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QUALITY AND EFFICACY OF EARLY INTENSIVE AUTISM INTERVENTION: A MATTER OF TRUST, KNOWLEDGE, AND SUPERVISION?

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ABSTRACT

In contrast to previous research, mainly focusing on child characteristics and on quantity of treatment as essential moderating and predicting outcome factors of Early Intensive Behavioral Intervention (EIBI), the overall aim of this thesis was to examine the importance of different aspects of quality of EIBI when conducted in a community setting.

In Study 1, EIBI knowledge and allegiance levels among supervisors, supervised preschool staff, non-supervised preschool staff, school staff and parents were examined through an online survey (N=294). We found that EIBI supervisors showed significantly higher knowledge and allegiance than all other groups in the study. EIBI supervised preschool staff had higher levels of knowledge than preschool staff in general but were not more allegiant to the treatment. The results highlight the impact of supervision as a means to increase knowledge among preschool staff and particularly the importance of addressing ethical questions and evidence based practices in order to arbitrate potential misconceptions and negative attitudes. Moreover, the markedly low levels of prerequisite knowledge among preschool staff probably hampers the EIBI quality at the onset of a child's program and could be rectified by adding components of Applied Behavior Analysis methods and procedures to educational curricula.

In Study 2, we conducted the first validation of the York Measure of Quality of Intensive Behavioural Intervention (YMQI) in a non-Anglo-Saxon country. In our evaluation of 97 video-recorded sequences in a sample of 34 preschool children, the YMQI demonstrated moderate to excellent psychometric properties. We concluded that the YMQI showed feasibility when used in a community setting, but noted that the manual would benefit from some clarifications and modifications.

In Study 3, we investigated whether supervision, enriched with information from the YMQI assessment together with a workshop focusing on quality factors would increase delivered EIBI quality compared to regular supervision. We found that the enriched supervision group improved significantly on overall quality as well as specifically on quality variables concerning organization and planning together with the intensity of teaching, which indicates an increased awareness of the intention of the program as well as the trainer role. We conclude that standard clinical supervision would benefit from including an evaluation of video clips of preschool training sessions and additional relevant quality factors, such as programming and intensity of supervision along with formal education of both supervisors and trainers.

In Study 4, the predictive power of EIBI quality on treatment outcome after a period of 4 to 6 months was examined in a community setting. Multinomial logistic regression showed that overall quality predicted global outcome, and especially basic language and learning skills as well as global functioning. Specific quality factors of predictive value were organization, teaching level, generalization and differential reinforcement. These findings endorse the importance of EIBI quality assurance for treatment success.

LIST OF SCIENTIFIC PAPERS

I. **Långh, U**., Hammar, M., Klintwall, L., & Bölte, S. (2016). Allegiance and knowledge level of professionals working with early intensive behavioural intervention in autism. *Early Intervention in Psychiatry*. Advance online publication. doi: 10.1111/eip.12335

II. **Långh, U**., Cauvet, E., Hammar, M., & Bölte, S. (2017). Cross-Cultural Validation of the York Measure of Quality of Intensive Behavioural Intervention (YMQI). *Behavior Modification*. Advance online publication. doi: 10.1177/0145445517719397

III. **Långh, U**., Cauvet, E., Perry, A., Eikeseth, S., & Bölte, S. (2017). A pragmatic randomized controlled trial of standardized supervision on quality of early intensive behavioral intervention in autism. Submitted.

IV. **Långh, U**., Perry, A., Eikeseth, S., & Bölte, S. (2017). Quality of Early Intensive Behavioral Intervention as a predictor of children's outcome. In manuscript.

CONTENTS

LIST OF ABBREVIATIONS

ABA	Applied Behavior Analysis
AAC	Augmentative and Alternative Communication
ADHD	Attention Deficit-Hyperactivity Disorder
ADI-R	Autism Diagnostic Interview-Revised
ADOS	Autism Diagnostic Observation Schedule
APA	American Psychiatric Association
ASD	Autism Spectrum Disorder
BACB	Behavior Analyst Certification Board
BCBA	Board Certified Behavior Analyst
BCaBA	Board Certified Assistant Behavior Analyst
BST	Basic Language and Learning Skill Test
CGI	Clinical Global Impression Scale
DSM	Diagnostic and Statistical Manual of Mental Disorders
DTT	Discrete Trial Training
EIBI	Early Intensive Behavioral Intervention
ICC	Intra Class Correlation
ICD	International Classification of Diseases and Related Health
ICF	International Classification of Functioning, Disability and Health
IRR	Interrater Reliability
IT	Incidental Teaching
IQ	Intelligence Quotient
M-CHAT	Modified Checklist for Autism in Toddlers
NET	Natural Environment Teaching
RBT	Registered Behavior Technician
SRS	Social Responsiveness Scale
SRS-P	Social Responsiveness Scale – Preschool version
TEACCH	Treatment and Education of Autistic and related Communication Handicapped Children
VABS	Vineland Adaptive Behavior Scales
WHO	World Health Organization

YMQI   The York Measure of Quality of Intensive Behavioural
              Intervention

YSQA   The York System of Quality Assurance for Intensive Behavioural
              Intervention programs

1 INTRODUCTION

1.1 AUTISM SPECTRUM DISORDER

1.1.1 Classification and phenotype

Autism spectrum disorder (ASD) is an early onset neurodevelopmental disorder characterized by difficulties verbal and non-verbal social communication and interaction and excesses in stereotyped behaviors and interests causing functional impairments (American Psychiatric Association [APA], 2013; WHO, 1993). Over the years, the definition and labels used to describe the condition have varied and in the updated Diagnostic and Statistical Manual of Mental Disorders Fifth Edition (DSM-5) several diagnoses have merged into one: *Autism Spectrum Disorder*. The severity of each criterion should be specified as well as the presence (or absence) of other known disabilities or disorders. Moreover, symptoms must cause impairments in daily functioning and have been present in the early developmental period (APA, 2013).

Studies have shown increasing rates of ASD diagnoses. Prevalence is now estimated to be approximately 1-2% (Baird et al., 2006; Kim et al., 2011) with a recent Swedish study showing a rate as high as 2.5% among teenagers in Stockholm County (Idring et al., 2015). However, research also indicates that prevalence is rather unchanged if phenotype assessment is held stable (Lundström, Reichenberg, Anckarsäter, Lichtenstein, & Gillberg, 2015). Suggested explanations for the escalation of ASD diagnoses are primarily the broadening of the diagnostic concept, an increased awareness of ASD, as well as improved access to services (Elsabbagh et al., 2012; Wazana, Bresnahan, & Kline, 2007).

Males are diagnosed 2 to 4 times more often than females (Fombonne, 2005). However, the sex ratio is reported to be close to equal in the subgroup with lower level of intellectual functioning (Gillberg, Cederlund, Lamberg, & Zeilon, 2006; Volkmar, Szatmari, & Sparro, 1993; Wing, 1981). There is a high degree of co-morbidity with medical and neuro-psychiatric conditions such as epilepsy, attention deficit-hyperactivity disorder (ADHD), genetic syndromes and gastrointestinal problems (Bölte, 2014; Lai, Lombardo, & Cohen, 2014). The co-morbidity with intellectual disability is estimated 45% and the one with ADHD 28 to 44% (Lai et al., 2014).

There is no known single cause explaining the occurrence of ASD. On the contrary, there appear to be multiple etiologies. Research indicates that genetic and heredity factors can be accounted for as the major causes and environmental factors have a more modest impact (Lichtenstein, Carlström, Råstam, Gillberg, & Anckarsäter, 2010; Sandin et al., 2014). Siblings to children with ASD have an increased risk compared to the general population and the prevalence among siblings has been reported to be 19.5% (Messinger et al., 2015). Twin studies in Sweden and elsewhere contribute to the understanding of the different genetic and environmental risk factors that are involved and interact in the etiology of ASD (Bölte, Willfors, et al., 2014; Hallmayer et al., 2011). Neurobiological research indicates that

individuals with ASD have an altered cortical organization and an atypical neural connectivity underpinning the cognitive and behavioral alterations (Lai et al., 2014; Rane et al., 2015).

In order to understand the early course and development of ASD, research projects have followed siblings to children with ASD from early age (Bölte et al., 2013). According to the literature, the early trajectories vary in terms of age of first symptoms and behavior patterns. Moreover, ASD has a gradual onset and affects many areas of development apart from the core clinical areas. However, there is still not enough evidence of the early risk markers identified in siblings and their prognostic value in more general terms population (Rogers, 2009; Zwaigenbaum et al., 2015; Zwaigenbaum et al., 2009).

Deficits in social communication may be detected early as children with ASD often show fewer attempts of joint attention (Elsabbagh et al., 2013). Joint attention refers to reciprocal eye contact as a part of communication. A reduction in this ability may influence the development of social interaction with adults and peers, affecting learning of basic as well as more advanced play skills (Charman, 2003; Mundy, Sigman, & Kasari, 1994). Research has shown that children with ASD demonstrate lesser preference to attend to other people when compared with typically developed children (Falck-Ytter & von Hofsten, 2011). Dawson (2008) suggested that ASD is primarily a social motivation disorder and that children with ASD are less reinforced by social mediated stimuli (Chevallier, Kohls, Troiani, Brodkin, & Schultz, 2012; Klintwall & Eikeseth, 2014; Klintwall, Macari, Eikeseth, & Chawarska, 2015).

The ASD diagnosis entails impairments in communication as well as in the comprehension and understanding of language. Some individuals never develop speech, are restricted to a few words, or may use speech in an idiosyncratic manner such as echolalia, which entails the immediate or delayed repetition of a word or sentence (Neely, Gerow, Rispoli, Lang, & Pullen, 2016). Others develop a fluent language but may be limited to a concrete understanding and struggle with the abstract interpretation of words (Maljaars, Noens, Scholte, & van Berckelaer-Onnes, 2012; McGregor et al., 2012).

As previously mentioned, the other core clinical feature of ASD are repetitive and stereotyped behaviors, limited interests and activities. In children, this type of inflexibility may be manifested in various ways such as rigidly following routines, flipping objects, unusual interests or fixations on specific items or topics as well as flapping hands. These behaviors often preoccupy the child for long periods of time, are difficult to interrupt and reduce the child's possibility to learn from the environment.

Hypersensitivity increases the vulnerability of development of fears, aversions and specific phobias such as towards loud sounds (Kerns et al., 2014). Sensitivity to food consistency and texture is prevalent and together with inflexibility and demand of sameness often results in odd or problematic eating habits. Difficulties to discriminate between sensorial stimuli as well as the reduced ability to regulate and adapt to sensations often lead to a high level of

stress and overwhelming tiredness among individuals with ASD. This may affect the circadian rhythm and consequently, sleep disturbances, such as insomnia and early awakenings occur commonly in the group (Quine, 1992).

The many challenges that arise in everyday often results in challenging behaviors, such as self-injuries, high screams or aggressive behavior. These behaviors can be understood as an effect of insufficient approaches in combination with the individual's deficiencies, especially the lack of communication skills (Carr, 1997; Didden, Korzilius, van Oorsouw, & Sturmey, 2006).

There are several cognitive theories aiming to explain the characteristics of ASD, of which *Theory of mind* (Baron-Cohen, Leslie, & Frith, 1985) is commonly referred. The Theory of mind hypothesis describes the ability of having a mental representation of other individual's feelings, thoughts and needs which is an important competence when relating to other people. Individuals with ASD tend to have a concrete interpretation of the context which leads to difficulties in understanding different situations, often leading to social problems and misunderstandings.

Another theory refers to a *weak central coherence*, which alludes to a tendency to attend to details while at the same time lacking global understanding of the situation at large. This theory might capture the non-social features of ASD, i.e., special and limited interests (Frith & Happé, 1994; Happé & Frith, 2006). Indeed, a bias for details and the risk of missing the whole picture can also lead to barriers in social communication and relations (Nuske & Bavin, 2011).

Furthermore, it is hypothesized that reduced *executive functions* may cause the persistence and inflexibility when transitioning between activities or in adapting to new environments (Ozonoff & McEvoy, 1994; Rosenthal et al., 2013). Additionally, many individuals with ASD have either a hyper- or hyposensitivity reaction to stimuli, this dimension is also included in the latest version of the diagnostic manual DSM (APA, 2013).

1.1.2 Impact of functioning and quality of life

While individuals with ASD are heterogenic in presentation (Reichow, Barton, & Hume, 2012) an ASD diagnosis by definition is associated with major obstacles in everyday life.

The importance of functioning, as a parallel and separate dimension to symptomatology is described in the International Classification of Functioning, Disability and Health (ICF/ICF-CY; WHO, 2001; 2007). ICF defines disabilities as a combination of impairments in body functions and structures together with activity, limitation and participation restriction. In order to find the essential categories associated with ASD diagnoses in the comprehensive ICF classification system, research has been done to develop core sets for ASD (Bölte, de Schipper, et al., 2014; Bölte et al., 2017; de Schipper et al., 2016). The result of this work

demonstrates that the impact of ASD on functioning is much broader than the core symptoms, such as cognitive and motor skills, digestion and an overall difficulty to handle everyday tasks (de Schipper et al., 2016).

Growing up with ASD often entails fewer peer relationships and many report experiencing bullying and teasing in school (Fisher & Lounds Taylor, 2016). Children and adolescents often develop psychiatric problems such as anxiety and depression and have lower levels of general health and quality of life (Barneveld, Swaab, Fagel, Engeland, & de Sonneville, 2014; Domellöf, Hedlund, & Ödman, 2014; Lai et al., 2014). Furthermore, life-span prognosis has shown to be poor (Mordre at al., 2012) and a recent study by Hirvikoski et al. (2016) found an increased risk for premature mortality among individuals with ASD.

Parenting a child with ASD puts additional demands on families in terms of upbringing, special solutions and adaptions in everyday life as well as countless contacts with healthcare services and other authorities, parents often ending up as the coordinators of the various instances (Hayes & Watson, 2013). Accordingly, the literature also report a higher level of stress among parents to children with ASD as compared to other disabilities as well as an increased risk for psychiatric problems, sick leaves and divorce (Mugno, Ruta, D'Arrigo, & Mazzone, 2007; Rao & Biedel, 2009).

The economical lifelong cost of ASD including healthcare, educational as well as family expenses and productivity loss was estimated by Järbrink and Knapp (2001) to exceed £2.4 million in the United Kingdom while a recent study by Buescher, Cidav, Knapp, and Mandell (2014) reported the life time cost to be £1.5 million in the United Kingdom and $ 2.4 million in the United states. These studies highlight the importance of effective interventions in order to reduce the economic consequences of ASD in the society.

1.1.3 Diagnosing, health care and support

Theoretically, ASD might be diagnosed as early as 12 months of age, but usually the child is older when the assessment is conducted, due to the limitations of the diagnostic instruments' sensitivity and specificity for younger ages (Volkmar, Chawarska, & Klin, 2005). The ASD diagnosis is considered to be reliable from 24 months of age (Roll-Petterson, Olsson, & Ala'i-Rosales, 2016) and it is of paramount importance to detect atypical development early since it affects the access to early intervention, health care and support (Roll-Petterson et al., 2016).

The most common instruments used to diagnose ASD are the Autism Diagnostic Interview-Revised (ADI-R; Rutter, Le Couteur, & Lord, 2003) in combination with the Autism Diagnostic Observation Schedule (ADOS; Lord, Rutter, DiLavore, & Risi, 1999) together with developmental tests, measures of adaptive behavior and medical examinations. If possible and preferably, the diagnostic procedure also includes assessments by a speech and language pathologist and an occupational therapist.

In order to identify ASD as early as possible screening instruments such as Modified Checklist for Autism in Toddlers (M-CHAT) have been developed and tested in primary health care. However, the literature is somewhat inconsistent in the reported usefulness of a broad screening of all children (Höglund Carlsson, Gillberg, Lannerö, & Blennow, 2010; Nygren et al., 2012; Robins et al., 2014).

In Sweden, children with an ASD diagnosis have the right to free support and services through the county council habilitation centers, providing treatment and counselling to families (Socialstyrelsen, 1994). Through the municipality, the child has the possibility to receive extra support at the preschool and school as well as assistance in the home (Skolverket, 2010; Skolverket, 2016; Socialstyrelsen, 1994).

1.2 INTERVENTION

ASD is a behaviorally defined disability, not a disease, and most interventions have consequently focused on facilitating everyday life, favoring development and preventing worse outcomes rather than attempting to cure (Bölte, 2014; Jonsson et al., 2016; Oono, Honey, & McConachie, 2013; Reichow et al., 2012). However, there are a plethora of treatments, lacking systematic scientific evaluation, that promise major improvements and remedy (Green et al., 2006). In reaction to this and in order to promote the usage of evidence based practice, several organizations, e.g. the May Institute (National Autism Center, 2014) and Frank Porter Graham Development Institute (Wong et al., 2015), have conducted systematic reviews of behavioral and educational interventions, available free of charge on their websites. The evidence-based approaches in these reviews are predominately based on applied behavior analysis, involving teaching strategies as well as aids and support. However, in some cases, pharmacological treatments are justified.

1.2.1 Pharmacological treatment

There is no specific medical treatment for ASD core symptoms (Bölte, 2014; Kaplan, & McCracken, 2012; Statens beredning för medicinsk utvärdering, 2013). Associated behaviors such as aggression, hyperactivity and stereotypies may be treated by atypical antipsychotics and methylphenidate (Ritalin) (Hirsch & Pringsheim, 2016; Kaplan & McCracken, 2012; Research Units in Pediatric Psychopharmacology Autism Network, 2005). Other common problems such as depression, anxiety and irregular sleep patterns can be medicated but the effects have not been thoroughly tested (Bölte, 2014; Kaplan & McCracken, 2012). Pharmacological approaches should be used carefully and monitored thoroughly because of the side effects and always as one component of a broader comprehensive treatment (Hirsch & Pringsheim, 2016; Kaplan & McCracken, 2012).

1.2.2 Educational and psychological intervention

Different pedagogical methods are used to help individuals with ASD to gain skills or adapt to the environment. Some approaches such as Treatment and Education of Autistic and related Communication Handicapped Children (TEACCH) focuses predominantly on adapting the environment in order to suit the person's difficulties and interests (Welterlin, Turner-Brown, Harris, Mesibov, & Delmolino, 2012). Several types of Augmentative and Alternative Communication (AAC) tools such as pictures and signs are recommended and widely used (Light & McNaughton, 2012). Other commonly used strategies are social stories and comic strip conversations where social events are clarified and visualized by text or images to facilitate understanding and information processing by the individual with ASD (Gray & Garand, 1993; Glaeser, Pierson, & Fritschmann, 2003)

In addition to these specific compensating methods, there are several procedures established within the field of Applied Behavior Analysis (ABA) that can be used to enhance the development of new skills (Cooper, Heron, & Heward, 2007). ABA is a field which evolved in the deinstitutionalization period, in order to help people with learning disabilities to obtain independent living skills (Aylon & Azrin, 1968; Azrin & Foxx, 1971; Matson, Tureck, Turygin, Beighely, & Rieske, 2012). The field was defined in a pioneering article in the first number of Journal of Applied Behavior Analysis (Baer, Wolf, & Risley, 1968).

Core ABA principles and procedures such as reinforcement, shaping, chaining, prompting, generalization and the use of functional analysis have been evaluated in a magnitude of studies, using both single case as well as group design research methodologies. These studies have been conducted in a variety of clinical and educational settings and the evidence of their usefulness is considered to be robust (Matson et al., 2012; Wong et al., 2015).

Regarding children with autism the use of procedures and principles based on ABA have been applied in both focused and comprehensive early intervention programs such as Early Intensive Behavioral Intervention (EIBI) to increase the child's learning rate and developmental trajectories (Dawson et al., 2009; Eldevik et al., 2009; Lovaas, 1987; Reichow et al., 2012).

1.2.3 Early Intensive Behavioral Intervention (EIBI)

1.2.3.1 Background

In the sixties, Lovaas, Koegel, Simmons, and Long (1973) at the University of California, Los Angeles (UCLA) conducted a study on 20 children with ASD of which 13 were living in institutions and the remaining were outpatients. During 12 months, the children participated in a teaching program based on ABA with the purpose to teach language, communication, social and everyday learning skills to manage and prevent problem behavior. At the offset of the study, the children had achieved new skills and abilities and furthermore, the problem behaviors declined dramatically. However, many of the children lost these new skills when

the intervention period ended and consequently their problem behaviors returned to the initial level. This was especially evident for the children living in institutions. Nevertheless, this study has contributed to the understanding that children with ASD can learn with this new systematic way of evaluating and using behavior analytic procedures and principles, and that these can have a long-term effect on development (Lovaas et al., 1973).

Based on these findings, Lovaas and colleagues designed a new study only including children under 46 months of age, with a prolonged intervention period of two-years as well as with higher treatment intensity in order to provide learning opportunities throughout the day addressing all deficits inherent in ASD (Lovaas, 1987). In addition, the intervention was home based and one of the parents was expected to be actively involved in the treatment. The nineteen children receiving 40 hours of therapy per week made up the high-intensity group. The two control groups consisted of in total forty children, who obtained 10 hours per week, or less, of treatment. Nine of the nineteen children in the most intensive group (47%) reached normal intellectual level and obtained regular school placement without special education support, compared to one of forty (2%) children in the control group. Due to parent protests and ethical considerations the study was quasi randomized rather than strictly randomized. A follow-up study showed that eight of the nine children when they were on average 13 years of age still had regular school placement and were indistinguishable from their peers (McEachin, Smith, & Lovaas, 1993).

Several authors have raised questions concerning the validity of the results in these two studies (Gresham & MacMillan, 1997a, b; Howlin, 1997; Jordan, Jones & Murry, 1998, Mesibov, 1993; Schopler, Short, & Mesibov, 1989) and rebuttals have been made by Lovaas and colleagues (Lovaas, Smith, & McEachin, 1989; Smith & Lovaas, 1997) as well as by Eikeseth (2001) to clarify agreements and disagreements on possible conclusions to be drawn by Lovaas' research. Nevertheless, despite criticism the promising results have led to the development of numerous research projects to improve methodological shortcomings and explore the possibility to replicate initial results.

1.2.3.2   Description of EIBI

EIBI programs are based on a teaching curriculum, divided in parts from early and basic skills to more complex and independent skills (Eikeseth, 2010). Several standardized manuals exist describing curriculums relevant for EIBI (Harris & Weiss, 2007; Leaf & McEachin, 1999; Lovaas, 2003; Maurice, Green, & Foxx, 2001; Maurice, Green, & Luce, 1996). The standardized curriculum is individualized and tailored to each child and setting based on assessment of the child's deficits and excesses and follow, when possible, typical development (Eikeseth, 2010). It is usually recommended that an EIBI program begin before the age of five and last at least two years, with an intensity of 20 to 40 hours per week (Klintwall & Eikeseth, 2014; Reichow et al., 2012).

Depending on the skill in focus together with an evaluation of where the child is in the learning process and potential need of adjustments, different learning arrangements are used

either in a one-to-one format or as group instructions together with other children (Maurice et al., 1996; Klintwall & Eikeseth, 2014). *Discrete Trial Training* (DTT) is a one-to-one format, characterized as a planned and clearly defined learning situation that enables teaching in small steps and repeated learning opportunities (Eikeseth, Smith, & Klintwall, 2014; Lovaas, 2003). *Incidental Teaching* (IT) is based on the child´s initiative and is especially useful when teaching communication skills (McGee, Morrier, & Daly, 1999; Hart & Risley, 1975). *Natural Environment Teaching* (NET) implies learning in natural and unstructured situations letting the child to in get in contact with the natural reinforcers of a behavior (Schreibman et al., 2015).

A basic element of all EIBI programs is structured delivery of positive reinforcement, grounded in a thorough reinforcement assessment. In addition, systematic prompts and prompt fading are used to show and help the child to succeed and carry out a task independently without having to fail in the process (Klintwall & Eikeseth, 2014; Lovaas, 2003). Approaches such as visual support and Augmentative and Alternative Communication (ACC) are incorporated in the treatment together with the different ABA procedures (Eikeseth & Jahr, 2001; Frost & Bondy, 2002, Wong et al, 2015).

An additional component is a functional assessment of challenging behavior including fears and other troublesome behaviors, which is conducted in order to choose appropriate actions and steps (Cooper et al., 2007; O'Neill, Albin, Storey, Horner, & Sprauge, 2015). Commonly occurring difficulties such as feeding problems and sleep disturbances are included and managed within the program (Leaf & McEachin, 1999).

EIBI programs can be delivered to children in different setting such as clinical-, home-, or community-based. In a clinically based setting, the interventions are implemented by well-experienced staff with formal ABA credentials and with the access to an in-house supervisor whereas in a home-based program the parents together with hired staff or students conduct the treatment in the home. School or preschool are the most common community-based training environments for children receiving EIBI (Eldevik, Hastings, Jahr, & Hughes, 2012; Klintwall & Eikeseth, 2014). Programs delivered at home or at preschool have the advantage of a natural learning environment and the benefit of typically developing peers being available, increasing the probability that skills acquired during the program will generalize to social situations beyond EIBI training (Lovaas, 2003). On the other hand, since regular preschool staff perform the treatment, a major challenge is to achieve and maintain sufficient EIBI quality and thus generate necessary prerequisites for optimal intervention outcomes (Eikeseth, 2010). In Sweden, the county council's habilitation centers are responsible for providing treatment services to children with ASD, including supervision, while the preschools in the municipalities have the day-to day care. Thus, in order to begin an EIBI program the habilitation center, the preschool and the parents need to reach a mutual agreement and specify everyone's commitments.

Taken together, implementation of EIBI programs place high demands on the supervisor responsible for assessments and programming as well as on the staff and parents

implementing the program (Eikeseth, 2010). Several authors have raised concerns and argue that without adequate competence, EIBI will be performed in an inflexible and rigid manner (Eikeseth, 2010; Leaf et al., 2016). Therefore, the Association of Behavior Analysis (ABAI; International (ABAI; https://www.abainternational.org/) advocates and promotes high standards for ABA treatments. Furthermore, to protect consumers of behavior analysis services on a global basis the Behavior Analyst Certification Board (BACB; https://bacb.com/) has established guidelines defining university course content and professional standards that are internationally recognized for certification purposes. These include Board Certified Behavior Analysts (BCBA) applicable for supervisors as well as Board Certified Assistant Behavior Analyst (BCaBA) and Registered Behavior Technician (RBT) available for preschool staff depending on educational degree. However, despite a growing interest concerning quality assurance internationally, this credential system is not officially recognized in Sweden, though the numbers of BCBAs are increasing (at present 15 BCBAs and one BCaBA). One explanation for this is the shortage of adequate university based graduate training programs in which Stockholm University is the only exception in Sweden, providing coursework meeting BACB standards with a focus on autism (Keenan et al., 2015) to professionals with backgrounds such as psychologists, special educators, speech and language pathologists, occupational therapists, and social workers.

In addition to the high level of competencies required of EIBI supervisors, staff and parents need sufficient understanding and experience in order to conduct and implement the program in daily practice (Leaf et al., 2016). Zakirova Engstrand and Roll-Pettersson (2014) found that only 14% of the preschool teachers in their sample received their knowledge during their university training, indicating a substantial lack of competence among preschool staff, and moreover, Eldevik et al. (2012) report negative attitudes by some staff participating in their study. To assure competence, some centers in the United States and Canada require that staff has BACB credentials, but these thresholds are still out of reach for many countries, including Sweden. Thus, currently, in Sweden EIBI quality management is not based on the BACB guidelines, since the services are mostly municipality-based (education services), with regulations and directions differing from those of the county council (health care) (Roll-Pettersson et al., 2016), in which health care recommendations by the association for habilitation directors (Föreningen Sveriges Habiliteringschefer, 2004), advocate EIBI, while preschool national curriculum does not (Lpfö 1998 revised 2016; Skolverket, 2016).

1.2.3.3 EIBI outcome research

Lovaas' pioneering study has been followed by subsequent research aiming to replicate previous findings. Below is a summary of original EIBI outcome studies and findings from meta-analyses and systematic reviews.

Birnbrauer and Leach (1993) conducted a study on children with severe developmental disabilities and autism comparing EIBI (n=9) with a matched control group (n=5). Although the EIBI group received lower intensity (28.7 h/week) compared to Lovaas study (1987), the

result showed substantial improvements, after 24 months of treatment, in four of the children in the EIBI group compared to one in the control group.

Sheinkopf and Siegel (1998) evaluated home-based delivered EIBI to 11 children compared to a matched group receiving conventional school based treatment. The EIBI group obtained significant higher IQ and decreased autism severity, however still fulfilled the diagnose criteria. The authors concluded that home-based EIBI assisted by community-based clinicians can be an alternative to an academic setting.

In a controlled randomized study by the research group at UCLA (Smith, Groen, &Wynn, 2000), results showed positive gains for the intensive group (24.5 h/week) compared to a parent mediated training group (5h/week). However, the best outcome group was 27% compared to 47% in Lovaas' (1987) study. Lower intensity is discussed to be one of the explanations to this result. Another finding in this study was that children with milder autism tended to have better outcome.

Bibby, Eikeseth, Martin, Mudford, and Reeves (2002) report data from parent-managed treatment of 66 children in the United Kingdom and concluded that this service model could not replicate earlier results since no child neither reached a normal IQ range nor attained non-assisted mainstream school. Although the children made some progress the authors raised concerns regarding the adequacy of this service model, among other things, because none of the supervising consultants were authorized to provide the UCLA intervention model and the amount of supervision was considerably reduced compared to Lovaas' study

In a Norwegian study, Eikeseth, Smith, and Jahr (2002) compared EIBI (n=13) with eclectic treatment (n=12) in preschools/schools among children 4 to7 years of age. Group placement was based on the availability of EIBI supervisors. An independent habilitation director allocated participants to either group and both groups received the same amount of treatment with a mean of 28.52 hours per week. The results showed that the EIBI group had better outcome on the standardized measurements after one year of treatment. Findings from a 2-year follow up (Eikeseth, Smith, Jahr, & Eldevik, 2007) revealed that the EIBI group in addition to previous results also demonstrated less deviant behaviors and fewer social problems. The result indicate that elements other than intensity are important to obtain substantial developmental gains, that children older than 4 years of age can benefit from intensive treatment, and that EIBI can be successfully implemented in a community based school setting.

Howard, Sparkman, Cohen, Green, and Stanislaw (2005) also evaluated community delivered EIBI compared to eclectic interventions. In their study the EIBI group (n=29) received 25 to 40 hours/week whereas control group 1 (n=16) and control group 2 (n=16) obtained 30 h/week and 15 h/week, respectively. Assignments to groups were parent-determined and not random. However, the three groups did not differ significantly at baseline while at follow-up the EIBI group scored higher on all measurements and had higher learning rates compared to the two control groups, which in turn, did not differ significantly.

Comparing a clinic based UCLA model (n=13) with parent-directed EIBI (n=10), Sallows and Graupner (2005) found no outcome differences between these two settings after 4 years. When combining the two groups they found that 48% of the children were considered as "rapid" learners, which is comparable with Lovaas' best outcome group. The best outcome group had a mean intensity of 34 h/week year one and 31 h/week year two.

Cohen, Almerine-Dickens, and Smith (2006) replicated the UCLA model in a community based preschool setting. The experimental group (n=21) had 35 to 40 h/week EIBI compared to an IQ matched group (n=21). They used a quasi-experimental design study, in which allocation to respective group was based on parental preference. The EIBI group obtained higher IQ and adaptive skills and six of the children (29%) obtained regular school placement without additional support (year 3) and eleven children (52%) were included with extra support compared to only one child in the control group (5%).

Remington et al. (2007) compared home-based EIBI (n=23), university directed or privately funded programs, with treatment as usual within the United Kingdom educational system (n=21). As in Cohen et al. (2006), participants were assigned to each group based on parents' preference. After two years the children in the intensive group had made significant higher gains compared with the control group on measures of intelligence, language, daily living skills and positive social behavior. Moreover, the home-based program did not increase parental stress ratings.

In a two year prospective follow-up study Magiati, Charman, and Howlin (2007) compared two groups of children enrolled in either home-based EIBI (n=28) or preschool based eclectic program (n=16). Results show that both group improved on age equivalent scores but change on standard scores were limited and they report no significant differences between groups.

Perry, Cummings and colleagues (2008) reported outcomes for 322 young children participating in a community-based program in Ontario, Canada. Although this study lack a comparison group the authors concluded that EIBI can be successfully implemented in a community setting with non-ideal circumstances, since 75% of the children showed gains and 11% reached average functioning.

Fernell et al. (2011) examined the outcomes in 208 children obtaining services in a Swedish habilitation center and found only limited evidence for EIBI compared to less intensive ABA interventions, particularly for low IQ autistic children. However, the authors do not report amount of treatment for the assumed less intensive group and thus hamper a comparison based on intensity.

An evaluation of EIBI in Norwegian mainstream preschools by Eldevik et al. (2012) in which the EIBI group (n=31) was compared to a control group receiving treatment as usual (n=12) group assignments were based on geographical location. Despite a relatively low intensity, overall 13.6 h/week, the EIBI group scored significantly higher than the treatment as usual group on IQ measures and adaptive level.

In addition to these outcome studies several reviews and meta-analyses have been published. In a review of different comprehensive treatments Rogers and Wismara (2008) stated that EIBI is "well established" but that no treatment met the criteria of "probably efficacious". A systematic review of 11 studies by Howlin, Magiati, and Charman (2009) concluded that there is support for EIBI effectiveness on a group level but the outcome is highly variable. Eikeseth (2009) identified 25 studies regarding comprehensive interventions, 20 of them consisted of behavioral treatment. Based on the studies' scientific values as well as magnitude of outcomes, the author concluded that there is sufficient evidence to recommend EIBI. However, Spreckley and Boyd (2009) found the evidence to be inadequate, and Parsons et al. (2011) stated that it is unlikely that one single treatment can produce best outcome for all children with ASD. Warren et al. (2011) noted improvements in cognitive level, language and adaptive behavior skills but raised methodological issues.

Reichow and Wolery (2009) included 12 studies in their comprehensive synthesis report of descriptive analysis, effect analysis and meta-analysis. Their findings reveal a mean effect size of 0.69, suggesting that EIBI is in overall effective for increasing IQ scores but not for all children. Eldevik et al. (2009) included nine studies in a meta-analysis and reported a large average effect size of 1.10 for change in full-scale intelligence, while they found a moderate effect size of 0.66 for change in adaptive behavior composite. They conclude that present data support EIBI to be the current treatment of choice. In a synthesis of 14 studies Makrygoanni and Reed (2010) found in their meta-analysis moderate to high effect sizes and that behavioral programs are effective in comparison to eclectic interventions. In another meta-analysis of 22 studies by Virués-Ortega (2010) medium to large positive effect of EIBI were noted, and findings from Peters-Scheffer, Didden, Korzilius, and Sturmey (2011) strongly support the effectiveness of EIBI in their analysis of eleven studies. Klintwall, Eldevik, and Eikeseth (2015) analyzed data from 453 children and reported learning rate instead of change in standard score. They found an effect size of 0.85 in IQ learning rate and a 75% faster learning rate in the EIBI group compared to controls. Corresponding findings for adaptive behavior was an effect size of 0.48 and a 38% higher learning rate in the EIBI group.

A systematic review from the Cochrane collaboration (Reichow et al., 2012) concluded that there is some evidence to support the use of EIBI, but that the support mostly relies on non-randomized studies. The Swedish Council on Health Technology Assessment (Statens beredning för medicinsk utvärdering, 2013), also stated in a systematic review that the evidence is still insufficient due to the lack of RCT research designs, but emphasize that despite the methodological problems with these type of studies there is promising ongoing well designed research. In addition, Bölte (2015) raised concerns about the hazards of systematic reviews in which too rigorous criteria could lead to the exclusion of important and informative studies, thereby falsely signaling "no evidence for anything". Most contemporary studies are conducted in community settings and the choice of interventions are based on parent's preference, which make it difficult to have a randomized procedure. Moreover, a prolonged treatment as in EIBI makes a waiting list control group ethically problematic, especially since most researchers and clinicians agree on the importance of early intervention.

Another issue, frequently discussed, is what should be considered as good or successful outcome. In Lovaas' study (1987) the main result, presented as school placements, was opposed and debated since that measure does not necessarily reflect changes in ASD core symptoms (Lovaas, 1987). Consequently, the follow-up study (McEachin et al., 1993) included measures regarding social functioning and the wording "recovery" was changed to "normal functioning". Matson and Goldin (2014) conclude that assessment of core symptoms, as well as challenging behavior and comorbid psychopathology are rarely included in EIBI studies and hence, the total effectiveness of the intervention cannot be evaluated.

Most outcome studies have measured children's mental age/intelligence and the level of adaptive behaviors (Eldevik et al., 2009; Reichow et al., 2012) and results are often reported as standard scores. Klintwall, Eldevik, et al. (2015) pointed out that outcome measured in standard scores could be misinterpreted as regression for children with slow learning rate and instead recommended the use of age-equivalents and calculated learning rate, presented as developmental trajectories. MacDonald, Piarry-Cruwys, Dupere, and Ahearn (2014) argued that children's progress can be difficult to detect with instruments measuring overall functioning and promoted direct and observational measures. Moreover, Dawson et al., (2012) used EEG as a secondary outcome measure and found a normalizing effect on brain activity associated with improvements in social behavior after two years of early behavioral intervention.

Lastly, aside from McEachin et al. (1993), there are very few follow-up studies evaluating the long-term effects of EIBI (Matson & Konst, 2013; Matson et al., 2012). Kovshoff, Hastings, and Remington (2011) found in a 2-year follow-up that previous group differences had weakened and underscore the importance of active programs and the need of an increased understanding of which children benefit from follow-up treatment.

1.2.3.4   Summary

Several outcome studies, with different angles and perspectives, have been published aiming to replicate Lovaas' findings on EIBI (Lovaas, 1987; McEaching et al., 1993). EIBI has been tested in different community settings with good results (Cohen et al., 2006; Eikeseth et al., 2002; Howard et al., 2005; Sallows & Graupner, 2005) and evaluated when implemented in the regular preschool system (Eldevik et al., 2012; Perry, Cummings, et al., 2008). Studies following Lovaas' research protocol as described features in Green, Brennan, and Fein, (2002) tend to have better outcome compared to more loosely implemented approaches (Bibby et al., 2002; Fernell et al., 2011; Magiati et al., 2007). In addition, findings show promising results with lower levels of intensity (Birnbrauer & Leach, 1993; Eikeseth et al., 2002; Eldevik et al., 2012; Remington et al., 2007; Smith et al., 2000) and that older children can benefit as well (Eikeseth et al., 2002). Smith et al. (2000) published the only study using a randomized control procedure, with most studies using a match group design.

Although the evidence is still limited, the research on EIBI shows promising overall outcome on a group level but the individual variability is considerable. Many factors can explain the outcome diversity between subjects and an important research area is to find outcome predicting elements and essential treatment ingredients as well as appropriate outcome measures.

1.2.4 Treatment outcome moderators and mediators

Outcome factors can be divided into characteristics of the child (e.g. age, severity of symptoms and developmental level), the family (e.g. socio-economic status, parental stress, number of siblings) and treatment (e.g. intensity, content, quality) (Perry & Freeman, 1996).

1.2.4.1 *Age at intake*

Harris and Handleman (2000) reported that younger age of intake predicted regular school placement. Flanagan, Perry, and Freeman (2012) concluded that early age of treatment onset increases the likelihood of better outcomes and Perry, Blacklock, and Dunn Geier (2013), comparing a younger group (2 to 5 years) with an older group (6 to14 years), found that the younger group made greater gains, suggesting the importance of an early start. Perry et al. (2011) also found that youngest children have the best outcome. These findings are consistent with Eldevik et al. (2012) who found that age at intake correlated positively with gains in adaptive scores, as well as with the recent study by Smith, Klorman, and Mruzek (2015) which also found that younger age predicted favorable outcome.

However, the findings are somewhat inconsistent. Eikeseth et al. (2007) did not find that intake age predicted outcome or treatment gains and neither Rogers and Wismara (2008) nor Howlin et al. (2009) found enough support for the importance of age as a critical factor in their reviews. Moreover, age did not predict learning rate in the study by Klintwall, Eldevik et al. (2015) but they conclude that very young children have a greater chance to reach the normal developmental zone since the developmental gap to typically developing peers is of yet not that great.

1.2.4.2 *Pretreatment developmental level*

Another predicting factor is the child's initial developmental level as defined by intelligence quotient (IQ) or age equivalence. Harris and Handleman (2000) report that in addition to younger age higher IQ at intake predicts regular school placement. Rogers and Wismara (2008) refer to several studies that have shown IQ to be a powerful predictor while on the other hand, Howlin et al. (2009), reported inconsistent support for IQ as a predictor. Perry et al. (2011) and Perry et al. (2013) found cognitive level at onset to be the strongest predictor as did Eldevik et al. (2012), who reported a positive correlation between IQ at intake and improvements in socialization. Finally, Smith et al. (2015) found that higher initial IQ predicts favorable outcome, as in the study by Klintwall, Eldevik, et al. (2015) where intake IQ predicted learning rate.

1.2.4.3 Pretreatment adaptive level and severity of symptoms

Smith et al. (2000) found that children with milder symptoms (not fully diagnosed with autism) tended to have better outcome and this was also reported by Eldevik et al. (2012) who detected a positive correlation with progress in adaptive domains such as communication and daily living skills. Sallows and Graupner (2005) concluded that outcome was best predicted by level of imitation, language and social responsiveness prior to treatment. Perry et al. (2011) reported both adaptive level and severity of symptoms as predicting factors. Klintwall, Eldevik et al. (2015) on the other hand did not find that initial level of adaptive behavior predicted learning rate. However, Klintwall, Macari, et al. (2015) observed that level of motivation as measured by interest in toys, social routines, and activities predicted skill acquisition, confirming earlier findings suggesting that the amount of socially mediated reinforcers positively correlates with outcome (Klintwall & Eikeseth, 2012). When controlling for other factors, Smith et al. (2015) found that social engagement predicted later IQ and adaptive levels. In contrast, repetitive behavior and rituals did not have similar predictive value.

1.2.4.4 Family factors

Family characteristics are under-reported in EIBI studies and hence there is little information of the predictive power (Wolery & Garfinkle, 2002). A family factor that has been found to have a negative impact on EIBI treatment outcome is parental stress (Robbins, Dunlap, & Plienis, 1991; Osborne, McHugh, Saunders, & Reed, 2008; Shine & Perry, 2010).

Robbins et al. (1991) assessed a number of different family variables at intake and the clearest relationship was between maternal stress and children's progress. Similar findings were made by Osborne et al. (2008) concluding that EIBI effectiveness was hampered by high levels of parental stress. Shine and Perry (2010) noted similar trends but the relationships were weaker and not significant. In a recent study by Eikeseth, Klintwall, Hayward, and Galeb (2015) the authors assessed high levels of parental stress at intake. However, while maternal stress decreased during the first year of treatment the authors did not detect any relationship between parental stress at intake and children's improvements in functioning.

Nevertheless, stress is an important aspect to address since parents often are extensively involved in delivering the treatment and parental stress has been found to influence self-efficacy and performance (Hastings & Symes, 2002).

1.2.4.5 Quantity factors

The importance of amount of treatment has been debated since the first study by Lovaas (1987) that compared two levels of intensity (40 hours and 10 hours) with prolonged treatments up to eight years for some of the participants (Lovaas, 1987). However, despite the emphasis on intensity in EIBI there is no consensus in how to measure the amount of treatment (Eldevik et al., 2012) and how to count learning opportunities outside the actual

training situation (Rogers & Wismara 2008). Thus, possible differences in measurements of training hours complicates comparison between studies and conclusions become a bit arbitrary.

Nevertheless, several studies report a positive effect on intensity and outcome (Eldevik et al., 2010; Reed, Osborne, & Corness, 2007; Virues-Ortega, 2010) and studies with higher intensity tend to have larger effect sizes (Virues-Ortega, 2010). Klintwall, Eldevik, et al. (2015) also found that intensity of treatment predicted the children's learning rate. In a recently published study by Linstead et al. (2017) high intensity was strongly related to mastery of learning objectives.

Studies by Eldevik and colleagues (Eldevik, Eikeseth, Jahr, & Smith, 2006; Eldevik et al., 2012) showed superior treatment progress with less intensity (12 hours and 13.6 hours per week, respectively) when compared to eclectic controls, even though the effect tend to be lower than to high-intensity treatment. Peters-Scheffer, Didden, Mulders, and Korzilius (2010) reported an intensity on average 6.5 hours per week and that the group receiving behavioral treatment had after 8 months higher developmental age and improved adaptive behavior skills compared to controls but no differences were found on autism severity or on emotional and behavioral problems.

The total amount of EIBI treatment also depends on the duration, that is, the length of the treatment period. The description of EIBI duration and recommended length varies from one to four years (Eldevik et al., 2009; Matson & Konst, 2013; Reichow et al., 2012). In a study by Virués-Ortega, Rodrígues, and Yu (2013), their longitudinal analysis indicates that total intervention time is an important outcome factor.

Eikeseth et al. (2007) found that the largest improvement on IQ and communication was made the first year of treatment whereas additional gains on overall adaptive level and socialization was made the second year highlighting the importance of at least two years of treatment.

1.2.4.6 EIBI program content

EIBI programs are comprehensive and address many developmental areas (Eikeseth, 2010; Föreningen Sveriges Habiliteringschefer, 2004). Several manuals and curriculum guides describe both appropriate order of skills and detailed examples of how to conduct each step (Leaf & McEachin, 1999; Lovaas, 2003; Maurice et al., 2001; Maurice et al., 1996).

Attempts have been made to define key learning behaviors, also labelled pivotal behavior (Koegel, Koegel, & Mcnerney, 2001) or behavioral cusps (Rosales-Ruiz & Baer, 1997) and areas such as self-initiation and responsiveness to multiple cues have been suggested as examples of core areas (Koegel et al., 2001). The concept of cusps is helpful to select and prioritize behavior and skills that give the child increased access to new reinforcers, contingencies and environments (Bosch & Fuqua, 2001).

There are however few studies comparing different program ingredients and their relation to learning outcomes (Kasari, 2002). In the study by Smith et al. (2000), they reported that early acquisition of verbal imitation and labeling was linked to better outcome, indicating the importance of teaching language. Sallows and Graupner (2005) found a positive correlation between gains in social skills and the amount of supervised peer play and highlight the importance of helping parents to arrange social meetings and play times with peers.

According to Kasari (2002), few studies have systematically evaluated interventions aiming to improve joint attention and symbolic play, despite the empirical support of their significance, and points out the need of methodological valid studies comparing different content and focus. Matson and Goldin (2014) raised the question of how to prioritize behaviors for intervention since children with ASD tend to have a number of comorbid problems accompanying the core deficits and noted that none of the studies in their review described the prioritizing process.

1.2.4.7 Quality factors

Another critical factor which has been raised concerns how well the interventions are performed, that is, the quality of treatment (Penn, Pritchard, & Perry, 2007). For example, Bibby et al. (2002) reported limited outcomes and discussed these shortcomings in terms of amount of supervision and supervisor competence affecting quality and treatment integrity.

Eldevik et al. (2012) experienced serious challenges when starting-up EIBI in mainstream preschools, owing to personnel being unfamiliar with, and possibly negative to EIBI methods. Practitioner's attitude towards EIBI has been found to be important with respect to treatment fidelity and the quality of performance (McLeod, 2009) and consistently Klintwall, Gillberg, Bölte, and Fernell (2011) showed a positive relation between attitude and treatment outcome.

Eikeseth and colleagues found a positive correlation between amount of staff supervision and outcome, indicating that intensity of supervision is an important outcome factor (Eikeseth, Hayward, Gale, Gitlesen, & Eldevik, 2009). Several authors have underscored the importance of the supervisor's tutorial style in order to increase both staff knowledge and allegiance (Davis, Smith, & Donahoe, 2002; Grey, Honan, McClean, & Daly, 2005; Hastings & Symes, 2002; Jahr, 1998; Symes, Remington, Brown, & Hastings., 2006). Findings indicate that effective supervision is characterized by a combination of theoretical and practical guidance (van Oorsouw, Embregts, Bosman, & Jahoda, 2009), together with a perceived personal support by the supervisee (Gibson, Grey, & Hastings, 2009).

A community-based program often involves preschool staff with no prior knowledge of ABA or EIBI, emphasizing the importance of an evaluation, not only of the child's progress, but also of the staff's implementation. In line with this, Eikeseth (2010) compiled the necessary components to become a qualified EIBI-professional including both advanced theoretical knowledge and extensive practical training. However, quality of implementation is also dependent on the performance of procedures and techniques by staff and parents actually conducting the training (Penn et al., 2007).

Few attempts have been made to link quality of treatment to outcome and there are no widely used standard instruments to evaluate EIBI performance (Penn et al., 2007). Koegel, Russo, and Rincover (1977) identified five categories of teacher competence which have been both assessed and successfully trained. They found that child's improvements depended on the teacher's training and furthermore that the teacher through the specific training learned generalized effective behavior. Davis et al. (2002) validated an assessment procedure aimed to evaluate supervisors on theoretical knowledge, programming as well as training performance. However, both of these assessments (Davis et al., 2002; Koegel et al., 1977) are limited to the UCLA intervention model and do not cover a broader variety of quality indicators.

In the absence of a common quality assessment instrument, Perry and colleagues developed the *York Measure of Quality of Intensive Behavioural Intervention* (YMQI) (Penn et al., 2007; Perry, Flanagan, & Pritchard, 2008). In order to assure the quality of EIBI delivered in Ontario, Canada, the YMQI evolved within a broader system (*The York System of Quality Assurance for Intensive Behavioural Intervention programs, YSQA)* including evaluations of programming and education levels as well as parent and staff surveys (Perry, Koudys, & Sheese, 2008). The YMQI is designed to measure performed EIBI quality on collected video clips and covers 31 quality factors. The psychometric properties have been established (Denne, Thomas, Hastings, & Hughes, 2015; Penn et al, 2007; Perry, Flanagan, et al, 2008; Whiteford, Blacklock, & Perry, 2012) and the instrument has been used in research (Denne et al., 2015) as well as a feedback tool in community school settings (Hoerger, 2011; Jaffrey & Hoerger, 2011).

1.3   SUMMARY AND CONCLUSIONS

ASD is one of the most common disabling developmental conditions often accompanied by additional difficulties and associated with poor prognosis. Hence, there is a demands for adequate services and interventions that address the behavioral deficits and excesses associated with ASD in order to improve quality of life for individuals and families as well as reducing societal costs.

During the last 50 years, approaches, methods and techniques have step-by-step been tested and evaluated within the field of behavior analysis. This cumulated knowledge has been applied in comprehensive EIBI programs for children with autism. Research has shown that most children benefit from intensive treatment, but the variability in response is substantial and it is still unclear to what extent achieved improvements will persist over time. More RCT studies are needed to strengthen the evidence but this is a challenge since EIBI is a prolonged treatment mainly conducted in clinical settings.

Based on research, EIBI is recommended to children younger than age 4 years, with an intensity of 20 to 40 hours/week and a duration of at least one year. The EIBI program is comprehensive, addressing all developmental areas and teaching is based on ABA procedures and principles. However, the findings are somewhat inconsistent and incomplete and

recommendations for practitioners can mainly be given on a group level and not for specific children.

It is important to identify moderating and mediating factors and increase the knowledge on how these components covary and interact, in order to answer questions such as whether certain child or family characteristics imply particular program content or a specific range of intensity.

Furthermore, there are limited data on which treatment factors interactive with each other and which can function compensatory, for example, if EIBI is performed with high quality could that factor result in good outcome even with lower level of intensity?

These questions become especially salient when EIBI is implemented in community settings with limited resources and insufficient competence. It is important to know how much and in what sense the treatment can deviate from the recommendations and still be called EIBI. Moreover, it is essential to have a way to ensure and monitor different aspects of treatment integrity.

These current knowledge gaps concerning EIBI quality and outcome in community settings resulted in the four studies comprising this thesis with corresponding research questions.

2 AIMS OF THE THESIS

The overall aim of this thesis is to study and highlight the importance of quality of delivered treatment when evaluating EIBI both in research and in clinical practice.

2.1 STUDY 1

The aim of this study was to survey levels of knowledge concerning allegiance towards EIBI in groups of individuals working with EIBI, who meet children with ASD in educational settings or living closely to them. These groups include supervised preschool staff conducting EIBI, unsupervised preschool staff not conducting EIBI, behavior modification experts, school staff, and parents of children with ASD.

2.2 STUDY 2

The objective of this study was to evaluate the cross-cultural validity of the York Measure of Intensive Behavioural Intervention (YMQI) in a community setting in Sweden and also to investigate additional psychometric properties such as item difficulties and item total correlations.

2.3 STUDY 3

The objective of this study was to examine whether EIBI supervision enriched with information and feedback from YMQI assessments would add value to delivered EIBI quality, when compared with EIBI supervision alone. We hypothesized that preschool staff receiving enriched supervision would improve on quality factors assessed by the YMQI compared to preschool staff obtaining supervision as usual.

2.4 STUDY 4

EIBI quality as predictor of treatment outcome has not been investigated in research and therefore the aim of this study was to examine this relationship. We expected that the quality of EIBI would predict treatment outcome in young children with autism in a preschool delivery model.

3   METHODS

3.1   SETTING

All studies presented in this thesis were conducted through or within natural clinical and community based setting, involving both the Autism Center for Young Children at Habilitation & Health, which is a part of Stockholm County Council and numerous preschools in the municipalities surrounding the Stockholm area.

In Stockholm County, children are diagnosed by the child and adolescent psychiatry services following the clinical guidelines for ASD assessment and treatment according to the criteria described in DSM-IV/ICD 10 and thereafter referred to the Autism Center for Young Children (APA, 2000; Stockholms läns landsting 2010; Stockholms läns landsting, 2015; WHO, 1993).

At the center, individual mutual agreements concerning type of interventions are made based upon the child's needs and level of severity along with family's preferences and abilities. In most cases, the common agreement also involves cooperation with and consultation to the child's preschool staff.

The majority of families receive treatments that focus on one or two learning objectives during a limited period of time but there is also a possibility to agree on a comprehensive and prolonged EIBI program. In these cases a more detailed assignment for each partner is formulated including the amount of treatment hours, the attendance of parents and preschool staff at meetings as well as the responsibility of the EIBI supervisor.

For parents and preschool staff, EIBI begins with a brief workshop introducing basic ABA principles and procedures, a joint start-up followed by weekly supervision meeting at the center. An initial evaluation takes place after a period of approximately two months, where decisions are made concerning whether the comprehensive program will continue or not and potential changes in the agreement are discussed. An EIBI program then proceeds for as long as two years with continuous weekly or biweekly supervising meetings, given that the child makes progress, evaluated every third month, and that involved parties follow agreed terms.

The EIBI supervisors at the Autism center have different professional backgrounds (e.g. psychologists, speech and language pathologists, special education teachers/pedagogues). In order to ensure high quality of supervision the center utilizes a model of internal consultation, which entails that a senior colleague with extensive EIBI experience accompanies more junior colleagues when supervising preschool staff, and is also available for consultation at other times. Three of the four internal consultants at the center were Board Certified Behavior Analysts (BCBA).

The Autism center has developed its own EIBI curriculum including descriptions and materials based upon national guidelines and recommendations (Föreningen Sveriges

Habiliteringschefer, 2004) together with available manuals in the field of ABA and EIBI and updated knowledge and procedures mediated on conferences and courses.

3.2 STUDY 1

3.2.1 Procedures and participants

In Study 1, a survey was distributed through information posted on webpages at the Autism Center for Young Children in Stockholm, via flyers in the waiting room and through oral information to visitors at the center. In addition, the Center of Neurodevelopmental Disorders at Karolinska Institutet (KIND) and the Autism and Asperger Society in Stockholm posted information on their web sites and circulated newsletter. EIBI supervisors in Stockholm and elsewhere in Sweden, were contacted via e-mail and telephone and encouraged to participate themselves as well as to spread the word to others that could be interested.

In total, 294 individuals responded to the survey and were divided into five subgroups due to reported belonging: (i) EIBI preschool staff (*n*=33), (ii) preschool staff (*n*=26), (iii) behavior modification experts (*n*=60), (iv) school staff (*n*=25), and (v) parents of children with ASD (*n*=150). All groups consisted predominately of women, especially the two preschool staff groups (See table 1).

Table 1. Participant characteristics in Study 1

Group *N*=294		*n* (%)*	Description
(i)	EIBI preschool staff	33 (91%)	Personnel in preschool having EIBI supervision
(ii)	Preschool staff	26 (96%)	Personnel in preschool not having EIBI supervision
(iii)	Behavior modification experts	60 (87%)	EIBI supervisors of different professions and CBT psychologists
(iv)	School staff	25 (80%)	Teachers and other personnel working in schools
(v)	Parents	150 (81%)	Parents to children with ASD

Note. * Percentage women

3.2.2 Instrument

The survey consisted of two separate questionnaires one aiming to measure amount of knowledge of ABA and EIBI and the other with purpose to assess attitude towards and allegiance to the intervention.

3.2.2.1 The knowledge questionnaire

The knowledge questionnaire contained 15 multiple choice questions consisting of five alternatives, one correct answer, three incorrect answers, plus the option "Do not know", in order to reduce correct answers by chance (see Appendix 1). Questions were derived from the content of various exams applied in university programs and elsewhere and was piloted for feasibility by a group of experts with broad ABA and EIBI experience. Cronbach's alpha for this scale was r_α=.83 (95% CI .81-.86). Item difficulties (I_p) varied between I_p =.25 and .83 (M =.52, SD =.20). Item-total correlations (I_{i-t}) varied between I_{i-t}=.43 and .70 (M =.55, SD =.08).

3.2.2.2 The allegiance questionnaire

The allegiance questionnaire consisted of 12 five-point Likert scaled items representing statements about ABA and EIBI (Appendix 2). Participants rated their agreement or disagreement with the given statements resulting in a maximum score of 60, implying a very positive allegiance. The items were selected to represent common criticism on ABA and EIBI, and stem from a similar scale, used in the study by Klintwall et al. (2011). Cronbach's alpha for the allegiance scale was r_α = .90 (95% CI .89-.92). Item difficulties varied between I_p = 3.13 and 4.25 (M =3.85, SD =0.39). Item-total correlations varied between I_{i-t} =53 and .78 (M =.71, SD =.07).

3.2.3 Data analysis

Group differences in knowledge and allegiance were computed using a one-way between-subjects multivariate analysis of variance (MANOVA), with group membership as independent variable and allegiance and knowledge total scores as dependent variables. Differences between individual groups were assessed using post hoc Tukey´s HSD tests. The results on the knowledge and allegiance scales were correlated using Pearson's product moment coefficient. A multivariate regression did not show any effect of age on knowledge or allegiance scores (*Pillai's trace* = 0.10*, F*(2,291) = 1.414 *, p* = .245). Moreover, Exact Mann-Whitney U tests did not detect any sex effects on allegiance (*U* = 5000, *p* = .252) or knowledge (*U* = 4721.5, *p* = .092)]. All statistics were performed using R 3.1.2.

3.3 STUDY 2, 3 AND 4

3.3.1 Participants

Participants in these three studies were recruited among preschool children with ongoing EIBI programs supervised through the Autism center for young children during two consecutive periods, with 19 participants in the first (April to October 2013) and 15 participants in the second (February to June 2014) ending up with a total of N=34. The children (30 boys and 4 girls) were between 2.5 and 6 years of age. Together with the children one of their preschool staff, who was primarily responsible for daily care and training, was also recruited. The EIBI trainers (32 women and 2 men) were of different professions such as preschool teachers, preschool nurses and assistants (see table 2).

All 34 participants were included in Study 2 while in Study 3 as well as in Study 4 the number of participants decreased to N=30 due to four dropouts. In two cases, EIBI was terminated as the child had reached its developmental goals and in the other two cases termination was due to preschool staff resigning from employment during the course of the studies.

Table 2. Participant characteristics in Study 2, 3 and 4

	Study 2 (*N*=34)		Study 3 & 4 (*N*=30)	
	n	Percentage	*n*	Percentage
Gender children				
Female	4	12	4	13
Male	30	88	26	87
Gender staff				
Female	32	94	28	93
Male	2	6	2	7
Staff profession				
Preschool teachers	10	29	8	26.7
Preschool nurses	19	56	17	56.7
Preschool assistants	5	15	5	16.7
	M	*SD* (range)	*M*	*SD* (range)
Children age (months)	50.9	8.9 (33-73	50.8	9.0 (33-73)
Staff age (years)	38.9	11.9 (20-62)	39.1	12.5 (20-62)
Years in profession	11.5	10.8 (0.1-37)	12.2	11.3 (0.1-37)
Months as EIBI trainer	8.9	13.1 (0.25-48)	7.6	11.6 (0.25-48)

3.3.2 Measures

3.3.2.1 *The York Measure of Quality of Intensive Behavioural Intervention (YMQI)*

The York Measure of Quality of Behavioural Intervention (YMQI) measures quality of performed EIBI on collected video clips (Penn et al., 2007; Perry, Flanagan, et al., 2008). Two 5-minute segments are randomly selected from a video clip of at least 20 minutes and coded on 31 quality items, structured in 9 categories (see table 3). Each item is scored on a 5-point Likert scale (1, 1.5, 2, 2.5, 3) with "1" representing poor quality and "3" indicating excellent quality. There is also a possibility on several items to code "not applicable" (N/A), to be used in the case the quality indicator cannot be rated (e.g. no reinforcers or prompts are used by the trainer alternatively the child does not exhibit a specified behavior). An EIBI summary score is calculated by the average of the summed ratings of the two 5-minute sequences (N/A not included). A summary score of less than 2.1 is considered to reflect poor quality, a score of 2.1 to 2.5 good quality and a score of 2.5 to 3 excellent quality. Aside from the summary score, the instrument includes four additional subdomains (Organization, Pacing, Teaching level, Generalization). Herein, only 15 of the 31 items are used to summarize trainer performance and the quality criteria for which values are viewed as poor, good and excellent differ across these subdomains (Perry, Flanagan, et al., 2008).

YMQI has been shown to have good psychometrics properties (Denne et al., 2015; Penn et al., 2007; Perry, Flanagan, et al., 2008; Whiteford et al., 2012) and has demonstrated an internal consistency (Cronbach's alpha) of r_α =.82 (Perry, Flanagan, et al., 2008) and r_α =.77 (Whiteford et al., 2012).

Interrater reliability (IRR) calculated with intraclass correlation (ICC) were found to be r_{icc}=.68 in Perry, Flanagan, et al. (2008). In the study by Whiteford et al. (2012), IRR was reported as percentage agreement across the 31 items demonstrating an average of 88.95% (range 74%-87%) and in the study by Denne et al. (2015) IRR was 99% (range = 81%-100%). The validity was examined in Perry, Flanagan, et al. (2008) by correlating YMQI scores with an consensus rating of a dyad of EIBI experts reaching a convergent validity of r=.58. The expert scale consisted of nine items corresponding to the YMQI categories and was scored in a 5-point Likert scale (1, 1.5, 2, 2.5, 3) where "1" indicated little evidence of appropriateness (significant concerns), "2" generally appropriate (moderate concerns) and "3" consistently appropriate (no concerns). In addition to the nine quality areas, the experts also conducted an overall judgment of quality using a 7-point Likert scale ("1" extremely poor quality to "7" exceptional quality). This expert scale was translated to Swedish by a bi-lingual expert and approved by the original authors in order to be used in Study 2.

The YMQI was used in all of the three studies. First, in Study 2 the objective was to examine its psychometric properties, in Study 3 as an outcome measure, and in Study 4 as a predictor of outcome. The instruments described below were used to measure child demographics in Study 3 and as outcome measures in Study 4.

Table 3. The York Measure of Quality of Intensive Early Behavioural Intervention (YMQI)

A. Discriminative Stimuli	**E. Pacing**
1. Attending during S^{D}s	16. Length of inter-trial intervals
2. Varying S^{D}s	17. Suitable pace for the child
B. Reinforcement	18. Intensive teaching
3. Rapid reinforce delivery	**F. Teaching Level**
4. Motivational reinforcers	19. Suitable task difficulty
5. Varying reinforcers	20. Evidence of skill acquisition
6. Relation of reinforcers to the task	**G. Instructional Control**
7. Sincere/motivating verbal reinforcers	21. On-task following requests
8. Differential reinforcement	22. Maintenance of child's focus
C. Prompting	**H. Generalization**
9. Effectiveness of prompts	23. Varying teaching materials
10. Fading and augmenting of prompts	24. Mixing tasks
11. Lack of prompting errors	25. Teaching away from table
12. Follow through	26. Teaching embedded in naturalistic activities
13. Implementation of error correction	27. Response generalization
D. Organization	28. Flexible teaching
14. Clear plan and teaching goals	**I. Problem Behaviour**
15. Accessible materials	29. Result of problem behaviour
	30. Reinforcement of appropriate behaviour
	31. Use of prevention strategies

Note. S^{D}s= Discriminative Stimuli

3.3.2.2   *Mullen Scales of Early Learning*

In order to measure developmental level *Mullen Scales of Early Learning* were used (Mullen, 1995). Mullen is a standardized, normed-referenced measurement including four subscales (Visual perception, Fine Motor, Receptive Language and Expressive Language) and in this study, we used the generated age equivalents

3.3.2.3 The Basic Language and Learning Skill Test

Level of skills was assessed by an instrument called *The Basic Language and Learning Skill Test* (BST; Jahr & Eldevik, 2009). This instrument was inspired by Early Learning Measure (ELM; Buch, 1985) and developed as a clinical tool to measure learning rate. BST consists of 216 tasks and questions covering several target area such as imitation, receptive and expressive language, pre-academic abilities, and play skills. Results are reported as percentage of correct responses. The instrument also includes three additional play scenarios not included in this study due to practical reasons.

3.3.2.4 Vineland Adaptive Behavior Scales

Vineland Adaptive Behavior Scales 2nd edition (VABS II; Sparrow, Cichetti, & Balla, 2005) measures adaptive behavior from birth to adulthood and is commonly used as an outcome measure in EIBI studies (Eldevik et al., 2012; Klintwall, Eldevik, et al., 2015; Perry Flanagan, Dunn Geier, & Freeman., 2009; Reichow et al., 2012). VABS yields standard scores on four subdomains (communication, daily living, socialization, and motor skills) together with an overall standardized behavior composite. VABS was administered as a survey and filled in by parents.

3.3.2.5 Social Responsiveness Scale

The *Social Responsiveness Scale* (SRS; Constantino & Gruber, 2005) is a parent-report scale assessing autistic traits in children aged 4 to 18 years. For the participants younger than 4 years, we used the version for preschool children (SRS-P). The SRS and SRS-P consist of 65 Likert-scaled items scored 0-3 (total score 195).

3.3.2.6 The Clinical Global Impression Scale

General clinical severity was measured by the *Clinical Global Impression Scale* (CGI; Kadouri, Corruble, & Falissard, 2007), completed as a consensus rating by preschool staff. The CGI is a seven point scale where 7 indicates extreme clinical severity and 1 showing no clinical symptomatology

3.3.3 Procedures and data analysis

3.3.3.1 Common set-up

We used a joint set-up in order to collect data for all three studies (see figure 1). Children and staff were video recorded at the preschool at three times and quality of delivered EIBI quality was assessed by using YMQI. In connection to the recordings at probe 1 (baseline) and 3 (follow up) the children were tested on Mullen and on BST and the preschool staff were asked to answer CGI and the parents filled in VABS and SRS.
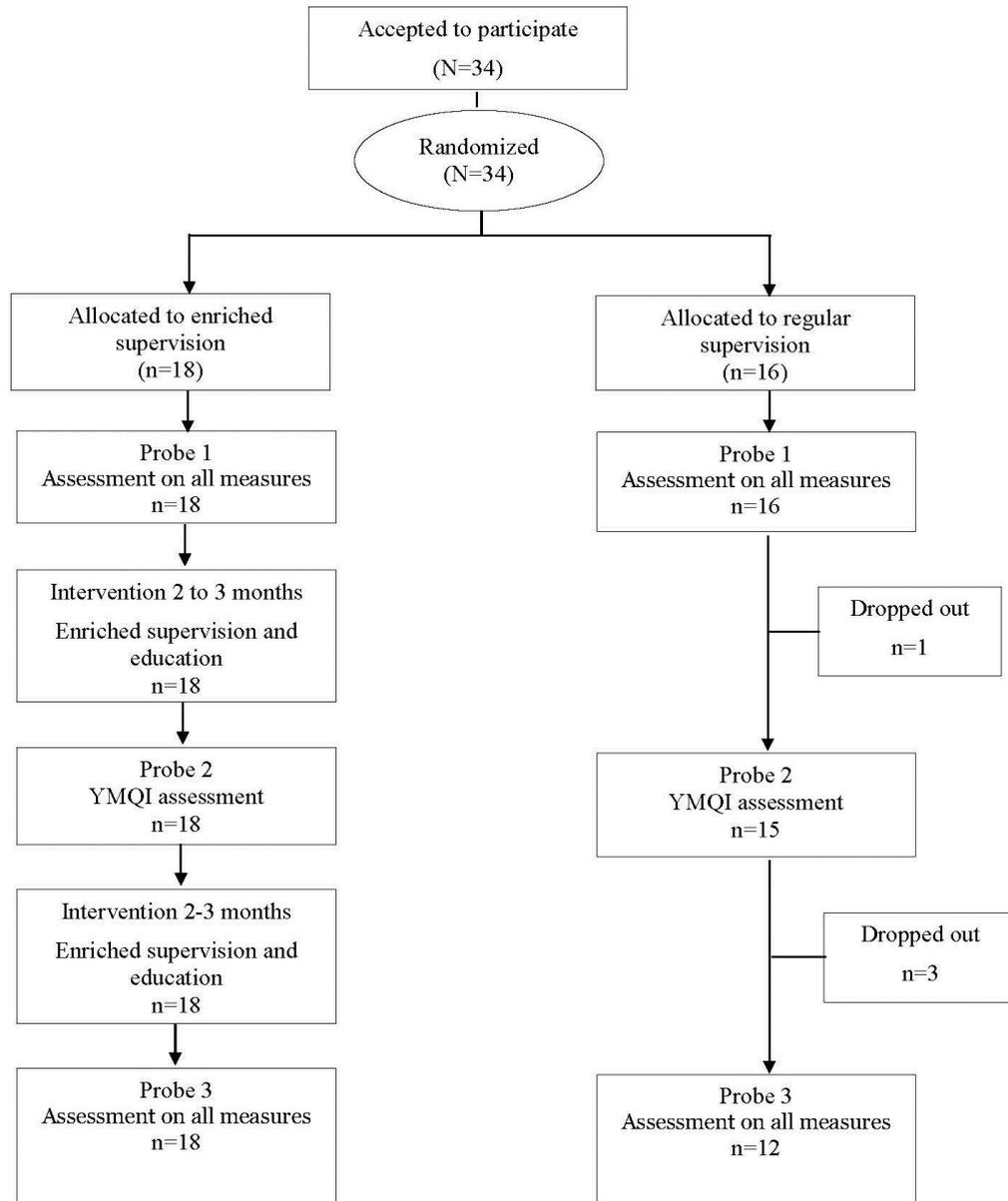
Figure 1. CONSORT flow diagram

3.3.3.2   Study 2

In this study, a cross-cultural validation of the YMQI was conducted. In order to measure *interrater* reliability the collected video clips of EIBI intervention (k=97) were assessed by two of a total of three raters and *intrarater* reliability was explored on 15 videos re-rated by two of the coders after 6 months. Validity was examined by comparing the YMQI scores on 30 videos with the quality assessment by ten EIBI supervisors using the less comprehensive expert scale. The supervisors (8 female, 2 male) had 10 to15 years of experience in the field and completed the expert scale as consensus rating of formed dyad.

The statistics program R 3.2.3 was used to determine the psychometric properties of the YMQI. Item-total correlations (I_{i-t}), indicating how well the specific quality factor predicts overall quality, were calculated by Pearson product moment correlations between YMQI item scores and the summary scores in the total sample. The trainers' average performance on an item reflects item difficulty ($I_{p)}$) and were provided as the mean of the items score in the total sample. Internal consistency was measured using Cronbach's alpha. Interrater reliability and intrarater reliability was presented as percentage agreement on items as well as Intra Class Correlation (ICC) for total score and subdomain scores. In accordance with previous studies (Perry, Flanagan, et al., 2008; Whiteford et al., 2012) percent agreement was calculated with a tolerance of 0,5 discrepancy and moreover we also included the agreement on choosing to score (1 to 3) or assessing an item as N/A (here converted to 0). In order to examine convergent validity the YMQI summary score, the four subdomains and the nine category scores were correlated with the expert ratings using Pearson's coefficient.

3.3.3.3   Study 3

This study examined whether supervision enriched with information from the YMQI quality assessment would increase performed quality compared to regular supervision.

The recruited triads of children, preschool staff and supervisors were stratified according to amount of supervision, i.e., weekly or bi-weekly and then randomized into enriched or regular supervision. The total number of triads that completed the study was N=30 of which 18 were distributed to the enriched supervision group and 12 to the regular supervision group.

Children with ASD and their EIBI trainers were stratified according to the amount of supervision, i.e., weekly or bi-weekly, and then randomized to either regular or standardized tailored EIBI supervision. At baseline we detected a difference between the groups regarding age where children in the regular group were significantly younger compared to the enriched supervision group (45.8 vs. 54.2 months, p =.001) as well as demonstrating lower developmental age on the Mullen subscale Visual Reception (age equivalent 22.5 vs. 35.2, p=.02). No other significant differences were detected on child or staff characteristics. Statistics on demographics were performed using IBM SPSS statistics version 22.

The supervisors in the enriched supervision group received written feedback on all the 31 quality factors and was shown the 10 minutes of video clips after probe 1 and 2, in order to use the information as a complement in the ordinary supervising meetings with preschool staff. The detailed feedback was delivered by the doctoral student of this thesis (who was not further involved in the treatments), and were based on the YMQI assessments as well as clinical experience. Additionally, in order to facilitate the implementation of the enriched supervision, the preschool staff in this group also participated in two half-day workshops focusing on important quality factors, illustrated by video examples together with practical exercises and role-play.

In order to analyze if enriched supervision yielded higher levels of EIBI quality over time compared to regular supervision we used a two-way repeated measures ANOVA. Child age

and Mullen visual reception scores were controlled for in the ANOVA since both measures initially differed between the groups. Moreover, group comparisons at probe 1 and 3 were also calculated using one-tailed Student t-tests and in order to investigate improvements on item level, analyses were conducted using one-tailed paired Student t-tests within subjects, comparing probe 1 and 3 based on the mean of the two segments and the two raters. P values were adjusted for multiple comparison using the Holm-Bonferroni method. These statistics were computed in R 3.2.3.

Table 4. Outcome measurements at baseline and follow-up, and improvement criteria

Measures	Baseline		Follow-up (4 to 6 months)		Moderate improvement	Major improvement
	M	SD (range)	M	SD (range)		
Mullen age equivalents (months)					6 to 12-months gain	>12-months gain
Visual Reception	30.1	15.1 (6-64)	35.1	15.7 (7-66)		
Fine Motor	31.0	14.5 (10-62)	34.8	15.8 (10-68)		
Receptive Language	26.9	17.5 (4-62)	32.2	18.5 (8-62)		
Expressive Language	25.2	14.6 (5-55)	31.2	18.5 (5-70)		
Vineland Adaptive Behavior Scales					7.5 standard points	15 standard points
Communication	59.8	15.6 (29-93)	63.3	18.4 (33-112)		
Daily Living	66.5	14.1 (37-95)	65.3	13.5 (35-91)		
Socialization	64.3	11.4 (46-100)	63.5	15.6 (41-105)		
Motor	63.4	17.5 (20-110)	62.9	14.9 (24-91)		
Social Responsiveness Scale	83.2	22.7 (33-129)	82.3	28.0 (35-154)	6.5 points	10 points
Clinical Global Impression	4.7	0.9 (2-6)	4.6	1.0 (2-6)	1 point	2 points
Basic Language and Learning Skill Test	41.9	33.8 (0-92)	55.8	35.6 (1-98)	18%	35%

3.3.3.4   Study 4

In this study, we examined EIBI quality at baseline as a predictor of the children's outcome. We used the YMQI (total score, 4 subscales and 31 item scores) assessment obtained at baseline for the 30 children and the outcome measurements conducted after 4 to 6 months of intervention. Behavioral change on the outcome measures was analyzed separately for each

of the instruments, using a derived global dichotomized estimate of clinically significant positive change taking into account the results on all measures. This was done by first categorizing the change on the single scales into no, moderate and major improvement based on reported psychometric properties and the assumption that a one standard deviation change is clinically relevant (see table 4). For each child the moderate or major successful change on the scales were then summarized into the global measure of outcome. We decided that moderate or major improvement on three or more of the 11 scales and no decline would indicate an overall clinically significant positive behavioral change for the given time window. As a result 8 (27%) of the 30 children were classified as exhibiting significant clinical change at follow-up.

The data were analyzed using a series of main effect model (stepwise) multinomial logistic regressions in IBM SPSS Statistics 23, with YMQI scores as quantitative predictors and the overall qualitative and the single quantitative behavioral outcome measures as dependent variables. An alpha level of 5% and trend level of 10% were applied and inference tests conducted one-sided, owing to directional hypotheses. Given our sample size (N=30), and alpha (5%), the power to detect a large effect was 78%, a medium effect 37%, and small effect 8% (GPower 3.1.9.2, χ^2 tests, post hoc). Thus, no correction for multiple testing was used to avoid conservative decisions in a sample of limited power.

3.4 ETHICAL CONSIDERATIONS

The local board of ethics in Stockholm approved all four sub-studies (protocol id. 2012/1734-31/4).

In Study 1 the responders provided informed consent in order to enter the survey and participation was anonymous.

Study 2, 3 and 4 were offered as a supplement to ordinary procedures of ongoing clinical intervention. Information, both oral and written, regarding the research project was given to potential participants in connection with supervising meetings at the Autism Centre. It was especially emphasized that a decision not to participate would not affect the ongoing treatment. Informed consent was obtained from parents as well as preschool staff. The additional intervention provided through the research project was conducted during the limited period of 4 to 6 months and was then offered to the regular supervision group, i.e. the supervisors received feedback and reviewed video clips after follow-up and the preschool staff attended the education on EIBI quality.

Participating in the study did not require extra intervention time for neither parents nor preschool staff, since the enriched supervision was implemented in the ordinary and scheduled meetings. However, the preschool staff was video recorded at three points in time, which could have resulted in some inconvenience, even though collecting video clips is a common way to document the treatment and would therefore in most cases have been

perceived as a usual procedure. The parents contributed by answering two questionnaires, and given the circumstances of having a child with ASD, it may have been difficult to find enough time to complete the forms, especially if Swedish was not the family's first language.

4 RESULTS

4.1 STUDY 1

Group differences were found in the survey as a whole [$F(8, 578) = 22,14$, $p < .001$, Pillai´s Trace = .47, partial η^2 = .24] as well as individually on both the knowledge questionnaire [$F(4,289) = 58.73$, $p < .001$, partial η^2 = .53] and the allegiance scale [$F(4, 289) = 18.19$, $p < .001$, partial η^2 = .20].

Post hoc investigation revealed that behavior modification experts had higher levels of knowledge and had more allegiance to the method than all the other groups participating in the survey ($p < .003$). Furthermore, the preschool staff receiving EIBI supervision scored higher on knowledge than preschool staff in general as well as parents ($p < .001$ and $p = .015$) but they did not show higher levels on allegiance compared to preschool staff in general ($p = .061$). In addition, school staff scored higher than both preschool staff in general and parents ($p < .001$ and $p = .026$) and lastly parents showed higher knowledge than preschool staff in general ($p = .038$) (see figure 2). Calculation demonstrated that knowledge and allegiance scores were correlated $r = .52$ ($p < .001$).
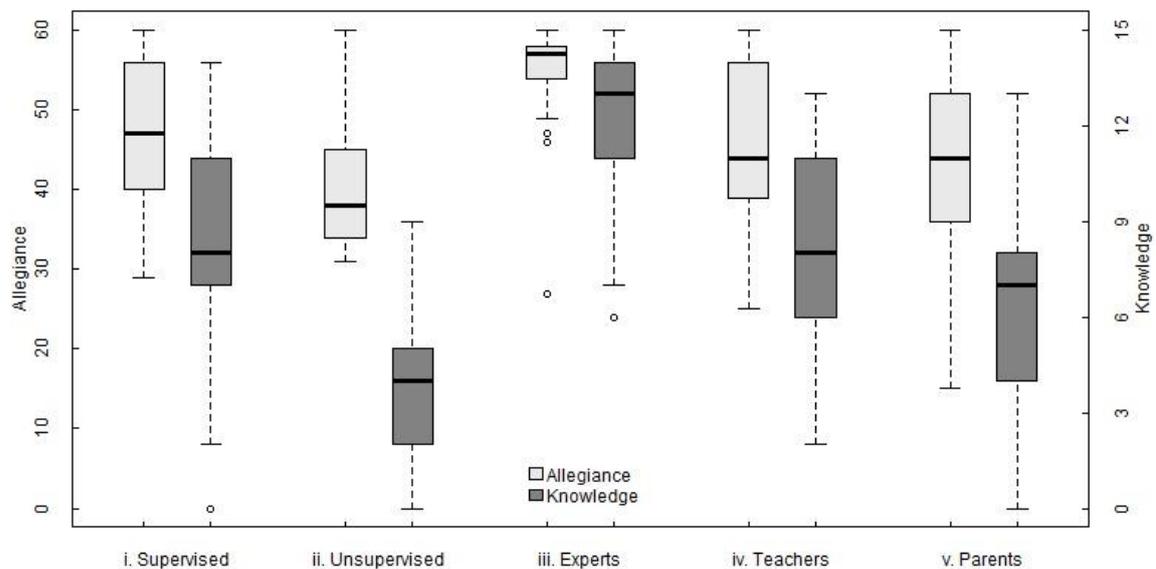

Figure 2. Box and whiskers plot for allegiance and knowledge between the groups

4.2 STUDY 2

Analysis shows that item difficulties were generally in the similar range of low average to average difficulties (I_p=2.04-2.67). Item-total correlations, on the other hand, varied considerably between I_{i-t}= -.13 and .74. Nine items had low item-totals ($I_{i-t} \leq .30$) while eight items reached high item-totals ($I_{i-t} \geq .60$) and thereby showed best prediction of total quality

(for details, see table 5). The internal consistency of all YMQI items was $r_\alpha = .87$ and $r_\alpha = .60$ for the domain "organization" (2 items), $r_\alpha = .80$ for the "pacing" (3 items), $r_\alpha = .53$ for "teaching level" (2 items), and $r_\alpha = .48$ for "generalization" (8 items).

Interrater reliability reported as ICC was .71 for total score and ICCs for the sub-scales were .70 for organization, .70 for pacing, .44 for teaching, and .64 for generalization. Overall percentage agreement across all items was 76% (range 56%-90%) and the median item agreement was 78%. The agreement between whether an item could be scored or considered as N/A was, on average, 83% (range = 62-100%). Intrarater reliability measured with ICC was .87 for the summary score and ICC's for the four domains were .75 for organization, .90 for pacing, .80 for teaching and .89 for generalization. Intra-rater reliability in terms of overall percentage agreement was 91%. Percentage of agreement on item level is presented in table 5.

The convergent correlation between the YMQI total score and the expert rating overall score was $r = .49$ ($p = .006$). Correlations between YMQI ratings and expert ratings of EIBI quality for the four subdomains were $r = .27$ ($p = .15$) for organization, $r = .53$ ($p = .0025$) for pacing, $r = .52$ ($p = .003$) for teaching level and $r = .73$ ($p < .0001$) for generalization.

4.3   STUDY 3

A main Group effect was found in which the enriched supervision group showed higher scores than the regular supervision group [$F(1,50) = 8.074$, $p < .007$]. However there were no significant main effect of Probe $F(1,50) = 1.035$ nor an interaction between Group and Probe $F(1,50) = 1.187$. The covariates age and visual score of Mullen were not significant, respectively $F(1,50) = 2.652$, p=.11 and $F(1,50) = .671$.

Further calculations demonstrated that while there were no significant quality differences between the groups at baseline [$t(23) = 1.74$, p=.19] the enriched group scored higher that the regular group at follow-up [$t(17) = 5.33$, $p = .0002$]. Moreover, the majority of the enriched group trainers (14 of 18) reached a higher level of quality than any of the trainers in the regular supervision group and the variability, as shown in the interquartile range, was reduced in the enriched supervision group compared to the regular supervision group (.05 vs. .21) (see figure 3).

In addition, we investigated the effect of supervision over time within each group and the analyses demonstrated EIBI quality improvements in the enriched supervision group from probe 1 to 3 [$t(17) = -3.83$, $p = .002$] whereas the regular group showed no significant changes in quality during the study [$t(11) = -1.30$, $p = .19$].

Table 5. YMQI item difficulties (I_p), item-totals (I_{i-t}), and item interrater and intrarater reliabilities

YMQI item	I_p	I_{i-t}	% agreement interrater	% agreement intrarater
1. Attending during S^Ds	2.33	.63	86	95
2. Varying S^Ds	2.26	.31	74	92
3. Rapid reinforce delivery	2.67	.28	85	95
4. Motivational reinforcers	2.65	.37	85	93
5. Varying reinforcers	2.37	.21	67	83
6. Relation of reinforcers to the task	2.13	.13	85	88
7. Sincere/motivating verbal reinforcers	2.66	.51	90	97
8. Differential reinforcement	2.12	.32	65	72
9. Effectiveness of prompts	2.47	.47	63	82
10. Fading and augmenting of prompts	2.22	.45	65	80
11. Lack of prompting errors	2.24	.53	56	85
12. Follow through	2.51	.62	81	93
13. Implementation of error correction	2.17	.52	63	78
14. Clear plan and teaching goals	2.36	.64	77	97
15. Accessible materials	2.3	.29	76	88
16. Length of inter-trial intervals	2.16	.54	68	100
17. Suitable pace for the child	2.23	.70	78	100
18. Intensive teaching	2.14	.55	88	98
19. Suitable task difficulty	2.09	.44	73	95
20. Evidence of skill acquisition	2.04	.43	84	95
21. On-task following requests	2.31	.62	90	98
22. Maintenance of child's focus	2.25	.72	87	97
23. Varying teaching materials	2.25	.26	75	97
24. Mixing tasks	2.05	.36	80	97
25. Teaching away from table	2.25	.09	80	98
26. Teaching embedded in naturalistic activities	2.16	.00	82	92
27. Response generalization	2.08	.12	86	87
28. Flexible teaching	2.12	.05	84	95
29. Result of problem behaviour	2.13	.74	66	90
30. Reinforcement of appropriate behaviour	2.13	.55	60	88
31. Use of prevention strategies	2.08	.66	57	90

Note. S^Ds= Discriminative Stimuli

Analyses on item level showed that the enriched supervision group specifically improved quality for *Clear plan and teaching goals* [item no. 14; Mean change $_{3-1}$ = .57, t (17) = - 4.86, p=.002], *Accessible materials* [item no. 15; mean $_{3-1}$ = .40, t(17) = - 4.08, p =.01], and *Intensive teaching* [item no. 18; mean$_{3-1}$= .42, t (17) = - 4.30, p=.007]. There were also trends for *Implementation of error correction* [item no 13. t(16) = - 3.07, p =.10] and *Length of inter-trial intervals* [item no 16 t(17) = - 3.31, p =.06]. There were no significant quality improvements on any YMQI item in the regular supervision group.
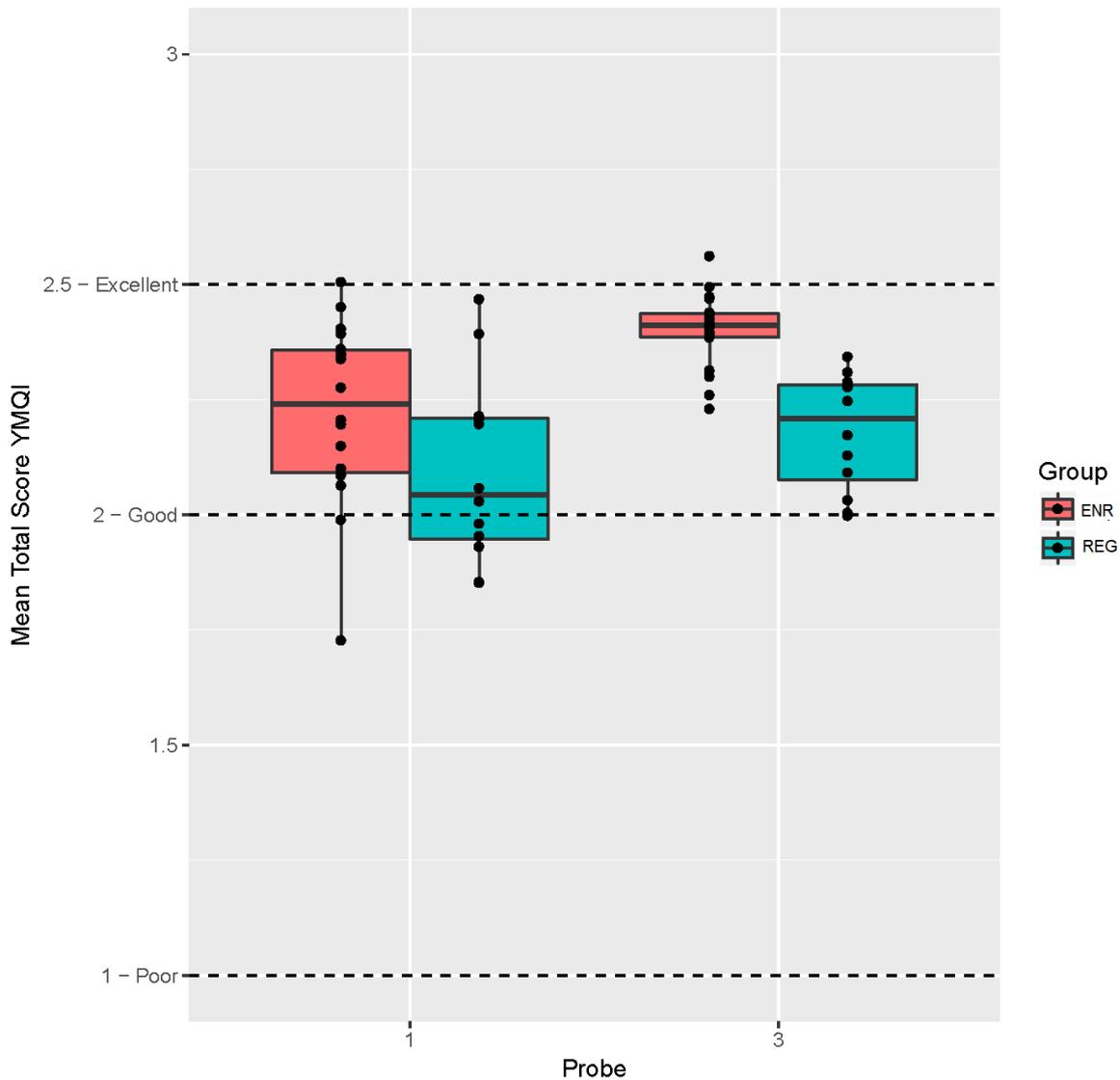

Figure 3. Box Plots for delivered EIBI quality (YMQI total scores) in the enriched (ENR) and regular (REG) supervision trainer groups.

4.4 STUDY 4

In this study we found that the YMQI average item score at baseline predicted the treatment success summary evaluation at 4 to 6 months follow-up ($\chi2$ $_{df=1}$=2.98, *p* =.042).

Moreover, two of the four YMQI subscales, Organization ($\chi2$ $_{df=1}$=3.43, *p* =.035) and Teaching level ($\chi2$ $_{df=1}$=3.15, *p* =.037), had a significant influence on treatment success summary evaluation.

In addition, six YMQI items had a predictive value: 6 "Relation of reinforcers to task" ($\chi2$ $_{df=1}$=9.22, *p* =.001), 8 "Differential reinforcement" ($\chi2$ $_{df=}$1=4.98, *p* =.013), 13 "Implementation of error correction" ($\chi2$ $_{df=1}$=7.16, *p* =.005), 24 "Mixing tasks" ($\chi2$ $_{df=1}$=4.74, *p* =.021), 26 "Teaching embedded in naturalistic activities" ($\chi2$ $_{df=1}$=7.05, *p* =.004), and 27 "Response generalization" ($\chi2$ $_{df=1}$=4.55, *p* =.019).

The YMQI average item score predicted improvement on the BST summary evaluation ($\chi2$ $_{df=1}$=4.88, *p* =.016), and showed a trend for behavior improvement on the Global Clinical Impression Scale ($\chi2$ $_{df=3}$=6.15, *p* =.079).

5 DISCUSSION

5.1 STUDY 1

In Sweden, as in many other countries, EIBI is conducted in a community based setting which indeed has several advantages over solely clinically based settings, including access to typically developing peers and numerous possibilities to generalize skills. However, a regular preschool often entails staff with little knowledge of ASD, evidence based practice, and with a preference to other teaching approaches than EIBI. Consistently, our findings show a low level of basic ABA knowledge among preschool staff in general, compared to other professionals as well as parents. Preschool staff receiving EIBI supervision showed a higher knowledge level, indicating the importance of high quality supervision as a way of conveying competence and as an assurance of quality. Still, the quality of performed treatment is probably insufficient when starting up a new EIBI program, running the risk of jeopardizing the child's chance of optimal progress. Moreover, the results did not find a corresponding supervision effect regarding the allegiance scale, revealing that some of the staff may have a negative attitude towards EIBI that could affect both treatment integrity and the quality of practice. In order to enhance the implementation of EIBI in the Swedish community system, a broadly increased level of basic ABA knowledge is essential on a basic level and could be achieved by adding these components in the university curricula for preschool teacher education. In clinical practice, an extra emphasis on staff education prior to treatment start-up would be preferable. Furthermore, supervisors should encourage discussions of evidence based practice, different learning strategies as well as ethical considerations during supervision meetings, in order to address possible skepticism.

5.1.1 Limitations

In this study, we only measured theoretical knowledge, which can be seen more as a prerequisite not necessarily reflecting actual quality of performed EIBI, where practical experience may be equally important. Moreover, since the survey was conducted in the Stockholm area the generalizability of the results to other parts of Sweden is relatively unknown. Nevertheless, we think that our findings are sufficiently representative due to the relatively standardized ASD services around the country and since Stockholm County includes both urban and rural areas. Lastly, we do not have true control over who completed the survey and the population can be biased. Yet, we estimate that a majority of the EIBI supervisors and approximately half of the EIBI preschool staff answered the survey, and we assume that the other groups answering the survey were extra interested and, if anything, with a higher knowledge base and positive attitude towards EIBI, making it unlikely that our findings overestimated the true between-group effect.

5.2 STUDY 2

It seems obvious that how well an intervention is performed is a critical component when evaluating an intervention. Nevertheless, this is an area which has received relatively little attention, which may be due to the absence of a commonly used instrument. The YMQI is a promising exception, but is not yet widely used in research or clinical practice and therefore more information about its feasibility is direly needed.

In this cross-cultural validation of the YMQI in a clinical setting in Sweden, we examined interrater reliability and validity in accordance to previous evaluations of the scale. In addition, we also explored item analysis and rater stability, i.e. intrarater reliability, providing new information about the instrument's properties. We found little variation of item difficulties, which seems logical for a criterion-based scale measuring performance where the goal is to reach high scores on each item. Nine items had low items-totals whereas one was even negatively correlated to total quality, indicating that the scale is not measuring one single construct. However, quality of EIBI is rather multidimensional and the aim of the scale is not to differentiate individuals on one dimension but to assess different aspects of quality. Our results are in line with previous psychometric evaluations but showed lower interrater reliability, which could be explained by the use of naïve raters and cultural differences in interpretation of the manual, and in the understanding of good EIBI. Some items had considerably low agreement and additional clarifications in the manual would be beneficial.

5.2.1 Limitations

The raters in our study also collected the video clips and therefore had further information than the selected segments on some of the children, which could have affected interrater reliability. In order to minimize the rater effect, raters were instructed not to influence the treatment session and they were not involved in the other processes related to data collection. We also used the original English version of the manual, which might have impeded an optimal administration of the instrument. Moreover, we only used three raters whereas a naturalistic clinical setting would probably include several raters with different professional backgrounds and levels of experience.

5.3 STUDY 3

In this study, in which we investigated the benefit of tailored feedback on quality factors to preschool staff, we found that the enriched supervision group improved significantly compared to the regular supervision group. The study was conducted in a naturalistic setting, including children with various developmental levels and ASD severity and furthermore, the added activities were well embedded in the usual clinical practice, enhancing the possibility of incorporating these in future supervision procedures.

We found that the enriched supervision group was strengthened in total quality and showed reduced variability. The improvements, particularly on the quality factors regarding organization planning, indicates an increased awareness of the intent and structure of the child's program and the trainer roll. The structured supervision approach, using video clips and the YMQI evaluation, provides the supervisor with concrete examples of trainer performance, enabling more accurate and personalized supervision, which seems to result in higher training quality and possibly to better outcome for the individual child.

Even though our results favor the use of the YMQI in supervision, it is important to keep in mind that the EIBI program is highly influenced by the supervisor's competence. Also, on a more general level, different child and treatment factors affect treatment outcome and therefore a broader quality assurance system such as the YSQA is recommendable. Moreover, the administration of the YMQI in a clinical setting could be too time-consuming and may need to be limited to fewer selected important items.

5.3.1 Limitations

Firstly, the sample size in our study was rather small and restricted to the Stockholm area, entailing an uncertainty of representativeness. Secondly, the groups differed on some child related characteristics, which could have had an impact on the trainer skills required by the staff. Thirdly, because the enriched supervision included both tailored feedback as well as education this study cannot determine the specific effect of these components. Fourthly, the YMQI focuses on certain quality factors, and the competence of a trainer with an EIBI program with less discrete trial training in favor of more child-initiated teaching may be underestimated due to the construction of the scale. Moreover, the YMQI is not constructed primarily to measure change and gives relatively little range for variability and observed quality change. Fifthly, our hypotheses was only partly supported since the results were limited to group differences at probe 3 and a within-group improvement in the enriched supervision group but no interaction effect on group by time. Finally, there might be a biasing effect of using the YMQI both as a tool for training and feedback as well as an outcome measure.

5.4 STUDY 4

In this fourth and final study, we explored EIBI quality as a predictor of successful outcome. Since EIBI in Sweden, as well as in many other countries, is mainly conducted by staff without formal ABA/EIBI education, monitoring and evaluation of quality is imperative to the securing of treatment adherence and integrity.

We found a positive relation between baseline overall quality and total outcome, as well as more specifically between overall quality and BST and CGI. This indicates the need of outcome measures that cover both detailed skills linked to the training and also to more global changes.

In addition to the predicted value of overall quality, we found that the subscales organization and teaching level had a positive influence on outcome, together with several items concerning generalization as well as differential reinforcement and error correction. Quality in organization might indicate skills of having a clear plan and being well prepared and thus optimizing the learning situation for the child, while quality on teaching level might represent an ability to act on an appropriate level of task difficulty and actually be teaching the child new skills.

The quality items of significant predictive power related to generalization all aim to conduct the training sessions as similar as possible to everyday life, thus enhancing the usefulness and applicability of learned skills beyond the training context. Thereby, endorsing the importance of incorporating and emphasizing incidental teaching and natural environmental training in order to promote better outcome. The other two quality items "Differential reinforcement" and "Implementing of error correction", demonstrate the significance of following the core procedures of reinforcement of desired behavior and knowledge of how to analyze contextual learning situations in order to help the child to reach the target behavior.

5.4.1 Limitations

Firstly, the dichotomous clinical change criterion based on the progress on the eleven outcome measures can be perceived as a crude and arbitrary choice of evaluation. Still, it enabled us to have one holistic measure, sensitive to the impact of YMQI scores and furthermore reduce the risk of false positive results. Secondly, the short EIBI intervention period as well as including ongoing programs restricted the possibility of large behavioral gains. Thirdly, there may be certain quality factors not covered by the YMQI that are crucial for favorable outcome, such as the learning environment per se, programming, documentation, cooperation with parents together with known quality factors such as amount of supervision as well as theoretical and practical knowledge among both supervisors and trainers. Fourthly, we did not collect data on amount of received treatment and intensity which might be correlated with quality. However, the usual agreement with the preschool is 15 hours/week complemented by the parents at home (5 hours/week) and thus we concluded that training intensity was most likely fulfilled. Lastly, since the recruitment of participants to this study, as to the thesis in its entirety, was restricted to on-going EIBI programs at the Autism Centre for Young Children in Stockholm County, the representativeness in relation to children trained elsewhere is unknown.

6 CONCLUSION AND FUTURE DIRECTIONS

In a series of studies presented in this thesis, we have focused on the quality of delivered EIBI when implemented in the community. We have described the low level of EIBI knowledge and skepticism among preschool staff, pointing out the need to address this issue when evaluating outcome. We have also conducted a cross-cultural evaluation of the psychometric properties of the YMQI and demonstrated that including information from the YMQI in an enriched supervision model resulted in an increased level of performed quality compared to regular supervision. Lastly, we have found that quality of performed EIBI predicts children's overall outcome.

Findings underscore the importance of addressing competence when examine EIBI outcome both in terms of prerequisites such as knowledge among staff as well as the necessity to monitor and evaluate performance based competencies, both in research and in clinical practice.

In a community based intervention setting, where the EIBI services depend on the collaboration between the county's habilitation centers and the municipalities' preschools, there is clearly a high risk of time-consuming discussions concerning division of responsibilities and organizational issues, delaying or hindering a start-up process, thereby probably broadening the developmental gap between children with ASD and typically developing peers. Moreover, when a child is enrolled in a program, the beginning period maybe hampered by the lack of competencies among preschool staff. The possibility of offering EIBI earlier and to more children would be greatly facilitated with an overall national agreement between counties and municipalities, defining terms of collaboration. Furthermore, as previously mentioned, the quality and integrity of delivered treatment would benefit from a higher level of prerequisite knowledge by preschool staff, by adding basic ABA components to the educational curriculums. In clinical practice, an assessed minimal standard of knowledge and practice prior to start-up could guide the supervisors to direct extra support to the staff not reaching the cut-off level. A common agreement among EIBI researchers concerning a gold standard of prerequisites would provide useful principles to clinicians as well as enable comparisons between studies conducted in naturalistic settings.

Another critical question which needs to be addressed is the level of supervisor competence. Supervisors at the habilitation centers are mostly recruited on the basis of their different professions, disregarding competence in ABA/EIBI. In reality, supervisors may obtain their first training while at the same time supervising on-going programs, posing a threat to quality implementation. In countries with center-based settings, a supervisor may have acquired several thousands of EIBI training hours along with competence through university courses, nowadays often supplemented with a BACB accreditation, prior to providing supervision to others. In Sweden there is only one course, offered by Stockholm University, that focuses on the science of behavior analysis (approved by the BACB), which limits the possibility of

increasing the supervisor expertise. However, during the last ten years the number of BCBAs in Sweden has increased from 0 to about 15 and there is a growing number of applicants of the existing ABA course. But of course, in order to reach a nation-wide competence and for a more equal allocation it would be preferable if ABA courses were offered at additional universities. Furthermore, as in the case of education for preschool staff, professions such as speech-language pathologists and teachers in special education, would be much better prepared for EIBI supervisor assignments with elements of ABA procedures in the regular university program curricula. In the recommendations from the association for habilitation directors (Föreningen Sveriges Habiliteringschefer, 2004) the necessary organizational conditions are stated as well as program content and evidence based procedures, but nothing is mentioned about recommended competence by the habilitations' supervisors and how to achieve such training or adherence. A complementary section on needed supervisor skills would be beneficial, in order to clarify the complexity of programming and supervising a comprehensive intervention, as well as guiding professionals how to obtain the adequate and sufficient competence.

In regard to the psychometric evaluation of YMQI we concluded that the instrument is feasible to Swedish conditions, but also suggested an update of the manual on some items and that a more comprehensive and broader assessment of quality is preferable. Future research should continue the promising research by Perry and colleagues (Perry, Flanagan, et al. 2008; Perry, Koudys, et al., 2008) and Denne et al. (2015) to establish a set of quality measurements to be used in research and practice, and also explore and agree on sufficient level of competence as mentioned above. In our study on quality as a predictor of outcome, we identified certain quality factors that specifically influenced the results which could be used as indicators of relevant components to focus on. However, there is a need of further research with larger sample sizes and a longer treatment period, in order to either confirm our findings or to detect other additional important predicting quality variables. Furthermore, it would be important to identify if different kinds of quality approaches are required, depending on child characteristics and developmental profiles.

Based on the findings in this thesis it is evident that enriched supervision, including the addition of specific and tailored feedback increased EIBI quality within a limited period of time. Usually, the information of trainer quality is restricted to what can be observed on supervision meetings and consequently, the lack of data on how the training actually is conducted and implemented on a daily bases may lead to inflexible and rigid applications of methods and procedures (Leaf et al., 2016). An extended and regular evaluation of video clips of preschool training would be a valuable supplemental information even without a formal quality assessment.

In conclusion, we hope that the studies included in this thesis will further stimulate assessment, monitoring and assurance of EIBI quality in treatment trials and clinical practice with children with ASD.

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