Early intervention programs for toddlers with Autism Spectrum Disorder: a systematic review

Programas de intervenção precoce para crianças com Transtorno do Espectro do Autismo: uma revisão sistemática

Programas de intervención temprana para niños pequeños con Trastorno del Espectro Autista: una revisión sistemática

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Abstract
This review identified 8 randomized controlled trials (RCTs) that evaluated early intervention (EI) programs for infants in the age-group 18-48 months who either had been diagnosed with, or were considered at risk for, autism spectrum disorder (ASD). The studies were summarized in terms of participant characteristics, intervention characteristics, rigor of study/research and outcomes. Intervention characteristics included the provision of training to parents. All the studies used RCT design, with control subjects who were either toddlers of typical development (TD) or toddlers with ASD following “treatment as usual” (TAU) or another treatment, and all were rated as strong in terms of quality/rigor. Positive results were recorded for parental acceptability and satisfaction, and reduction of parenting stress. In most of the studies, the social communication and developmental skills of the toddlers were enhanced. We conclude that EI programs for ASD show promise, and may be beneficial for both the toddlers and the parents, but the limited number of RCTs and the wide variety in intervention programs and assessment instruments used indicates the need for additional research to evaluate the specific benefits.

Keywords: Autism Spectrum Disorder; Autism; Early intervention; Toddlers; Parents.

Resumo
Esta revisão identificou 8 ensaios clínicos randomizados (RCTs) que avaliaram programas de intervenção precoce (EI) para bebês na faixa etária de 18-48 meses que foram diagnosticados com, ou foram considerados em risco de transtorno do espectro do autismo (ASD). Os estudos foram resumidos em termos de características dos participantes, características da intervenção, rigor do estudo / pesquisa e resultados. As características da intervenção incluíram o fornecimento de treinamento aos pais. Todos os estudos usaram um design de RCT, com indivíduos de controle que eram bebês de desenvolvimento típico (TD) ou bebês com ASD após “tratamento usual” (TAU) ou outro tratamento, e todos foram classificados como fortes em termos de qualidade / rigor. Resultados positivos foram registrados para aceitação e satisfação dos pais e redução do estresse parental. Na maioria dos estudos, a comunicação social e as habilidades de desenvolvimento das crianças foram aprimoradas. Concluímos que os programas de EI para TEA são promissores e podem ser benéficos para crianças e pais, mas o número limitado de ensaios clínicos randomizados e a
amplified variability of programs of intervention and instruments of evaluation used indicate the need for additional research to evaluate specific benefits.

**Palavras-chave:** Transtorno do Espectro do Autismo; Autismo; Intervenção precoce; Crianças; Pais.

**Resumen**
Esta revisión identificó ocho ensayos controlados aleatorios (ECA) que evaluaron los programas de intervención temprana (IE) para lactantes en el grupo de edad de 18 a 48 meses que habían sido diagnosticados o se consideraban en riesgo de tener un trastorno del espectro autista (TEA). Los estudios se resumieron en términos de características de los participantes, características de la intervención, rigor del estudio / investigación y resultados. Las características de la intervención incluyeron la provisión de capacitación a los padres. Todos los estudios utilizaron un diseño de ECA, con sujetos de control que eran lactantes de desarrollo típico (DT) o lactantes con TEA que seguían el "tratamiento habitual" (TAU) u otro tratamiento, y todos se calificaron como sólidos en términos de calidad / rigor. Se registraron resultados positivos para la aceptabilidad y satisfacción de los padres y la reducción del estrés parental. En la mayoría de los estudios, se mejoraron las habilidades de comunicación social y desarrollo de los niños pequeños. Concluimos que los programas de IE para los TEA son prometedores y pueden ser beneficiosos tanto para los niños pequeños como para los padres, pero el número limitado de ECA y la amplia variedad de programas de intervención e instrumentos de evaluación utilizados indica la necesidad de realizar investigaciones adicionales para evaluar los beneficios específicos.

**Palabras clave:** Trastorno del Espectro Autista; Autismo; Intervención temprana; Niños pequeños; Padres.

### 1. Introduction

It is currently possible to diagnose autism spectrum disorder (ASD) at a very early stage, namely when the toddlers are aged around 2 years. This early identification of toddlers with ASD facilitates the implementation of early intervention (EI) for children even before they attend school (Mottron, 2017), which is considered to be “essential to achieving the best outcomes” (Pierce et al., 2016). Several studies have shown improved outcomes for toddlers and children with ASD after EI (Granpeesheh et al., 2009; Rogers et al., 2012; Zachor et al., 2007).

EI is addressed to toddlers and young children with disabilities and/or developmental delay, and their families, and can help them to cope with the difficulties that their condition causes in their everyday lives. Behavioral interventions are not aimed at “curing” ASD, which is a neurodevelopmental disorder already established in infancy (Landa et al., 2018), but one of the main goals of EI is to reduce the manifestation of ASD symptoms to a minimum. Other intervention goals include the development of social, language, cognitive, adaptive, and play skills (Green et al., 2017; Landa & Kalb, 2012).

The age of enrollment of children in EI programs is a factor that affects significantly their effectiveness and long-term outcomes, because the first two years of a child’s life are characterized by rapid changes in many areas, especially in social, cognitive and language development. This means that the introduction of EI at around two years of age, when the developmental gaps between toddlers of typical development (TD) and those with ASD are still small, should bring the best results (Bradshaw et al., 2015). In their review, Granpeesheh and colleagues (2009) found that EI was more effective for younger participants (2.55.15 years) than for those who were older (5.157.14 years).

An EI program can be either evidence-based or associated with empirical data that validates its effectiveness (Stahmer et al. 2005). While many EI programs were based on applied behavior analysis (ABA) in the past, a method that was strongly supported by the research community (Lovaas 1987; Reichow 2012), contemporary EI tends to follow the principles of developmental psychology and other naturalistic methods. Thus, interventions have become directed more towards the child itself and are now conducted in more natural environments, such as the child’s home (Schreibman 2014).

Several EI programs that are popular among researchers and clinicians have one thing in common, which is the integration of behavioral, naturalistic, and developmental strategies, and they are labeled “naturalistic developmental behavioral interventions” (NDBIs) (Bradshaw et al., 2015). Some of these are the Early Start Denver Model (ESDM) (Rogers and Dawson 2010), the Enhanced Milieu Teaching (Kaiser and Hester 1994), and the Pivotal Response Treatmen” (PRT)
Systematic reviews of EI conducted to date include studies of a variety of EI programs (Bradshaw et al., 2015; Landa, 2018) or several studies implementing only one type of EI, e.g., the ESDM (Waddington et al., 2016). The present review was focused on currently available studies of EI programs applied to children with ASD between the ages of 18 and 48 months. The effectiveness of these programs was evaluated through RCTs, where participants were assigned randomly to either a treatment group or a control group. The results of the eligible studies were synthesized, and the most relevant findings are presented. Critical elements that were explored in this review were: a) the types of EI programs that were implemented, b) infant and parent outcomes, c) intensity and duration, and d) maintenance and generalization.

2. Methodology

The review focused on interventions for toddlers aged 18-48 months with ASD. The review methodology was based on the guidelines of Ahn & Kang (2018). The first step was the formulation of research questions. Next, the authors determined the inclusion and exclusion criteria for the studies that were to be analyzed, and conducted a rigorous literature search. The study selection was made with the application of the eligibility criteria, and the quality of the presented evidence was discussed. The final steps were data extraction and analysis, and presentation of results. Each study that met the predetermined criteria was analyzed and summarized in terms of a) participant characteristics, b) intervention approach, c) toddler and parent outcomes.

2.1 Research questions

1. What were the most popular early intervention programs for toddlers with ASD?
2. What were the intervention outcomes for toddlers with ASD after EI? Were they positive or negative?
3. Did the parents feel that they had benefited from the EI programs? What were the main effects on them?

2.2 Inclusion and exclusion criteria

Only RCTs were included in the review. To be included, a study had to meet the following criteria: a) empirical research evaluating the effects of an EI program, b) the participants of the EI were toddlers with an age of above 18 months and below 48 months at entry to the program, c) the toddlers had been diagnosed with ASD, d) the results of the study included at least one objective child measurement and one parent outcome measurement.

Articles were excluded from the review if they: a) were non-experimental (e.g., literature reviews, meta-analyses, case reports); b) did not include an EI program; c) did not include toddlers aged 18 to 48 months. Studies were included that primarily, but not exclusively, targeted children aged <48 months or whose mean age was <48 months at the start of the intervention. Grey literature (i.e., dissertations, chapters, etc.) was excluded.

To determine whether a study met the inclusion criteria, the first and the last authors independently completed the search and evaluated all the studies. The selected articles were then compared for reliability, which was calculated using percent agreement on the articles each author identified as meeting the inclusion criteria. Disagreement between the two authors was discussed until they came to an agreement.

2.3 Search procedure

The research papers were found by a search in the PubMed, Education Resources Information Centre (ERIC), Science Direct and Scopus databases for papers published in English, appearing in peer-reviewed journals since 2010. The keywords used were: ASD, autism, autistic, early intervention program, toddler. The initial search resulted in 4,348 studies after
Duplicates were removed. The researchers read the titles and the abstracts to exclude studies that did not incorporate experimental results of EI programs, and/or referred to disabilities not including ASD. The remaining articles were independently screened by the authors for the inclusion criteria.

An ancestral search was conducted using the reference lists of the studies that met the inclusion criteria and the “cited in” feature in Scholar Google, and a hand search was made in peer-reviewed articles. Finally, eight studies were identified that fulfilled the criteria (Figure 1). The overall interrater agreement (IRA) was 90% and consensus was reached to resolve the few disagreements.

**Figure 1.** Review of randomized controlled trials of early intervention for toddlers with autism spectrum disorder: Selection of research papers based on PRISMA flowchart

Source: Authors.
2.4 Coding procedures

To map and synthesize the included studies, the following coding categories were used: a) child characteristics (number, age, and diagnosis), b) parent characteristics (number of parent participants), c) intervention approach (i.e., empirical and theoretical basis extracted from the description of the intervention; intensity and duration of the intervention, in terms of number of individual sessions over a set period of time, the length of each session), d) quality of the study/research rigor, e) child outcome measurements (e.g., scores on cognitive, language, and/or adaptive behavior assessment), and f) parental outcome measurements (e.g., changes in parenting stress, skills, responsivity, parental use of evidence-based strategies).

Finally, each study was coded to assess its quality based on the evaluative method for determining evidence-based practices in autism, which has been reported to have good to excellent reliability and validity (Reichow et al. 2008). To evaluate the rigor of the studies, two rubrics were developed; one for group research and one for single-subject research. These rubrics include two levels of methodological elements: primary quality indicators and secondary quality indicators. Three levels of rating were given to each study: strong, acceptable/adequate, weak; and demonstrating concrete evidence of quality, strong evidence in most, but not all areas, missing elements, and/or fatal flows. Primary quality indicators for group research include the quality of the description of participant characteristics, independent variable, comparison condition, dependent variable, the link between research question and data analysis, use of statistical tests. Secondary quality indicators were not deemed necessary for the establishment of the validity of the study and are related to random assignment, interobserver agreement, blind raters, fidelity, attrition, generalization and/or maintenance, effect size, social validity.

The second, the third and the fourth authors reviewed independently the included studies to determine whether each of them met the coding categories and the evaluative method for determining evidence-based practices in autism. They extracted data from each of the eight studies and created a summary, as shown in Tables 1 and 2. The authors compared the results for the coding, and any disagreement between the authors was discussed until they came to an agreement. Overall, the IRA for all the coding categories was 100%.
Table 1. Review of randomized controlled trials of early intervention for toddlers with autism spectrum disorder: Characteristics of the children, intervention approach, quality/rigor of the studies.

<table>
<thead>
<tr>
<th>Study</th>
<th>Child characteristics</th>
<th>Parental Characteristics</th>
<th>Intervention approach</th>
<th>Maintenance/Generalization</th>
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<tbody>
<tr>
<td></td>
<td>N</td>
<td>Age (months)</td>
<td>Diagnosis</td>
<td></td>
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<tr>
<td>Carter et al. (2011)</td>
<td>N=32 (intervention group) and N=30 (no treatment group)</td>
<td>15-25 (mean21.11 treatment group; mean 29.98 control group)</td>
<td>ASD NS</td>
<td>HMTW: a parent-mediated communication-focused treatment in preschool-aged children with ASD</td>
</tr>
<tr>
<td>Ibanez et al. (2018)</td>
<td>N=52 (control group), N=52 (tutorial group)</td>
<td>18-60 (mean44.77, control group; mean42.83, tutorial group)</td>
<td>ASD N=52 (control group), N=52 (tutorial group)</td>
<td>Self-directed web-based parenting tutorial Enhancing Interactions tutorial Parent behavior survey, child behavior survey, PIA-CV, PES, PSI/SF</td>
</tr>
<tr>
<td>Kasari et al. (2014)</td>
<td>N=34 (control group) and N=32 (treatment group)</td>
<td>15-31 (mean22.37)</td>
<td>High risk for ASD N=43 (control group), N=32 (treatment group)</td>
<td>Parent-education intervention Participants’ home Standardized manual CSEFEL</td>
</tr>
<tr>
<td>Study</td>
<td>Sample Size</td>
<td>Type</td>
<td>Intervention</td>
<td>Goals</td>
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<tr>
<td>Kasari et al. (2015)</td>
<td>N=43 (JASPER), N=43 (PEI)</td>
<td>ASD</td>
<td>PEI: Psycho educational intervention</td>
<td>Provide individual education and support to parents of young children with ASD</td>
</tr>
<tr>
<td>Oosterling et al. (2010)</td>
<td>N=36 (experiment group), N=31 (control group)</td>
<td>ASD</td>
<td>Focus parent training: using a professional-as-consultant and parent-as-therapist model and adopting an eclectic approach within a social [1] pragmatic and developmental context</td>
<td>Sustain periods of joint engagement and increasing joint attention gestures and play skills</td>
</tr>
<tr>
<td>Study</td>
<td>Sample Size</td>
<td>Age Range</td>
<td>Diagnosis</td>
<td>Goal</td>
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<tr>
<td>Rogers et al. (2018)</td>
<td>N=24 (P-ESDM group), N=21 (P-ESDM++)</td>
<td>12-30 mean 25</td>
<td>ASD</td>
<td>Test the effects of an enhanced version on parent and child learning, and evaluate the sensitivity to change of proximal versus distal measures of child behavior</td>
</tr>
<tr>
<td>Turner-Brown et al. (2019)</td>
<td>N=32 (FITT group) and N=17 (SAU group)</td>
<td>17-35 for FITT group (mean 29.6), 22-35 months for TAU group (mean 29.7)</td>
<td>ASD</td>
<td>To examine the efficacy of the FITT program in enhancing the developmental and social communication skills of toddlers with ASD and also to reduce parenting stress and promote their well-being</td>
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</table>
Welterlin et al., 2012) 

| N= 3 (HTP group) and N= 3 (WL group) that completed the intervention | 24-37 for HTP group (mean 30.5), 24-39 for WL group (mean 30.5) | N= 3 (HTP group), N= 3 (WL group) that completed the intervention | Home TEACCHing Program for toddlers and their parents and comparison with a Waitlist group. Sessions were videotaped | Goal: the study aimed to reduce parental stress and to instruct parents on how to implement structured teaching with their children. Also, the HTP intervention group children were expected to have improved behavior both during sessions and on developmental outcome measures as compared with the WL control group. Strategies: Families were paired according to their children’s developmental age and then randomly assigned to a group (either intervention or control group). Three pairs (six families) completed the multiple-baseline single-subject design phase. Data were collected through videotaping a small part of each session |
| HTP group: 1.5h/week for 12 weeks where parents met with a specialist, each treatment session had several 5-10 min teaching times (parent training sessions) | No/No |

3. Results

The first search yielded in 4,348 papers, from which the final selection resulted in 8 studies that met the inclusion criteria and the coding procedures. Tables 1 and 2 provide a summary of the studies in this review in terms of a) participant characteristics, b) intervention characteristics, c) quality/rigor, and d) outcomes.

Child characteristics

The eight studies included a total of 485 participants aged between 17 and 48 months, of which 251 received EI and 234 were control subjects, who were either children of TD or children with ASD who received “treatment as usual” (TAU) or another treatment. All the studies required that the ASD participants either had been diagnosed with ASD or were considered to be at risk for ASD (i.e., they presented behavioral symptoms of ASD) prior to participating in the intervention. The study of Ibanez and colleagues (2018) did not have any children as direct participants in EI, as this study reported on training of the parents and its effect on their children.

The children had a diagnosis of ASD or high risk for ASD in all eight studies, although in one study, one child included in the experimental group had a diagnosis of pervasive developmental disorder, not otherwise specified (PDD-NOS) (Oosterling et al., 2010). The ASD diagnosis was based on the Autism Diagnostic Interview-Revised (ADI-R) and the Autism Diagnostic Observation Schedule (ADOS) (Kasari et al., 2015). In one study the tool that was used was not defined, but a copy of the child's diagnostic report confirming an ASD diagnosis was one of the criteria for inclusion in the research (Ibanez et al., 2018). In three studies the participants were described as being at high risk of ASD, based on the Screening Tool for Autism in two-year-olds (STAT) (Carter et al., 2011), the Modified Checklist for Autism in Toddlers (M-CHAT), and the social composite score of the Communication and Symbolic Behavior Scales Developmental Profile (CSBS DP) (Kasari et al., 2014), the Early Screening of Autistic Traits Questionnaire (ESAT) (Oosterling et al., 2010).

In 6/8 studies, exclusion of children was reported based on other medical, physical, genetic, or neurological conditions, specifically, a genetic disorder (Carter et al., 2011, Kasari et al., 2014, Turner-Brown et al., 2019), or severe auditory, visual, or motor impairments (Ibanez et al., 2018, Carter et al., 2011, Rogers et al., 2018, Turner Brown et al., 2019). In one study the researchers included only children with either a diagnosis of ASD in combination with a developmental age of at least 12 months or children with a diagnosis of PDD-NOS in combination with a developmental age of at least 12 months and a developmental quotient (DQ) below 80 (Oosterling et al., 2010). The gender of the participants is reported in all the studies; most of the participants were male (80%).

Parental characteristics

Almost all the interventions (6/8) used parent-mediated procedures; the parents were taught specific procedures, which they were expected to use with their children during the intervention sessions and in everyday life. The intervention strategies involved didactic sessions about treatment techniques, and a feedback session in which parents and their toddlers practiced the intervention while a therapist provided feedback about implementation. One study examined the effects of an interactive web-based tutorial for improving children’s engagement in daily routines and social communication, and parenting efficacy and parental stress (Ibanez et al., 2018).

Intervention approach

The empirical and theoretical basis

Most of the studies adapted intervention models that had been previously applied for toddlers. These included
Hanen’s More Than Words (HMTW) (Carter et al., 2011), psychoeducational intervention (PEI) (Kasari et al., 2015), Joint Attention Symbolic Play, Engagement, and Regulation (JASPER) (Kasari et al., 2015), focus parent training (Oosterling et al., 2010) and the ESDM (Rogers et al., 2018), which have all been used with toddlers and preschool-aged children. Two studies (Welterlin et al., 2012; Turner-Brown et al., 2019) applied similar intervention models: the Family Implemented TEACCH for toddlers (FITT) and the Home TEACCHing Program for toddlers and their families. These two early intervention programs follow the basic principles of the TEACCH program which was modified and altered to be implemented for toddlers and their families, mainly in at-home settings.

Two studies provided, respectively, an interactive, web-based parenting tutorial (Ibanez et al., 2018) and a self-directed, web-based training course (online course/tutorial; Kasari et al., 2014), which included 24-hour accessibility, standardization of training, personalization/individualization (e.g., self-paced), risk-free environment, and the opportunity for interactive exercises and multimedia components. Self-directed, web-based parent training programs appear to be cost-effective and easily available to the parents.

### Intensity and duration

The duration of treatment ranged from 4 to 12 weeks in most of the interventions, and all were low-intensity, totaling no more than 2 hours of intervention per week. In one study (Turner-Brown et al., 2019), the sessions were carried out for 24 weeks, and in two studies the duration is not specified; in that of Carter and colleagues (2011), the intervention involved 8 group sessions with parents only, and 3 in-home individualized parent-child sessions, and in that of Ibanez and colleagues (2018), the entire tutorial was approximately 6 hours, with the parents reviewing the tutorial across at least 4 or 5 sessions.

All the studies reported data collection for evaluation at either two or three time-points, specifically at baseline and 1 year after the start of the intervention (Oosterling et al., 2018); baseline and approximately 7 months after (Turner-Brown et al., 2019), or time1: prior to randomization/baseline/pre-treatment time 2: 5/1/3/ months, post-treatment; time3: 9/2/12/6 months post-enrollment. In one study data were collected every 4 months on child and parent mastering of skills, and long-term observations of of child change were made (Rogers et al., 2018). One study included four data collection points, specifically prior to intervention, and at the fourth, eighth and twelfth weeks, the last being post-intervention (Welterlin et al., 2012).

### Research rigor

In terms of research rigor, all eight studies were rated as having a strong research design, according to the criteria developed by Reichow et al. (2008) and Reichow (2011) (Table 3). High quality was observed on all primary quality indicators (i.e., participant characteristics, independent variable, comparison condition, dependent variable, a link between research question and data analysis, use of statistical tests), and the studies showed evidence of four or more secondary quality indicators (i.e., random assignment, interobserver agreement, blind raters, fidelity, attrition, generalization and/or maintenance, effect size, social validity).

### Child outcome measurements

The Mullen Scales of Early Learning (MSEL) (Mullen, 1995), the Vineland Adaptive Behavior Scales, Second Edition (Vineland II) (Sparrow et al., 2005), ADOS (Lord et al., 2000), the early social communication scale (ESCS) (Seibert et al., 1982), the Reynell Developmental Language scales (Reynell & Curwen, 1977), the Parenting Stress Index (PSI; Loyd & Abidin, 1985), a Dutch version of the MacArthur Communicative Development Inventory (N-CDI) (Fenson et al., 1993; Zink...
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& Lejaegere, 2002), the Clinical Global Impression—Improvement scale (CGI-I) (Guy 1976), the 7-point Erickson rating scales (Erickson et al., 1985), the ESDM Fidelity Rating System (Rogers & Dawson, 2010), a Likert-based, 5-point rating system of 13 adult behaviors, the PATH Curriculum Checklist (PATH CC; Rogers et al., 2013), the Child Behavior Checklist for 1 ½–5 Years (CBCL) (Achenbach & Rescorla, 2000), Child Intervention History (Version 6-10-13), adapted from the CPEA Network Intervention History form (Rogers et al., 2012b), a demographic information form and a services/intervention questionnaire, the Parent Implementation Rating Form (PIRF), The FITT Fidelity Forms (Turner-Brown et al., 2019), the Parent Interview for Autism-Clinical Version (PIA-CV) (Stone et al. 2003), the RAND-36 (Ware and Sherbourne 1992), and the Scales of Independent Behavior-Revised (SIB-R) Bruininks et al. 1996), and self/parent-report measurements using two routine-specific surveys; one describing the behavioral strategies the parents used and the other describing the child’s engagement (or participation) behaviors (Ibanez et al., 2018).

The child and parental outcomes of EI programs are presented in Table 2. Five studies reported at least one child outcome measurement of the child's social interaction and communication skills (Carter et al., 2011; Kasari et al., 2014; Kasari et al., 2015; Rogers et al., 2018, Turner-Brown et al., 2019). One study focused on improving children’s engagement in daily routines and social communication (Ibanez et al., 2018), and another on language development, engagement, and social communication (Oosterling et al., 2010). The outcome measurements included joint attention, initiating behavior requests, intentional and/or nonverbal communication, expressive and language skills, visual perception, functional and symbolic play, engagement during daily routines, and compliance and willingness to join in mutual activities. Three studies reported positive results for these outcome measurements, but Carter and colleagues (2011) found no major effects of treatment on child outcomes, either immediately after the parent-implemented treatment or at the follow-up assessment. Kasari and colleagues (2014) and Oosterling and colleagues (2010), also, found no significant differences between the two groups on joint attention and language skills. The study of Welterlin and colleagues (2012) focused on different types of skills, specifically on children’s independent living skills, which they reported to be enhanced after the treatment, for most of the children that participated.
<table>
<thead>
<tr>
<th>Study</th>
<th>Child outcomes</th>
<th>Parental outcomes</th>
</tr>
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</table>
| Carter et al. (2011)  | No main effects of the HMTW intervention on children’s communication immediately after the parent-implemented treatment or 5 months after treatment  
There were treatment effects on child communication gains to Time 3 that were moderated by children’s Time 1 object interest. Children with lower levels of Time 1 object interest exhibited facilitated growth in communication; children with higher levels of object interest exhibited growth attenuation. | No main effects of the HMTW intervention on parental responsivity: the effect size immediately after treatment was medium to large (0.71) and was moderate even at the follow-up period (0.50) |
| Ibanez et al. (2018)  | Children in the Tutorial group exhibited increased engagement during routines at T2, which was sustained at T3. Children improved in their ability to tolerate and transition during routines, which may include exhibiting fewer externalizing behaviors (e.g., physically struggling, leaving the area, fussing). Social-communication improvements coincided temporally with children’s increased ability to engage during routines and may suggest that improvements in routine-specific behaviors generalized to broader contexts and interactions. | The Tutorial group exhibited: (a) increased parental use of evidence-based strategies at T2 and T3 (large effect sizes); (b) decreased parenting stress and increased parenting efficacy at T3 (medium effect sizes), and (c) improved child engagement during routines and broader social communication at T2 and T3 (medium to large effect sizes). The Control group did not exhibit any such gains.  
The tutorial had a direct effect on the immediate dynamics of the interactions during routines Parenting stress related to challenges in the parent-child relationship (i.e., PSI/SF PCDI scale) declined significantly for parents in the Tutorial group, |
<p>| Kasari et al. (2014)  | Although visual reception, expressive and receptive language scores significantly increased from treatment start to follow-up, there was no significant group by time interaction effects. No significant changes were noted in joint attention. | A significant effect of the intervention on parental responsiveness from start to the end of treatment was maintained at follow-up, no significant difference between groups on children’s joint attention and language skills |</p>
<table>
<thead>
<tr>
<th>Kasari et al. (2015)</th>
<th>Joint engagement more than doubled from entry to week 10 for the JASPER group, with a large effect size. The increase in the length of time spent jointly engaged was maintained at the 6-month follow-up and significant for the JASPER group compared with the PEI group. The JASPER group increased more in types of functional play than the PEI group; however, these skills did not maintain at follow-up. Children in the JASPER condition engaged with their teachers more in their early intervention classroom. These findings may be among the first indicating generalization of joint engagement skills from a parent-mediated intervention to new partners and contexts.</th>
<th>Parents coached in specific JASPER strategies were significantly more effective at engaging their children in play at post-treatment and follow-up than parents who received information about specific strategies through the PEI. Effect sizes were moderate to large. Results indicated a reduction in parenting stress for families in the PEI condition.</th>
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<tr>
<td>Oosterling et al. (2010)</td>
<td>Language skills of children and engagement in both groups improved with time. Clinical global improvement from baseline to endpoint was not different between the two groups. Regarding engagement and early precursors of social communication, no intervention effects were found.</td>
<td>Concerning parental skills, no significant improvement with time was found. The mothers in the experimental group did not show an improvement in parenting skills relative to the mothers in the control group.</td>
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<td>Rogers et al. (2018)</td>
<td>While there were significant gains for both groups over time, there were no group differences in the degree of improvement in children’s skills after 12 weeks of intervention. There was a significant positive relationship between the degree of improvement in parental fidelity of implementation and increases in child social communication and decreases in autism symptoms on the proximal measure of change.</td>
<td>The rate of parental learning of the intervention was improved. Parents in the P-ESDM++ group demonstrated significantly increased sensitivity and skill in supporting child social-communicative development measured by increases in parent fidelity of implementation scores compared to the parents in the P-ESDM group. Parents in both groups were extremely satisfied with the intervention that they received. This is important in allaying concerns about parent-implemented interventions and their potential for increasing parent stress.</td>
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Turner-Brown et al. (2019)  | There were no significant differences for FITT and SAU groups at baseline. Children from the FITT group had higher PIA scores (a measure of autism symptom severity) and PIA imitation scores than children from the SAU group. The evaluation form that was filled in by the parents revealed no regression in social interaction, cognitive skills, and communication of children from both groups. 43% of the children from the FITT group were reported to have made “a lot of progress” in social interaction skills (with a statistically significant difference from the SAU group). Parents from the FITT group had lower levels of stress and parental distress. They also had better results regarding their quality of life (with the RAND-36 tool) and they reported high levels of satisfaction with the program. All parents exhibited high levels of engagement according to therapist evaluations.

| Welterlin et al. (2012) | Independent functioning skills were enhanced for two of three pairs of participants. Some differences were noted for young participants from each pair, specifically from pair 2. Subject HT-C2 had a decrease in their target skills, whereas WL-C2 had better results. Regarding the first pair, both subjects (HT-C1 and WL-C1) had an increase in their outcomes, but variation was larger for WL-C1 after the treatment. Regarding the third group, subject HT-C3 showed a great response to the treatment, whereas subject WL-C3’s response was smaller. Children from the HTP group made progress in expressive language, as well as children from the WL group. Differences between children from the two groups were not statistically significant. An increase in setup behavior was observed for all parents that participated in the program. Also, there was an increase in effective prompts and a decrease in ineffective prompts for all parents, but variations were more significant for those that participated in the treatment group. There were no statistically significant differences between the HTP and the WL groups. Parent stress was had a slight decrease for HTP and a slight increase for WL participants, but again, group differences were not statistically significant.

FITT= Family Implemented TEACCH for Toddlers, HMTW= Hanen’s “More Than Words”, HTP= Home TEACCHing Program, JASPER= Joint Attention Symbolic Play Engagement and Regulation, PIA-CV= Parent Interview for Autism–Clinical Version, PSI/SF= Parenting Stress Index/Short Form, TAU= treatment as usual Source: Authors.
Table 3. Review of randomized controlled trials of early intervention for toddlers with autism spectrum disorder: Research quality indicators.

<table>
<thead>
<tr>
<th>Studies</th>
<th>Quality Indicators</th>
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<tr>
<td></td>
<td>Primary Quality Indicators</td>
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<td>Participant characteristics</td>
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<tr>
<td>Carter et al. (2011)</td>
<td>HQ</td>
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<td>Ibanez et al. (2018)</td>
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<td>Welterlin et al. (2012)</td>
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Note: AQ=Acceptable Quality, E=Evidence, HQ=High Quality, NE=No Evidence, UQ=Unacceptable Quality.
Source: Authors.
Parent outcomes measurements

Positive results in parental responsivity were reported in 6/8 studies. The effect size was medium to large immediately after treatment, and moderate at the follow-up assessment (Carter et al., 2011). Nearly 80% of parents in the focused playtime intervention improved in their responsiveness (Kasari et al., 2014), in parental use of evidence-based strategies, such as providing simple verbal instructions, using visual schedules, modifying routine steps (Ibanez et al., 2018), use of effective prompting during structured teaching implementation (Welterlin et al., 2012), and learning about the intervention by showing increasing sensitivity and skill in supporting child social-communicative development (Rogers et al., 2018). Parenting stress related either to challenges in the parent-child relationship or to the disorder itself declined significantly in the treatment group (Ibanez et al., 2018, Turner-Brown et al., 2019) and the parent education program (Kasari et al., 2015), although the decrease in parental stress in the treatment group was not significant in the studies of Welterlin and colleagues, (2012), and Oosterling and colleagues (2010) reported that the training program did not significantly influence parental skills.

Social validity

A questionnaire about parent satisfaction was included in 4/8 studies. Positive results regarding feasibility, acceptability or satisfaction with the intervention were reported in three studies (Carter et al., 2011; Ibanez et al., 2018; Rogers et al., 2018). In the study of Ibanez and colleagues (2018), the parents in the tutorial group indicated high levels of satisfaction with the technical aspects and the clinical content. Rogers and colleagues (2018) administered the Intervention Evaluation Form for Parents, a Likert-type scale of 14 questions, at the end of the treatment period, and other researchers monitored treatment integrity by measuring fidelity of implementation (Kasari et al., 2014; Kasari et al., 2015). Oosterling and colleagues (2010) used professional observation, parent reports, and video recording of data collection. Turner-Brown and colleagues (2019) reported positive scores on parental satisfaction with the “Family Implemented TEACCH for Toddlers”; specifically, high ratings in the domains of satisfaction in general, and satisfaction with the goals set, with the intervention procedures, and with the outcomes.

Maintenance/Generalization

Maintenance and/or generalization probes were conducted in 6/8 studies. In three, only maintenance was reported on (Carter et al., 2011, Ibanez et al., 2018, Kasari et al., 2014), and in two studies both maintenance and generalization phases were included (Kasari et al., 2015, Rogers et al., 2018). In the study of Carter and colleagues (2011) the parents exhibited a moderate decrease in their responsivity during the follow-up period, but the children’s increase in communication was moderate to large (weighted frequency of intentional communication) and very large (nonverbal communication). Ibanez and colleagues (2018) could not refer to long-term sustainability in gains demonstrated by parents and children, because of the relatively short follow-up period. Kasari and colleagues (2014) conducted a long-term follow-up, which showed lack of parental responsiveness; only those parents who showed responsiveness at baseline maintained their responsiveness to follow-up. Mixed results were reported by Kasari and colleagues (2015), since maintenance of joint engagement was limited, and the children’s improvements in functional-play diversity and overall play level were not maintained at follow-up. The lack of follow-up data in the study of Rogers and colleagues (2018) prevents determination of the extent to which the treatment resulted in stable changes in parent delivery, or whether the results are generalizable to community settings. Kasari and colleagues (2015) explored the generalization of joint engagement in the classroom and reported that children in the JASPER program engaged more in their early intervention classroom.
Moderators of outcome

Two studies included moderator variables to uncover the effects of specific child and intervention characteristics on child and parent outcomes. Carter and colleagues (2011) identified limited object interest as a moderator for facilitating growth in communication for the HMTW group. Ibanéz and colleagues (2018) identified the tutorial itself as leading to changes in the routine-specific strategies used by parents, and improvement in the behaviors exhibited by children. Kasari and colleagues (2014) revealed a possible relationship between the durability of the treatment and the long-term outcomes. Extending the duration of the intervention, or supplying “booster” sessions, may improve responsiveness and maintain positive changes in parental behavior. Kasari and colleagues (2015) indicated a reduction in parenting stress for families in the PEI program, who consulted with an expert about their children and gained greater knowledge about ASD. Oosterling and colleagues (2010) reported that the DQ may affect language improvement, engagement, and precursors of social communication. Welterlin et al. (2012) noted that parents may need more time and practice opportunities to be more effective in implementing structured teaching, and that the implementation of only one baseline probe could not lead to potent conclusions. Lastly, Turner-Brown and colleagues (2019) discussed factors such as therapist consultation, in-home implementation of the early intervention program with particular emphasis given to understanding ASD, and implementation of parent groups, which may have a positive impact on parent outcomes.

4. Discussion

The purpose of this review was to evaluate EI programs for toddlers with, or at risk for, ASD. Using stringent criteria, eight relevant studies were identified, all of which were published since 2010. All of the studies included in the review were RCTs, and they examined EI for toddlers in the age range 18-48 months at enrolment. The interventions varied in intensity and duration, ranging from 4 to 12 weeks, with no more than 2 hours per week. Mixed findings were reported regarding enhanced effectiveness over the TAU comparison groups in a range of outcome measures, including social skills in the children, and parenting stress.

A wide variety of EI programs was implemented in the eligible studies, including HMTW, JASPER, the Focus Parent Training, the Parent-implemented ESDM (P-ESDM, FITT, and others. No two (or more) studies implemented the same EI program, so each investigated the effectiveness of a different program, and therefore conclusions cannot be drawn on which program is more popular, simply based on the findings of this review.

Similarly, a various different instruments were used for outcome measurement in the children and the parents. Most of the studies reported some positive outcomes for the participating parents and/or toddlers, which is encouraging, and serves to demonstrate the need for further research. Positive parental outcomes were reported by 4/8 studies, which were maintained at follow-up, related to the use of evidence-based strategies, reduced parenting stress, increased parental sensitivity, and skills in supporting their children (Ibanez et al., 2010; Kasari et al., 2014; Kasari et al., 2015; Rogers et al., 2018), although two studies detected no major effects on parental responsivity and skills (Carter et al., 2011; Oosterling et al., 2010). Most studies (Ibanez et al., 2018; Kasari et al., 2015; Oosterling et al., 2010) also reported significant positive child outcomes, particularly in engagement during daily routines, communication skills, joint engagement, and language skills,. but others recorded no significant group differences in the degree of improvement in children’s skills post-intervention (Carter et al., 2011; Kasari et al., 2014; Rogers et al., 2018).

Exploration of the moderator variables associated with outcome data may provide useful information about factors that can influence the effectiveness of an intervention. HMTW appears to be more effective with children who show less interest in objects, whereas children who had a high interest in objects exhibited growth attenuation (Carter et al., 2011). Other
moderators appear to be the intensity of the intervention and the tutorial itself. Rogers and colleagues (2012) found that children who received more intervention hours appeared to benefit more.

It is common to find inconsistencies in the outcomes of studies of EI (Landa et al., 2018), which can be attributed, among other factors, to the individual differences and characteristics of the children (Howlin et al., 2009). Clinicians and therapists should therefore consider each child’s strengths and weaknesses, and the family environment, very carefully before suggesting an EI program. Following enrolment, there should be constant contact with the family and meticulous gathering and evaluation of information.

The current review suggests that the various EI programs used in the reviewed studies, based on the high ratings for research rigor, offer promising treatment for toddlers with or at risk for ASD. All the studies included in the review were rated as being methodologically strong, which increases the certainty of the evidence. Two studies with strong ratings, however, did not report significant improvement for either the toddlers or the parents (Carter et al., 2011; Oosterling et al., 2010), but positive results from six of the eight studies support the EI programs that were used, as promising interventions for toddlers with ASD and their families.

It should be noted that EI is aimed at facilitating the participation of children with ASD in more inclusive settings, minimizing the developmental and behavioral obstacles that these children face (Landa et al., 2018). To this end, parents and clinicians should collaborate and decide on the best approach that fits their child’s needs, as each program could have different effects on different children. The age of enrollment, the goals that are set before enrolling in an EI program, and the intensity and duration, must be tailored to the individual circumstances.

5. Final Considerations

This review has several limitations, and the conclusions presented are based upon a relatively small sample size. It is possible that some relevant studies were excluded based on the stringent criteria related to experimental design and/or publication in English-language peer-reviewed journals. Research groups investigating the effectiveness of EI must consider the impact of moderator variables and their effect on outcomes, so careful identification of the factors that might have an influence on the results is essential. Particular attention should be given to conducting maintenance and generalization probes, in order to examine the long-term benefits of an EI program. For future studies of EI programs for toddlers with ASD, larger sample sizes and application of various different intervention approaches would provide useful evidence.

References
(References marked with an asterisk indicate studies included in the literature review)


