Number and Controllability of Reinforcers as Predictors of Individual Outcome for Children with Autism Receiving Early and Intensive Behavioral Intervention:

A Preliminary Study

There is growing interest in Early and Intensive Behavioral Intervention (EIBI) as a treatment for children with autism. A EIBI program is constituted by a large number of techniques based on learning principles from Applied Behavior Analysis (ABA) that are used with a high intensity (typically more than 20 hours per week) with children under the age of 5 years (Eikeseth, 2009). The aim of EIBI is to teach the child adaptive skills such as verbal behavior, independent and social play, everyday functioning such as dressing, eating and personal hygiene, and any other behavior that is lacking in the child's behavioral repertoire but is necessary for an independent life.

Effects of EIBI

A large number of studies have investigated the therapeutic effects of EIBI. One meta analysis have reported effect size for Adaptive Behavior and IQ to be 0.66 and 1.09, respectively (Eldevik et al., 2009). A meta analysis with more generous inclusion criteria (Virués-Ortega, 2010) reported an effect size on adaptive behavior to 0.81 and for IQ to be 1.31.

Even if the scientific support for EIBI as a treatment for children with autism appears solid, the scientific quality of published studies varies greatly, making an exact assessment of treatment efficacy and effect size difficult. According to a review by Eikeseth (2009), only one properly randomized controlled trial has been published to date (Smith, Groen, & Wynn, 2000), evaluating EIBI for children with autism and PDD-NOS. This study included both independent assessment in pre- and post-tests and randomized assignment to an ABA-treatment (n=15) or a

parent training control group (n=13). Although the two groups did not differ significantly at intake, after two years of treatment the EIBI group scored significantly higher on IQ, visual–spatial skills, language and school placement. The groups, however, did not differ significantly in measures of adaptive functioning. It is worth noting that in this study, children with PDD-NOS exhibited larger gains in IQ than children with autism.

Notable in all published outcome studies for EIBI is the large individual variation in outcome (Eldevik et al., 2010). Some children benefit greatly from treatment, while some children seem to gain little or nothing at all. For example, Hayward, Eikeseth, Gale, and Morgan (2009) found that 50% of children receiving one year of EIBI gained 15 IQ points or more, 39% showed a gain of less than 15 IQ-points, 2 % had a stable IQ; and 9 % showed a decrease. Note that a decrease in IQ-point does not necessarily reflect decrease in actual functioning but could be an effect of applying age norms for older children.

Predictors of individual outcome

Several explanations for the variation in individual gains have been suggested. For instance, treatment factors such as intensity of supervision (Eikeseth, Hayward, Gale, Gitlesen, & Eldevik, 2009), intensity of treatment (Eldevik et al., 2010) and therapist allegiance to the treatment (Klintwall, Gillberg, Bölte, & Fernell, 2011) have been linked to treatment outcome. Also, high levels of parental stress have been reported to be detrimental to treatment effects (Osborne, McHugh, Saunders, & Reed, 2008).

Perhaps more importantly, different child characteristics have been found to be beneficial for individual outcome. A low age at start of treatment has been reported as a beneficial factor (Fenske, Zalenski, Krantz, & McClannahan, 1985), as have level of social interest (Sallows, Graupner, & MacLean Jr, 2005). In a study by Ben-Itzchak and Zachor (2007), contrasting

children with high and low IQ at intake, children with high intellectual functioning was reported to have better outcomes in treatment. In an interesting study by Stoelb, Yarnal, Miles, Takahashi, Farmer, and McCathren (2004), children with dysmorphic features was found to gain less from treatment than children without such characteristics.

Even though these individual reports are interesting, they might be spurious. A recent study conducted by Eldevik et al. (2010) analyzed outcome for 309 children receiving EIBI with a multiple regression including age at intake, IQ at intake, autism severity at intake and intensity of treatment. Of these putative predictors, intervention intensity was the only one that independently predicted gains in both IQ and adaptive functioning. Age at intake did not predict IQ gains or gains in adaptive behavior. Intake IQ and intake adaptive behavior predicted gains in adaptive behavior.

Further exploration of child characteristics that may influence treatment outcome is a priority because there is need to improve treatment techniques to help those children who show little or no gains from present applications of EIBI.

Reinforcement

In EIBI, *reinforcement* is a fundamental treatment principle. Reinforcement is used in at least three different ways: Firstly, it is used to reduce aberrant behavior using differential reinforcement and noncontingent reinforcement procedures (Carr, Severtson, & Lepper, 2009). Secondly, it is used to motivate appropriate behavior, for example, by keeping favorite items out of the child's reach so that the child can request them to access them (Maurice, Green, & Foxx, 2001). Thirdly, reinforcement is used during teaching to facilitate acquisition of important skills such as communication, play, academic, social and daily living skills (Lovaas, 2003).

Even in the natural environment, reinforcement controls behavior, development and skill acquisition. For example, a child may request a favorite item by asking, pointing or using the adult's hand as a tool, or tantrum to escape from an unpleasant situation (such as a classroom). Even stereotyped behavior has been shown to be maintained by reinforcement (Lovaas, Newsom, & Hickman, 1987; Rapp & Vollmer, 2005; Rincover, 1978; Rogers & Ozonoff, 2005). In the case of stereotyped behavior, the reinforcing stimuli maintaining the behavior may be the sensory and/or perceptual stimuli produced directly by the behavior itself (Lovaas et al., 1987). For example, body rocking may be reinforced by the proprioceptive stimuli this behavior produces. Skinner (1965) termed such sensory and/or perceptual reinforcers automatic reinforcers, in contrast to social reinforcers (a better term is socially mediated reinforcers, as to not confuse it with social stimuli such as smiles and verbal praise, which may or may not be a reinforcer for a specific child), where, for example, the child is naming a favorite object to gain access to it. This latter is a socially mediated reinforcer because the reinforcer (i.e., the favorite object) is given to the child by another person. In the case of stereotyped behavior, the reinforcer is produced directly by the behavior itself, and hence the term automatic reinforcement.

By engaging in stereotyped behavior, a child produces automatic stimuli which might be reinforcing in themselves. These reinforcers would then compete with the socially mediated reinforcers presented by the therapists and parents, and hence may hamper development of communication, social and other important skills (Cunningham & Schreibman, 2008; Rapp & Vollmer, 2005). A child for whom a large number of socially mediated reinforcers are effective may, in contrast, be more engaged in learning because of the availability of highly potent reinforcers in the teaching environment. Also, the child's behavior may be influenced to a wider range of reinforcers overall, reducing the likelihood of satiation (Egel, 1981).

It is worth noting that stereotypic behaviors need not be maintained by automatic reinforcement. On the contrary the majority seem to be maintained by socially mediated reinforcers such as escape from demands and attention (Cunningham & Schreibman, 2008). However, even if a stereotypic behavior is found to be maintained by socially mediated stimuli the acquisition of the behavior may have started with automatic reinforcers. A behavior which first appears with one function may then shift or add functions, i.e. the stereotypic behavior is first developed due to automatic reinforcers and then the socially mediated function is added to that. In either way, the child was once reinforced by the automatic stimulus produced by the behavior. Note that all children and adults engage many behaviors to produce automatic reinforcement; it is not an "autistic feature" in itself.

In summary, socially mediated reinforcers are any reinforcing stimuli which likely are delivered to a child by other people. Examples of this are food, verbal praise, soap bubbles, hugs, or smiles. Being able to communicate improves the child chances of gaining access to these stimuli. Children with autism are often reported to be reinforced by fewer stimuli, i.e. these children often have a narrow reinforcer repertoire.

In contrast, automatic reinforcers are reinforcing stimuli which the child produces herself, often with very little effort. Examples of this are the proprioceptive feedback from body-rocking, the sight of one's own fingers flapping in front of eyes, the visual distortion from squinting one's eyes or the sound of oneself whistling.

Aim of present study

Based on this analysis, a possible predictor for treatment outcome is the number and type of stimuli, automatic or socially mediated, that function as a reinforcer for the child's behavior. The current study addressed the following questions: Firstly, do children with a larger number of socially exhibit larger gains in EIBI? Secondly, do children with a larger number of automatic reinforcers benefit less from EIBI? Lastly, do these two measures taken together explain even more of the variation in treatment outcome?

Method

The design of the study was a retrospective correlational study, investigating the connection between individual treatment outcome after the first year in treatment, and number and type of reinforcers as measured at anywhere between 6 and 18 months in treatment.

Participants

Twenty-one children (five girls) with a diagnosis of autism were recruited from the Banyancenter EIBI clinic in Stockholm, Sweden. All children at the center were included in the study. One child was excluded from the study due to a diagnosis of Rett syndrome, and three parents declined participation in the study. Two of the children at the center were excluded because the therapist had not been working with the child more than a few weeks and was judged to be unable to complete the questionnaire accurately. The children had received their diagnosis from a state psychiatric outpatient unit and were referred to the clinic for treatment. The mean chronological age at intake was 3 years and 7 months (range: 2 years and 3 months to 4 years and 11 months). Mean Vineland standard score at intake was 64.6 (range 45 to 78), and the scores on the Childhood Autism Rating Scales (CARS), for which scores were available for 17 of the participants, were elevated with a mean of 39.5 (range 29 to 55). Thus, the children displayed significant delays in adaptive behavior and autism classifications on the CARS (Schopler, Reichler, & Renner, 1986). A majority of the children lived in middle-class families, with all except two families being ethnically Swedish. The parents of all children were asked for written consent to participate in the study.

Treatment

All children received EIBI supervised by the clinic and carried out by parents and therapists. All children attended mainstream kindergartens, often being the only child with a diagnosis in the class. An employee at the kindergarten was chosen as a principal therapist. All treatment was carried out by a team consisting of the kindergarten therapist, the child's parents, a supervisor from the clinic and any other significant others such as older siblings and grandparents. The team received weekly or bi-weekly supervision. The intensity of the treatment was aimed to be 30 hours per week, although the actual intensity achieved varied with a mean of 20 hours per week. Supervisors from the EIBI clinic were psychologists, speech pathologists, or paraprofessionals who held a minimum of bachelor's degree in psychology. The supervisors, in turn, were supervised by the second author of this paper, who also saw all participating children. The treatment followed published manuals for comprehensive ABA treatment (Leaf & McEachin, 1999; Lovaas, 2003; Maurice, 1996).

Outcome Measure

The parents of all children were interviewed using the Vineland Adaptive Behavior Scales-II (Sparrow, Cicchetti, & Balla, 2005) at intake and after approximately one year of treatment. These interviews were carried out by the staff at the EIBI clinic and were scored using a computerized scoring system. Norm data for American children were used, as no norm data for Swedish children are yet available. To adjust for variation in intake age and a slight variation in time between measurements, the figure used as an outcome measure was change in age equivalent, computed as follows: The mean age equivalent for all subdomains except "Written" was calculated for all interviews (The written subdomain were excluded as it gives a minimum age equivalent of 2 years and 11 months, which adds irrelevant data to the outcome measure). The change in total age equivalent, divided by number of months in between measurements, was then calculated; yielding a learning rate (zero learning rate meaning that child has shown no progress). This method assures that neither difference in chronological age nor time between measurements adds any irrelevant variation to the outcome measure. All subsequent statistical operations were also carried out using change in Vineland standard scores, with no difference in results obtained. The mean learning rate for the group was 1.08 (slightly more than one developmental year per chronological year), with the expected large variation (range 0.07 to 2.04). Although some of the children had received more than one year of treatment, only outcome data from the first year of treatment were used for this study.

Questionnaire

A novel questionnaire was developed for this study: The Socially Mediated and Automatic Reinforcer Questionnaire (SMARQ; see Appendix 1). This questionnaire was completed by a parent and a therapist after the child had been in a minimum of 6 months in treatment. Note that follow-up outcome data for these children were collected 6 months later.

The questionnaire was constructed to cover both socially mediated and automatic reinforcers. The first page of the questionnaire lists 38 behaviors often reported to be in excess in children with autism (for example "Shaking one's head side to side" and "Spinning or twirling objects or pieces of string"). The items were selected from Lovaas (2003) and out of clinical experience. The presentation any of these behaviors in excess were assumed to be automatically reinforced (at least to some extent). The items were scored as either present or not present, with a cut-off frequency of "at least once every day". Even though such binary scoring of the frequency of the stereotypic behaviors is obviously a simplification of the data, this is necessary to be able to add the number of stereotypic behaviors (and assumed reinforcers) together. The second page of the questionnaire lists 26 stimuli that are commonly used as reinforcers for young children in EIBI (for example "Candy, chocolate or ice-cream" and "soap bubbles"). The respondents were asked to indicate whether or not the child "enjoys gaining access to" each stimulus. This questionnaire was completed by both a parent of the child and the principal therapist in the kindergarten, independently. Only items on which the parent and therapist independently agreed were scored as functioning as reinforcers for the child's behavior in question. The questionnaire does not differentiate between different reinforcing strengths of the stimuli, which is a gross simplification. In reality, stimuli that have been scored as reinforcers will very likely be very different in their ability to reinforce the behavior of the child. However, such binary scoring is necessary to be able to add the reinforcers up to one single number.

It is important to note that the questionnaire presented in this article should not be viewed as indicating some underlying latent variable ("reinforceability"). On the contrary, the items in the questionnaire define the total score. Thus, the SMARQ should be viewed as an example of a *formative model* (Borsboom, Mellenbergh, & Van Heerden, 2003).

Psychometrics of the Questionnaire

All items in the questionnaire were checked for inter-rater reliability. For this analysis, SMARQ-questionnaires from 4 children without outcome data was included. Items for which the inter-rater reliability was deemed unacceptable were excluded from further analysis, the cut-off for which was arbitrarily set at a minimum of Cohen's Kappa at 0.3 (admittedly a rather low level). Cohen's Kappa is preferable to percent agreement as the frequency of positive answers vary greatly between the different items in the questionnaire. Applying this rule, 2 of the socially mediated and 8 of the automatic reinforcers were excluded. Since no one of the children scored present on 7 of the items for automatic reinforcers, these also were excluded from further

analysis (i.e. inter-rater agreements). The mean Cohen's Kappa was 0.59 for socially mediated reinforcers and 0.52 for automatic reinforcers (moderate agreement). As noted above, items were only scored as a reinforcer if parent and therapist both had designated it as such. This was an extra precaution to ensure validity, i.e. that the stimuli is indeed a powerful reinforcer for the child, even across different settings.

The mean number of socially mediated reinforcers was found to be 13.4 (range 8 to 19), and the mean number of automatic reinforcers to be 4.9 (range none to 14). A combined score (socially mediated minus automatic reinforcers) had a mean of 8.6 (range 1 to 16). The top five most commonly reported socially mediated reinforcers were: (1) being tickled (88 %), (2) smiles or verbal praise (84 %), (3) hugs or cuddling (80 %), (4) candy, chocolate or ice-cream (80 %), and (5) fruits or raisins (76 %). The top five most commonly reported behaviors, assumed to be automatically reinforced were: (1) Flapping or clapping one's hands (40 %), (2) repeating certain questions, expecting an answer (32 %), (3) repeating a certain nonsense sound (32 %), (4) opening and closing doors or drawers (28 %), and (5) spinning around, jumping up and down or walking in circles (24 %).

Statistical Methods

The questionnaire yields two figures for every child: number of socially mediated reinforcers and number of automatic reinforcers. A stepwise multiple regression was carried out in order to investigate the best predictors of individual therapy outcome (learning rate). Predictors tested for inclusion in the model were number of socially mediated reinforcers, number of automatic reinforcers, age at intake, and Vineland standard score at intake.

Results

In the stepwise multiple regression, the number of socially mediated reinforcers was found to be a significant predictor (p=0.001). For every socially mediated stimuli scored as a reinforcer for a child, the learning rate increased by more than a month per year (B=0.115; standardized B=0.560; t=3.858). Number of automatic reinforcers was also found to be a significant predictor (p=0.031). For every automatic stimuli scored as a reinforcer, the learning rate decreased by half a month per year (B=0.055; standardized B=-0.354; t=-2.359). Age at intake was also found to be a significant predictor of learning rate (p=0.032). Surprisingly, older children exhibited larger treatment gains (B=0.234; standardized B=0.362; t=2.352). Vineland score at intake did not predict learning rate (p=0.449). A linear regression model comprised only of socially mediated and automatic reinforcers explained more than half of the variation in treatment outcome (R^2 =0.499).

Applying a pearson correlation, the number of socially mediated and automatic reinforcers was found to be almost completely independent of each other (r = -0.107; p = 0.609). Thus, the two figures can be reduced to one with a simple subtraction without losing much predictive power. Such a subtracted figure and its relation to learning rate are plotted in figure 1.

Discussion

The present study reports a positive relation between treatment gains by the end of the first year of EIBI and retrospective reports of the number of socially mediated reinforcers, and a negative relation between treatment gains and concurrent reports of automatic reinforcers. These two figures taken together explained 49.9 % of the variation in treatment gains for children after one year of EIBI.

These results might explain why some children benefit little or nothing at all from EIBI. A child who responds only to a few socially mediated reinforcers may satiate quickly on these reinforcers and thus lose motivation for learning (Egel, 1981). Moreover, lack of motivation for learning may arise from the potent automatic reinforcers produced by the stereotyped behaviors. That is, the child may access potent reinforcers with stereotyped behaviors requiring a minimum of response effort. However, to obtain the socially mediated reinforcers provided by the therapist, the child has to perform rather complex and demanding tasks. Another reason why stereotyped behavior can hinder learning is the mere incompatibility of attending to the teacher and the teaching materials while engaging in stereotypic behavior.

Children with PDD-NOS and mental retardation without autism seem to benefit more from EIBI than children with autism (Eldevik, Jahr, Eikeseth, Hastings, & Hughes, 2010; Smith et al., 2000). The present study might help to explain this, as these groups of children likely exhibit fewer stereotypies and a larger repertoire of socially mediated reinforcers.

The present study has several methodological limitations which should be considered. The stimuli assumed to be reinforcers were identified through a questionnaire filled out by parents and therapists rather than through direct preference assessments or through evaluations of whether the stimuli actually functioned as reinforcers. Hence, the function of these stimuli was assessed only indirectly, rather than through systematic behavior observations, which by definition are needed to show that they indeed were reinforcers (Skinner, 1965). In fact, even people who know a client well may not necessarily be good judges of which stimuli functions as reinforcers (Logan & Gast, 2001). However, all respondents had been involved in the behavioral treatment for at least 6 months and had been explicitly trained to identify effective reinforcers to be used in training.

Although stereotyped behaviors are often maintained by automatic reinforcement, some research has demonstrated that such behaviors sometimes also can be maintained by socially mediated reinforcement (Cunningham & Schreibman, 2008). The SMARQ does not distinguish between stereotyped behavior maintained by automatic reinforcement and socially mediated reinforcement. Rather it is assumed that all stereotyped behavior is maintained by automatic reinforcement, and hence this may limit the validity of the inventory. However, stereotyped behavior currently maintained by socially mediated reinforcement will most likely originally have been shaped and maintained by automatic reinforcement, and whenever socially mediated reinforcement is involved in a particular stereotyped behavior, automatic contingences often operates concurrently.

Another limitation of the present study is the lack of control of other variables which might also affect treatment outcome. Such factors were not investigated in this study.

Future studies could investigate etiologies of these differing reinforcer repertoires. Although very the differences very likely are due to differences in conditioned reinforcers, it cannot be ruled out that the differences reflect differences in primary reinforcers, perhaps due to deviant sensory processing (Klintwall et al., 2011). This could be a putative endophenotype for genetic studies. Also, future studies could assess experimentally whether there is a causal relationship between number and type of reinforcers and treatment outcome for children receiving EIBI. If so, an important next step would be to develop techniques to increase the number of stimuli that can function as socially mediated reinforcers for a particular child. Techniques for this are being developed (Holth, Vandbakk, Finstad, Grønnerud, & Akselsen-Sørensen, 2009). If such techniques are successful in increasing number of such reinforcers, this could lead to larger treatment gains for treatment resistant children.

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