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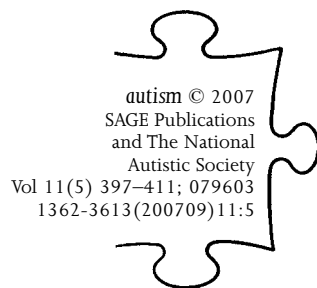
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Evaluation of the Relationship Development Intervention Program



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ABSTRACT This study is the second in a series evaluating the effectiveness of Relationship Development Intervention (RDI) to address unique deficits inherent in autism spectrum disorders. RDI is a parent-based, cognitive-developmental approach, in which primary caregivers are trained to provide daily opportunities for successful functioning in increasingly challenging dynamic systems. This study reviewed the progress of 16 children who participated in RDI between 2000 and 2005. Changes in the Autism Diagnostic Observation Schedule (ADOS) and Autism Diagnostic Interview-Revised (ADI-R), flexibility, and school placement were compared prior to treatment and at a minimum 30 month follow-up period. While all children met ADOS/ADI-R criteria for autism prior to treatment, no child met criteria at follow-up. Similar positive results were found in relation to flexibility and educational placement. Generalizability of current findings is limited by the lack of a control or comparison group, constraints on age and IQ of treated children, parent self-selection, and parent education conducted through a single clinic setting.

KEYWORDS

autism;
caregiver
training;
Relationship
Development
Intervention

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Introduction

Autism is widely perceived as a heterogeneous disorder, impacting each individual in different ways across language use, behavior, and intelligence. Despite this heterogeneity, there are certain aspects of the disorder which are universally present in the autism spectrum (Bacon et al., 1998; Charman et al., 1997; Hobson, 1993; Mundy and Crowson, 1997). Measures of experience sharing and flexible-adaptive thinking strongly discriminate between affected and non-affected samples (Bacon et al., 1998; Mundy and

Crowson, 1997; Osterling et al., 2002). Experience sharing refers to dynamic social-emotional and social-cognitive abilities, employed for sharing and integrating unique experiences with others (Hobson, 1993; Sigman and Ruskin, 1999; Trevarthen, 2001). Joint attention, sharing enjoyment with others, conjoint pretend play, declarative communication, social referencing, and perspective taking are examples of experience-sharing abilities found to be deficient in individuals on the autism spectrum (Baron-Cohen et al., 1996; Gutstein, 2001; Lord, 1995; Osterling and Dawson, 1994).

This study examines the effectiveness of a cognitive-developmental parent-training model, Relationship Development Intervention (RDI) (Gutstein, 2001), and is a follow-up to a preliminary evaluation study (Gutstein, in press). In that study, children diagnosed with an autism spectrum disorder who participated in RDI demonstrated a significant reduction in experience-sharing related symptoms, as measured by the Autism Diagnostic Observation Schedule (ADOS: Lord et al., 2002). Following an average of 16 months of treatment, over 50 percent of children in the study no longer met ADOS criteria for autism spectrum disorders. In contrast, a comparison group of children treated with more traditional intensive intervention methods failed to show significant improvement in these core deficit areas, despite receiving up to five times more therapist involvement than the RDI group. The children in the RDI group were also significantly more likely to function without support in typical classrooms.

The current study examines the durability of these changes and provides a more thorough evaluation of the effectiveness of RDI. Participants were evaluated prior to treatment and again after a minimum of 30 months in RDI; many received more than two follow-up evaluations. Evaluations included (1) the Autism Diagnostic Interview-Revised (ADI-R: Rutter et al., 2003), (2) the ADOS, (3) a measure of parental perception of their child's flexibility and adaptation, and (4) evaluation of school placement. The major hypothesis proposed that the majority of children participating in RDI would demonstrate a decrease in autism-related symptoms to the extent that they no longer met combined ADOS/ADI-R diagnostic criteria. We also hypothesized that children treated with RDI would be rated by primary caregivers as highly flexible and adaptive and have a low incidence of special education placement. Additionally, we hypothesized that improvement would not be related to IQ, concurrent treatments, or age at treatment onset.

Relationship Development Intervention

RDI attempts to address the distinct patterns of perceptual, cognitive, and emotional difficulties unique to individuals on the autism spectrum (Gutstein, 2001; Gutstein and Sheely 2002a; 2002b). The primary agents

of change in RDI are the child's parents, who begin their training by attending 6 days of intensive workshops in the theory, principles, and components of RDI, followed by thorough planning and regular weekly or biweekly consultation meetings with a certified RDI consultant. Meetings include progress updates, discussion of goals, program planning, and review of specific videotaped segments of primary caregivers working with their child. Parents and children participate in intensive re-evaluation approximately every 6 months.

At the core of the program, parents learn how to perceive and scaffold opportunities for their child to respond in more flexible, thoughtful ways to novel, challenging and increasingly unpredictable settings and problems. Parents are trained to incorporate these opportunities into their lifestyle, so that each day involves frequent, carefully framed opportunities presented seamlessly into the child's routine.

Method

Subjects

Data were collected from chart reviews of children whose families initiated RDI at the Connections Center in Houston, Texas, the clinic where RDI was developed, between January 2000 and May 2003. Children included in this study met the following criteria: (1) an interval of at least 30 months between initial and follow-up testing, (2) a previous diagnosis of autism, Asperger syndrome, or PDD-NOS, (3) participation in the RDI protocol, (4) age at time of RDI initiation between 20 and 96 months and (5) pre-treatment IQ score of at least 70. Sixteen children met these criteria.

Pre-treatment information is summarized in Table 1. At time of treatment onset, ages ranged between 21 and 94 months, while gender was overwhelmingly male (15:1). Five children met diagnostic criteria for autism, while seven were diagnosed with Asperger's disorder and four with PDD-NOS. Secondary diagnoses included language disorder (eight), ADHD (five), bipolar disorder (one), and food allergies (one). IQ scores ranged from 70 to 118, placing this population in the intellectually 'high-functioning' segment of the ASD population.

Regarding pre-RDI treatment history, one child participated in treatment at our clinic prior to 2000 (the year that RDI was systematized). Five of the 16 had participated in biomedical treatment and 12 subjects had participated in behavioral interventions prior to RDI.

In terms of educational settings, children were placed into one of four categories: (1) mainstream classroom placement with no special services required, (2) mainstream with pullout services, where the child spent parts

Table 1 Subject characteristics at treatment onset

Months in RDI	median: 41.5	range: 33–79
IQ standard score	mean (SD): 90.50 (13.23)	range: 70–118
Initial age (months)	mean (SD): 60.50 (20.43)	range: 20–94

<i>Diagnosis</i>	<i>Frequency</i>
<i>Primary diagnostic category</i>	
Autism	5
Asperger's disorder	7
PDD-NOS	4
<i>Secondary diagnoses</i>	
Language delays	8
ADHD	5
Bipolar disorder	1
Food allergy	1
<i>Initial educational placement</i>	
Mainstream full-time	1
Mainstream with pullout services	1
Mainstream, partial special education	4
Special education or full-time aide	8
Not yet school age	2

of each day in a resource room to help with non-behavioral issues, such as reading, (3) partial special education in mainstream placement, with the child spending part of each day in a special education environment due to behavioral and adaptational concerns, and (4) full-time special education placement due to difficulties in behavior and adaptation, or placement in a typical class with a full-time aide. Two children were not yet school-aged at treatment onset.

Evaluation measures

Test administrators completed ADI-R and ADOS training at the University of Michigan, a training site for research use of the instruments. To control for bias effects, the same administrator was never used twice in testing a single child.

Autism Diagnostic Observation Schedule (ADOS) The Autism Diagnostic Observation Schedule (ADOS: Lord et al., 2002) is considered the 'gold standard' in distinguishing individuals with autism from other populations (Lord et al., 2002; Robertson et al., 1999). The ADOS appears to be highly

stable and relatively unaffected by age or time (Lord et al., 2002; McGovern and Sigman, 2005) and has been used in studies of treatment effectiveness (Belsito et al., 2001; Owley et al., 2001). The ADOS provides subscale scores on four dimensions, as well as a summary diagnostic rating of autism, autism spectrum, or non-autism. Scores of 0 represent no impairment, while scores of 1 indicate some impairment and scores of 2 indicate significant or severe impairment. The variables of particular interest in this study were the diagnostic subscales (communication and social interaction) which strongly correlate with core social-emotional difficulties of autism, intersubjective engagement and changes in diagnostic classification (Robertson et al., 1999; Tanguay et al., 1998). The ADOS was administered after an average of 15.33 months ($SD = 6.63$) and again after an average of 21 months ($SD = 7.73$). ADOS inter-rater reliability was computed by comparing ratings of two trained raters, both of whom were not involved in the child's care. Both raters had extensive training in the ADOS. Intra-class correlation (ICC) was utilized to compute reliability (Shrout and Fleiss, 1979) and was found to be very good ($\alpha = 0.86$).

Autism Diagnostic Interview—Revised (ADI-R) The Autism Diagnostic Interview—Revised (ADI-R) is a diagnostic interview for autism, developed as a complement to the ADOS. The ADI-R examines 'qualitative abnormalities in reciprocal social interaction', 'qualitative abnormalities in communication', and 'restricted, repetitive, and stereotyped patterns of behavior' in the past and at the time of the interview. As with the ADOS, scores of 0 represent no impairment, 1 some impairment, and 2 significant impairment. The interrelationship between the ADI-R, the ADOS, and the *Diagnostic and Statistical Manual of Mental Disorders* fourth edition (DSM-IV-TR) has been found to be good (de Bildt et al., 2004). The ADI-R has also been used successfully as a change measure (Fecteau et al., 2003; Howlin et al., 2004; McGovern and Sigman, 2005), and has shown excellent inter-rater reliability (Hill et al., 2001). The communication, social interaction, and repetitive behaviors of the 'current' domain served as outcome measures. The mean scores for the social and communication domains were utilized in order to control for variance due to age of subject.

Critical ADI-R/ADOS items Thirteen items determined to be most representative of experience sharing were selected from the ADI-R and ADOS. Table 2 provides a summary of these critical items.

Flexibility interview This semi-structured interview, developed by the authors, comprises 10 items related to the child's ability to adapt to change and transition. Parents rated the flexibility of their child across five

Table 2 ADOS/ADI-R items selected for analysis of experience sharing

ADI-R	
1	Peer relationships
2	Range of non-verbal behavior
3	Shared enjoyment
4	Social/emotional reciprocity
5	Reciprocal conversation
ADOS	
1	Conversation (modules 2 and 3 only)
2	Facial expression
3	Gesture
4	Imagination/creativity
5	Intonation
6	Quality of social overtures
7	Shared enjoyment
8	Spontaneous initiation of joint attention (modules 1 and 2 only)

categories: 'completely rigid', 'some difficulty', 'somewhat flexible', 'mostly flexible', and 'age appropriate' flexibility. For purposes of this study, changes in 'age appropriate' ratings were used. The items included seven related to adaptation and three related to flexible thinking.

Educational placement Educational placement is a frequently used measure of intervention effectiveness (Lovaas, 1987; Smith et al., 2000). In this study, educational placement was ascertained through parent and teacher interview. Along with the four criteria mentioned earlier, a fifth category – home schooling – was added to include families who, following involvement in RDI, chose to provide home schooling for their children

Results

ADOS

In the initial study, the RDI group demonstrated significantly greater improvement in ADOS diagnosis after an average 18 months in treatment, with 70 percent of children in the RDI group improving at least one diagnostic category, while no children in the non-RDI treatment group improved.

In the current study, the data were examined using both a qualitative approach, based on changes in ADOS diagnostic category, and a quantitative approach, analyzing changes in ADOS scores. Initial ADOS scores were available for 12 of 16 children and follow-up scores were available for all

16 children. Prior to treatment, 10 children received an initial ADOS 'autism' rating, while two received an 'autism spectrum' rating. After a median of 41.5 months in treatment, no child met ADOS criteria for an autism diagnosis, six children met criteria for autism spectrum, and 10 children were rated in the 'non-autism' diagnostic category. Five children initially rated in the 'autism' category achieved a 'non-autism' rating. Figure 1 summarizes these changes.

A repeated measures analysis of variance (ANOVA) was conducted, with ADOS score as the within-subjects factor, to examine the stability of the improvement in ADOS social interaction and communication score changes over time. Table 3 presents a summary of these findings. First, the ADOS communication domain total score was examined. The results indicated a significant time effect, Wilks's $\Lambda = 0.165$, $F(2, 10) = 35.4$, $p < 0.0001$. Follow-up contrasts indicated a significant linear effect between ADOS 1 and 2, $t(11) = 5.1$, $p < 0.0001$, but not between ADOS 2 and 3, $t(15) = 1.51$, $p = 0.152$. These findings suggest that initial improvements made in the first year of treatment were maintained over time. Next, the

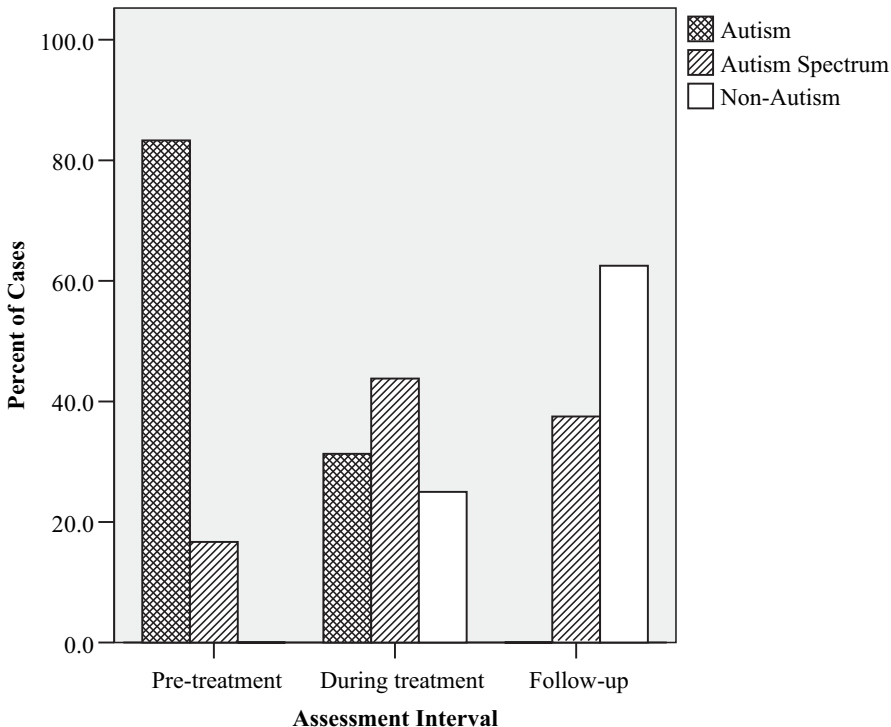


Figure 1 Percentage of cases in each ADOS diagnostic category, pre-treatment and follow-up

Table 3 Means and standard deviations of ADOS scores

	<i>N</i>	<i>Min.</i>	<i>Max.</i>	<i>Mean</i>	<i>SD</i>
ADOS 1 communication score	12	2	10	5.92	1.975
ADOS 2 communication score	12	0	10	2.58	2.28
ADOS 3 communication score	12	0	4	2.67	0.99
ADOS 1 social interaction score	12	3	13	9.58	2.968
ADOS 2 social interaction score	12	1	13	5.83	3.27
ADOS 3 social interaction score	12	0	9	4.17	2.44

ADOS social interaction domain total score was examined. The results indicated a significant time effect, Wilks's $\Lambda = 0.34$, $F(2, 10) = 9.8$, $p < 0.004$. Follow-up analyses indicated a significant declining trend over time, ADOS 1 and 2 $t(11) = 3.9$, $p < 0.01$ and ADOS 2 and 3 $t(15) = 2.8$, $p < 0.05$, suggesting that children continued to make significant improvement in social interaction. Figure 2 presents findings in the social interaction and communication domains.

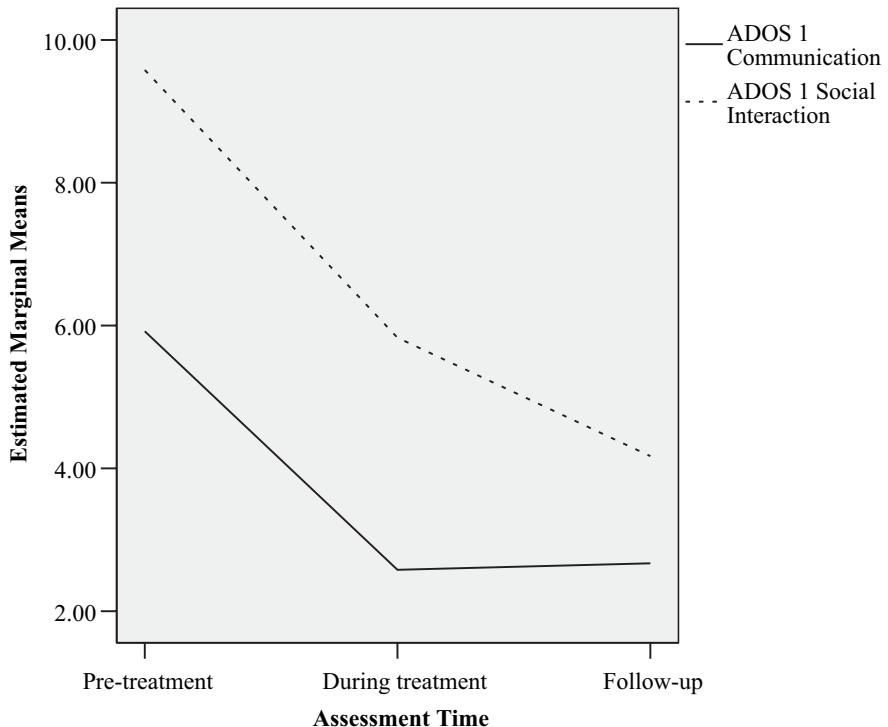


Figure 2 Results for repeated measures ANOVA in ADOS communication and social interaction domain scores

ADI-R

To examine changes in ADI-R algorithms, the scores for the communication and social interaction domains were summed (C + S) and a mean score for each child was determined. Pre-treatment, mean ADI-R C + S score was 10.6, with a minimum score of 7.9 and a maximum of 14.3. At follow-up, scores fell to a mean score of 2.4, with a minimum of 0 and a maximum of 5.7. As with the ADOS, the difference between pre-treatment and follow-up ADI-R scores was dramatic and significant, $t(12) = -19.2$, $p < 0.0001$). Figure 3 provides a summary of changes.

ADI-R and ADOS critical items

Table 2 presents the items which were selected from the ADI-R and ADOS. Analysis of the ADI-R experience-sharing items indicated a dramatic improvement in scores. Prior to treatment, only 17 percent of children received a 0 score on the ADI-R critical items, while at follow-up two-thirds of children earned a 0 score. Prior to treatment, 54 percent of children received a 2 score on the ADI-R while at follow-up only 5 percent earned a score of 2, suggesting a dramatic improvement in functioning as rated by parents.

On the ADOS, prior to treatment only 13 percent of subjects received a 0 score while 32 percent received a 0 at follow-up. Finally, 53 percent received 2 scores prior to treatment while at follow-up this number decreased dramatically to 11 percent, suggesting significant improvement in observed functioning which was consistent with the parent ratings. Figure 4 presents these findings.

Flexibility

The percentage of children falling in the 'age appropriate flexibility' category as rated by their primary caregiver is presented in Table 4. The average percentage of age appropriate flexibility ratings increased from a pre-treatment mean of 16 percent to 71 percent at follow-up. The greatest change was uncovered in three items, 'unexpected change to familiar routines', 'unexpected omission of a routine activity', and 'changes to activities without preparation' (81%, 88% and 81%, respectively), followed by 'anticipating an event and encountering another', and 'unexpected actions by familiar people' (both 75%). Twelve children improved from the least flexible two categories (completely rigid, some difficulty) to the most flexible two categories (mostly flexible, age appropriate) in at least one area.

Educational placement

Prior to treatment, more than half of the children attended special education classrooms while four were in partially mainstreamed classes. Only two participants attended mainstream classrooms without an aide

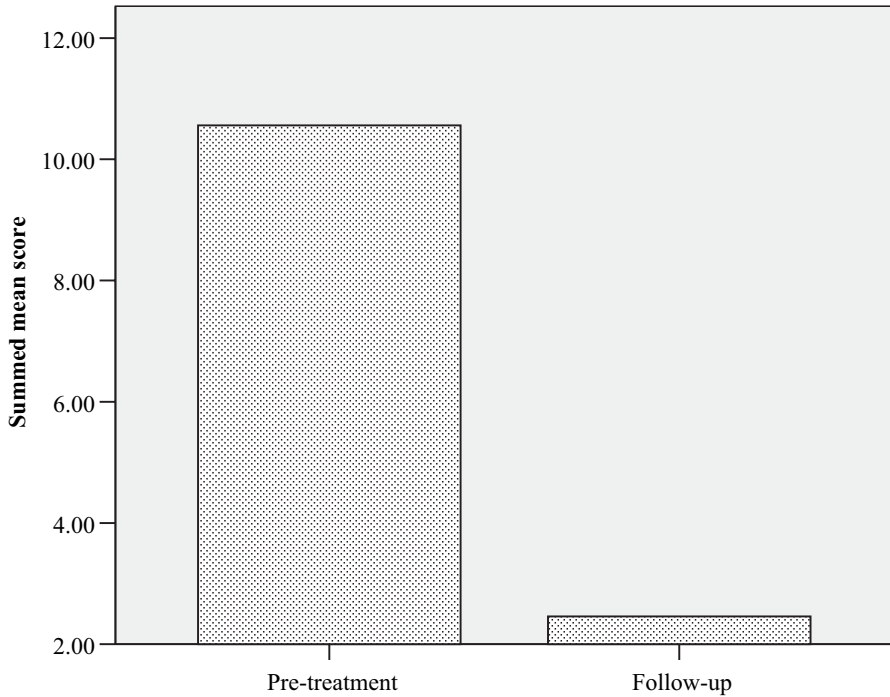


Figure 3 Comparison of ADI-R summed domain scores

Table 4 Percentage of participants rated as demonstrating ‘age appropriate flexibility’ by item

	<i>Pre-treatment</i> %	<i>Follow-up</i> %
Unexpected change to familiar routines	12.5	81.3
Unexpected omission of a routine activity	18.8	87.5
Changes to activities without preparation	25.0	81.3
Anticipating an event and encountering another	31.3	75.0
Unexpected actions by familiar people	25.0	75.0
Interruption during a highly favored activity	6.3	68.8
Stopping a task before it is finished	12.5	68.8
Planning for things that might go wrong	00.0	43.8
Adapting when original plans don't work out	18.8	75.0
Using familiar objects in a novel way	12.5	56.3
Grand mean	16.3	71.3

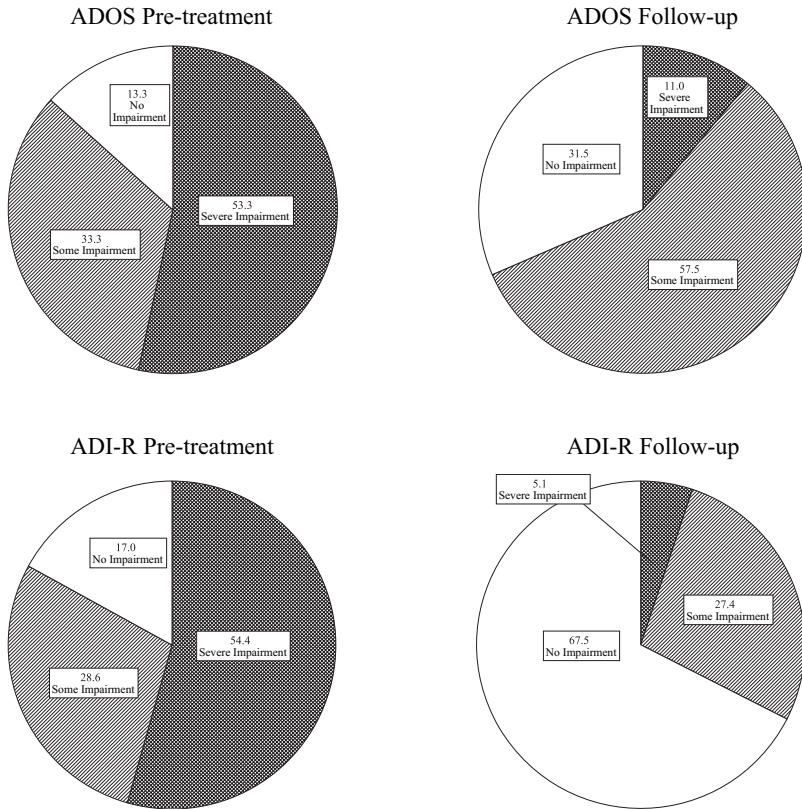


Figure 4 Percentage of subjects in each descriptive category on ADI-R and ADOS critical items

prior to treatment. At follow-up, 10 of 16 children were functioning in mainstream settings without an aide (categories 3 and 4), while only one child remained in a special education classroom. Table 5 and Figure 5 present these findings.

Table 5 Percentage of participants in each educational placement category

Category	Pre-treatment %	Follow-up %
1 Special education	57.1	12.5
2 Partial mainstream	28.6	18.8
3 Mainstream with pullout	7.1	31.3
4 Mainstream	7.1	31.3
5 Home school	0	6.3

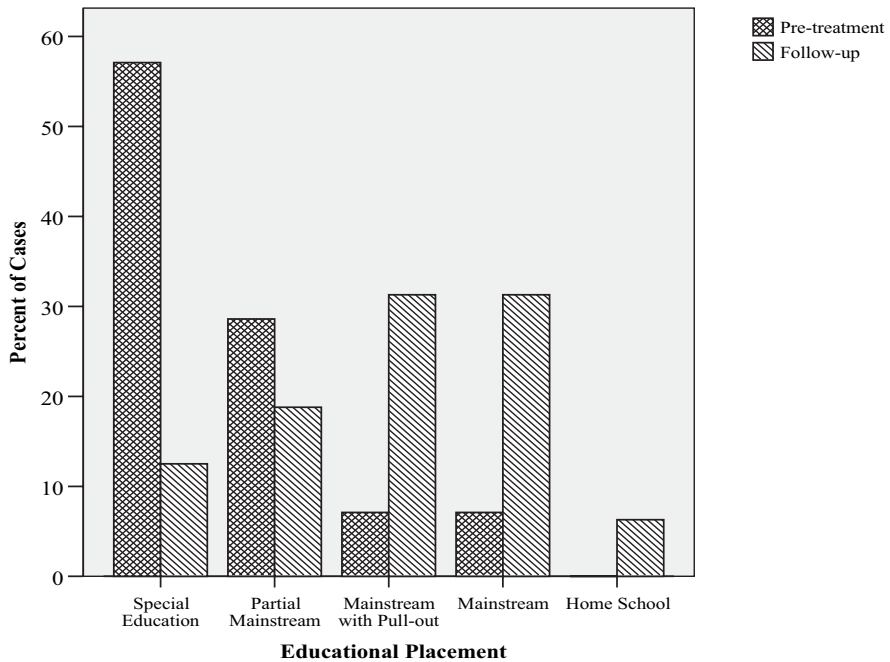


Figure 5 Educational setting prior to treatment and at follow-up

Factors impacting treatment success

To determine whether IQ score was related to outcome, initial IQ score (IIQS) was compared to ADOS change scores in the combined communication and social interaction score (C + S) ($n = 13$, $r = -0.01$, $p = 0.98$), and ADI-R combined change scores (C + S) ($n = 13$, $r = -0.08$, $p = 0.78$). No significant correlations were uncovered. Similarly, age at onset of participation in treatment was not significantly correlated with ADOS or ADI-R change scores (ADOS $n = 13$, $r = 0.24$, $p = 0.4$; ADI-R $n = 13$, $r = 0.21$, $p = 0.73$).

With regard to prior and concurrent treatments, no significant correlation was found between amount of behavioral treatments prior to (ADOS $n = 13$, $r = 0.27$, $p = 0.39$; ADI-R $n = 13$, $r = 0.35$, $p = 0.23$) or concurrent with treatment and treatment outcome (ADOS $n = 13$, $r = -0.41$, $p = 0.11$; ADI-R $n = 13$, $r = 0.34$, $p = 0.26$). Similarly, no significant relationship was found between amount of previous biomedical treatment and outcome (ADOS $n = 12$, $r = -0.10$, $p = 0.75$; ADI-R $n = 13$, $r = 0.17$, $p = 0.58$) or amount of biomedical treatment during treatment and outcome (ADOS $n = 12$, $r = 0.12$, $p = 0.70$; ADI-R $n = 13$, $r = 0.16$, $p = 0.60$). Similar non-significant relationships were found between prior or concurrent amount and type of non-RDI treatment and school placement and flexibility ratings.

Discussion

As in our initial evaluation, current results support RDI as a promising program for remediating critical experience-sharing difficulties of children on the autism spectrum. Children who participated in RDI became significantly more socially related, engaged in more reciprocal communication, functioned in school settings with less adult participation, and also were perceived by parents as behaving in a dramatically more flexible and adaptive manner.

The current study strengthens initial findings in several ways. Data were obtained from observation (ADOS), objective findings (school placement) and parent perception (ADIR and flexibility). Results from all three perspectives are consistent. Not only did the children demonstrate dramatic gains in functioning, but those gains remained stable, with children retaining their improvement for an average of over 3 years. Parental perceptions of their child's increased flexibility and adaptive functioning were consistent with their improved communication and social interaction.

Involvement in the RDI program appeared to be the primary factor responsible for improvement. The majority of families in this study were involved only in RDI during the treatment time period. Participation in other interventions, either prior to or during RDI, was not significantly related to improvement.

The most significant limitation of the study is the lack of a control or comparison group. There is an urgent need for independent clinical researchers to conduct neutral evaluation of the relative efficacy of the most commonly employed intervention methods.

While IQ scores were not found to relate to treatment success, evaluation did not include children with more severe measured cognitive difficulties. Similarly, while age and outcome were not related, we did not study children who were older than 9 years when treatment was initiated. However, the fact that children of elementary ages appeared to make as much improvement as younger children is an important finding that we hope to extend to older children in the future. We are currently analyzing a larger sample that is more diverse in IQ and age. A final factor limiting generalization of these findings is that they are restricted to families treated at The Connections Center, the primary training and development site for RDI. The sample of families is therefore somewhat 'self-selected', as they deliberately sought out RDI treatment and were already committed to the model. Research is needed to evaluate the ability of the program to be 'ported' to other sites with clients who may have less initial commitment.

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