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ABSTRACT

The number of children diagnosed with Autism Spectrum Disorder (ASD) is on the rise. Evidence-based early intervention is one of the keys to improve outcomes. This article briefly presents the history of Naturalistic Developmental Behavioral Interventions (NDBI) and uses the Early Start Denver Model (ESDM) as an exemplary NDBI to illustrate the current evidence and future directions of NDBI. At the end, we call for more efforts to disseminate NDBI across the globe.

KEYWORDS: EARY INTERVENTION, EARLY START DENVER MODEL, ESDM.

RÉSUMÉ

Il est temps de changer. Le modèle de Denver pour jeunes enfants

Le nombre d'enfants diagnostiqués avec un trouble du spectre de l'autisme (TSA) est en progression. L'intervention précoce fondée sur les preuves est l'une des clés pour améliorer l'évolution. Cet article présente brièvement l'histoire des Interventions Comportementales et Développementales en milieu naturel (Naturalistic Developmental Behavioral Interventions NDBI) et utilise le Modèle de Denver pour jeunes enfants (ESDM) comme exemple d'une intervention NDBI pour illustrer les données scientifiques actuelles et les orientations futures de ces interventions. Finalement, nous appelons à plus d'efforts pour diffuser ce type d'intervention à travers le monde.

MOTS-CLÉS : INTERVENTION PRECOCE, MODÈLE DE DENVER POUR JEUNES ENFANTS, ESDM.

Societies and cultures have long used education as a vehicle to prepare children to become contributing members of society. A high-quality early childhood

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program can produce such "life-cycle benefits" (Garcia, Heckman, Leaf, & Prados, 2016; Heckman, 2008). Studies demonstrate that children who participate in a quality early childhood program have more desirable outcomes than those who do not in terms of health, education, and employment in adulthood (Camilli, Vargas, Ryan, & Barnett, 2010; Knudsen, Heckman, Cameron, & Shonkoff, 2006; Reynold, Temple, & Robertson, 2001; Reynolds *et al.*, 2007). Such effects then extend to their own offspring, producing a rate of return of up to 13.7 % per annum for the community (Garcia *et al.*, 2016). Thus, early education makes positive changes in children's lives, and these findings suggest that early intervention may also have long-term effects on children with disabilities (Guralnick, 2005).

Autism spectrum disorder (ASD) is a neurodevelopmental disorder characterized by social and communication deficits and restricted, repetitive, and stereotypical behaviors, interests, and activities (American Psychiatric Association, 2013). Such difficulties emerge early during the developmental phase and limit an individual's ability to fulfill age-expected social roles throughout life. The impacts of ASD are multilevel (e.g., individual, familial, and societal), multifaceted (e.g., biological, psychological, and behavioral), and long-standing. Caring for a child with ASD can take a large toll on families' emotional and financial resources. Many parents experience a high level of stress, negative emotion, and health-related problems (Benson & Kersh, 2011; Bristol, 1987; Hayes & Watson, 2013). As individuals with ASD age, many of them experience deteriorating health, co-morbid mental conditions, limited learning opportunities, unemployment, dependent living, and lack of participation in their communities (Collins, 2011). The intensive needs for care also create expenses at a societal level. Autism services cost $236-262 billion annually in the U.S. (Buescher *et al.*, 2014). The medical expenditures of individuals with ASD are three to nine times higher than those without ASD (Peacock, Amendah, Ouyang, & Grosse, 2012). These long-term impacts on persons, families, and society as a whole have ignited great interest in interventions for ASD to improve long-term outcomes and independence, and many efforts have been made to develop and test interventions for people with ASD.

A range of early biological and environmental risk factors have been found to be associated both with atypical brain features and also with the behavioral developmental trajectories seen in children with ASD (e.g., Auranen *et al.*, 2002; Dawson, Carver, & Meltzoff, 2002; Friedman *et al.*, 2006). These altered behavioral/developmental patterns result in altered patterns of interaction between a child and his/her social and nonsocial environment. Dawson (2008) coined these atypical patterns of interaction "risk processes" and hypothesized that such risk processes distance children from activities essential to the development of early social and linguistic brain circuitry and behavior and also mediate the relationship between these early susceptibilities and later outcomes. The eventual development of the full ASD syndrome is thus a product of the cumulative risk processes – genetic and environmental – over time. This model highlights the flexibility of a young child's brain and emphasizes the role of

environmental input in brain and behavioral development during this sensitive period involving first few years of life. This is a time of rapid learning which provides a particularly fruitful time for improving the course of development for children with ASD. The importance of early learning has catalyzed many efforts to develop ASD-effective early intervention.

Early intensive behavioral intervention (EIBI) and naturalistic developmental behavioral intervention (NDBI)

Early intensive behavioral intervention (EIBI) was the first comprehensive treatment program for young children with ASD to publish a controlled trial of EIBI. Lovaas's (1987) study comparing a 40-hour, weekly, one-to-one intensive behavioral treatment, grounded in operant learning theory and discrimination-learning data and methods, with a 40-hour, weekly, one-to-one treatment convenience comparison group. The participants in this study were 3 years or younger at the start and received multiple years of intervention. Follow-up of the children revealed that 47 % of those in the intensive treatment group successfully passed through first grade in a public school without any special supports and obtained an average or above average score on IQ tests, while only 2 % of those in the control group reached such a high level of cognitive and educational functioning. Furthermore, these gains were maintained after six years of the study (McEachin, Smith, & Lovaas, 1993). Of the 53 % of children who did not have this level of outcome, they nevertheless demonstrated better outcomes proportionately in terms of school placements and IQ scores than did the comparison group.

Importantly, neither of the two randomized controlled trials of this method replicated the extent of these outcomes. Smith, Klorman, & Mruzek (2015) replicated the finding that the group receiving EIBI showed significant IQ advantages compared to the control group, but did not replicate the finding concerning the level of IQ and educational gains that the first study reported. A community-implemented randomized controlled trial in Britain (Magiati, Charman, & Howlin, 2007) failed to find significant effects of home-based EIBI over regular community-based early support. Nevertheless, the 1987 paper has had tremendous influence on the field of early intervention for ASD. The overwhelmingly positive results of the initial study not only defined the long tradition of the use of principles and procedures from Applied Behavior Analysis (ABA), but also informed more naturalistic and child-directed approaches that have in turn demonstrated positive effects on generalizability of skills, spontaneity of learning, and child engagement during treatment (Schreibman *et al.*, 2015). In addition to behaviorally-based research, research in child development, both of typically and atypically developing populations, has informed ASD early intervention practice in relation to the development of social

communication (Bates, 2014), differences that arise in cognition, play, and social skills (Carpenter, Pennington, & Rogers, 2002; Dawson & Adams, 1984; Rogers, Hepburn, Stackhouse, & Wehner, 2003), and in early symptomology of toddlers with ASD (Dominick, Davis, Lainhart, Tager-Flusberg, & Folstein, 2007; Sigman, Dijamco, Gratier, & Rozga, 2004; Siller & Sigman, 2008).

In some ways, this research has challenged some basic tenets of EIBI, especially those concerning the nature of language development and the degree to which young children with ASD learn through very different processes than do typically developing children. New models and findings from studies of early language/communication, cognitive, and social development shed light on some of the foundational skills (e.g., joint attention, imitation, and active social engagement) necessary for the later acquisition of language, social, and cognitive learning (Kasari, Sigman, Mundy, & Yirmiya, 1990; Rogers & Lewis, 1989) in both typically and atypically developing children, including ASD. They have also demonstrated the effectiveness of naturalistic teaching methods, including child directedness, reciprocal communication exchanges, and the use of natural, embedded reinforcers based on children's accomplishments of their action goals (Koegel & Williams, 1980). In the thirty years following the Lovaas article in 1987, many intervention scientists have worked to integrate the principles of the science of learning and developmental science findings in interventions for early ASD. The resulting treatment approaches are now known as Naturalistic Developmental Behavioral Interventions (NDBI, Schreibman *et al.*, 2015). Examples of these efforts include Early Start Denver Model (ESDM, Dawson et al, 2010), Wetherby's studies using Joint Social Communication/ Emotion Regulation/Transactional Support (SCERTS, Prizant Wetherby, Rubin, & Laurent, 2003) and Kasari's studies of Attention Symbolic Play Engagement and Regulation (JASPER; Kasari, Freeman, & Paparella, 2006). Some interventions first built from ABA models have incorporated developmental features into their approaches, including Pivotal Response Treatment (PRT, Koegel & Koegel, 2006), Enhanced Milieu Teaching (EMT, Kaiser & Hester, 1994), and Project ImPACT (Improving Parents As Communication Teachers (Ingersoll & Wainer, 2013).

Schreibman and colleagues summarized the 12 core features of NDBI:

(a) three part contingency of antecedent-response-consequence; (b) manualized practices; (c) fidelity of implementation criteria; (d) individualized treatment goals based on developmental sequences; (e) ongoing measurement of progress to examine program effectiveness and individualize treatments; (f) child-initiated teaching episodes; (g) environmental arrangement to promote child initiation of interaction; (h) natural reinforcement and related methods for enhancing motivation of the child; (i) use of prompting and prompt fading, (j) balanced turns within object or social play routines; (k) modeling; and (l) broadening the attentional focus of the child through varied stimuli.

In the following section, the Early Start Denver Model will be used as an example to illustrate further the nature of one NBDI, its evidence, and its impact on children, families and larger society.

Early start denver model (ESDM)

The Early Start Denver Model (Rogers & Dawson, 2010) is a comprehensive early intervention for toddlers as young as 12-months. It evolved from the Denver Model, a preschool program for young children aged 24-60 months with autism (Rogers & Lewis, 1989), and Pivotal Response Training (Schreibman & Koegel, 1996), a naturalistic ABA-based intervention, and it is rooted in the Rogers and Pennington's Model of Interpersonal Development (Rogers & Pennington, 1991), and Dawson's Social Motivational Hypothesis (Dawson *et al.*, 2004), and extensive research by both of its developers,

The ESDM is built from sound educational science principles. It is assessment-based, with all areas of development and problem behaviors assessed. When a child first enters treatment, he/she is evaluated in the areas of receptive communication, expressive communication, joint attention, imitation, social skills, play skills, cognitive skills, fine motor skills, gross motor skills, and self-care skills using the developmental ESDM Curriculum Checklist (Rogers & Dawson, 2010). A functional assessment of behavior problems is also carried out for severe behavioral problems with the first few weeks of intervention. From these developmental and behavioral data, the child's primary therapist and the child's parents form short-term (12 week) learning objectives and behavior plans. Objectives are broken down into 5-6 smaller steps to be taught in sequence, and data on all objectives are taken throughout each treatment session to measure progress. Changes to the treatment plan based on daily data are made to maximize the child's rate of progress, including the use of a decision tree that defines the teaching approach to be used based on child progress on each objective. The child is re-assessed at 12-week intervals and new objectives and plans are created from the data. This 12-week cycle continues throughout the child's treatment.

Several aspects of this intervention approach stand out. First, the inclusion of parents in the development of the treatment plan ensures that the learning objectives are not only fitted to the child's current developmental skills and profile of strengths and weaknesses, but it also assures that the treatment plan is socially valid for the family and fits their own priorities as parents. This partnership of the primary therapist (team leader) and parents continues throughout the treatment deliver. Therapists use strategies based on learning science (e.g. ABA: three-part contingencies, reinforcement strategies, maintenance of learned skills and repeated practice at new skills, assuring child motivation and attention for learning, task analysis, prompting, shaping, and chaining procedures, Premack principle, etc.; Koegel, O'Dell, & Koegel, 1987; Premack, 1959; Schreibman *et al.*, 2015) to teach skills systematically in a developmentally-informed sequence based on the curriculum, following children's interests and leads, and embedded in everyday materials and routines. Parents are coached in how to support children's learning using ESDM strategies during everyday activities at home, and parent coaching continues regularly throughout the entire course of the child's treatment. Parents are part of treatment sessions as is feasible for them.

ESDM is a highly flexible intervention and is constructed to be delivered by a range of professionals, as well as paraprofessionals and parents working under the supervision of a professional lead therapist. It can be carried out in a variety of settings: 1:1 treatment from a therapist or supervised paraprofessional, parent-implemented intervention at home and in the community, or group interventions in both specialized and inclusive settings (Rogers *et al.*, 2012; Dawson *et al.*, 2012; Vismara, Young, Stahmer, Griffith, & Rogers, 2009; Vismara, Young, & Rogers, 2013; Vismara, Colombi, & Rogers, 2009; Vivanti *et al.*, 2014). There are treatment manuals that describe procedures for each of these settings, and measurement tools for both the adult and child to assure the adult delivers the intervention with fidelity and to assure that children are progressing well on all of their objectives. Child progress data is taken in set intervals throughout all types of treatment sessions. While the primary therapist and parents act as the team leaders and primary service deliverers, an interdisciplinary team with expertise in developmental/clinical psychology, ABA, early childhood special education, speech-language pathology, and occupational therapy provide oversight of child treatment plans and progress, evaluation, and consultation, as well as direct treatment, as needed.

Evidence for efficacy of ESDM

Dawson and colleagues (2010) carried out a landmark randomized controlled trial of intensive delivery of ESDM. Children in the ESDM group received intervention by trained therapists for two-hour sessions, twice per day, five days per week, for 2 years and ongoing coaching in PRT. Children in the Community treatment group received on average 9.1 hours of individual therapy and an average of 9.3 hours per week of group interventions (e.g., developmental preschool), and their parents received treatment recommendations annually. After two years, the ESDM group showed a 17.6 point increase in cognitive ability, as measured by the Mullen Scales of Early Learning composite standard scores, compared with 7.0 points in the control group. In terms of adaptive skills, as measured by the Vineland Adaptive Behavior Scales, the ESDM group showed stable standard scores at the one- and two-year after the receipt of intervention, while the control group on average showed an 11.2-point decline.

Such improvement is not only statistically significant, but is also clinically meaningful and long-lasting, and maintained two years after the intervention (Estes *et al.*, 2014). Estes and colleagues followed this original sample for two years and published the following findings: Compared to the community group, the ESDM group had a non-significant, 6.4-point verbal IQ advantage. The children in the ESDM group also demonstrated a significantly lower Restricted and Repetitive Behavior score and total score on the ADOS. These findings suggest that ESDM can improve the core symptoms of ASD, which were originally thought to be stable traits (Eaves & Ho, 1996).

A rigorous replication study (Rogers et al, in press) used a randomized, multisite, single blind, intent-to-treat design, and assigned 118 children ages 12-24 months to either ESDM or Community treatment in three different university communities. The experimental group of children received three months of parent-implemented ESDM followed by 24 months of intensive intervention (averaging 15 hours per week) and biweekly parent coaching. The Community group received a wide variety of interventions based on available services and family choice in their communities. Results revealed a partial replication of the results reported in the 2010 paper. While both groups of children accelerated development significantly in all areas assessed, the ESDM group as a whole made significantly greater overall language gains than the community group. This was also true in two of the three sites, but in the third site there was no significant group difference in language, though both groups showed significant advances.

The effects of ESDM treatment appear to be more than "skin-deep". A secondary analysis (Dawson et al, 2012) involving EEG measures of social and nonsocial preference for static photos revealed that those children from the 2010 study who had received two years of intensive ESDM showed normalized brain activity in response to looking at photos of people and of toys when compared to a typically developing control group. Both of these groups showed preference for social pictures over toy pictures. The Nc component from the prefrontal cortex and anterior cingulate cortex, which is usually used to reflect attention engagement with stimuli, was used to measure latency and intensity of visual attention. Both a shorter Nc latency and increased cortical activation were found when viewing photos of faces (social) among the both ESDM group and typically developing children, revealing faster and stronger responses to faces than to objects. However, the children in the Community treatment groups demonstrated the opposite pattern: faster and stronger responses to objects than to faces (Dawson *et al.*, 2012).

There are also supportive data from other methods of delivering ESDM. Parent-implemented ESDM (P-ESDM) has been examined in a series of single subject and group designs (Estes *et al.*, 2014; Rogers *et al.*, 2012; Rogers, et al, 2018; Vismara, McCormick, Young, Nadhan, & Monluz, 2013). In order to disseminate ESDM to children as early as possible after diagnosis, Vismara, Rogers, and Colombi (2009) developed a series of procedures for coaching parents to implement ESDM strategies during everyday activities at home. This low-intensity treatment (1-1.5 hour sessions, once or twice a week, for 12 weeks) has yielded a series of papers demonstrating parents' rapid learning of ESDM techniques, children's acquisition of language, social, and play skills, and most importantly, the positive relationship between parent mastery of ESDM techniques and child learning (Rogers et al, 2018). In addition, a randomized controlled trial of 12 P-ESDM coaching sessions compared to Community treatment revealed two important findings. First, both groups of children showed similar rates of acceleration in all developmental areas measured (Rogers et al, 2012), even though the Community group received, on average,

three times the number of intervention hours as did the P-ESDM children. Second, parents from the P-ESDM group reported stronger alliances with their therapists (Rogers *et al.*, 2012) and lower parental stress than parents in the Community treatment group (Estes *et al.*, 2014) Thus, parents can implement ESDM at home, and the skill with which they use the techniques appears to influence their children's learning rates (Rogers et al, 2018). This is good news for all the families who cannot receive more intensive services for their children. However, the extent to which parent implementation can approach the same outcomes as intensive delivery is unknown.

The third delivery approach to be studied is community-based group ESDM (G-ESDM), developed and tested by Vivanti, Dissanayake, Zierhut, Rogers, & Victorian ASELCC Team (2013) at the University of LaTrobe and by Fulton , Eapen, Črnčec, Walter, & Rogers

(2014) in Sydney, Australia. Vivanti's group implemented 15 to 25 hours of group-based ESDM, with a staff-child ratio of 1:3, for 21 preschoolers for one year. The group-based ESDM followed the procedures detailed by Rogers and Dawson (2009), but activities were modified to offer more opportunities for peer interaction. A pre-post test revealed that children whom received the group-based ESDM, on average, gained improvement that is equal to 10 months, 5.5 months, and 8 months of development in visual reception domain, fine motor domain, and receptive and expressive language respectively. Similarly, Fulton and colleagues also provided 15 to 20 hours of group based ESDM with one hour of one-on-one intensive ESDM therapy for almost one year (Fulton *et al.*, 2014). They found that children's early learning abilities measured by the Mullen Scales of Early Learning was improved significantly after treatment, while 68 % of children showed decreased maladaptive behaviors by 12 weeks after the treatment started.

Vivanti and colleagues (2018) took another step to evaluate the effectiveness of group-based ESDM between an inclusive and autism-specific classrooms. This RCT found that group-based ESDM is feasible to be implemented in inclusive education classrooms and no differences in child-related or parent-related outcomes were found between the inclusive or autism-specific classrooms. Not only did the aforementioned studies find positive results in children's development in the same areas as Dawson and colleagues (2010) reported, but they also reported acceptable fidelity when community providers and parents implemented ESDM. (Fulton *et al.*, 2014; Vivanti *et al.*, 2014)

In an effort to reach beyond clinical settings, ESDM scientists have also begun to use telehealth delivery directly with parents. Vismara and colleagues (2018) conducted a randomized controlled trial and delivered 12 weekly 1.5-hr videoconferencing sessions. During the intervention, parents had access to the Parent-Mediated ESDM (P-ESDM) online learning modules and received real-time coaching through telehealth. Thirty-two parents and their children with autism who were between 18 to 48 months old participated in this study. This randomized control study found that, compared to the control group, the parents in the telehealth P-ESDM group significantly used more intervention skills (e.g., management of child attention) and had higher program satisfaction.

While more community-based, telehealth studies are on the way, these studies show that ESDM is able to be adopted by community-based providers and thus has a clear scalability potential (Vismara *et al.*, 2013).

The efficacy data, the theoretical framework, and the accessible published treatment manuals and measures of ESDM have created a great deal of interest in the intervention internationally. Many different research groups have studied ESDM in other locations, including France (Zelmar *et al.*, 2018), Italy (Colombi *et al.*, 2018), and China (Zhou *et al.*, 2018). A meta-analysis completed by Baril and Humphreys (2017) of five ESDM studies between 2009 to 2012, which included a total of 149 children and toddlers, showed that the effect sizes of language, visual reception, fine motor, communication, socialization, daily living skills, and motor skills ranged from small to large across the studies. ESDM is a research-based, comprehensive treatment with a large evidence base. Its effect is particularly large for those who entered the program at an earlier age and who received more intensive ESDM.

ESDM does not only impact the lives of children and parents; it also reduces the societal cost of caring for individuals with ASD. Autism is a complex condition and its services cost $236-262 billion in the U.S. (Buescher *et al.*, 2014), while early identification and treatment is estimated to save up to approximately one million dollars per person across life time (Peters-Scheffer, Didden, Korzilius, & Matson, 2012). Insufficient early intervention imposes an additional $208,500 later educational cost later on (Chasson *et al.*, 2007). Two cost-effectiveness studies of ESDM have been conducted in order to evaluate the economics of implementation at a societal level. In Canada, the ESDM-Parent Delivered Model produced an additional 0.17 dependency-free life years for $8600 less than typical early intervention programs. On the other hand, ESDM-Intensive generated an additional 0.53 dependency-free life years for $45,000 less than typical early intervention programs (Penner *et al.*, 2015). In another study in the United States (Cidav *et al.*, 2017), it was found that even though the cost for ESDM was higher than community-based treatment by about $14,000, there was a significant decrease in cost ($19,000 per child per year) in the post-intervention period for those in the ESDM group as they received fewer services (e.g., ABA/EIBI, occupational therapy, physical therapy, and speech therapy).

Summary and future directions

After 30 years of studies in early intervention for children with ASD, it appears that sufficient evidence is now accumulating to support the use of ESDM and of NDBIs in general as a viable alternative to discrete trial teaching approaches and ABA-only based treatments. Among the next steps of NDBI related research, two in particular stand out: (1) the need to understand the active ingredients of such interventions and (2) the need to conduct implementation/

dissemination studies of such services to determine their impact when delivered in typical communities by community providers (Schreibman *et al.*, 2015). While a number of such studies are in progress, the dissemination and implementation of effective treatments does not solely rely on programmatic-level improvement, but also on a structural- and societal-level leap in terms of commitment and capacity to deliver high quality, evidence-based early education programs for all children with disabilities and developmental risk, and also to those without such risks to their outcomes. Access to services is often compromised by such factors as race (e.g., African American & Latino), low social economic status (Liptak *et al.*, 2007), non-metropolitan areas (Thomas *et al.*, 2007), insurance types (Wang, Mandell, Lawer, Cidav, & Leslie, 2013), a shortage of professionals (Murphy & Ruble, 2012), and parental advocacy. Compared to caregivers of children with other developmental disabilities and/ or mental health issues, those caregivers of children with ASD experienced more difficulty using services, insufficient insurance coverage/ financial support, and lack of care coordination (Vohra, Madhavan, Sambamoorthi, & St. Peter, 2014). Without structural-and societal-level changes in the commitment and the capacity to deliver high quality early childhood education, the benefits of ESDM and other effective intervention approaches will be unable to find a way to families in need. Intervention supports are needed that can be provided directly to parents using easy to access formats and learning approaches, to help provide some help for children and families who cannot access professional intervention services. Policies that allow public funding and service infrastructure are as critical to the wide-spread implementation of high-quality early intervention (Cernius, 2016) as are ongoing research studies that demonstrate culturally-adapted NDBI intervention approaches that fit within the economic service and delivery structure of various nations. Such structural changes and resource development activities necessitate collaboration among individuals with ASD, family members, advocates, professionals, researchers, governmental agencies, and policy makers. Efficacy data showing the power of early intervention to help young children with ASD and the rapid flow of autism-related information across the globe are mobilizing parents, professionals, and policy makers to improve delivery of efficacious early intervention to young children with ASD and their families worldwide.

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