognitive perspective formulated by Vygotsky and Bandura, proposing the child's innate ability to learn, versus Skinner's behavioristic theory stressing the importance of environmental stimuli and children's learning by operant conditioning. Piaget, in turn, examined the individualistic perspective of cognition in his systematic studies of children's developmental processes.

Social constructs of Learning

The Social Learning Theory (SLT), as presented by Bandura (1977), states that behaviour is learned from direct experience or the observation of others through the process of observational learning, i.e. it is a process of external influences on the responsiveness to other people's actions. A further analysis clarifies two roles or phases of imitation behaviour. The acquisition of the modeled behaviour is followed by a performance of imitative responses (Baldwin & Baldwin, 2001). Either roles, or phases, are influenced by the competence and likeability of the model, the nature and complexity of the modeled behaviour, as well as contextual cues. Bandura (1971) proposed that the human capacity for learning by observation has contributed to skills such as symbolic representation, self-regulation processes, and the cognitive capacity to acquire large units of behaviour patterns. Identification occurs with another person (the model) and involves taking on (or adopting) the behaviours, values, beliefs and attitudes observed in this person. For this reason, Bandura (1971) stressed the balance of similarity between observer and model. Visual similarity, such as shape and color, or gender, has to be present to some extent and is of major significance for young children. In his view, the basic function of reinforcement is to strengthen responses by informative and motivational aspects that can be both external and internal and contain positive and negative values. Self-reinforcement systems are developed by the child in interaction with others and become autonomous with development and thus available to use independently in future social experiences.

According to Social Learning Theory (Bandura, 1977), the environment is an arena that inevitably permeates any individual and to which children have to adapt their responses. The emitted behaviour, in turn, partly shapes the environment, which in turn will

influence the behaviour. Thus, a two-way dynamic process of causality is created with the environment being as influential as the behaviour it controls.

Vygotsky's theoretical work emanated from his belief that children are cognitively active and use imitation to discover and conquer their social world (Vygotsky, 1978), "In learning to speak, as in learning school subjects, imitation is indispensable" (Vygotsky 1934/1986, p. 188). He was concerned with cultural and historical contexts and with their relation to interpersonal (micro-level) interactions and to intra-psychological concepts. According to Vygotsky's (1978) first aspect of cultural development, any psychological function of children's development appears simultaneously on a social, a psychological, and a biological trajectory. These trajectories influence attention, memory, and the formation of concepts of language and cognition. A second important concept in Vygotsky's theory (1978) is the "zone of proximal development" (ZPD), which involves a potential for cognitive development from birth. ZPD is a social process where competent adults promote children's social behaviour. The optimal development for a child depends on the quality of this social interaction. According to Vygotsky (1978), developmental processes first occur as relations between adult and child and are then internalized and develop into the language of the child. Imitation strategies support children to perform actions that increase their own capacities but nevertheless out of ZPD (Vygotsky, 1978).

A third major theorist is Piaget. He examined cognitive development as a progressive reorganization connected to biological maturation and environmental experience (Inhelder & Piaget, 1958). Piaget's (1951) main theories refer to the mental representation of intelligence, with 'schema' defined as a set of linked mental representations of the child's environment enabling the child to understand and respond to situations and stimuli. He designated a cognitive theory where the transition from new learning to the generalization of skills at the individual level is presented as a series of steps between the accommodation and assimilation levels. Piaget (1962/1951) linked imitation development to the ability of inner representation. He further proposed that the infant's ability of systematic imitation starts at the age of 4-8 months, a proposal which has been questioned by e.g. Meltzoff and Moore (1983), and Nagy and Molnar (2004). At that time of development Piaget found imitation to contain both new observed and already accommodated (learned and spontaneous) behaviours. In its later development the child's own interest in achieving a goal can be used as a "starter" for imitations of movements. During the child's first year, intentionality was found to be the activator of imitation, and children develops, according to Piaget, imitation from trial and error explorations.

Behavioristic constructs of learning

Behaviorism emphasizes observable and concrete behaviour in the study of humans. The hypothesis, which is based on the logical positivism position of knowledge, was endorsed by Skinner (1958), who advocated direct observation, experiments and present empirical

results. Interpretations were rejected as speculations, while stimulus-response learning was presented as the basis of all behaviour, including thoughts and emotions.

Skinner (1958) postulates that stimuli play a major role, both as reinforcing and as creating the opportunity or signal, for subsequent behaviour. Skinner's behavioural theory is essential for understanding the intervention of Intensive Learning for young children with autism (IL), especially the way imitation is implemented in this thesis. Skinner argued that instruction, as in learning, should be defined as stimuli, which he placed on an equal footing with motivation, drive, and creativity. He claimed that achieving mastery of models helps new learners or observers to learn. Further, Skinner (1953) considered imitation to occur when the observer performed a response similar to the models using the term "inverse imitation" when a reinforcement straightens the child's behaviour to respond differently from the action of the model by, for instance, taking the other part in a dance. Since learning implies a change in behaviour, such a change is related to the consequences. In further reading expressed as Imitation on Demand (Nadel, 2014): this imitation could be compared with eliciting imitation in Rogers and Williams (2006). In a meta-study of imitation Edwards (2014) explains eliciting imitation as imitation where also prompt strategies could be used, and where also not verbalized or directed instructions could be used for example in experiments of structured interactions with a demonstrator. The difference between elicit and demand is then explained as, during the Imitation on Demand the child had to relate to a known person during daily sessions, whereas elicited imitation mostly are used in laboratory setting with an unknown experimenter. Both builds on the ability to respond on demand or respond on command of a demonstration. To make changes possible, manipulating consequences is primary. Skinner elaborated the terms of reinforcement, as positive or negative, primary or secondary reinforces, and set up rules for how to use these (Baldwin & Baldwin, 2001). Through the proper use of teaching strategies like shaping (adding new behaviour to fit a situation by approximation), the teacher can promote the development of new skills.

Sociocognitive theories of learning

Attention

Attentional capacities are fundamental for what we perceive and how we decide to act. Without attention, a child is hardly or never able to learn. Typical infants' attendance is based on their face processing, on visual and auditory perception. Posner and Rothbart (2007) identify three main aspects of attention. All three processes – alertness, orientation, and executive attention - are found to be part of the ability of self-regulation and control: *Alertness* is the primary condition, indicating readiness for reacting to incoming stimuli, while *orientation* is the response due to the selection of incoming sensory signals, and *executive attention* is the ability to monitor responses, thoughts and feelings. These three processes of attention develops early in life, with alertness and orientation becoming

functional within the first year, while executive attention develops more slowly (Posner & Rothbart, 2007). An example of alertness and orienting response in early infancy is the observation that infants prefer human-like faces, which is seen as an innate mechanism (Cassia, Turati, & Simion, 2004). Another example is that infants at birth recognize human voices and select their mother’s voice on account of its melody (Gomes, Molholm, Christodoulou, Ritter, & Cowan, 2000). At six months an infant is able to detect the prosodic pattern of human language and at twelve months a child reacts to phonemes that are not typical of the language it is usually exposed to (Kuhl, 2004).

Habituation

Thompson (2009) and Thompson and Spencer (1966) describe habituation as the process of ignoring familiar and predictable stimuli with a general preference for novelty. The brain works as a filter whose selection processes are built on the recognition of stimuli. Stimuli that are perceived as predicted, by previous experience or predominating environmental features draw no further attention, in contrast to novel stimuli (Gollisch & Meister, 2010). The process of habituation is always more rapid when stimuli are simple and slower when faced with more complex or varied stimulus patterns. Dishabituation of a stimulus reduces the probability to become intrinsically learned. According to Rankin et al. (2009), behavioural habituation of stimulus decreases with the use of external reinforcement.

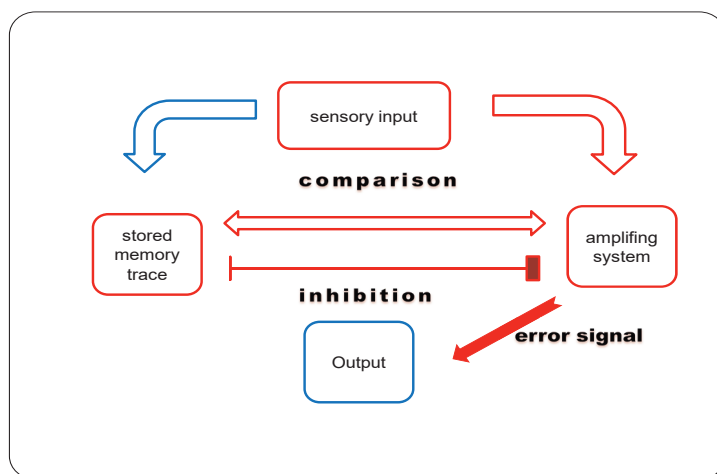


Figure 1. A model of the role of inhibitory feedback of habituation (Sokolov, 1963; Rankin et al, 2009).

The model of inhibitory feedback in habituation, see Figure 1, outlines the way a new sensory signal takes to become a stored, specific stimulus. This model contains a feedback inhibition which differentiates, as a neural comparator, new stimuli from familiar ones. Only the parts that differ between the new stimulus and the stored memory are transmitted

by afferent signals to subsequent layers in the brain. This systematized comparison starts the activation of the amplifying system whereby new and familiar stimuli are compared. The amplifying system also controls for the activation of storing new stimuli as a centrifugal inhibition and contains an error signal that avoids the restoring of familiar and already stored stimuli (Ramaswami, 2014). Habituation is closely connected to cognition (Ramaswami, 2014) and hence to interventions. This model of habituation, which is related to attention and predictive coding, allows interventions to control the mapping of stored memory that involves learning from others and enables motivation for sharing attention with others as in imitation (Vivanti & Rogers, 2014). They mention imitation as a foundational process for developing understanding and social learning in young children.

Social perception

Intersubjectivity has been formulated by Delafield-Butt and Trevarthen (2015) as the rapid cultural development of newborn infants, involving self-other awareness and engagement in imitation, joint attention and communication. Early psychologists like Vygotsky (1978) and Piaget (Inhelder & Piaget, 1958; Piaget, 1962) underlined the potential of learning by imitation as something unique for human beings. Intersubjectivity is inspired by research utilizing observations and experiments of the non-verbal communication of infants, young children, and their parents.

This section presents five levels of social connectedness in early development (Rochat & Passos-Ferreira, 2009) and the assumed chronological ages associated with the actual context, behaviour and process of these levels (see, Table 1). Behaviours and processes start at “typical ages”, and behaviours develops over time, some of which remain unequal to their former variant.

The first level of social connectedness (Rochat & Passos-Ferreira, 2009) relates to neonatal mirroring. Trevarthen & Aitken (2001) proposed the theory of intersubjectivity as neonatal imitation incurred by innate motives and mirroring face-to-face actions which lie beyond instinctive behaviours for communicating intricately with other humans. Through the ability of imitation infants are closely connected to the social world and relate to learning, recognition and imaging. Experimental evidence for the ability to perform various facial gestures, i.e. tongue protrusion and mouth opening, was first found by Meltzoff and Moore (1977).

Trevarthen (1979) and Trevarthen and Aiken (2001) included newborns as being especially motivated, and Meltzoff (2005) proposed the “like me” hypothesis based on an intrinsic neural mechanism. He claimed that children’s learning about other people and their behaviour was equal to their learning of themselves and their own behaviour. Gallese (2003) argued that these basic mirroring processes are foundations connected with social binding mechanisms for the purpose of engaging the correspondence of others already from birth. Meltzoff and Moore (1997) suggested that infants were able to coordinate three processes to produce an imitative response: a) the correspondence of body parts, b)

active intermodal mapping as a representational system, and c) an action system enabling infants younger than one month to begin imitations (Nagy, 2008). As these activities lead children to make their first active correspondence to perceptual stimulation, Meltzoff and Moore (1997) argued that a modal or supramodal coding exist at birth as an “Esperanto of perception” (Nadel, 2014, page 28) converting all kinds of sensory information into terms of movement.

Table 1. Presentation of five levels of social connectedness in early development, with their typical ages, actual contexts, behaviours and processes (adapted from Rochat & Passos-Ferreira, 2009).

Levels of Intersubjectivity					
	Mirroring	Primary	Secondary	Tertiary	Ethical stance
Age	Birth	2 months	9 months	20 months	From 4 years
Context	Face-to-face engagement	Reciprocal dyadic changes	Triadic exchanges about things	Triadic exchanges about the value of things	Decision regarding the value of things, what is right vs. what is wrong
Behaviour	Imitation	Proto-conversation, social expectations	Joint attention, social referencing	Self-recognition, embarrassment, use of possessives, claim of ownership, prosocial behaviours	Sense of property, sharing, distributive justice, ToM
Process	Automatic stimulation	Emotional co-regulation	Intentional communication and intentional co-experiences	Projection and identification of self onto others	Valuing negotiation with others, narration, meta-representation of reputation

The second level of social connectedness (Rochat & Passos-Ferreira, 2009) starts around the age of two months. This is comparable to what Trevarthen (2011) refers to as primary intersubjectivity. He (Trevarthen, 1979; 2011) formulated “proto-conversation” as the behaviour that emanates from the consciousness of both self-acquaintance and the acquaintance of others. Trevarthen and Aiken (2001) stated that the ability of infants to imagine and reflect the thoughts and emotions of others is developed long before they understand language fully. During this phase the infants’ interest in and conversation with others develop further. Their gradual understanding of sharing experiences with others, as well as their imitative and mutual responses, increases (Rochat & Passos-Ferreira, 2009; Trevarthen, 2011). These responses include the interactive detection of embodied and

interpersonal experiences from the attunement of the choreographed and facial expressions, bodily movements, motor signature and embodied emotional expressions of others (Rochat, 2001; Gallagher, 2005). Three-month-old children are able to a higher degree to regulate and coordinate head, arm and hand movements and to use these body parts more actively for imitation.

Meltzoff and Moore (1992) stated that during their third month children has a drop-off in their imitative behaviour, due to new social abilities such as vocalizing and smiling. Heimann (1998) proposed that this pattern could be connected to the development of more controlled behaviour responses. Later, at six months of age, children recur to performing imitations of single and isolated actions with familiar objects (Meltzoff, 1985).

The third level of social connectedness (Rochat & Passos-Ferreira, 2009) starts approximately at nine months of age, which coincides with Trevarthen's secondary phase of intersubjectivity (2011). At this age the child's attention changes from a dyadic relation to include an interest in the surroundings and in other children. This entails an established relationship between action and object (Meltzoff, 1995). The imitation of unfamiliar (not previously experienced) actions with objects is observable by the age of nine months (Meltzoff, 1985), although some claim that early steps of this ability can be observed already by six months, as evidenced by studies of deferred imitation (e.g., Barr, Dowden, & Hayne, 1996; Heimann & Nilheim, 2004). This more elaborated step of imitation (Meltzoff, 1995) comprises the child's use of goals, where simple goals will at first be followed by imitation of actions in two steps (for example using a truck - pulling the truck and then putting another object onto the truck) or multi-steps of familiar actions (clothing, washing or eating rituals). The ability of imitation develops strongly during the first to the second year of life, comprehending development in general and spontaneous imitation in particular (Delafield-Butt & Trevarthen, 2015). Masur and Olson (2008), who examined imitation in the play of mothers and children, found the highest level of imitation to occur between 13 and 21 months of age. This finding is in line with Nadel and Dumas (2014), who defined the same age as forming the peak of imitative interactions. These children were eager learners of meaning and experiences with others, as in creative manipulations of objects in new combinations. Trevarthen (2011) dedicated "confident and confiding kind of mutual attention and sharing of experiences with the aid of others' guidance" (page 184) as the potential of ensuring developmental changes in self-representation and interaffect-related activities during this period. At 14 months the child can manage the imitation of interrupted actions, followed by the complex imitation of chained actions in three steps at 18 months of age (Bauer, 2004). Furthermore, Trevarthen (2005) argues, in line with Piaget (1962), that children's development of symbolization starts with representations of their own and other people's communicational acts, comprehending both intentional and affect-regulation abilities.

Joint attention constitutes the ability of two individuals to share focus on an outside object or event. This skill is achieved when a child alerts another individual to an object by eye-gazing, pointing or other verbal or non-verbal indications (Scaife & Bruner, 1975). The first sign of the joint attention ability can be found when infants are around eight to ten months old (Bakeman & Adamson, 1984; Charman & Campbell, 1997; Tomasello, 1995). Joint attention is described as a two-way loop with the capability to represent both one's own and another person's goal-related behaviour (Tomasello, Carpenter, Call, & Behne, 2005). This process of interaction by compiling information from internal and external monitoring functions as an expander of cognitive development. Bakeman and Adamson (1984) have proposed that joint attention consists of two forms. The first, supported joint attention, refers to an event where the child is more passive and where parents attend to engage their child in toys or other interesting objects. The second form, coordinated joint attention, involving activity from the child in both the adult and the object, is developed three months later about the time of the child's first birthday (Bakeman & Adamson, 1984; Mundy & Newell, 2007). This ability is based on orientation and perceptual attention skills. A specific aspect of the development is the child's ability to initiate joint attention, which is important for developing communication skills. Initiating joint attention develops attention and communication in mutual harmony (Mundy & Newell, 2007), following the child's experiences of social interactions in incidental situations during the first nine months of its life. In a recently published study by Brooks and Meltzoff (2015) it was demonstrated that a child's ability to follow someone's gaze plays a major role in its linguistic and social-cognitive development. It was also found that this ability predicted children's production in terms of mental states, which in turn can be related to the development of mentalizing. These findings support the hypothesis that preverbal features such as joint attention and requesting behaviour, like pointing, play a significant role to achieve the development of children's language skills.

The *fourth level of social connectedness* develops from 20 months of age, according to Rochat and Passos-Ferreira (2009), and is parallel to Trevarthen's and Aiken's concept of tertiary intersubjectivity (2001). Now children have developed a beginning sense of self in relation to other people and are constantly comparing new situations to earlier ones (Rogers & Williams, 2006). During this period children develop their ability for compromising within the negotiation and reward values of proposals. They are also able to begin to make comparisons of subjectivity, intentionality and affectionality. Children's ability to pretend also begins to develop when they start to disguise their own emotions. In addition, pro-social behaviours increase with acts of giving or providing help (Rochat & Passos-Ferreira, 2009).

Both Rogers and Williams (2006) and Nadel (2014) state that children at 18 months of age use imitation of a more complicated nature and more frequently in the social context. Imitation serves two functions, as a learning vehicle and as a means of nonverbal

and preverbal communication (Nadel, 2014). Nadel adds that children reduce their use of imitative behaviour at about the age of 42 months, since imitation is no longer used for communication to the same extent, and children sometimes perceive other people's imitation as mocking.

During this period children are found to be intentional imitators. From being focused on behaviours alone, they are now also capable of understanding cause and effect (the goal) of imitation behaviour (Meltzoff, 1995). At this age both responding to and initiating joint attention in conjunction with experiences can be organized to support intention and interpreting goal understanding (Mundy, 2007). Initiating joint attention explores children's spontaneous social communication, i.e. a triadic intentional communication as well as the ability to negotiate about the values of how things develop. The advantage of effective joint attention can be described as using the adult's gaze to identify objects, while the adult's labelling of the connection between object and word will evolve to an insight into the symbolic function of language. Beuker, Rommelse, Donders and Buitelaar (2013) reported that the earlier the children handled joint attention skills, the more positive was the observed effect on language skills.

The *fifth level of social connectedness*, also called Ethical Stance by Rochat and Passos-Ferreira (2009), starts approximately between three and five years of age, when children have captured an explicit understanding of morals by the value of objects and situations. They have now achieved a new level of self-reciprocity concerning the inner representation of others and themselves, namely a Theory of Mind (Baron-Cohen, 1991). Imitation learning is still useful, especially with playmates. Nadel-Brulfert and Baudonnire (1982) found that during this period children were able to spontaneously combine pretend play, joint attention, and imitation in play interactions. For typical developing children this period also signifies rapid language development (Bates, Benigni, Bretherton, Camaioni, & Volterra, 1975).

Imitation and ASD

As early as 1972 DeMeyer and colleagues identified imitative impairments for children with autism. Since then imitation and imitation deficits have been further investigated with the result that today's research possesses a broader perspective of the typical and atypical aspects of imitation and autism. To analyze and classify imitation it is important to distinguish between different types of copying behaviour (Byrne & Rosson, 1998; Want & Harris, 2002).

This structure (Table 2) for analyzing copying behaviour and summarizing it into categories (Vivanti & Hamilton, 2014) uses distinctions between the goal and the means of an action. Where emulation involves copying the goal of an action, mimicry means copying an action, and imitation requires copying both the goal and its means. Imitation as the manipulation of objects and attention to the model was investigated by Amano, Kezuka and Yamamoto (2004), who found that typical infants had a higher preference for looking at

the model's hands than at the model's face and eyes. Senju and Csibra (2008) state that both children's attention and degree of precision increased by direct gaze, while both Trevarthen and Aiken (2001) and Vivanti and Rogers (2014) report that the degree of precision in imitation is less important than timing.

Table 2. Different copying behaviour in typically developing children, according to Vivanti and Hamilton (2014, page 280)

	Emulation	Imitation	Mimicry
Typology	The observer copies the goals of an action.	The observer copies both the goals and the means of the observed action.	The observed acts are un-intentionally and spontaneously matched by the model.
Behaviour	Observing a person sitting on the floor listening leads to pretending to sit in a chair listening. The observer wants to achieve the same end state as the model (listening).	Performing similar actions as those being modelled.	For instance, changing arm positions when sitting in front of somebody.

Three topics of imitation have been investigated in children with autism. The first aspect encompassing attention and imitation was studied by Vivanti, Nadig, Ozonoff and Rogers (2008), who found that children with ASD looked less at the demonstrator than typical children. Vivanti and Rogers (2011) reported that children with ASD were found to devote less time to looking at conditions of non-meaningful actions than to meaningful actions, while typical children looked longer at the demonstrator's face. Vivanti and Rogers (2011) suggest that children with ASD have problems with selective attention by failing to detect cues from the demonstrator's face, which complicates the imitation process. This will affect their possibility to understand complex goal-directed actions. Vivanti and Rogers (2011) also argue that an impaired ability to interpret the demonstrator's social cues will lead to less imitation performance. The second topic, frequency of imitation, has been referred to in several studies. Most studies report lower rates of spontaneous imitation of actions involving objects and of gestures to children with ASD compared to typical children (e.g. DeMyer et al., 1972; Ingersoll, 2008; Brown & Whiten, 2000), but there also exist studies with the opposite findings (Nielsen, Slaughter, & Dissanayake, 2013; Rogers, Young, Cook, Giolzetti, & Ozonoff, 2008). The third topic encompasses studies investigating children with ASD and their spontaneous imitation. This is investigated by a retrospective analysis of home videos produced before children had received their diagnosis. The reported findings are that children show lower rates during their first two years (Maestro et al., 2001; Osterling & Dawson, 1994). In an experimental study by Heimann, Nordqvist, Strid, Connant Almrot and Tjus (2016) it was found that children with ASD showed less elicited

imitation compared to typically developing children matched for mental and language age, but that no difference was noted for spontaneous imitation.

Joint Attention and ASD

Children with low or absent joint attention are found to be at risk for autism (Bruinsma, Koegel, & Kern Koegel, 2004). Rogers and Dawson (2010) argue that joint attention is related to four processes (visual perception - shifting attention - motor behaviour - emotion) that are highly coordinated, and if the biological base behind these processes becomes afflicted it can have a negative influence on its development. Liebal, Colombi, Rogers, Warneken and Tomasello (2008) found that children with autism showed a weaker performance on joint attention and imitation tasks, which prevented them from fully participating in cooperative exercises. Strid, Heimann, Gillberg, Smith and Tjus (2013) reported evident problems of both types of Initiating Joint Attention (IJA) behaviours such as gaze shift and point/show in a group of young children with autism, which confirmed earlier observations by Charman et al. (2003). In contrast, Pickard and Ingersoll (2014) discovered that gaze shift and point/show of children in the age of 22 - 93 months measured by ESCS were not associated with each other. Instead, IJA point/show and responding JA were both associated with and predictors of imitation. IJA behaviors point/show were more connected to social-communication skills than gaze-shift.

Strategies of Imitation

Intervention studies using imitation with the aim to increase social communication in children with autism tend to differ in how imitation is implemented as an educational or therapeutic tool. Children who have less ability to engage in imitation acts, like play and games, have a higher risk to develop less social-cognitive skills (Vivanti & Rogers, 2014).

Two main different approaches of utilizing imitation are discerned when used in experiments or in clinical training: either the therapist imitates the child's behaviour - *Being Imitated strategy* - or the adult (supervisor or trainer) asks the child to repeat a modelled behaviour – an *Imitation on Demand* or *Elicited imitation strategy*.

The *Being Imitated* is a strategy aimed to increase social interest among children with autism developed by Nadel (2000). She argued for an imitating the child strategy, where the experimenter will respond and imitate the child during play. A recent article by Contaldo, Colombi, Narzisi and Muratori (2016) provided renewed support for the Being- imitated strategy for improving social gazes, proximal social behaviors and play skills.



Picture 1. The trainer imitates the child in all his actions: manipulation of body and objects, sounds, words, gestures, mimicry, or other expressions of importance.

In the Being Imitated strategy the child leads and the adult follows the child's intentions (goals) (see Picture 1) such as manipulations with objects, gestures, vocals and affects (Nadel, 2000; Ingersoll, 2008). The adult does not imitate behaviour that can hurt the child or the adult. The main aim of this strategy is to increase the child's ability to initiate communication and responsiveness to other people's actions. This strategy is supported by experimental studies that have investigated the effect of brief but focused interventions (Field, Field, Sanders & Nadel, 2001; Escalona, Field, Nadel, & Lundy, 2002; Heimann, Laberg & Nordøen, 2006). The Being Imitated strategy used in these studies increased children's eye contact, coordinated joint attention, communication and turn-taking as well as social imitation (e.g. Field et al., 2001; Ingersoll, 2008). The Being Imitated strategy used in interventions include some targeted programs like *The Scottish Center for Autism Preschool Treatment Programme* (Salt, et al., 2001, 2002), *Reciprocal Imitation Training* (RIT) (Ingersoll, 2010), and *L'imitation au service de l'autisme* (Nadel, 2014). Another related type of intervention, *Intensive Interacting*, presented by Nind and Hewett (2001), makes use of a similar approach for elderly people who are deaf-blind or suffer from dementia, accompanied with impaired communication.

In the other strategy, *Imitation on Demand or Elicited imitation* (Lovaas, 1981/2003), the trainers model their actions with the instruction: "do like me" (stimulus) to enable the child to follow (response). Imitation on Demand is often the initial strategy of imitation in Applied Behaviour Analyses (ABA) programs, used for scaffolding goals to teach the child a skill. It increases children's skills to learn from models so that they can develop their ability to manipulate their object use as well as their adaptive and language skills. The goal

of using Imitation on Demand is to increase children's learning from listening to instructions and making the requisite decisions. Ingersoll and Meyer (2011) found this strategy to have a positive effect on vocabulary. Comprehensive programs developed from ABA programs include, e.g., Lovaas (1981/2003, *The ME book*), *Companion* (Fovel, 2002), *A work in progress* (Leaf & McEachin, 1999), and *Behavioural Intervention for Young Children With Autism* (Maurice, Green, & Luce, 1996), all including Imitation on Demand as part of their training schedule. Whereas Leaf & McEachin (1999) recommended imitation with object manipulation and Lovaas (1981/2003) suggested the use of gross motor movements.

Early Interventions

The main purpose of early intervention is to provide learning opportunities to increase social, cognitive and adaptation skills for young children with developmental problems during a period of maximal brain plasticity (Volkmar et al., 2014). A variety of educationally based interventions have been developed as means of remediating the characteristic deficits of autism. In this thesis the main focus is on Imitation Responding (IR) and Intensive Learning (IL).

Start of learning processes to deal with generalization

The generalization of skills is a highly valued and expected ability, as an effect of the child's increased capacity and of the efficacy of the intervention. The relation between Discrete Trail Training (DTT) (Smith, 2010) and Natural Environment Training (NET) (Sundberg & Partington, 1999) could be explained as the variation of format that increases the teaching of new tasks by instruction to include spontaneous actions and generalized knowledge (Sundberg & Partington, 1999). The DTT format, which has been used and developed by Lovaas (1981/2003), is supposed to be a start-up format. It is intended to establish good learning behaviour, such as attention, appropriate responding, and good learning positions for the child like staying or sitting down while receiving instructions, to develop tolerance to demands from other's and respond positively to other's initiatives (Sundberg & Partington, 1999). In this format the therapist could arrange and control the stimuli and reinforcement. DDT also includes organizing a room where all necessary stimuli can be exposed with less stimulus distraction. The easiest DTT procedure is based on tasks presented and planned together with a trainer (parents or teacher) for a demarcated and allocated period of time. There is another aspect of the use of dedicated room for children's training, considered by Salt et al. (2001) that concerns the role of the adult. He argued that adults are more open-minded towards following the child without restrictions by the use of a dedicated room. The aim of making the environment optimally available for learning is the required need for repetition in a situation where the offered stimuli can be controlled independently of which model of intervention the child is offered.

NET is focused on communication and comprises a strategy where the adult gives an adequate "teaching moment" to child initiations and allows teaching across contexts and in interactions. NET could be used to explore the child's own motivation for or interest in facilitating therapy (Sundberg & Partington, 1999). The NET format should be offered when skills are to be generalized, using naturalistic procedures like the incidental teaching method, the time-delay prompt, and milieu language teaching (Sundberg & Partington, 1999). During the initial stages of NET, the trainer focuses on exercises or rigged situations that develop the child's needed repertoire by offering interesting objects or activities (Sundberg & Partington, 1999). Rogers and Williams (2006) and Nadel (2014) have found that children, when growing up, learn to act in new environments more similarly to the way

they act in familiar ones. NET also enables children with autism to develop spontaneous verbal behaviour in the context of motivating activities (Sundberg, Partington, & Sundberg, 1998). By combining the DTT and NET formats individualized teaching or interventions can be offered. NET focuses on everyday events and is used to support strategies that scaffold the child's communication abilities with the aim to provide positive experiences. In supporting the child a balance has to be achieved between DTT and NET (Sundberg et al., 1998). The process of learning could be more effective if the therapist support parents and preschools teachers to use the DTT format restrictively. This means to change format with respect to the child's social and cognitive capacities.

How to stimulate and motivate young children

When working with toddlers and young children the choice of objects is essential in handling or promoting the child's attention, social interest and task performance. Dumas, Nadel, Soussignan, Martinerie and Garnero (2010) who examined the use of objects to support synchronic hand movements during social interaction, reported synchronization of cerebral rhythms between child and adult. Vivanti, Dissanayake, Zierhut and Rogers' (2012) research of predictive variables explaining success with the Early Start Denver Model (ESDM) showed that the use of functional objects accounted for 70% of the variance in visual reception gains, whereas the ability to infer another person's goal-directed actions represented only 30% of the variance of receptive language gains.

Imitation Responding

During sessions of Imitation Responding the selection of objects will address the child's initiating of hand manipulations and further developing of its manipulation skill (Nadel & Dumas, 2014). To this end, a "special order" is used, with one third containing objects that are familiar by being previously manipulated by the child (Scarpa et al., 2013). One third comes from a group of typical toys such as balls, cars, animals and dolls. The last third should contain objects that are new to the child with the view to encourage the child to make new experiences.

In Imitation Responding (IR) the child's progress is analyzed from its noted selection and performance of manipulations of the objects offered (Nadel, 2000). The process of manipulation is expected to be more frequent and more varied compared to when the therapist and child use one and the same object, for example by both "driving a car" at high and low speed, forward and reverse, and doing it in different ways depending on which toy is manipulated. The child's attention and responses to the trainer's acting would increase the width and variety of the child's motor repertoire. Such experiences are expected to assist the child's cognitive and social development (Nadel, 2015). Using objects in two samples synchronized is expected to lead to the appearance of shared representations of actions (Nadel, 2014), depending on the trainer's mastery of playfulness. The child's experienced imitative exchange to remain reciprocal and to perceive a social reward is related to the

trainer's response (Nadel, 2015). In this response the therapist intuitively grade the child's behaviour by interpretations of the child's intentionality of communication or manipulation or both (Byrne & Russon, 1998; Vivanti & Rogers, 2011) based on his or her attention to and sensitivity to the child's action. Another aspect is the trainer's timing of the child's action. By focusing on the child's attention to the adult and to the objects, the ability to initiate joint attention could be developed. This is especially relevant for non-speaking children with ASD, since initiating joint attention has been found to be at a low level (Strid, Heimann, Gillberg, Smith, & Tjus, 2013).

Imitation on Demand

Objects or stimuli in Imitation on Demand are used to increase the child's repertoire of behaviours (Smith, 2010). Objects are selected according to a planned agenda where the child's actual manipulation level has been identified, where stimuli are delivered related to children's cognitive level on a concrete-to-abstract scale (Leaf & McEachin, 1999), and where repetition proceeds from the stage of assimilation to that of accommodation (Inhelder & Piaget, 1958). Imitation on Demand is considered to be a DTT format (Smith, 2010). In this format the three teaching contingencies of stimuli, response, and consequence are maintained at the level of the child's existing abilities and with as little support as possible. The individual teaching comprises an organized level of information, instructions and objects related to the child's capacities of understanding, communication and manipulation (Leaf & McEachin, 1999). Support is offered as prompts, for example as slow talk, few words, signs, gestures or pictures. Objects/stimuli are selected and presented by the trainer, with the aim to increase interest and motivation in the child. To increase the child's responding, prompts and prompt fading should be used (Leaf & McEachin, 1999). When offering Natural Environment Training (NET) the child will mostly respond to familiar objects, with time-delayed prompting as the appropriate support strategy (Sundberg et al., 1998).

In Imitation on Demand in the DTT format, the strategy for reinforcing the child's behaviour does not need to be directly related to individual responses (Smith, 2010). NET includes natural contingencies of reinforcement operating simultaneously as part of the learning activity itself. Reinforcement within Imitation on Demand should encourage the child's further use of responses (Sundberg et al., 1998), underline the child's successful response and support repetition. This way of teaching also allows the development of socio-cognitive skills like social gestures, eye contact and eye pointing (Sundberg et al., 1998) although this, in the adult- directed way of inducing stimuli, will make greater demands on the child's own willingness to respond than their own initiating skills.

Effects of IBT

Warren et al. (2011) reported significant gains of language and cognitive skills with parents as trainers in an overview of IBT studies published from 2000 to 2010. They drew attention to subgroups of children with greater benefits of IBT.

Strauss, Mancini and the SPC Group Fava (2013) presented an overview of six meta-analyses of IBT published from 2009 to 2011. Their summary of IBT as an intervention method was an increased intellectual functioning, language skills and adaptive behavior with medium-to-large effects sizes (ES). They also reported that including parents in the intervention had a positive effect. With the purpose to investigate the effectiveness of IBT, Peter-Scheffer, Didden, Korzilius and Sturmey (2010) found that experimental groups who received IBT compared to control groups gained 4.96–15.21 points on standardized tests of IQ, non-verbal IQ, expressive and receptive language and adaptive behavior. An earlier meta-study by Spreckley and Boyd (2009) based on only four Randomized Controlled Trial (RCT) or quasi-randomized controlled studies reported that IBT compared with standard care, did not significantly improve the cognitive, language or adaptive outcomes of children in the IBT group.

The Cochrane report by Reichow et al. (2012) presented ES for the IBT group of 203 children younger than six years. They report medium ES for communication and language skills, IQ and daily living skills. Low ES were found measures for socialization. Their weighted mean ES (*g*) across IQ and adaptive behavior ranged from small (.30) to large (1.19). They suggest clinical guidelines that the use of IBT for children should be used on a case by case basis and upon prior clinical experience. Reichow et al. (2012) concluded in their meta-analysis of five articles from 2009 and 2010 that IBT is an effective intervention for many children with ASD.

Effects of interventions with the Being Imitated strategy

Outcome of the Being Imitated strategy was analyzed by Contaldo et al. (2016) in their meta-analysis of seven studies published between 2000 and 2014. Contaldo et al. (2016) concluded the Being imitated strategy to be a useful tool to improve social attention and responsiveness.

Other studies with the Being -imitated strategy, not included in Contaldo et al. (2016), are Nind and Hewett (1994; 2001) and Salt et al. (2001;2002). Nind and Hewett reported positive development during 30-week of intervention with schoolchildren. The children increased in joint focus, positive affect and interactive involvement. Salt et al. (2001; 2002) reported results from a one-year intervention combining with ABA strategies during pre- to post-treatment conditions. The treated group showed gains of large ES of imitation, joint attention and of medium size of social interaction (Rogers & Vismara, 2008).

Effects of comparison groups with no or little treatment

An interesting finding of a comparison group of children in the waitlist control condition is reported by Salt et al. (2002). They found that the experimental group showed a stable development over time with a tendency to maintain their initial standard scores. A negative outcome was found for the children in the comparison group who showed a low frequency of joint attention episodes and the imitation tasks. In a study by Howard, Sparkman, Green, Cohen and Stansilaw (2005) a declined development of standard scores was observed among children participating in a 15-hr-week generic preschool program. Rogers and Vismara discussed in their overview (2008) the former article and proposed that the children's slow outcome was an effect of eclectic delivery and underscores the importance of the quality in hours of training instead of quantity to young children with ASD. A study of community based IBT by Flanagan, Perry and Freeman (2012) compared a treatment group with a waitlist control (122 children matched in the two groups). The outcome of the control group showed higher levels of autism severity, lower adaptive and cognitive functioning.

Process of evaluation and intervention implementation

Efficacy research and effectiveness research are the main approaches when examining outcome of psychotherapy. These two different research strategies are created for different research questions and intended to verify the implementation process of intervention and transfer from experiment to ordinary clinical practice (Hunsley & Lee, 2007).

Efficacy studies examine whether the outcome is caused by the intervention or not. Veerman and van Yperen (2007) presented a model with four different levels of evidence of effects of intervention. The highest value of evidence of effectiveness was found in RCT and in repeated case studies. The next level, Indicate, verified outcome of functional effectiveness and includes for example quasi-experimental studies, norm reference or benchmark studies, client satisfaction-, and quality assurance studies. The third level is the theoretical level with the possibility to produce plausible effectiveness of outcome in reviews, meta-analysis or expert knowledge studies. The lowest value of evidence according to Veerman and van Yperen (2007) is the descriptive level with a potential ability to review effectiveness of intervention in descriptive, observational or interview studies or analysis of document.

Efficacy research is characterized by examining a new intervention with the aim to specify the optimal conditions of delivery and is focused on the measureable effects of the specific intervention (Flay et al., 2005; Nathan, Stuart, & Dolan, 2000). This examination is reliant of the design of the experimental situation such as well-defined interventions and homogeneous patient groups with restricted criteria of inclusion. Another feature of efficacy studies is the concern of replication. Flay et al. (2005) established standards for guidance of replication in efficacy studies. The four main standards comprise: specificity of program, clarity of causal inferences, and generalizability of findings and precision of

outcome. Efficacy research is mostly planned as short-termed studies because of the costs when examining a new and unproved intervention (Hunsley & Lee, 2007).

The criticism of RCT has been summed up by Veerman and van Yperen (2007) as too few interventions are evaluated. The studies are not representative of typical practice, the outcome is usually not found to be desirable and there will be no guarantee for any further replication or evaluation. Veerman and colleague (2007) also point to the problem with manuals and “fixed features” in the clinical practice where the ordinary task is to meet patients with individual special needs.

Research of effectiveness is aimed to investigate the level of beneficial effects of interventions during typical circumstances (Fixsen, Naoom, Blasé, Friedman, & Wallace, 2005). Relevant collected data must be related to treatment outcome and answer essential clinical questions. The overall aim of outcome studies is to collect and analyze data to increase the knowledge of the potential impact of treatment for the relevant group of patients. In this design of effectiveness studies the hypothesis will be formulated due to practice and clinical decisions. Flay et al. (2005) proposed effectiveness standards to comprise (1) manuals, education and technical support to implement intervention, (2) evaluation under clinical settings, (3) reproduce practical information of outcome effects, (4) clearly inform to whom an intervention could be generalized.

Evaluation of effectiveness studies is aimed to answer if the intervention in the clinical settings with ordinary therapists will benefit groups of patients and if the implementation of strategies works. This type of design often increases generalization since it involves clinicians who are routinely providing psychological services and patients who have been referred to the clinical settings. Common weaknesses of outcome research are often due to selection of collected data (Gilbody, House, & Sheldon, 2002). The goal is to maximize external validity while trying to maintain an acceptable controlled level of internal validity (Hunsley & Lee, 2007).

A model of five steps in the implementation process with the goal to increase validity, generalizability and replicability in psychotherapeutic research has been introduced by Onken, Carroll, Shoham, Cuthbert and Riddle (2014). Their model encompasses the following steps:

- The first step will provide the basis for generating a new intervention or the modification of an existing one, for example from one country to another. This step emphasizes basic understanding of the important strategies, therapeutic roles and materials for creating an intervention to a more potent and implementable method.
- The second step comprises the development of the researcher’s knowledge and experiences of the new intervention and measurement to maximize the internal validity.

- In step three the act of “real-world testing” will emphasize mechanisms that facilitate the ability to manage the intervention.
- Step four will be the important change from experience and efficacy research to clinical practice and effectiveness research of an intervention. At this point the implementation process should include both the intervention- and the supportive part of the organization. Fixsen et al. (2005) argue that top management support and access to dedicated resources are directly related to implementation outcome.
- At step five it is generally considered necessary to show that an intervention is effective before implementation. This process refers to the distribution of knowledge such as education of therapists and supervisors and information to relevant groups (Onken et al., 2014). Components of this part of implementation have been disseminated by Fixsen et al. (2005). They defined staff selection, preservice and in-service training, ongoing consulting and coaching, staff and program evaluation to be part of how to maximize the implementation process.

General aims

The aim of this thesis was to investigate the effectiveness of interventions offered to children with autism attending the Habilitation service. Another aim was to investigate the efficacy of a new intervention Imitation Responding within the setting of Swedish Habilitation services.

In Study I the aim was to investigate if interventions, after early screening, showed a positive outcome. The interventions were the comprehensive IBT- method Intensive Learning for young children with ASD (IL/IBT) (Spjut Jansson, Lindahl, & Rutberg Nilsson, 2011) and Eclectic Interventions encompassing a selection of methods based on TEACCH or communication methods.

In Study II the aim was to explore if a focal intervention Imitation Responding (IR), could function as a complement to IL/IBT, which is treatment as usual at the Habilitation services.

In Study III the aim was to examine if IR and IL/IBT had any effect on Joint Attention skills during short or long term evaluation.

Study I

The main aim of this naturalistic study was to study the two-year outcome in children who had screened positive for autism in a general population setting at about 2.5 years and who had then received intervention after having been diagnosed with autism spectrum disorder (ASD).

All interventions were offered at Habilitation service due to parental request that their child should receive either Eclectic intervention or IL/IBT. The 71 children were enrolled in interventions provided by Habilitation services at a mean age of 3.5 years. They were offered two types of the Intensive Learning program (IL/IBT) as a Regular IL/IBT or Modified IL/IBT, or Eclectic interventions groups.

Study II

The aim of the second study was to explore the efficacy of IR compared with IL/IBT during a twelve-week period occurring directly after the children received their autism diagnosis. In this Randomized Controlled Study (RCT), 40 children younger than 48 months and with an ASD diagnosis were voluntary respondents. In the IR group, a Being imitated strategy was applied inspired by the empirical research on imitation reported by Nadel (2000; 2004) and Heimann et al. (2006). This initial treatment was offered during the first three months. The IL/IBT group received Intensive learning for young children with autism (IL). All participating children were randomly assigned to either IR or IL/IBT directly after completion of baseline measurement (T1). At baseline (T1) and after the intervention (T2) the children were assessed with the Psycho Educational Profile (PEP-R) (Schopler, 1992) and the Vineland Behavior Adaptive Scales, second edition (VABS-II) (Sparrow, Cicchetti,

& Balla, 2005). We hypothesized that children receiving IL/IBT would show significant gains in language and in interpersonal relations (Eikeseth, Smith, Jahr, & Eldevik, 2002, Eikeseth, 2008; Eikeseth, Klintwall, Jahr, & Karlsson, 2012; Remington et al., 2007). For children receiving IR we expected significant increases within play, interpersonal relations and imitation based on previous reports (Heimann et al., 2006; Nadel, 2000).

Study III

This study aimed to compare the efficacy of Imitation Responding (IR) and IL/IBT to develop initiating joint attention (IJA) skills for 30 children receiving long-term IL/IBT after the initial 12 weeks intervention. The final sample consisted of 22 children (three children did not manage to complete the final assessment and data for five other children were lost due to technical problems). This resulted in two treatment groups, one IR+ IL/IBT with 13 children, and one IL/IBT with 9 children.

Summary of studies

Clinical context

At the Habilitation centers there are teams responsible for interventions to children with ASD consisting of different professionals; neuro-pediatrician, psychologist, social worker, occupational therapist, physiotherapist, speech- and language pathologist and dietician. The team offers different types of interventions and variation of intensities.

Participants in Study I

Between 2009 and 2011, a total of 134 children under four years of age with suspected ASD were referred to the Child Neuropsychiatric Clinic (CNC) in Gothenburg for further ASD assessment (Time 1=T1). Parents of 129 (102 boys, 27 girls) of the 134 children provided written informed consent to have their child participating in the assessment program at the CNC. One hundred of the children met criteria for a diagnosis of ASD at T1, and were referred to a Habilitation center in Gothenburg for intervention.

Attrition in Study I

Of the 100 children referred to two Habilitation centers in Gothenburg all had received interventions, but only 71 participated in a follow-up at the CNC two years later. Two families had moved from Gothenburg, two families did not take part in any of the interventions programs, five families declined contact with the Habilitation center and 20 families declined the two-year follow-up at the clinic. No child was excluded from the intervention or follow-up because of low IQ or presence of comorbidities, or the parent(s) speaking another native language than Swedish.

Interventions groups in Study I

The Regular IL/IBT group consisted of four girls and 27 boys, a total of 31 children, with a mean chronological age at their first assessment at 35.2 months. Their average cognitive level at start was 78 IQ, where 11 children had IDD. Twelve children had both their parents born in Sweden, one parent not born in Sweden was reported for six children and 13 of the children had parents who both were born in other countries.

The Modified IL/IBT group comprehended of 19 children, whereof seven were girls and 12 boys, with a mean chronological age of 35.7 months. Their mean cognitive level at start was 69 IQ, and 12 children had IDD. Five of the children had parents born in Sweden, five children had one parent born in another country and nine children had parents who were born in other countries.

The Eclectic group comprised four girls and 17 boys, in total 21 children with a mean chronological age of 37.4 months, and a mean cognitive level of 82 IQ. In this group seven children had IDD. Ten children had both parents born in Sweden, two children had one parent born in another country and nine children had both their parents born outside Sweden.

Participants in Study II

In Study II, all children referred between March 2011 and December 2012 to Child and Adolescent Habilitation services in Gothenburg, Sweden, and meeting the inclusion criteria of (1) a chronological age (CA) between 24 - 48 months, and (b) a recently confirmed diagnosis of Autism Spectrum Disorder according to DSM-IV-TR (American Psychiatric Association, 2000) were invited to participate. Exclusion criterion was severe EP judged to hinder therapy. The final studied group comprised 40 children. A majority (n=34) of the children had been diagnosed at the CNC, Sahlgrenska University Hospital, four had been diagnosed at Children's Health Care units, one child at Angered Specialist Hospital, and one child by the autism team at the Child and Adolescent Habilitation Services. The neuropsychiatric work-up encompassed clinically validated instruments; the Diagnostic Interview for Social and Communication Disorders (Wing, Leekam, Libby, Gould, & Locombe, 2002), Autism Diagnostic Review-Revised (Lord, Rutter, & Le Coteur, 1994), Autism Diagnostic Observation Schedule (Lord, et al., 2000), and the Social Communication Questionnaire (Rutter, Bailey, & Lord, 2003).

Attrition in Study II

Three children withdrew due to family reasons (one child in the IR group and two children in the IL/IBT group) before treatment started and for one child the preschool decided not to participate in the study. No children withdrew from the study after treatment.

Intervention groups in Study II

Group characteristics. The IR group consisted of 21 children (4 girls, 17 boys) with a mean CA of 41.6 months (SD = 6.2 months, range 27-49). The average mental age was 19.3 months (SD = 7.3 months, range 3-29) based on a Swedish version of the Bayley Scales of Infant and Toddler Development, Third Edition (Bayley, III) (Bayley, 2005). A majority of the children (n = 16) lived with both parents, while five lived in single-parent households. Five children had no siblings, twelve children had one sibling and four had two siblings or more. Three children had a sibling with ASD. Ten families were native Swedish speakers.

The IL/IBT group consisted of 19 children (4 girls, 15 boys) with a mean CA of 40.1 months (SD 5.8 months, range 30 - 48), and a mean mental age of 19.6 (SD 9.1 months, range 2- 42). All children lived with both their parents. Six of the children had no sibling, nine children had one sibling, and four had two siblings or more. Five of the children had a sibling with ASD. Nine families were native Swedish speakers.

Participants in Study III

In Study III all children are referred with the same conditions as in study II (see, study II) and 30 children were included. The final studied group comprised 22 children, see attrition paragraph.

Attrition in Study III

We limited our study sample to include only children who had complete ESCS data at all three time-points (see table 3). The attrition after randomization was due to children's refusal to participate at assessment ($n = 3$) or technical problems ($n = 5$). In total eight children, three children in the IR+IL/IBT group and five in the IL/IBT group were excluded from outcome analysis. The final sample consisted of 22 children. An Analysis of Variance (ANOVA) was used to examine if the attrition had led to any significant differences between the sample before and after attrition, (intervention groups collapsed). Results showed no significant differences between any of the variables as shown in Table 3.

Table 3. Characteristics of participants in the original sample, attrition and final sample.

	Original sample N=30		Attrition N=8	Final sample N=22		Original sample vs. final sample
Treatment groups	IR+IL/IBT N=16 M _i (SD)	IL/IBT N=14 M _i (SD)	Attrition N=8 M _i (SD)	IR+IL/IBT N=13 M _i (SD)	IL/IBT N=9 M _i (SD)	
Age in months	42.8 (5.7)	39.7 (6.5)	44.4 (3.2)	43.2 (4.5)	38.7 (7.1)	ns
Girls/boys	2/14	1/13	2/6	2/11	1/8	ns
Mental age	20.1 (7.8)	21.4 (8.9)	20.4 (6.7)	21.6 (6.2)	19.8 (10.1)	ns
Children with MR	4	3	2	4	1	ns
Bilingual families	6	7	2	6	5	ns
Two-parent family	12	14	1	10	9	ns
Siblings w. NPD	1	4	1	1	3	ns
<i>PEP-R</i>						
Expressive- Language	14.8 (4.2)	18.1 (7.6)	20.5 (7.5)	13.8 (4.4)	16.5 (6.9)	ns
Receptive language	14.9 (5.6)	17.0 (7.3)	19.9 (7.5)	14.2 (3.5)	15.9 (7.1)	ns
<i>VABS-II</i>						
Expressive language	14.8 (9.2)	16.5 (9.1)	18.0 (9.6)	12.9 (7.4)	13.9 (9.0)	ns
Receptive language	15.4 (14.6)	18.71 (12.3)	18.6 (17.0)	11.4 (6.1)	14.8 (7.8)	ns
<i>ESCS</i>						
Eye contact	7.44 (7.53)	7.23 (9.65)	-	7.46 (8.04)	7.89 (11.12)	ns
Alternate	3.50 (6.05)	3.62 (4.90)	-	3.77 (7.59)	2.67 (4.47)	ns

Final intervention group characteristics

The final group with IR+IL/IBT comprised 13 children (2 girls, 11 boys) with an average CA of 43.2 months (range, 29-49 months). A majority of the children ($n = 10$) lived with both parents, while three lived in single-parent households. One child had a sibling with ASD. Seven families were native Swedish speakers. The group's mean mental age was 21.6 months (range, 3-29 months).

The IL/IBT group comprised 9 children (1 girl, 8 boys) with a mean CA of 38.7 (range, 30-47 months). All children lived with both of their parents. Four children had a sibling with ASD. Four families were native Swedish speakers. The group's mental age at start was 19.8 (range, 6-42 months).

Representation of the interventions groups in Study III

Group characteristics in the final group of IR+IL/IBT comprised of 13 children (2 girls, 11 boys) with an average CA of 43.2 months (SD 4.5) (Table 3). A majority of the children ($n = 10$) lived with both parents, while three lived in single-parent households. One child had a sibling with ASD. Seven families were native Swedish speakers. The mean mental age was 21.6 months (SD = 6.2). The children's expressive language had a mean of 15.2 months (SD = 4.3), and receptive language had a mean age of 15.1 months (SD = 5.8) measured by with PEP-R. The same variables reported in VABS-II interview scored 15.3 months (SD = 9.1) and 14.5 months (SD = 10.2).

The IL/IBT group comprised 9 children (1 girl, 8 boys) with a mean CA of 38.7 months (SD = 7.1). All children lived with both of their parents. Four children had a sibling with ASD. Four families were native Swedish speakers. The mean mental age was 19.8 months (SD = 10.1). The children's expressive language had a mean of 15.9 months (SD = 6.7), and receptive language had a mean age of 15.3 months (SD = 6.9) measured by with PEP-R. The same variables reported in VABS -II interview scored 13.2 months (SD = 8.7) and 14.3 months (SD = 7.5).

Between groups analysis of the final groups (IR+IL/IBT vs IL/IBT) showed no significant differences of variables in Table 3.

Materials

All assessments had been selected to their possibility to be used for non-speakers or speakers of Swedish or a foreign language. However, if needed, assessments were conducted by help from certified interpreters for parents and their children.

Procedure and Instruments

Study I

In Study I, all children were assessed at the neuropsychiatric clinic two years after their first assessment with regard to autistic behavior/ASD, intellectual function, adaptive and

global functioning. The tests were administered according to standard procedures. All professionals were blinded to type of intervention the children had received.

Adaptive function was used as an outcome variable and was assessed by an interview with parents utilizing the Vineland Adaptive Behavior Scales (VABS-II) (Sparrow et al. 2005). This variable includes an adaptive composite score and three subdomains scores; Communication Skills, Daily living Skills and Socialization. All results are given as normed scores.

Global functioning was measured with The Children's Global Assessment Scale (C-GAS) (Schorre & Vandvik, 2004; Shaffer et al., 1983). The scale was originally developed for use with 4 - 20-year-old children and adolescents, and has been adapted for younger children by the CNC assessment team (Kantzer, Fernell, Gillberg, & Miniscalco, 2013).

Comprehensive programs in Study I The specific comprehensive intervention IBT program, used in the western parts of Sweden, was the "Intensive Learning for young children with autism" (IL/IBT). IL/IBT is a manualized comprehensive program (Spjut Janson et al., 2011) which includes a theoretical part and information to parents and preschool teachers on how to perform training and sessions. IL/IBT was used in two different settings; 1) the regular form, implemented for parallel use at home and at the child's preschool. This program included sessions twice a month with supervisors, the "1:1 intervention" was provided ten hours at home and 15 hours at preschool/each week (totally 25 h/ each week). 2) the modified form was implemented only at home and offered regularly sessions once a month with supervisors, the "1:1 intervention" was provided ten hours at home/week (totally 10 h/week).

Eclectic interventions in Study I

Habilitation services are mainly organized as education to parents or preschool staff, where the Habilitation staff acts as supervisors. For a summary of the habilitation interventions, see Appendix I. Eclectic interventions, in total 24 methods that were found to prove scientific criteria of having an effect, have been identified by Odom, Collet-Klingenberg, Rogers and Hatton (2010) and one of the methods is similar to an intervention used by Swedish Habilitation services. This is a focused program, the Picture Exchange Communication System (PECS; Frost & Bondy, 2002). A parent-implemented intervention evaluated according to parents and course leaders' experiences of the program (Ferm, Andersson, Broberg, Liljegren, & Thunberg, 2011), but not included in the compilation made by Odom et al. (2010), is ComAlong, a Swedish early intervention parent course utilizing Augmentative and Alternative Communication (AAC) (Thunberg, Carlstrand, Claesson, & Rensfeldt Flink, 2011). Other interventions were implemented during the two years as parts of the comprehensive program TEACCH (Treatment and Education of Autistic and Related Communication-Handicapped Children; Ozonoff & Cathcart, 1998).

Assessment of variables and instruments in Study II

In study II, all children were assessed with the same test battery both before and after intervention.

Cognition was measured with The Bayley Scales of Infant and Toddler Development, third edition, Swedish version 2009 (Bayley-III, 2005).

Expressive and Receptive Language, and Visuo Motor Imitation were all measured by Psychoeducational Profile Revised, vol. I., Swedish version, 2005 (PEP-R) (Schopler, 1992). Expressive and Receptive Language, Play and Interpersonal Relations had been examined by interviews with preschool teachers with the Vineland Behavior Adaptive Scales, second edition (VABS-II) (Sparrow et al., 2005). The variables of PEP-R and VABS-II are reported as age equivalents (AE).

Intervention Procedure. IR was implemented in a specific room at the preschools. The setting of double toys offered the child an open social and solitary situation with a wide combination of possible choices of manipulation objects. The child's choices are simultaneously performed with co-regulation of synchrony by the therapist. Based on synchrony a system of communication and exploratory learning emerges from the manipulation of the objects. This intervention model had a main goal to develop the child's initiating ability of joint attention and responding to other's manipulation and to other's initiation of joint attention. Sessions were offered the children every day for at least 30 minutes and a maximum of 40 minutes each session at the preschool. Supervision from Habilitation center took place at the preschools once a week with preschool teacher and parents to evaluate sessions and to plan the new program.

IL/IBT Learning for young children with autism (IL/IBT). This intervention training is planned for 25 hours each week, this standard of time applies independent who is responsible for training. If only parents are responsible they do all training (100%). When both parents and preschools are responsible for training, parents are planned for ten hours (40%) and the preschool teachers for fifteen hours (60%) each week. Protocols are used to register exercises and training time. Clinical experience indicate that training in the start-up-phase rarely exceed to more than 15 hours/week. After six months most of the children usually will receive 25 hours/week. Supervision sessions of parents and preschool teachers lasted between 1 and 1.5 hours and took place once a fortnight, or more often to parents when they are "only-trainers", at the Child and Adolescent Habilitation Center. During the start-up period there were one or two informative workshops with the aim to teach parents and preschool teachers about the method. This method is focused on generalization to typical milieus as soon as children have prerequisites to use this format and that trainers could use it successfully.

Study III assessed variables and instruments

In study III, all children were assessed with the same test battery three times, before and after both the short- and the long-term intervention. Initiating Joint Attention (IJA) has

been assessed with ESCS (Mundy et al., 2003). This is a structured assessment in which the experimenter presents different toys to a child who is seated at a table. The observation procedure took approximately 12-14 minutes, it was videotaped and later coded.

For this study two variables were used to measure IJA with ESCS. The variable Eye - gaze comprehended eye contact noted if the child held an inactive toy and looked back and forth between the tester and an active object (ESCS; altering). The other variable was to measure of child-initiated Point/show noted if the child pointed to, or showed, a toy. For description of IL/IBT see study I.

Description of effect –considerations and measures

In Study I it was found that outcomes presented with means and standard deviations were affected by large variations in the observed standard deviations. This finding was reported as an individual variance of outcome.

In study II and III we used effect sizes (Cohen's *d* and Hedge's *g*) to describe the outcome of the intervention.

In Study II students' t-tests was used to compare differences between T1 and T2. While examining the standard deviations we noted a variation from 6.61 to 13.27 units (months) across all variables. We computed the effect size with the original standard deviations at T1 and T2 for each treatment group and pooled within-group standard deviation (Löfgren, 2006).

In Study III we calculated ES using Hedge's *g* (Hedges & Olkin, 1985), correcting for small sample size and reducing the effect size by about 4% (Durlak, 2009).

Assignment

Study I

In Study I, all assignments of program and interventions are due to children, parents and professionals cooperation and suggestion of the best selected intervention.

Study II

The children were randomized to the IR group or to IL/IBT groups (home training and home and pre-school training) immediately after T1. The IL/IBT subgroups encompassing home-training or both home and preschool training was later collapsed since no statistical difference in outcome between the two groups was found.

In the IR group twenty preschools were involved in the study, and one preschool teacher was assigned for each child in the group conducting the treatment. Two children attended the same preschool but different teachers were responsible for their treatment. In the IL/IBT group there were nine preschools involved with one preschool teacher per child. Two children who attended the same preschool had different responsible staff. In this group two of the children's parents were familiar with the method, since they had older children who had been trained with the method. Nine children were trained by their parents at home.

Study III

After training with either IR or IL/IBT during three months, 30 children participated in the long-term interventions (12 months) with only IL/IBT.

Main results

Study I

The main finding in study I was that the type of intervention was not critical for outcome of adaptive and global functioning according to the results on VABS-II adaptive composite score and the C-GAS. Thus, treatment based on ABA did not differ from other types of interventions provided from the Habilitation center or preschool. This is somewhat in contrast to low gains utilizing TEACCH method reported in a meta-analysis by Virúes-Ortega, Julio and Pastor-Barriuso (2013). Similar findings are reported by Flippin, Reszka and Watson (2010) in their meta-study of PECS. Both these methods involved strategies of ABA.

The only effect that was significant with regard to adaptive functioning outcome was the child's intellectual level, i.e., the group of children with average intellectual functioning and children with borderline intellectual functioning had higher VABS-II adaptive composite scores and C-GAS scores compared to the group with intellectual developmental disorder (IDD).

Although, the mean adaptive composite score did not change significantly between T1 and T2 the variance increased significantly. This probably reflects that gains and loss of adaptive skills varied considerably between children at the follow-up. This finding underscores the need for regular monitoring of the child's functioning and developmental progress during the intervention period and the necessity to consider a change of or modification of the program in use.

Study II

The time of training offered to the children in the IR group (study II) was on average 2.2 hours/week carried out by the child's preschool teacher/therapist. To the children in the IL/IBT group 14.4 hours/week were carried out by a preschool teacher and the parents. This time of training follows the planned time in the manuals to both intervention types.

Seven subscales from PEP-R and VABS-II, covering language, visuo- imitation, play and interpersonal relations skills, were used as outcome measures. Results showed a significant increase in 6 out of 7 subscales for both groups. Only the IR group had a significant gain on the Interpersonal relations subscale ($d = 0.66$).

Study III

Study III examined how Imitation Responding (IR), affected the development of IJA among children with autism. ESCS is an instrument designed to measure joint attention and related behaviors in typically developing toddlers (Mundy et al., 2003). Two variables of IJA were studied. The first variable, named eye gaze, includes the child's eye-gaze and referring to an object with another person. The second variable, pointing/showing, contained pointing or showing behavior in order to coordinate attention with another person.

No variable of IJA showed any change during the first three months of treatment, nor were any significant between-group differences noted when comparing IR and IL/IBT groups. However, the group that had received IR (IR+IL/IBT) as first intervention showed a significant increased gaze shift score in contrast to the group that had only received IBT-training. No significant change was noted for the pointing and showing score for any of the intervention groups. These results indicate that early implementation of a Being Imitated strategy might be useful to increase joint attention as measured by eye-gaze shift if it is implemented as a “start-up” intervention that is followed by IL/IBT treatment.

General discussion

Study I

The first study presents a naturalistic study over two years with the aim of making a comparison between IL/IBT and eclectic interventions for children with ASD. IL/IBT was offered in two settings. Regular IL/IBT, involving both parents and preschools, comprised planned training 25 h/week, and Modified IL/IBT only with parents, planned training of 10 h/week. Children in the eclectic intervention group receive for example, ComAlong, TEACCH, or treatment in groups of children or parents aiming at increasing children's skill improvement or minimizing their behaviour problems. These children were offered more than two interventions each. The outcome was measured by Adaptive Composite scores (VABS) and global functioning (C-GAS).

The main finding was that initial IQ accounted for the outcome variance of both IL/IBT and eclectic interventions. The result is representative of interventions based on children's learning abilities, including both IBT and TEACCH as well as some of the eclectic interventions. This relation between IQ and the outcomes of interventions is in line with findings from the Stockholm study (Fennell et al., 2011). The IQ assessment has mainly been developed to measure abilities related to learning (Jacobson & Traux, 1991; Volkmar et al., 2014; Trembath & Vivanti, 2014). It is not surprising that children with low IQ scores had a lower outcome on IBT, since these interventions are aimed to compensate for learning dysfunctions. Trembath and Vivanti (2014) stated that behavioural characteristics are sensitive in early differential diagnosis, for example avoidance or ritualized behaviour, may be low predictors of treatment outcome. They suggest that different factors are associated with different ASD subgroups across different types of interventions. Vivanti and Rogers (2014) argue against more replication of the "known fact" of IQ and outcome, claiming that a high IQ gives a good and a low IQ a poor outcome. Instead, they highlights the need of research that examines which methods are beneficial and to which subgroups.

The results of Study I differ from the findings presented by Howard and colleagues (2005, 2014) comparing IBT and eclectic interventions. Their conclusion was that IBT could produce substantial improvements in young children with autism compared to common eclectic interventions, such as intensive TEACCH and sensory integration therapy. Howard et al. (2005, 2014) reported a positive outcome of IQ, language and adaptive behavior for the IBT with more than 20 hours weekly training in comparison with treatment delivered in multiple settings. Regular IL/IBT in Study I comprised home and regular preschool training. However, no registration of weekly delivered training had been present in Study I. Therefore we could not compare with the reported time of Howard et al. of 20 hours weekly. The planned time of IL/IBT in Study I was 25 hours weekly.

Cognitive level could be preferred as a reliable level of change over a period of two years of treatment. Howard et al. (2014) presented a "learning rate scale" based on intake age

equivalent scores as an outcome to avoid evaluation on the group level (mean scores). This is almost equal with Jacobson and Traux (1991), who recommended one standard deviation of a normed and measured variable to be regarded as “good effect” of an intervention. In Study I, statistical analyses of standard scores on a composite variable were used instead of a description of age equivalent scores. However, our findings, using group level mean, are in accordance with conclusions in the review by Odom, Hume, Boyd and Stabel (2012). They compared IBT programs and eclectic interventions and found that eclectic programs which were well planned according to individual needs will have a positive impact on the development, learning and life outcomes of children and youth with autism.

The problem of low outcome in clinical groups has sometimes affected the selection of participants to studies. Children with a low cognitive level or other problems are therefore excluded, and the external validity is restricted. A naturalistic study could address this problem. Since children with ASD are heterogeneous as a group, with associated medical conditions, seizures or low IQ, it is necessary to increase the knowledge of how these circumstances will influence treatment. A low IQ and a low score on adaptive behaviours reflect a variety of underlying processes, which often affect responses to interventions. The mean adaptive composite score did not change significantly from the start of the intervention to the follow-up two years later, but the variance increased significantly. This investigation indicates the existence of sub-groups of responders and non-responders, which will be an argument for further research.

Study II

The main findings from Study II demonstrated the efficacy of both interventions when used as a brief first choice for young children newly diagnosed with ASD. The RCT study confirmed that both IL/IBT and IR promoted the children’s social and communicative development. The IL/IBT method promoted a broad area of skills training. All training was made by modelling or was instruction-based, and the children were offered a stimulus to react or respond to (operant learning). The planned training has to be extensive, ideally around 20 h/week (Volkmar et al., 2014). Fewer training hours, only 15 hours/week, were conducted in the initial parts by parents and preschool teachers, as reported in protocols.

The results showed significant gains on six out of seven measures for both IR and IL/IBT in Study II. In a meta-analysis of IBT by Reichow et al. (2012), which includes both RCT and clinical studies encompassing participants with similar ages, could therefore be used as a comparison to the results from Study II (see Figure I). This study used Hedge’s g as an effect size measure, while in the study presented here Cohen’s d was used. Both effect sizes pool variances on the assumption of equal population variances, but Hedge’s g pools use $n - 1$ for each sample instead of n only. As with Cohen’s d , a comparison between these different effect sizes had to be made with care. This is relevant, since the samples are relatively small.

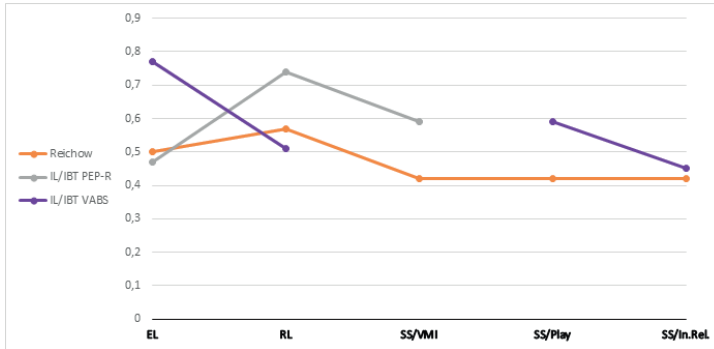


Figure 1. ES of all measured variables of IL/IBT (study II) in comparison reported ES of IBT (Reichow et al., 2012). (EL= Expressive Language; ES= Effect Size; RL=Receptive Language; SS=Social Skills, VMI= Visuo Motor Imitation; In. Rel. = Interpersonal Relations).

Variables and their ES of IL/IBT (grey and purple) from Study II compared to ES (orange lines) reported by Reichow et al. (2012). When comparing the outcomes (Figure 1) of IL/IBT with reported values of outcome by Reichow et al., it appears that the ES is lower for IL/IBT when measuring expressive language with PEP-R. Equal or somewhat higher effect sizes were shown for expressive language reported with both PEP-R and VABS-II. Social skills (PEP-R), such as Visuo-Motor Imitation or Play and Interpersonal relations (VABS-II), had ES values equal to those reported by Reichow and colleagues (2012).

The children's outcome in Study II verified that IL/IBT should be used further on. The outcome of the implementation with parents, solely or together with preschool teachers, was in line with Strauss et al. (2013), who also reported positive training effects after cooperation with parents.

Study II involved a controlled implementation of parental inclusion in IL/IBT by using two such subgroups. Both the procedures with parents alone and with parents and preschool teachers in cooperation had been shown to fit both the organization and the families. Due to parents' central role in their child's life the approaches of IL/IBT, with parents alone or in cooperation with preschools, are essential. The latter approach had some advantages, for example that children could be together in preschools with peers of the same age. The involvement of and cooperation with parents as trainers will add to the number of treatment hours that can be provided for each child. Furthermore, the children's natural settings could be used for training, which might lead to fewer behaviour problems (Scheibman et al., 2015). A conclusion, verified by analyses of subgroup outcomes and actual training time (Study II), is that using parents as the only trainers is comparable with having parents and preschool teachers cooperate during the first three months of the programme. This is in

contrast to the implementation in Study I, where some parents had to wait to be introduced to IL/IBT and all of them had to go through an introduction course.

Imitation strategies and children's outcome

The Being Imitated strategy showed changes of communication verified by increased expressed language and children's social skills as imitation, play and interpersonal relations, even though the daily training time was brief. Nadel (2015) points to the fact that repetitions are important for effective learning, which is well reflected in both imitation strategies. As the intensity for young children, and hence the opportunity for repetition, could be as low as twice or less each week, this could decrease motivation.

All language variables, plus play and visuo-motor imitation showed a medium or nearly medium effect size for children in the IL/IBT group with Imitations on Demand. Interpersonal relations had no significant outcome.

The circumstance that IR, with its low amount of weekly training time (with a mean time of 2.5 hours) had significant and comparable results in all skills (but not all variables) with IL/IBT is notable. This may be interpreted to the effect that the children in the IR group obtained a strategy that promoted their interest in other people. Daily reports from the preschool teachers' as descriptions of children showed an increased interest in participating in and responding to games, listening to books and looking at other people, like other children's parents.

In the IR group, we found children who had passed the level of using imitation as their main communication mode, a behaviour that was not found in the Imitation on Demand group. They showed motivation and interest in speech and were negative to receiving non-verbal answers. As a result of experiences in this study and Nadel's theoretical comment "...as soon as language is mastered, the imitative communication is not observable any more" (Nadel, 2015, page 57) the guidelines for IR will be changed.

From our results and observations of the children who participated, we would like to recommend that, in further guidelines to IR, all children whose spoken language is on a typical developmental level should be excluded. For this group Imitation on Demand or access to IL/IBT would be a better alternative.

Imitation strategies in Study II

The comparison between different imitation strategies reflects the idea that imitative performances like Imitation on Demand do not reflect all imitation capacities (Nadel, 2014). The actual imitation strategy applied is a Being Imitated strategy, used in the IR intervention, compared to the use of an Imitation on Demand strategy in the IL/IBT group. The Being Imitated strategy mainly aims at promoting children's capacity to initiate actions and respond to adults' imitative behaviour, i.e., timing within the imitation-based interaction and taking an interest in another person (read: therapist) through this process (Nadel, 2000; 2014). The optimal time for using the Being Imitated strategy depends

on children's development of communication, language and cognition and entails an established relationship between action and object (Meltzoff, 1995).

The Imitation on Demand strategy is mainly used in IBT programs as Fovel, 2002; Leaf & McEachin, 1999; Lovaas, 1981/2003; Maurice et al., 1996) from the start of the training. It occurs in exercises such as imitation with objects, motor imitation and oral or verbal imitation. In Imitation on Demand the adult has an agenda for which behaviour of the child is the correct response.

Both the Imitation on Demand and the Being Imitated strategies entail that the observer can copy both the goals and the means of the observed action (Vivanti & Rogers, 2014) as well as requiring that the child attends to action and to object (Meltzoff, 1995). Nadel (2015) and Trevarthen (2005) both argue in line with Piaget (1962) that children's development of symbolization starts with representations of their own and other people's communication acts, comprehending both intentional and affect - regulation abilities. In contrast to Imitation on Demand, the Being Imitated strategy are less demanding on children's level of motor behaviours as the movement or actions are part of the child's repertoire (Nadel, 2015).

Our finding of increased interpersonal relations in the IR group agrees with the study of Heimann et al. (2006) and with Berger and Ingersoll's (2013) findings of an increased ability to engage with other people. This suggests that there is a direct link between the communication format when the adult imitates the children's behavior and children's social initiatives as well as initiatives concerning themselves and their environment. The outcome of the IR group supports theoretical claims (Nadel, 2000; 2014) that imitation is a potent vehicle for social and cognitive development.

Study III

In the third study, the hypothesis was that the IR group would show an increased development in IJA over time, evidenced by the children's looking pattern (eye gaze shifts) and gestures (pointing and/or showing), compared with the group receiving only IL/IBT. This hypothesis builds on the joint attention theories of Bates et al. (1975), who postulate that joint attention may be developed through methods aiming to increase children's awareness of being the object of other people's social attention.

Our hypothesis was partly confirmed. Children who were first offered IR during three months, followed by IL/IBT during a period of one year, increased their amount of generalized initiated joint attention. An increased generalized initiated eye gaze was found with a high effect size in the IR group at the final evaluation after the children had got both IR and IL/IBT. This effect was not found in the group that participated in IL/IBT throughout the period. However, no gains in showing/pointing behaviour were found in either of the intervention groups. The improvement of IJA by eye-gaze shifting towards the adult and

the object required the specific IR method for three months as a precursor to the one-year training with IL/IBT.

A well-researched method is Joint Attention, Symbolic Play, Engagement, and Regulation (JASPER) (Kasari, Freeman, & Paparella, 2006). In an RCT study, Kasari et al. (2006) examined the outcome of 20 children who participated simultaneously in both JASPER and IBT. They found a significant increase in joint attention behaviours, both showing (IJA) and responding in JA, but no significant eye-gaze and pointing at the evaluation, made after three months of training (Kasari et al., 2006). In their RCT study, the JASPER method was offered for 5-6 weeks with 30 minutes' daily training and all children's participation in IBT. The therapists were graduate students in psychology with experience of children with autism. However, when trying to replicate the findings reported by Kasari et al. (2006) in preschools in Norway, Kaale, Smith and Sponheim (2012) reported no changes in any variable of ESCS. Furthermore, in a study using JASPER with parents as trainers (Kasari, Gulsrud, Paparella, Helleman, & Berry, 2015), no changes of IJA were observed while the joint engagement increased. Kaale et al. (2012) discussed their negative results as an effect of not using professionals educated in psychology. Similar findings were confirmed by Kasari and her team in the "Parents as trainers study" from 2015. Neither could Study III in this thesis confirm any changes of IJA with preschool teachers as trainers during the first three months of IR. It therefore seems that the conclusion drawn by Kasari et al. (2006) holds, that only therapists educated in psychology can improve IJA in a short-time treatment.

Another explanation of the significantly increased variables of IJA in both Kasari's (2006) RCT study and Study III could be the use of two kinds of treatment. Such an explanation may also shed light on the finding in Study III of increasing eye gaze at T3.

The outcome of IR plus IL/IBT showed a stable eye gaze with an ES of 1.10, while for pointing/showing the effect size was very small. The JASPER+IBT training took place during three months, while IBT seems to have been used before JASPER started. This combination of training methods was reported to have an impact on eye gaze with a very small effect size, as well as a medium effect size of pointing and showing.

IR plus IL/IBT produced a higher effect size on eye gaze than JASPER+IBT, which, in contrast, had the highest effect size on both showing and pointing. While the approach of IBT is designed to increase children's responses to adult instructions, the approach in IR and JASPER was to support the child's spontaneous initiating behaviours. Both Kasari et al. (2006) and Study III support the assumption that only a combination of treatments could provide for a generalized outcome of IJA variables. These findings are in line with reports from Salt et al. (2001) and Ingersoll and Schreibmann (2006). In their studies they all combined IBT and Being imitated strategies simultaneously.

A model by Mundy, Sullivan and Mastergeorge (2009) places imitation as a basic experience for both typical and atypical children's development of JA. The integration

of the information processing of social-problem solving from the first level “learning to JA” could successfully progress to the next level “learning from JA”. In this phase, a child has the capacity to give attention to another person and to objects by processing multiple information. Using this model in Study III fits with the children having some basic experiences during IR sessions that partly increased their eye gaze shifts, but only with known persons and objects. This period resulted in further learning with the IL/IBT and an increase in the children’s eye gaze ability, which was generalized to other persons and objects.

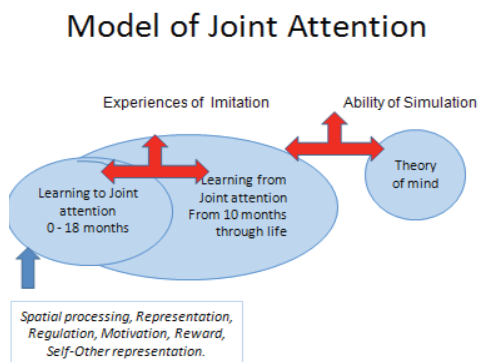


Figure 2. A model by Mundy, Sullivan and Mastergeorge (2009) to explain the steps in JA behaviour (modified by Birgitta Spjut Janson)

Behaviour or developmental approach?

In a study by Kaale et al. (2012) it was discussed if there had been a conflict of interest between different methodologies such as more developmental-based strategies and behavioral strategies, whereas Kasari et al. (2006) do not report such issues. Another issue to further examine is to follow the implementation of Kasari and colleagues and incorporate IR in IL/ IBT as a simultaneous package, with the guidelines that one and the same supervisor should be responsible for the child’s training in both methods.

Schreibman et al. (2015) stated that behavioural and developmental treatments are particularly well suited for infants and toddlers with ASD by allowing a more substantial and accelerated learning and behavioural change to take place.

Further studies of IR as a focused model and also as part of the comprehensive IL/ IBT program need to be conducted to give a more complete understanding of the methods’ strengths and weaknesses.

Impact on outcome

Internal validity is an issue in the present thesis. The sample in all three studies consisted of children with confirmed ASD. They had low abilities in both cognitive, social and

language skills compared to typically developing children. To create a homogenous group was important in studies II and III, where the link between the different interventions and the diagnostic group would be the main task, as argued by Nathan et al. (2000). Limitations such as invalid, unsystematic and irrelevant information of the participants' status at baseline could both complicate and overturn the possibility of replicating studies II and III and also of making valid interpretations in Study I. The shortcomings of internal validity depend on the naturalistic study's aims to review intervention in a real practice setting (Hunsley & Lee, 2007).

Fidelity, i.e. to what level the interventions in the studies were conducted as claimed, was critical, as this has an effect on the outcome in all interventions. This represents, for example, information of concrete components of the therapeutic strategies during the intervention sessions. Such knowledge is fundamental to enable the generalization of the examined methods to clinical practice. In RCT studies, treatment integrity includes the use of empirical measurements of outcome variables (Fixsen et al., 2005). Manuals for interventions, such as written and filmed information to support preschool teachers and parents in workshops are also vital. The everyday training of the child in IR and IBT implied the training of different skills by parents or preschool teachers. In both comprehensive and focal programs for children with ASD, where supervision by parents and preschool teachers is the main approach, guaranteeing the integrity of the intervention is fraught with problems. Earlier evaluation of IBT programs had been delivered in specialized clinical settings or with specially educated trainers (Lovaas, 1987; Eikeseth et al., 2002; 2007). Such controlled and experimental situations were not feasible in any of the studies in this dissertation, since health services, preschools and universities are organized as units with different missions and legislations. A new developmental approach of IBT (Schreibman et al., 2015) assumed that children's learning is a complex and dynamic process, involving interactions between child and family and their environment. An aspect of this is everyday life, where most families having young children with autism also are responsible for the children's training. Parents as trainers were examined by Bibby, Eikeseth, Martin, Mudford and Reeves (2001), who concluded that children with a low IQ also needed additional specialized training as a supplement to parent training with IBT.

In more recent IBT and in IL/IBT programs, supervisors have the competence to train parents and preschool teachers in delivering IBT or IL/IBT. In Study II we tried to control the "delivery of method", since IR with the Being Imitated strategy had not been previously practised in preschools. For this reason, videotapes were analyzed (IR) during supervision. When failures were noted they were discussed and an agreement of how to proceed was reached. All training of IBT and IR was documented by parents and preschool staff by daily written reports with the aim to control for fidelity.

All participating habilitation professionals had a university degree in the areas of special education, speech and language pathology or psychology. For the IR method, they

all received special training including both the theory of early social skills and how to implement the imitation strategy. Many of the supervisors had undergone an IBT education including the IL method, especially training formats and strategies of ABA principles and functional behaviour analysis.

In all efficacy research, the focus is on measureable outcome, and the reliability of instruments could affect the internal validity (Kazdin, 2008). Child studies, pre-post designs and long-term design studies are especially sensitive to instrumentation threats (Shadish, Cook, & Campbell, 2002). The level of threat depends on the stability of measurement units and scales over the age range. In Study II, and partly in Study III, the floor level of PEP-R included too many raw scores on the first level to demonstrate difference of low development, which affected our results. This measurability of children with low ability was not found for Bayley-III, VABS-II or ESCS.

Hypothesis testing as in studies II and III was essential for the construct of measurement. All measurements used in the three studies are well-established and documented clinical and research instruments. In Study I, the naturalistic study, the children's outcome was reported later, while children's cognitive level was measured with different tests, such as, Bayley scales of Infant and Toddler Development - third edition (Bayley, 2005), Griffiths Mental Development Scales I and II (Alin-Åkerman & Nordberg, 1980), Merrill-Palmer Revised Scales of Development (Roid & Sampers 2005), and Wechsler preschool and Primary Scale of Intelligence third edition (WPPSI-III) (Wechsler, 2004). To minimize the problem with comparability of different scales, standard scores were used. The particular test used depended on the ability of the child at the time of assessment and on the selection made by the psychologist.

The children were assessed with different tests at T1 and T2, which is something we could not control. This threat to internal validity complicated the interpretations of the outcome. In contrast to Study I, the naturalistic study, studies II and III were planned in advance and all children employed the same tests at all three measurement points.

The testing effect, or to what level the effect is a result of exposure to a test and not of the intervention, is an issue when repeating tests or interviews. For studies II and III this threat was partly controlled by the use of tests like Bayley-III and PEP-R. Both these measurements are standardized for persistent and repeated evaluations and for the actual age group (Shadish et al., 2002). For ESCS, we controlled that all toys were only used in the test situation and were not part of any training sessions for any child who participated. A smaller effect of repeated measurement could be expected for VABS-II due to its semi-structured way of questioning informants.

It is also important to consider whether the observed gains were "true" gains or just an effect of the children's maturation. Maturation surfacing as developmental changes occurs in all children. For young children, maturation and effects of interventions such as IR and IBT were of course difficult to disentangle. In a pre- and post-design for young children,

the effect of intervention could be controlled by analyzing changes in developmental speed before and after. In Study I, the children's outcome from the intervention and differences in changes between the groups were found to relate to the children's IQ before start of intervention and not to the type of intervention. A problem with naturalistic studies is the lack of control. A non treatment group would have increased the possibility to verify the outcome of an intervention. This study design could in theory have been used in study II and III. However, this was not possible to perform because of ethical reasons since all children receiving an ASD diagnosis should be offered treatment as early as possible.

In Study II, no significant differences between the study groups were found over a 12 -week period. On the other hand, we found significant changes between pre- and post-measures for both the IR/IBT and the IBT groups. It seems as if both intervention groups gained from the interventions as hypothesized. As an imitation-based intervention, IR was found to have an overall effect on the children's development. The intention of IBT was to change a number of the children's developmental domains as an effect of the comprehensive program.

Through statistical control we could see that both groups increased in their language and social development during the intervention period. The effect sizes found are comparable with the meta-studies of Reichow et al. (2012).

In the final Study III we found changes in the treatment group. These changes are in line with our hypothesis, but could only be verified at T3 and not at T2. Our findings at T3 are also in line with Salt et al. (2002) and Kasari et al. (2006).

Study III is limited by the low number of participants. The loss of participants as well as technical problems should have been anticipated. A larger group of 20 children at T3 in both intervention groups would have increased the strength of Study III.

External validity is the main issue in this thesis. The implementation process generated different research questions for studies I, II and III. Different outcomes of the IL/IBT method were also found in studies II and III. While randomized conditions and RCT especially try to examine an intervention during highly controlled circumstances to reflect the efficacy during "best circumstances", a naturalistic study like Study I evaluates the intervention in typical clinical settings with ordinary therapists.

Furthermore, in Study I, the question of participation asked from the parents only concerned whether they gave consent or not to the researchers using their child's assessment results from ordinary services without any further obligations of parents, child, or the child neurological health care or habilitation services. In the naturalistic study, the threats to validity are due to uncontrolled defection. There were parents who refused contact with the habilitation services and instead took part in external and private organizations to obtain interventions, and those who asked for a second opinion at another clinic.

In studies II and III all children who met the inclusion criteria were invited to participate in a specified research project that could generate positive expectations of both

a more qualified care and outcome. Nathan et al. (2000) suggest single-blinded trials of psychotherapy to reduce such impact. However in study II and III, it was impossible to withhold information about type of intervention due to parents and preschool teachers' roles as trainers, and spontaneous discussions between parents and measurement staff.

In study II and III, only children with severe EP were excluded, and some children did not participate due to their parents declining participation. Such restrictions will affect generalization in clinical practice. In addition, we do not know whether the effect holds for toddlers or children older than six years.

There is a limited applicability to individual children or subgroups of young children with ASD, since they form a heterogeneous group. For better guidelines and to increase the applicability of the methods used in this dissertation, a further examination of subgroups of children will be needed. Such knowledge could help the professionals to give advice to parents and preschools while also facilitating interventions to individuals. The subgroups could consist of children with varying intellectual functioning or different language levels. These are categories specified in DSM-5. Information on subgroups could improve accountability to clients, policy makers and financiers (Veerman & van Yperen, 2007).

The difference in findings in this thesis is also influenced by the measurements and scales reported. In Study I the assessment was based on ordinary clinical assessment tools such as Bayley-III, Griffith I and II, WPPSI-III, the VABS-II interview and the C-GAS scale. The measurements used in Study II were PEP-R and VABS-II, all of which reported outcomes presented as normed scores. Outcomes presented with subscales of valid instruments such as VABS have been reported in studies of Eikeseth (2008), Salt (2002) and Nadel (2014) for both IR and IBT interventions, as well as in a meta study of IBT (Reichow et al., 2012). Subscores and age equivalents or measurements specialized for children with an uneven or atypical development would better reflect how children develops. Cognitive ability is often reflected in earlier studies of IBT (Remington et al., 2007; Eikeseth, 2008) but not usually in Being Imitated studies with the exception of the study of Salt et al. (2002). Therefore, in studies II and III the results are presented with outcome measures that may have a social and communicative impact on the intervention on children with ASD. PEP-R, VABS-II and ESCS have all been used in earlier intervention studies (Nadel, 2014; Salt et al., 2002; Eikeseth et al., 2002; 2007; Eikeseth et al., 2012).

The outcome of the naturalistic study with IQ as a variable of importance is not surprising. The particular reliance on IQ in Study I is related to earlier IBT studies using IQ as an outcome measure (Lovaas, 1981/2002; Eikeseth et al., 2002). The IQ test has strong agreement with the ability of all kinds of learning i.e. experiential (cause and effect), social (observing other people in social situations), and academic learning (knowledge and skills learned through formal education). Trevarthen and Aitken (2003) did not find any correlation between IQ and communication and social development. They underline that social deficits in autism should not be attributed to cognitive deficits, since such severities

are primarily manifestations of a neurological disorder. In line with this argument, we excluded IQ as a primary variable of outcome in studies II and III.

Parents as trainers were examined by Bibby et al. (2001) who concluded that children performing low on cognitive tests also needed additional specialized training as a supplement to parent-training with IBT. Another measure that might supplement the outcome variables is parents' reports of family impact. These estimates could give support to interventions for children, especially for children with a low level of IQ and a high level of behaviour problems that can rarely be verified by standardized instruments. Both Salt et al. (2002) and Remington et al. (2007) found a no significant reduction of parental stress when using interventions such as the Being Imitated strategy or IBT. Grindle et al. (2009) reported increased family stress when implementing IBT programs. They also pointed to factors such as problems with access to professionally trained IBT therapists and disturbances of privacy due to receiving therapists at home (Grindle et al., 2009).

Clinical implications

All interventions must be controlled by follow-up assessments.

Supporting newly diagnosed children with ASD is more effective when IR and IL are combined. This means that habilitation services need to consider to provide resources for such implementation.

Delivery of treatments for children with ASD is most effective if professionals can change method when needed. This underlines the importance of a wider knowledge of methods among professionals.

The starting point for young children with ASD should always be their home or preschool, which enables training to be included in their everyday routines. This could strengthen the alliance between child, parents, preschool teacher and supervisor, since it would improve the chance of learning how the child acts and reacts at the place where the child spends most of its time.

Methods of both assessment and treatment should be offered to children by a “team” of professionals – adapted to parents’ and children’s wellbeing and development. Through such an extended assessment, less information will be lost and more time could be spent to support the child and their parents.

Evaluation of the child’s development during interventions should be verified by valid instruments. Study II found both PEP-R and VABS-II to be sensitive. The reported information given from preschool teachers was found to be reliable for the language and social variables used.

Supervision emphasizes parents as active agents in the planning process, training, analyses and self-evaluation of outcome in order to give them tools for further experiences.

Ethical considerations

The studies in this thesis were approved by the Regional Ethical Board in Gothenburg (numbers 494-08 and 418-10). Research with young children and their parents as part of clinical work means that ethical issues and concerns cannot be addressed only by adhering to the legal framework. An ethical issue before taking part of any information regarding studies II and III is that children must have been identified as having special needs before being invited to participate in the research. No child included in studies II and III had been offered treatment exclusively as taking part in a trial. No child had been given any placebo or inert intervention.

This research is based on therapeutic relations between participants and the research team in a way that was characterized by common sense and ethical commitment. The Declaration of Helsinki of the World Medical Association (WMA, 1964) has developed ethical principles for medical research. These standards could partly be applicable to this study of interventions in habilitation services of health organizations. In the fourth paragraph of WMA the intention of a professional had been formulated as to first of all considering the wellbeing and health of the patient (VMA, 1948). This was a benchmark for the research team.

All parents received both written and oral information about the study and all parents gave written consent but were also checked by the research staff during the ongoing intervention on whether they still wanted to participate voluntarily. The parents had been informed that withdrawal from the study would not have an impact on any further contacts with the habilitation organization. No incentives had been used.

The information about the study was given when parents have had an appointment with habilitation professionals. They were asked to take the leaflet home and send back to the researcher. A particular issue was to ensure that both parents were informed about the study before participation. The aim of the participation was to enable the inclusion of all children and their families, regardless of social, educational or cultural background. We used interpreters for all non-Swedish speakers.

All activities during the studies were planned with the aim to avoid personal and social harm to participants and researchers. All assessments and sessions were interrupted if the children showed fear or anxiety. Children and parents were supported and protected in such situations. When risks of any kind were found we took into consideration whether to continue, modify or immediately stop the intended part of the intervention or assessment.

Personal information and identity should not be disclosed. Every precaution must be taken to protect the privacy of research subjects and the confidentiality of their personal information. Beyond the habilitation organization and the University, as partners of the study, data were anonymous.

It was arranged for the participants to take contact with Head Manager of the Health & Habilitation, the VG Region, if they had any complaints, ethical or other, regarding how the study was performed.

All ethical principles above are compatible with every child's protection according to the Swedish Health Care Act (Hälso- och sjukvårdslagen, 1982:763) and the United Nations Convention on the Rights of the Child (1989).

Future directions of research

Important issues for future research include both replication of study II and III and longitudinal studies of the clinical groups in focus in this thesis.

Another important future project is to carry out a detailed analysis of subgroups, both investigating which children responded to intervention and identifying groups of non-responders to develop a better understanding of the learning abilities of ASD and their association with cognitive and language skills.

It is also important to analyze children's joint attention skills. Such studies could contribute to a deeper understanding of the development of social behaviours in IR and IL/IBT.

Another important issue to examine is whether an increased dosage of treatment has a positive effect or not, since the total time of treatment was low for the IR method. Further areas for studies include predictors and mediators of treatment.

Another urgent matter would be to provide education to professionals such as psychologists, special education teachers, speech and language specialists and preschool teachers concerning how to use imitation strategies and ABA strategies for children with special needs.

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