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Am J Intellect Dev Disabil. 2014 July ; 119(4): 351–370. doi:10.1352/1944-7558-119.4.351.**Risk Factors for Self-Injury, Aggression, and Stereotyped Behavior Among Young Children At Risk for Intellectual and Developmental Disabilities****Stephen R. Schroeder,**
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University of Kansas, Medical Center, Ophthalmology**Abstract**

Before the 1990s, research on the early identification and prevention of severe behavior disorders (SBDs), such as aggression, self-injury, and stereotyped behavior, among young children with intellectual and developmental disabilities (IDD), was mostly done with children 3 years or older. More recent work suggests that signs of SBDs may occur as early as 6 months in some infants. The present study combined a cross-sectional and longitudinal approach to examine SBDs in 180

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young children aged 4–48 months recruited through mass screening, then receiving an interdisciplinary evaluation and six-month follow-ups for one year. Twelve potential risk factors related to SBDs were examined. Eight of these risk factors, including age, gender, diagnosis, intellectual and communication levels, visual impairment, parent education, family income, were differentially related to scores for Aggression, SIB, and Stereotyped Behavior subscales on the Behavior Problems Inventory (BPI-01) at initial interdisciplinary evaluation. BPI-01 scores decreased over the year for 57% of the children and increased for 43%. The amount of decrease on each BPI-01 subscale varied with age, gender, and diagnosis.

Keywords

aggression; self-injurious behavior; stereotyped behavior; infants; toddlers; intellectual and developmental disabilities

Research on the early identification and prevention of severe behavior disorders (SBDs), such as aggression, self-injurious behavior (SIB), and stereotyped behavior among young children with intellectual and developmental disabilities (IDD), has been a long time in coming, although its importance has been acknowledged repeatedly in both the animal and the human literature (see Rojahn, Schroeder, & Hoch, 2008; Schroeder, Loupe, & Tessel, 2008 for extensive reviews). These behaviors appear together in a significant number of cases.

Most of the early research on SBDs has been with children 3 years or older, for example, with children with autism (Lovaas, 1987); with children with no or with mild intellectual disabilities (Baker, Blacher, Crnic, & Edelbrock, 2002); with a wide range of children in a pediatric outpatient setting (Wacker et al., 1998); and in children with IDD in school settings in the United Kingdom (Murphy, Hall, Oliver, & Kissi-Debra, 1999).

Only a few assessment instruments have been validated for children under age 3. Among them are the Aberrant Behavior Checklist (ABC: Karabekiroglu & Aman, 2009); the Baby and Infant Screen for Children with Autism Traits (BISCUIT: Matson, Mahan, Sipes, & Koslowski, 2010), and the Behavior Problems Inventory (Rojahn et al., 2012). They measure all three SBDs plus other less severe behaviors, and they have shown that all three SBDs occurred below age 3.

Matson's review (2009) showed that, until recently, little work has been done on the development of aggression among infants and toddlers at risk for IDD, although it is often linked with risk factors for SIB and abnormal stereotyped behaviors in older children and adults with IDD (Matson, Cooper, Malone, & Moskow, 2008; Matson, Dixon & Matson, 2005; McClintock, Hall, & Oliver, 2003; Oliver, Petty, Ruddick, & Bacarese-Hamilton, 2012). Most of these studies indicate a higher frequency of aggression, SIB, stereotypy, and hyperactivity, tantrums, and so forth, especially more among children with autism spectrum disorders (ASD) and IDD than among those with language delay or typical development. Risk factors identified were lower IQ, lower communication skills, more medical conditions, more than 15 genetic syndromes, presence of more than one related SBD (Arron, Oliver, Moss, Berg, & Burbridge, 2011; McClintock et al., 2003; Oliver et al., 2012), and lower

family income and parent education (Keller, & Fox, 2009). These and other risk factors remain to be examined among young children under 3 years of age at risk for SBDs and IDD.

The research literature on early development of stereotypy is more plentiful than research on aggression in IDD. Stereotyped movements of typically developing infants (Kravitz & Boehm, 1971; Thelen, 1979; Thelen, 1996) have shown that rhythmic repetitive behaviors occur in all infants; and some of these behaviors may appear to be self-injurious. Usually, these behaviors decrease by 5 years of age. Some forms of stereotyped acts, however, are abnormal; and they have been identified as such for many years (Berkson, 1967; Bodfish, Symons, Parker, & Lewis, 2000; Lewis & Baumeister, 1982). We define abnormal stereotyped behavior as repetitive, invariant acts or action sequences whose reinforcement contingencies are unspecified or noncontingent and whose performance is considered to be related to pathology (Schroeder, 1970). Guess and Carr (1991) published a model for the development of such stereotyped behaviors into SIB at an early age, but as yet, there is not much direct evidence for this model (Oliver et al., 2012). Emergence of repetitive and stereotyped movements as young as 1 year of age as an early predictor of later ASD has been of particular interest to researchers on ASD and has been a topic of several recent papers (see Damiano, Nahmias, Hogan-Brown, & Stone, 2013 for a review).

Recent cross-sectional studies among infants and toddlers at risk for SIB and IDD have cast light on the prevalence of SIBs as early as 6 months in some infants (see Richman, 2008; Symons, Sperry, Dropik, & Bodfish, 2005 for reviews). Risk factors for SIB, however, among children below the age of 5 years are less clear than they are among adolescents and adults. In a retrospective chart review of 196 children with developmental delays (aged 18–72 months; $M = 42.7$ months), evaluated in a comprehensive neuro-developmental clinic, Maclean, Tervo, Hoch, Tervo, and Symons (2010) found that those with and without SIB did not differ in vision or hearing impairment, seizure disorder, autism diagnosis, mobility, or cerebral palsy; but they did show higher rates of aggression, destructive behavior, and “unusual habits” on the Inventory For Client and Agency Planning (ICAP) (Bruininks, Hill, Weatherman, & Woodcock, 1986). MacLean and Dornbush (2012) repeated this survey with a similarly aged statewide sample of 257 children being considered for the Developmental Disabilities Home and Community Based Waiver and found a lower prevalence rate of SIB than in the MacLean et al. (2010) study, but a similar pattern of no difference in risk factors for SIB except for more aggression (“hurtful to others”) among the SIB group.

Matson and Tureck (2012) reviewed over 80 studies using their Baby and Infant Screen for Children with Autism Traits (BISCUIT) and the Batelle Developmental Inventory, second edition (BDI-2: Newborg, 2005) in assessing more than 2,000 children enrolled in the Early Steps Birth-to-Three population of the state of Louisiana for early signs of autism. The BISCUIT is an extensive, well-validated parent interview instrument with three parts that assesses (a) the core features of autism (62 items), (b) psychopathology (57 items), and (c) challenging behaviors (15 items) among infants and toddlers. Although the primary interest of Matson and colleagues was in assessing ASD at an early age, they have shown high rates of SBDs (Fodstad, Rojahn, & Matson, 2012) and emerging psychopathology (Matson, Hess, & Boisjoli, 2010) comorbid with children with autism, pervasive developmental disorder

(PDD), and developmental delays without autism. Symptom frequency was highly correlated with severity and increased across ages grouped at 6-month intervals from 12 to 39 months (Fodstad et al., 2012). Medeiros, Kozlowski, Beighly, Rojahn, and Matson (2012) found an interaction between developmental quotient on the BDI-2, communication and diagnoses. Toddlers with ASD or PDD-NOS had higher BDI-2 scores and more SBDs, whereas those with only atypical development had lower BDI-2 scores and fewer SBDs. The large number of children in these studies probably helped to reveal these differences.

Few longitudinal studies of SBDs among very young children have been conducted. Studies by Berkson and colleagues (Berkson, 2002; Berkson & Tupa, 2000; Berkson, Tupa, & Sherman, 2001) followed 48 of 64 at-risk children referred for stereotyped behavior and SIB by the staff in five Birth-to-Three Programs of 457 children in Chicago. Prevalence of SIB was 21 of 457 (4.6%) in the total sample, and 56% in the 39 high-risk children in the study sample. These children had a variety of syndromes, the most frequent of which were autism, Down syndrome, and epilepsy. Virtually all of them had stereotyped behaviors, which appeared to be related to onset of four-point crawling and, eventually, walking and which began to subside by 15–36 months of age for the majority of participants. SIB typically subsided later at around 24–36 months, although three children persisted for another 36 months.

We define SIB as acts directed toward oneself that are likely to result in tissue damage (Tate & Baroff, 1968). Berkson et al. (2001) further introduced a distinction between “proto-SIB” (i.e., SIB of the same form which was not causing tissue damage) versus SIB that was causing tissue damage. Others have not found this to be a useful classification (Kurtz, Huete, Cataldo, & Chin, 2012) because not all kinds of tissue damage are readily observable or measured. Richman and Lindauer (2005) did find the proto-SIB term useful in their study of 12 children aged 14–32 months with profound developmental delays. They found proto-SIB to emerge at 22.6 months, whereas tissue damage emerged at 24.8 months. They were not able to observe the onset of proto-SIB, however. Berkson also did not record aggression or potential risk factors related to other SBDs. Abnormal stereotyped movements were also not distinguishable from typical stereotyped movements by Berkson et al. (2001) with their observation system.

Kurtz et al. (2012) reported on an ongoing intensive observational study of 29 children ranging in age from 8 to 51 months ($M = 23.4$ months), 14 of whom had significant delays and 15 of whom had average development. They found no differences in SIB frequency or topography between children with and without language delays, but they did find an increase of other SBDs, along with emerging multiple topographies of SIB with age. Future outcomes await completion of this study. Our group (Mayo-Ortega et al., 2012) reported on early identification and prevention of SBDs in a total population study among 233 infants and toddlers aged 4–48 months (mean = 27.3 months), at risk for IDD in Peru, in collaboration with the Centro Ann Sullivan del Peru (CASP), using a mass screening method. CASP is a state-of-the-art school for people with autism and IDD sponsored by the Life Span Institute at the University of Kansas. The goal was to establish a set of risk factors for parents and caregivers to use in deciding to intervene with their children before these behaviors develop a lengthy history in their response repertoires. Rojahn et al. (2008)

published the model, and it postulates an ontogenetic model of gene-brain-behavior relationships affecting development and interaction of SBDs. Initially, we screened for nine categories of a long list of risk factors based on the literature (Mayo-Ortega et al., 2012). If a child exhibited any of these risk factors, they were included in an in-depth interdisciplinary evaluation, to confirm which of these risk factors occurred most frequently in the present study.

The current report has two parts. In Study 1, we present a cross-sectional study of risk factors found at entry into the study. By risk factors we mean those variables that contribute, either alone or cumulatively, to the probability of the occurrence of an SBD. The goal is to evaluate the prevalence and relative contributions of these risk factors to the performance of children as young as possible who show the first signs of developing SBDs and IDD. In Study 2, we report on a longitudinal study in which these young children from Study 1 were followed every 6 months for a year, in order to evaluate factors affecting their increasing or decreasing SBDs.

In Study 1, the risk factors studied included those found in Mayo-Ortega et al. (2012), those found in the other studies cited previously and in the literature of biological risk (Sameroff, 2009), and in our recent review of the literature (Schroeder & Courtemanche, 2012). From these sources, we selected 12 potential risk factors to examine. The five most commonly mentioned factors (age, gender, clinical diagnosis, intellectual level, communication level) are the primary focus of the cross-sectional part of this report. The other seven factors, which are often omitted in studies of SBDs in IDD, but which might be important to consider, are included in Study 1 as factors of secondary interest. These factors are: family income, parental education, vision impairment, hearing impairment, dental problems, nutritional deficiency, and seizures.

For the five factors of primary interest, we proposed the following hypotheses: (1) The children in the older age groups would have higher levels of SBDs than the children in the younger groups; (2) The males would have higher levels of SBDs than the females; (3) The levels of SBDs would differ significantly among the three different clinical diagnosis groups, with children at risk for autism having more SBDs; (4) The children in the higher IQ groups would have lower levels of SBDs than the children in the lower IQ group; and (5) The children in the group with higher levels of communication would have lower levels of SBDs than the children in the lower communication level group. For the other seven factors, we focused on exploring the prevalence of these possible risk factors and the relationships (if any) between the factors and the levels of SBDs.

In Study 2, the longitudinal study, our primary question focused on the patterns of change across the two 6-month periods for the three risk factors of age, gender, and diagnosis. Our primary question was whether the patterns of change in the levels of SBDs would be the same for each group in the factor or whether those patterns would differ. For example, we expected that the level of SBDs for the group of younger children would start with no or fewer SBDs and increase over 12 months, whereas the group of older children would start at a higher level of SBDs and generally decrease over 12 months.

Method

Participants and Ascertainment

The project occurred in three phases: Study 1 involved (a) Screening, (b) In-depth Interdisciplinary Evaluation at entry to the project. Study 2, involved 6-month follow-ups for a year and comparing performance across three 6-month time points.

Study 1: Screening and Interdisciplinary Evaluation

Screening—The details of the screening of the children can be found in Mayo-Ortega et al. (2012) and are summarized here. Children aged 6–36 months who might be at risk for SIB, stereotyped behavior, or aggression were solicited by television, radio, and newspaper announcements in Lima and throughout Peru. By “at risk” we mean that a child is showing topographical approximations or signs of a behavior that may raise the probability of its full-blown occurrence as a SBD in the future. CASP has a Telephone Triage Service (TTS) that answers inquiries from parents about services available to their children in Lima and all of Peru. If parents and the TTS agreed, then the child and his family were invited for a screening interview. After the TTS received 1,000 calls, 341 were invited for screening by trained veteran parents from the Center, who were trained in using The Parental Concerns Questionnaire (PCQ), a 15-item screening questionnaire (Mayo-Ortega et al., 2012; Schroeder et al., 2013), which took 15–30 min. Veteran parents had had a child with SBDs who was enrolled at CASP and had received 185 hr of training per year for several years. Using veteran parents was a strength in the screening process, since they were highly experienced in recognizing signs of SBD and they could empathize with the families and encourage them. The PCQ was based on an extensive list of nine categories of risk factors that had been associated with SBDs in the research literature (Dawson, 1996; Dunlap et al., 2006; Rojahn et al., 2008) (i.e., intellectual disability; communication impairment, genetic factors, family history of other brain disorders, several medical conditions, psychiatric factors, neurochemical and metabolic factors, neuropsychological factors, motor factors [see Mayo-Ortega et al., 2012]). These factors were based both on parental concerns and previous assessments by their family physicians in most cases, and they were not measured during screening, for example, neurochemical factors. They could also be found at the subsequent developmental pediatric exam and could be referred for appropriate assessments by relevant specialists if indicated. Any one of these concerns on the PCQ qualified a child to participate in the study. Only a limited number of all of the possible risk factors was reported by parents. The most frequently endorsed concerns by parents on the PCQ were Delayed Language Development (82%), Aggression (68%), Self-Injury (61%), Stereotyped Behavior (60%), Autism Diagnosis (63%), Medical/Neurological Diagnoses (54%), and Delayed Cognitive Development (58%) (Mayo, et al., 2012). After screening, 262 families were invited for an in-depth interdisciplinary evaluation at CASP.

Interdisciplinary evaluation—Of the 262 families invited, 233 attended and received a full interdisciplinary evaluation. The first evaluation (Time 1) began with informed consent and enrollment in the study. Then a family history and profile and height, weight, and head circumference were taken by teams of trained nurses. Developmental pediatric exams were conducted by U.S. board certified developmental pediatricians, and other Peruvian

physicians under their supervision. Cognitive assessments used the Cognitive Subscale of the Bayley Scales of Infant Development (BSID III [Bayley, 2006]); communication assessment used the Communication, Symbolic, and Behavior Assessment Scale (CSBS [Wetherby & Prizant, 2002]) completed by CASP staff trained by one of the authors who is a certified speech language pathologist and an expert in early communication; behavioral assessments used the Behavior Problems Inventory (BPI-01 [Rojahn, Matson, Lott, Esbensen, & Smalls, 2001]). These assessments were conducted by CASP staff trained and supervised by the senior author; visual assessment was done by a pediatric ophthalmologist expert in developmental disabilities and by other pediatric ophthalmologists from Peru; auditory screening was done by a U.S. certified audiologist and co-author and CASP staff under her supervision; dental screening was done by a Peruvian orthodontist and his residents, who were expert in dental treatment of people with disabilities. In all, over 100 U.S. and Peruvian professionals were involved in assessments. Autism screening by experienced senior CASP staff used the Child Autism Rating Scale (CARS [Schopler, Reichler, & Renner, 1988]). All staff were trained and monitored by eight expert U.S. consultants for fidelity of their assessments. Consultations (43 genetic, 42 neurological, 23 nutrition) were also given, as referred by the developmental pediatricians. Buccal cheek swabs for DNA microarray analysis were also taken on each child, in order to find possible genetic syndromes that might not have been observed clinically. DNA has been isolated on a subset of children for DNA structural microarray analysis following established protocol (Rethmeyer, Tan, Manzardo, Schroeder, & Butler, 2012). Results have been submitted for separate publication (Usrey, Manzardo, Roberts, Schroeder, & Butler, under review).

The primary dependent measure examined was the BPI-01 by Rojahn et al. (2001). This is a well-validated informant-based rating scale that assesses frequency and intensity of SIB (14 items), Stereotyped behavior (24 items) and Aggression (11 items) among people with developmental disabilities. Each item is scored on a 3-point Likert scale for severity (1 = slight, 2 = moderate, and 3 = severe) and a 5-point Likert scale for frequency (0 = never, 1 = monthly, 2 = weekly, 3 = daily, 4 = hourly). It has excellent psychometric characteristics (Rojahn et al., 2001) and recently published norms (Rojahn et al., 2012).

Secondary dependent behavioral measures included two widely used scales: the Aberrant Behavior Checklist (ABC [Aman & Singh, 1994; Aman, Singh, Stewart, & Field, 1985; Marshburn & Aman, 1992]) and the Repetitive Behavior Scale-Revised (RBS-R [Bodfish et al., 2000]), each of which overlapped with the BPI-01, but which also contained additional subscales of behavior disorders of interest. The ABC and RBS-R were only administered at Time 2 because of time limitations on the initial evaluation days. Because these scales have not been used in a population so young, a paper cross-validating these scales in the present study has recently been published (Rojahn et al., 2013) and will not be repeated here.

During the 6-month period following evaluation days, children received functional analyses (Iwata et al., 1994). This was an analog functional analysis conducted at the Center. Another functional analysis adapted for home use (Wacker et al., 1998) was also conducted in the home. Because this evaluation was labor intensive and expensive, analyses for only 17 children were completed.

Study 2: Six-month Follow-Ups

Data were collected at three time points (i.e., at birth, 6 months, and 12 months [Times, 1, 2, 3]). Six-month follow-ups were conducted by the same trained staff at CASP for continuity and consistency. Because of time constraints on evaluation days and availability of professionals in Peru, a reduced, streamlined protocol was used at Time 2, during which dental, visual, and hearing screenings continued to be conducted. All children received the BPI-01 and the CSBS-Caregiver Scale at all three evaluations. At the third evaluation (Time 3), children received the BPI-01, the CSBS, and a follow-up Developmental Pediatric exam.

Overall, 53 of 233 children failed to complete one or two of the three evaluations for a wide variety of reasons (e.g., illness, death, another childbirth, schedule conflicts, travel distance). Their mean BPI-01 scores were virtually identical to those of the 180 who did complete the study and on whom the statistical analyses were performed. There did not appear to be a systematic source of bias influencing drop out in relation to behavior problems.

Data Analysis Plan

All data analyses reported here were performed only on the 180 children who completed both studies.

Study 1—Initially, descriptive statistics of the participants enrolled in the study were calculated. Correlation matrices were also examined for potential relationships among the BPI-01 measures of SIB, Aggression, and Stereotyped behavior as well as the measures for potential risk factors.

To address the five hypotheses in the cross-sectional study, appropriate groups within each risk factor were formed. The Clinical Diagnosis factor comprised three groups: At-Risk for Autism, Down Syndrome, and Atypical Development. Four age groups were defined: 4–12 months, 13–24 months, 25–36 months, and 37–48 months. Age grouping by the year (1,2,3,4) was based upon our desire to observe the emergence and possible changes in the developmental trajectories of SBDs during this period of the children's rapid development. High and low IQ groups were formed with the low group comprising children whose Bayley scores were more than 1 SD below the mean (a commonly accepted cut point for risk of IDD). CSBS scores were formed with the upper quartile forming one group and the lowest quartile forming the second group. Univariate analyses of variance (ANOVAs) were conducted to test the primary hypotheses. In each analysis the risk factor was the between-subjects variable and one of the three SBDs was the dependent variable. For significant results, and when appropriate, post hoc tests were conducted to determine which groups differed significantly. The post hoc procedure to control Type 1 error was Games-Howell, a recommended procedure for unequal variances and unequal sample sizes. Effect sizes are reported as eta-squared. An often used interpretation of eta-squared in the social sciences is .01, small; .06, medium; and .14, large (Cohen, 1988). The seven factors that were of secondary interest (vision, hearing, dental health, family income, mother education, malnutrition, and seizures) were examined with descriptive statistics (means and frequencies) to aid in assessing prevalence for each factor.

We also explored the relation of the proposed set of risk factors to each SBD by conducting a multiple regression analysis for each behavior. Included as independent variables for each analysis were the five primary factors that had been examined previously plus three additional factors (vision, family income, and parent education); prior descriptive analyses and literature indicated these three factors might be relevant for at least one of the SBDs. Our first step was a regression analysis for each SBD to determine the total variance accounted for by the set of risk factors. Our next step was to determine the unique contribution of each factor for the specific SBD, i.e., the proportion of criterion variance associated with that specific factor, given that all the other factors were included in the model. The unique contribution is given by the square of the part correlation or, in the case of a categorical variable such as Diagnosis, by the sum of the squares of the part correlations of each of the dummy variables.

Study 2—To address the second hypothesis (i.e., that children with different levels or types of risk factors would have different patterns of change across time periods) a general linear mixed model was used, with time as a within-subjects variable and specific risk factors (age group, gender, and clinical diagnosis) as between-subjects variables. The initial focus was on the interaction between the risk factor and the time variable. If the interaction is significant, then the patterns of change over time are different for at least two of the groups (or for some combination of the groups) within the risk factor. For significant interaction effects, we examined each group to determine the particular pattern of change for that group and how the patterns differed among the groups. If the interaction was not significant, then our focus was on the significance of the time variable. If that was significant, then a general pattern of change was applicable to all groups, and we examined that pattern. If the time variable was not significant, then the mean level for that SBD across all groups at each time point was relatively unchanged. A third statistic to examine, after looking at the interaction and the time effects, was the effect for group. A significant group effect indicates that the group means (calculated across all time points) are different. We report this effect, but it is not of primary interest since it does not reflect any longitudinal effects. The analyses were conducted using SAS PROC MIXED with KR degrees of freedom.

Results

Descriptive Statistics

Of the 180 children who had complete data at all three evaluation periods, 62% were male. Mean age was 27.4 months ($SD = 10.13$, range = 4–48 months) (in a few cases, parents also asked that a sibling who concerned them be evaluated; so, we expanded the age range for admission to the study slightly). Distribution of age groups (12-month grouping) was: 4–12, 10%; 13–24, 27%; 25–36, 43%; 37–48, 20%, similar to the 6-month grouping in the Fodstad et al. (2012) study. Many children had had major illnesses (47%), injuries (17%), or hospitalizations (45%); had received a previous diagnosis (55%); and were receiving medications (29%) or other therapies (74%). They were seeking a second opinion and help. A few were in special education programs (18%).

Developmental Pediatric exams yielded a variety of primary clinical diagnoses: Down syndrome, ($n = 45$) (4 were also given a secondary diagnosis as At-Risk for Autism); Autism or At-Risk for Autism ($n = 74$); Atypical Development, that is, Blindness ($n = 4$); Strabismus ($n = 2$); Seizures ($n = 2$); Global Developmental Delay ($n = 4$); Hypotonia ($n = 2$); Hydrocephaly ($n = 2$); Schizocephaly ($n = 51$); Microcephaly ($n = 1$); Macrocephaly ($n = 1$); Cerebral Palsy ($n = 1$); Meningitis ($n = 1$); Agenesis of the Corpus Callosum ($n = 2$); Joubert syndrome ($n = 1$); West syndrome ($n = 2$); Moebius syndrome ($n = 2$); Russell Silver syndrome ($n = 1$); Delays with Unknown Causes ($n = 61$).

SBDs exhibited during the interdisciplinary evaluations were mild for all except two children, who were performing SIB likely to cause tissue damage. They were removed from the study and put directly into an intensive behavior intervention program at CASP. Thus, all of the SIB cases in the study were exhibiting proto-SIB, according to Berkson's classification (Berkson et al., 2001). Mean item frequencies based on time (monthly, weekly, daily, hourly) of SBD topographies can be seen in Table 1.

Study 1: Cross-Sectional Results at Time 1—Primary Analyses

BPI-01 frequency and intensity were highly correlated throughout all of the data; correlations ranged from .413 to .948. As a result, only BPI-01 frequency scores are presented in this paper, for the sake of brevity. A detailed analysis of the relationship of BPI frequency and intensity can be found in our companion paper on BPI-01 validity in this population by Rojahn et al. (2013). Initially, univariate ANOVAs were conducted to determine which risk factors were related to specific SBDs and the size of the effects. Results are given here for each factor separately and for each behavior.

Cross-sectional results—Primary Analyses

Gender—The means for the boys were generally higher than for the girls, but only significantly so for Stereotypy, $F(1, 177) = 5.79$, $p = .017$ (See Table 2).

Age—There was a large effect of age at Time 1. Table 3 shows that the mean BPI-01 scores were highest with the oldest children at entry at Time 1 and lowest with the youngest children; the two groups with ages in the middle had similar scores between the two extremes. SIB was the only SBD that did not differ significantly between the age groups. In addition to having statistically significant differences among the groups, both Stereotypy and Aggression had medium to large effect sizes. To determine which age groups differed, post hoc comparisons were conducted using the Games-Howell method for adjusting the p-values and taking into account the heterogeneity of variance that was present. For both Stereotypy and Aggression, the differences between the youngest age group and each of the other older age groups were statistically significant. No other age-group differences were significant.

Primary clinical diagnosis—The main clinically diagnosed syndromes by the developmental pediatricians were Autism or At-Risk for Autism (74) and Down syndrome (45). All but seven of the 74 children clinically identified as At-Risk for Autism also independently received CARS total scores of 20 or above (Mean = 35, Range = 15–53). The typical 4:1 ratio of boys-to-girls existed. Of the remaining medical clinically diagnosed

syndromes, none have been known to be associated with SIB, stereotyped behavior, or aggression in the literature. These were collected all into an “Atypical Development” category. Table 4 shows this result. There were statistically significant differences between the groups on all three of the BPI-01 measures with medium effects for SIB and Aggression and a large effect for Stereotypy. The variances were unequal for all three measures; so, the post hoc tests were conducted using the Games-Howell method. These tests showed significant differences between Autism and each of the other two groups (Down Syndrome and Atypical Development), but no significant differences between the Down Syndrome group and the Atypical Development group (see Table 4).

Bayley Cognitive Composite Scale—Of the 180 children who had complete BPI data, 159 had scores on the Bayley Cognitive Scale. Twenty-one were untestable due to severe global delays, blindness, or noncompliance. One hundred forty-six children had scores below the standard mean of 100. We were hesitant to attribute intellectual disability to them at such a young age. Using the standardized norms (mean = 100, standard deviation = 15), 63% (100 children) were more than one standard deviation below the mean; these children formed one subgroup (Low). The remaining 59 children (scores above the one standard deviation below the mean) formed a second subgroup (High). When these two groups were compared on BPI-01 subscale scores, there were significant differences only on the Aggression subscale, $F(1,157) = 6.24, p = .013$. Children in the High Bayley group had a higher mean aggression scores; the effect size was small-medium (see Table 5).

Communication and symbolic behavior scale (CSBS)—The BPI-01 scores in the top quartile vs. the bottom quartile of the CSBS raw scores were compared. Aggression was the only subscale with a statistically significant difference, $F(1, 75) = 10.15, p = .002$; the effect size was medium-large. The mean aggression score was larger for children with high CSBS scores than the mean for children with low CSBS scores, as Table 6 shows. Means for SIB and Stereotyped Behavior were virtually identical.

Cross-sectional results—Exploratory analyses

Vision—Vision assessment of 179 children was conducted by an expert U.S. pediatric ophthalmologist. On the World Health Organization 4-point vision scale of 1–4, 73% had normal vision; 16% had mild impairment; 4% had severe impairment; and 7% were blind. The mean BPI-01 score of the 48 (27%) children who had vision problems was compared to the mean of the children who had no vision impairment. The means were nearly the same for SIB in the two groups, but were somewhat higher for Aggression and Stereotypy in the no-vision-problems group. Visual impairment is a problem that may often be overlooked as a risk factor for SBD among children with DD.

Family income and parent education—Demographic data were collected by the nurses at intake regarding family income and education (both mother and father). Because of the close relationship between mother and father education, only mother education was examined. The three education groups were secondary or less, post-secondary technical, and superior/university. Children whose mothers had the highest education level had the highest Aggression scores. Means for SIB and Stereotypy were relatively similar. The four income

groups were poverty, medium/lower, medium/higher, and high. The means for the four income groups were quite similar for all the BPI-01 scores. It should be noted that the parental education and family income levels in this study were relatively evenly distributed over lower class to the upper middle class. Very few were either among the indigent or among the very high income groups.

Hearing—Hearing was screened by a U.S. expert audiologist. In the time available, 90 children were examined and were classified into two groups: “pass” and “fail.” Of the 57% who failed, the majority had middle ear disorders, which is common among children of this age, particularly in cities with air pollution, such as Lima. Middle ear infections may be painful and may be the occasion for behavior problems, as has been documented for otitis media (Carr & Smith, 1995).

Dental problems, malnutrition, seizure disorders—There were minimal differences in BPI-01 total scores due to dental problems (36%), malnutrition (1%), seizures and neurological diagnoses (5%). The most prevalent dental problem was dental caries, probably due to lack of fluoridation in the water in Peru.

Differential Contributions of Risk Factors to BPI Subscale Scores

For the final cross-sectional analysis we examined the variance in each behavior that is accounted for uniquely by each risk factor. For this analysis we included the five factors of primary interest (gender, age, diagnosis, IQ, and communication level) and included three variables from the exploratory analyses: Vision, mother education, and family income. We chose these three because of the relatively high correlations with one or more of the BPI-01 subscales or with a risk factor. Also, data on these factors was available for nearly all the participants in the 1-year study. Because hearing records were available for only about half of the sample, including that factor would have reduced the sample to a size too small for conducting the multiple regression analyses. Each BPI-01 subscale was the dependent variable in a multiple regression analysis with the set of risk factors as the independent variables. The R-square gives the value for the variance in the BPI-01 score that is accounted for by the set of variables in the model, and the square of the semi-partial (or part) correlation for a specific (or the set of dummy variables for nominal variables) gives the unique proportion of the total variance of the BPI-01 score that is accounted for by that variable. Table 7 contains the total R-square and the unique contribution made by each of the eight risk factors to the R-square for each BPI-01 subscale. The unique proportion of variance accounted for by each of the eight factors varied among the three behavior types. One purpose of these analyses was to determine which factors would be most relevant to include in the analyses in the longitudinal study.

For Stereotypy, Age, Gender, Family Income, Communication, and Diagnosis accounted for the most variance; Bayley, Visual Impairment and Mother Education accounted for very little. The size of the contribution for some of these factors mirrored the effect sizes, which ranged from large for Diagnosis, to medium-large for Age, and small-medium for Gender. For *SIB*, the largest unique contributions were made by Diagnosis, Income, and Vision; the other five variables each accounted for less than 1% of the variance. The effect sizes also

suggested that Diagnosis (medium effect), Gender (small-medium), and Age (small effects) were relevant factors. For *Aggression*, Diagnosis, Gender, Visual Impairment, and Mother Education were the primary factors that made the largest unique contributions; Family Income, Age, the Bayley scale, and the CSBS made minor contributions. The effect sizes for these factors ranged from medium to large for Age, and medium for Diagnosis. Thus, the unique contributions of the risk factors varied for SIB, Stereotyped Behavior, and Aggression, but Diagnosis was always an important factor, with Gender and Age consistently making contributions. Because of the preceding results, Age, Diagnosis, and Gender were the primary risk factors examined in the longitudinal analyses.

Study 2: Longitudinal Analysis Across Times 1, 2, and 3

General linear mixed models were used to determine the longitudinal effects of the factors across Times 1, 2, and 3 for each subscale (SIB, Stereotyped Behavior, and Aggression) with Age, Primary Diagnosis, and Gender as risk factors. As noted in the methods section, our primary focus is the Interaction effect indicating different patterns of change for the groups. In the absence of an interaction effect, we examined the effect for time, where a significant effect would indicate a common general pattern for change. Of lesser interest is the significant effect for group, which indicates that the groups have different means (averaging across time points); often this is a reflection of large differences at the first time point.

Age groups—Four age groups were defined based on the age of the child at entry into the study. There was a significant interaction between Age group and Time for the Aggression BPI-01 subscale score, $F(6,528) = 3.21, p = .004$. Follow-up tests indicated that the youngest age group had significant differences in the mean Aggression subscale scores at the three assessments, $F(2,528) = 5.08, p = .0065$, as did the oldest age group, $F(2,528) = 4.55, p = .0110$. The changes for the youngest group, however, were different from the changes for the oldest group: older children had higher group mean scores at entry and decreased over 12 months, whereas younger children were lower at entry and increased as a group over the 12 months although the rate of increase slowed in the last 6 months. The mid-range age groups did not change much over the 12 months (see Figure 1).

For SIB, the interaction between Time and Age group was not significant, nor were the main effects of Time or Age group. For Stereotyped Behavior, the interaction effect between Time and Age group was not significant, nor was the Time effect. The effect for group (collapsing across time), however, was significant, $F(3,528) = 5.39, p = .0012$, indicating that the means (averaged over the three time points) for each group differed. Follow-up tests showed that those differences were due to the mean for the oldest age group, which differed significantly from the means of each of the other age groups (Table 8).

Primary clinical diagnosis—When diagnosis was the factor, the Time \times Diagnosis interaction effect was not significant for any of the BPI-01 subscales. SIB was the only SBD that had a significant time effect, ($F(2, 531) = 3.22, p = .041$). A significant Time effect indicates that the time point means, calculated across all diagnostic groups, were different; in the case of SIB this was due primarily to significant differences between Time 1 and Time 3, $t(531) = 2.52, p = .012$. The At-Risk for Autism group started at a high level of SIB and

continually decreased over both time periods; the Atypically Developing group maintained a medium level and then decreased after Time 2, while the Down Syndrome group maintained a low level and then increased slightly from Time 2 to Time 3. For all three BPI-01 subscales, Primary Diagnosis was significant indicating that the group means, averaged across the three time points, are significantly different: SIB, $F(2, 531) = 9.56, p < .0001$; Stereotypy, $F(2, 531) = 41.72, p < .0001$; and Aggression, $F(2, 531) = 11.70, p < .0001$. Follow-up tests indicated that the At-risk for Autism group had significantly higher means (averaged across all time points) than the Down syndrome group and the Atypically Developing group, and this was true for all three SBDs. In general the At-Risk for Autism group started with high levels of each of the behaviors, and, although they decreased steadily over time, their mean levels were still higher than the other two groups who started with lower levels that didn't change too much over time (see Table 9).

Gender—When gender was the factor, there were no significant Time \times Gender interaction effects for any of the BPI subscales. SIB was the only SBD with a significant effect for Time, $F(2, 531) = 4.18, p = .0158$; for both boys and girls the SIB behavior decreased rather quickly over the 12 months with the frequency always somewhat higher for the boys. The lack of a significant time effect for Stereotypy and Aggression indicates that the means did not change much over time; however, a significant effect did occur for gender for both of these variables: Stereotypy, $F(1, 531) = 23.36, p < .0001$ and Aggression, $F(1, 531) = 11.35, p = .0008$. So, although the means did not change much for either group, those means were significantly different for the boys compared to the girls with the boys having much higher frequencies than the girls. There was not a significant gender effect for SIB.

General change patterns for the 12 months—While the majority (102 of the 180 children) had lower BPI-01 total scores 12 months later at Time 3, 78 of them (43%) continued to increase their BPI total scores and each of the three subscale scores. Age, Gender, and Diagnosis were strong predictors for the three SBDs. An increased number of older children than younger children and more females than males showed decreased BPI-01 scores. More children with Atypical Development and those At-Risk for Autism decreased their scores, while children with Down syndrome remained the same.

Discussion

As far as we know, this is the only cross-sectional and longitudinal large-scale population study which directly solicited a sample of young children across a whole country who were mostly below the age of 3 years for risk of SBDs and IDD. Participant ascertainment involved screening with the PCQ using the same methods for all children. They then all received the same comprehensive interdisciplinary evaluations using a well-validated instrument for assessing SBDs (BPI-01: Rojahn et al., 2013) in their age group by interviews with the same trained testers, so as to avoid issues with illiteracy or inconsistency of administration. The BPI-01 contains 14 SIB items, 24 Stereotyped Behavior items, and 11 aggression items. Some other instruments that have been used in the past, for example, ICAP, ABC, and RBS-R, have only a few items covering some of the SBDs of interest in the present study. Finally, 6-month longitudinal follow-up of participants by the same testers for

a year provided complete data set for 180. The only comparable longitudinal study by Berkson et al. (2001) had only 21 participants who were high risk for SIB only.

Study 1: Cross-sectional Analysis of Risk Factors for SBDs

The cross-sectional analysis of the present study examined the contribution of 12 risk factors that might affect the frequency of SIB, aggression, and stereotyped behavior in this young age-group, and it found complex interactions among them. For primary risk factors, i.e. older children, boys more than girls showed more stereotyped behavior, but did not differ significantly with SIB or aggression. Children At-Risk for Autism showed more frequent SIB, aggression, and stereotyped behavior than children in the Down syndrome or Atypical Development group. IQ and Communication Level were positively related to frequency of aggression, but not SIB or stereotyped behavior. Recently, Medeiros et al. (2012) also found that toddlers diagnosed with autism or PDD-NOS showed a positive relationship between total Developmental Quotients and challenging behaviors than did atypically developing toddlers. So there may be a different relationship among IQ, communication, and age and frequency of SBDs in the very young population depending upon a diagnosis of ASD (Richards, Oliver, Nelson, & Moss, 2012).

The relative contributions of risk factors varied, depending on the type of SBD. Risk factors for SIB differed from those for Aggression and Stereotyped Behavior (see Table 7). Overall, Age, Diagnosis, and Gender were the most influential risk factors. Secondary risk factors, i.e. Visual Impairments, Higher Parent Education and Higher Family Income, were also related to SBDs, but failed to reach statistical significance. Hearing problems, dental problems, nutrition deficiency, and seizures were elevated but also not substantially related to SBDs in this age group in this population. They can, however be an important factor when pain is involved (Carr & Smith, 1995).

The above risk factors may not have the same weight at a very young age as they might have at an older age. In adults, the most influential factors have usually been found to be severity of Intellectual Impairment and severity of Communication Deficits (McClintock et al., 2003). In the few available studies of children, the samples in the MacLean and colleagues studies were older than in the Matson and colleagues studies and in the present study.

The work of Sameroff (2009) with young children at biological risk for milder disabilities suggests that parent education, socioeconomic status, and the home caregiver environment are important social risk factors in early child development. Similar interactions were found in the present study with Parent Education and Family Income and aggression, but less for SIB and stereotyped behavior. In prevalence studies of SBD among people with IDD, social factors have rarely been considered in the past and might be examined in future studies.

Study 2: Longitudinal Analysis of SBDs

In the longitudinal analysis part of the study, the present findings replicate and extend several findings of Berkson et al. (2001), but in a Latin American population, using a different sampling technique than through teacher referral in a Birth-to-Three Program where 21 of 457 (4.6%) were determined to be high risk for SIB (Berkson et al., 2001). Solicitation of the total population of Peru for screening through public advertisements by

Mayo et al. (2012) was used in the current study, and 96% of responders were considered high risk for SBD. Only 4 of 180 children failed to meet inclusion criteria after interdisciplinary evaluation. The two studies also used different screening and evaluation methods including observation of videotapes, staff and parental interviews in the Berkson et al. (2001) study versus parental report on the BPI-01 rating scale during staff interviews in the present study. In the case of the present study, independent observations of videotapes from functional analyses of 17 children were also used, to corroborate the parent's ratings (Schroeder, Richman, Abby, Courtemanche, & Oyama-Ganiko, 2014).

Signs of aggression, abnormal stereotyped behavior, and SIB were found in some children as young as 6 months. The majority (67%) were performing all three of these behaviors, as was found by Kurtz et al. (2012). Each of these behaviors occurred alone in only 5% of the cases. Berkson et al. (2001) found similar results for SIB and stereotyped behavior, but these investigators were not recording aggression. The difference in SIB prevalence in these studies was likely due to the difference in sampling techniques. Sampling methods may make a large difference in prevalence rates in studies such as this one, as has been noted by MacLean and Dornbush (2012) and others.

There may be some question as to the nature of SBDs in children so young, since some definitions of SBDs imply intent to harm a target, stimulate one's self, or injure one's self. Subjective parental reports may impute the intents of the child. As Table 1 shows, however, items on the BPI-01 are objective; and they do not impute intent.

Berkson et al. (2001) found that stereotyped behavior and SIB of most of their children decreased over a one year period. In the present study, SBDs tended to decrease among the older children, but to increase among infants below 12 months, before they started coming down later (see Figure 1). In the case of aggression, the infant group was still increasing at 1 year. Berkson et al. (2001) did not record aggression. Future studies might investigate how SIB, stereotyped behavior, and aggression are linked to development in young children at risk for IDD, with a view to prevention. As Sandman, Kemp, Mabini, Pincus, and Magnusson (2012) have noted, the best predictor of SIB in adults is past performance of SIB. The same may be true for SBDs in young children with IDD.

In the Berkson et al. (2001) study, only three of their 21 SIB cases persisted after a year. In the current study, 57% of the children were decreasing, but 43% were still increasing their SBDs at the end of a year of follow-up. There may be several reasons for this difference. In the Berkson et al. (2001) study children were being followed and cared for regularly in a Birth-to-Three Program, so that interventions were likely occurring to decrease their SBDs. The importance of simply participating in such structured programs for young children at risk for SBD cannot be overemphasized. Parents and caregivers of such children are often under high degrees of stress (Hastings, 2002). Having a resource system to help them is often crucial. In the present study, older children at entry at Time 1 had the highest BPI-01 scores, and they did not decrease until they were in the study at CASP. This was the case, even though over half of them had sought professional help. Many parents told us on evaluation days that they had sought help, but had been unsuccessful in receiving assistance. Only 18% of the children were in special education programs at the beginning of the study.

A subset of these children is still being followed since the end of the grant funding, and it will be possible to return to the question of long-term follow-up later.

In the case of the present study, parents demanded help immediately. To prevent dropout during the study, CASP held six bi-monthly workshops for the families, entitled, “The ABCs of Developmental Disabilities,” which covered the broad aspects of raising a child with IDD and which were well attended by families in the study; and they helped maintain enrollment in the study. CASP staff also made monthly telephone calls to the parents, checking on their child and also coaching them on daily living skills, eating, sleeping, language training, discipline, and how to handle difficult situations surrounding behavior problems. Workshop and telephone follow-up data showed a relationship between number of workshops attended by the family and children's BPI-01 total scores, as well as reduction of family stress (Oyama-Ganiko, Mayo-Ortega, Schroeder, & LeBlanc, 2013).

Limitations

Because SBD was found so early among many children in the present study, only a few (4 of 180) entered the study before they began their SBDs. So we were unable to address the questions concerning the emergence of SBDs from a zero baseline condition. Our results fail to support the Guess and Carr (1991) model or the Berkson et al. (2001) findings that SIB appears to emerge from stereotyped behavior. Some appears to do so (Richman & Lindauer, 2005), and some does not (Kurtz et al., 2012; Schroeder, Mulick, & Rojahn, 1980). Even at this age, most of our children were engaging in several SBDs which appeared to develop in a complex fashion, perhaps with comorbid psychopathology, such as hyperactivity, tantrums, fears, and compulsions, as suggested by the research of Matson and colleagues (Matson, 2009; Matson, Dempsey, & Fodstad, 2009; Matson, Fodstad, Mahan, & Rojahn, 2010; Matson, Hess, & Biosjoli, 2010; Matson, Mahan, Sipes, & Kolowski, 2010) in the Birth-to-Three population. This is an important area for future research.

Another limitation was that, because of time constraints and financial resources, children in the present study did not receive a full diagnostic workup for ASD or the full Bayley Intelligence test. For this reason, we labeled them At-Risk for Autism and At-Risk for IDD. Children's intellectual status and their signs of ASD may change rapidly at this young age.

The categories of clinical diagnoses were not formed a priori, but after the data were collected, since this was the first attempt to collect such a nationwide total-population survey on this topic. This procedure may have been a limitation of the present study. It was also encountered by Matson et al. (2009), in his Early Steps Birth-to-Three population in Louisiana. It can be avoided now in future studies in this population.

The reasons for the large decrease in SBDs over the 1-year follow-up period are unclear due to our lack of a control or comparison group that did not receive training workshops and telephone follow-up. There was a need to balance the ethics of withholding treatments for such a high-risk group. In fact, two children who were performing dangerous SIB were dropped from the study and put into intensive behavioral programs immediately at CASP. A new study is under way, comparing two interventions for SBDs in a subset of the children from the present study. The design of this study will address the limitations discussed here.

Only a small number of children ($n = 17$) were videotaped during functional analyses for direct observational coding because of time constraints and lack of financial resources. In future studies, behavioral interventions via telehealth could be used which have been demonstrated to be an effective, long-term, and less expensive modality for delivering intervention (Wacker et al., 2011) than the usual standard therapist-led behavioral treatments. Such strategies could be used to reduce the cost of intensive behavior intervention and follow-up.

Conclusions

The present study had two parts: Study 1, the cross-sectional investigation, found five of 12 potential primary risk factors and two secondary social risk factors to influence the frequency of SBDs among at-risk infants and toddlers in a complex fashion. Study 2, the longitudinal analysis, replicated and extended several of the Berkson et al. (2001) findings of early development of SBD among infants and toddlers at risk for IDD.

There is also mounting evidence among older people with IDD that these behaviors tend to persist among a subset of chronic cases (Schroeder et al., 1982; Taylor, Oliver, & Murphy, 2011). The same result is likely to be the case in young children (Wacker et al., 1998; Wacker et al., 2011). It is likely that some of these children, perhaps 25% to 40% (Schroeder et al., 1982), may need to be followed long-term; and their interventions may need review and periodic renewal indefinitely (Wacker et al., 2013).

All of the results discussed here point to the importance of early identification and intervention before these behaviors become deeply ingrained and treatment resistant. Modest preventive efforts may avert later, more severe, behavior problems in a majority of cases. The study of intensity of early intervention for SBD needed would be a fruitful direction for future research, as has occurred in the research on early intervention for skill acquisition for children with autism.

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References

- Aman, MG.; Singh, NN. Aberrant Behavior Checklist-Community. East Aurora, NY: Slosson Education Publications; 1994. Supplementary Manual
- Aman MG, Singh NN, Stewart AW, Field CJ. The Aberrant Behavior Checklist: A behavior rating scale for the assessment of treatment effects. *American Journal of Mental Deficiency*. 1985; 89:485–491. [PubMed: 3993694]
- Arron K, Oliver CC, Moss J, Berg K, Burbridge C. The prevalence and phenomenology of self-injurious behavior in genetic syndromes. *Journal of Intellectual Disability Research*. 2011; 55:109–120. [PubMed: 20977515]
- Baker BL, Blacher J, Crnic KA, Edelbrock C. Behavior problems and parenting stress in families of three-year-old children with and without developmental delays. *American Journal on Mental Retardation*. 2002; 107:433–444. [PubMed: 12323068]

- Bayley, N. Manual of the Bayley Scales of Infant and Toddler Development. Third. San Antonio, TX: NCS Pearson, Inc; 2006.
- Berkson, G. Abnormal stereotyped motor acts. In: Zubin, J.; Hunt, HF., editors. Comparative psychopathology. New York, NY: Grune & Stratten; 1967. p. 76-91.
- Berkson G. Early development of stereotyped and self-injurious behaviors: II. Age trends. American Journal on Mental Retardation. 2002; 107:468–477. [PubMed: 12323071]
- Berkson G, Tupa M. Early development of stereotyped and self-injurious behaviors. Journal of Early Intervention. 2000; 23:1–19.
- Berkson G, Tupa M, Sherman L. Early development of stereotyped and self-injurious behaviors: I. Incidence. American Journal on Mental Retardation. 2001; 106:539–547. [PubMed: 11708939]
- Bodfish JW, Symons FW, Parker DE, Lewis MH. Varieties of repetitive behavior in autism: Comparisons to mental retardation. Journal of Autism and Developmental Disorders. 2000; 30:237–243. [PubMed: 11055459]
- Bruininks, RH.; Hill, BK.; Weatherman, RF.; Woodcock, RW. Inventory for Client and Agency Planning. Itasca, IL: Riverside; 1986.
- Carr EG, Smith CE. Biological setting events for self-injury. Mental Retardation and Developmental Disabilities Research Reviews. 1995; 1:94–98.
- Cohen, J. Statistical power for the behavioral sciences. 2nd. Hillsdale, NJ: Erlbaum; 1988.
- Damiano CR, Nahmias A, Hogan-Brown A, Stone WL. What do repetitive and stereotyped movements mean for infant siblings of children with autism spectrum disorders? Journal of Autism and Developmental Disorders. 2013; 43:1326–1335. [PubMed: 23080207]
- Dawson G. Brief report: Neuropsychology of autism: A report on the state of the science. Journal of Autism and Developmental Disorders. 1996; 26:179–184. [PubMed: 8744481]
- Dunlap G, Strain PS, Fox L, Carta JJ, Conroy M, Smith BJ, et al. Sowell C. Prevention and intervention with young children's challenging behavior: Perspectives regarding current knowledge. Behavior Disorders. 2006; 32:29–45.
- Fodstad JC, Rojahn J, Matson JL. The emergence of challenging behaviors in at-risk toddlers with and without Autism Spectrum Disorders: A cross-sectional study. Journal of Developmental and Physical Disabilities. 2012; 24:217–234. <http://dx.doi.org/10.1007/s10882-011-9266-9>.
- Guess D, Carr E. Emergence and maintenance of stereotypy and self-injury. American Journal on Mental Retardation. 1991; 96:299–320. [PubMed: 1756034]
- Hastings RP. Do challenging behaviors affect staff psychological well-being? Issues of causality and mechanism. American Journal on Mental Retardation. 2002; 107:455–467. [PubMed: 12323070]
- Iwata BA, Pace GM, Dorsey MF, Zarcone JR, Vollmer TR, Smith RG, et al. Willis KD. The functions of self-injurious behavior: An experimental-epidemiological analysis. Journal of Applied Behavior Analysis. 1994; 27:215–240. [PubMed: 8063623]
- Karabekiroglu K, Aman MG. Validity of the Aberrant Behavior Checklist in a clinical sampler of toddlers. Child Psychiatry and Human Development. 2009; 40:99–110. <http://dx.doi.org/10.1007/s10578-008-0108-7>. [PubMed: 18600444]
- Keller KM, Fox RA. Toddlers with developmental delays and challenging behaviors. Early Child Development and Care. 2009; 170:87–92. <http://dx.doi.org/10.1080/03004430600987126>.
- Kravitz H, Boehm JJ. Rhythmic habit patterns in infancy: Their sequence, age of onset, and frequency. Child Development. 1971; 42:399–413. [PubMed: 4104217]
- Kurtz PF, Huete JM, Cataldo MF, Chin MD. Identification of emerging self-injurious behavior in young children: A preliminary study. Journal of Mental Health Research in Intellectual Disabilities. 2012; 5:260–285. [PubMed: 22844389]
- Lewis MH, Baumeister AA. Stereotyped mannerisms in mentally retarded persons: animal models and theoretical analysis. International Review of Research in Mental Retardation. 1982; 11:123–161.
- Lovaas OI. Behavioral treatment and normal educational and intellectual functioning in young autistic children. Journal of Consulting and Clinical Psychology. 1987; 55:3–9. [PubMed: 3571656]
- MacLean WE, Dornbush K. Self-injury in a state-wide sample of young children with developmental disabilities. Journal of Mental Health Research in Intellectual Disabilities. 2012; 5:236–245. <http://dx.doi.org/10.1080/19315864.2011.590627>.

- MacLean WE, Tervo RC, Hoch J, Tervo M, Symons FJ. Self-injury among a community cohort of young children at risk for intellectual and developmental disabilities. *The Journal of Pediatrics*. 2010; 157:979–983. [PubMed: 20630541]
- Marshburn EC, Aman MG. Factor validity and norms for the Aberrant Behavior Checklist in a community sample of children with mental retardation. *Journal of Autism and Developmental Disorders*. 1992; 22:357–373. [PubMed: 1383187]
- Matson JL. Aggression and tantrums in children with autism: A review of behavioral treatments and maintaining variables. *Journal of Mental Health Research in Intellectual Disabilities*. 2009; 2:169–187. <http://dx.doi.org/10.1080/19315760902725675>.
- Matson JL, Cooper C, Malone CJ, Moskow SL. The relationship of self-injurious behavior and other maladaptive behaviors among individuals with severe and profound intellectual disability. *Research in Developmental Disabilities*. 2008; 29:141–148. <http://dx.doi.org/10.1016/j.ridd.2007.02.001>. [PubMed: 17350800]
- Matson JL, Dempsey T, Fodstad JC. Stereotypies and repetitive/restrictive behaviors in infants with autism and pervasive developmental disorder. *Developmental Neurorehabilitation*. 2009; 12:122–127. [PubMed: 19466619]
- Matson JL, Dixon DR, Matson ML. Assessing and treating aggression in children and adolescents with developmental disabilities: A 20-year review. *Educational Psychology*. 2005; 25:151–181. <http://dx.doi.org/10.1080/0144341042000301148>.
- Matson JL, Fodstad JC, Dempsey T. What symptoms predict the diagnosis of autism or PDD-NOS in infants and toddlers with developmental delays using the Baby Infant Screen for aUtism Traits. *Developmental Neurorehabilitation*. 2009; 12:381–388. <http://dx.doi.org/10.31/17518420903029501>. [PubMed: 20205546]
- Matson JL, Fodstad JC, Mahan S, Rojahn J. Cut-off norms and patterns of problem behavior in children with developmental disabilities Baby and Infant Screen for Children with aUtism Traits (BISCUIT-Part 3). *Developmental Neurorehabilitation*. 2010; 13:3–9. [PubMed: 20067340]
- Matson JL, Hess JA, Boisjoli JA. Comorbid psychopathology in infants and toddlers with autism and pervasive developmental disorders-not otherwise specified (PDD-NOS). *Research in Autism Spectrum Disorders*. 2010; 4:300–304. <http://dx.doi.org/10.1016/j.rasd.2009.10.00>.
- Matson JL, Mahan S, Sipes M, Kozlowski AM. Effects of symptoms of comorbid psychopathology on challenging behaviors among typically developing infants and toddlers as assessed with the Baby and Infant Screen for Children with aUtism Traits (BISCUIT). *Journal of Mental Health Research in Intellectual Disabilities*. 2010; 3:164–176. <http://dx.doi.org/10.1080/19315864.21010.495920>.
- Matson JL, Tureck K. Early diagnosis of autism: Current status of the Baby and Infant Screen for Children with aUtism Traits (BISCUIT-Parts 1,2,3). *Research in Autism Spectrum Disorders*. 2012; 6:1135–1141. <http://dx.doi.org/10.1016/j.rasd.2012.02.009>.
- Mayo-Ortega L, Oyama-Ganiko R, LeBlanc J, Schroeder SR, Brady N, Butler MG, et al. Marquis J. Mass screening for severe problem behavior among infants and toddlers in Peru. *Journal of Mental Health Research in Intellectual Disabilities*. 2012; 5:246–259. <http://dx.doi.org/10.1080/1935864.2011.590626>. [PubMed: 22962573]
- McClintock K, Hall S, Oliver C. Risk markers associated with challenging behaviors in people with intellectual disabilities: A meta-analytic study. *Journal of Intellectual Disability Research*. 2003; 47:405–416. [PubMed: 12919191]
- Medeiros K, Kozlowski AM, Beighly JS, Rojahn J, Matson JL. The effects of developmental quotient and diagnostic criteria on challenging behaviors in toddlers with developmental disabilities. *Research in Developmental Disabilities*. 2012; 33:1110–1116. <http://dx.doi.org/10.1016/j.ridd.2012.02.005>. [PubMed: 22502836]
- Murphy G, Hall S, Oliver C, Kissi-Debra R. Identification of early self-injurious behavior in young children with intellectual disability. *Journal of Intellectual Disability Research*. 1999; 43:149–163. [PubMed: 10392602]
- Newborg, J. *Batelle Development Inventory*. 2nd. Itasca, IL: Riverside; 2005.
- Oliver C, Petty, Ruddick L, Bacarese-Hamilton M. The relationship between repetitive, self-injurious and aggressive behavior in children with severe intellectual disabilities. *Journal of Autism and Developmental Disorders*. 2012; 49:910–919.

- Oyama-Ganiko R, Mayo-Ortega L, Schroeder SR, LeBlanc JM. Early distance intervention and follow-up for families of young children at risk for developmental disabilities and severe behavior problems in Peru. *Revista Educacao Especial*. 2013; 26:541–555.
- Rethmeyer JA, Tan X, Manzardo A, Schroeder SR, Butler MG. Comparison of biological specimens and DNA collection methods for PCR amplification and microarray analysis. *Clinical Chemical Laboratory Medicine*. 2012; 1505:1–5.
- Richards C, Oliver C, Nelson L, Moss J. Self-injurious behavior in individuals with autism spectrum disorders and intellectual disability. *Journal of Intellectual Disability Research*. 2012; 56:476–489. [PubMed: 22404122]
- Richman DM. Early intervention and prevention of self-injurious behavior exhibited by young children with developmental disabilities. *Journal of Intellectual Disability Research*. 2008; 52:3–17. [PubMed: 18173568]
- Richman D, Lindauer S. Stereotypies, proto-injurious and self-injurious behavior in young children with developmental delays. *American Journal on Mental Retardation*. 2005; 110:439–450. [PubMed: 16212447]
- Rojahn J, Matson JL, Lott D, Esbensen AJ, Smalls Y. The Behavior Problem Inventory: An instrument for the assessment of self-injury, stereotyped behavior, and aggression/destruction in individuals with developmental disabilities. *Journal of Autism and Developmental Disorders*. 2001; 31:577–588. [PubMed: 11814269]
- Rojahn J, Rowe EW, Sharber AC, Hastings RP, Matson JL, Didden DBH, et al. Dumont ELM. Behavior Problems Inventory-Short form (BPI-S) for individuals with intellectual disabilities I: development and clinical norms. *Journal of Intellectual Disability Research*. 2012; 56:527–545. [PubMed: 22151184]
- Rojahn, J.; Schroeder, SR.; Hoch, TA. Self-injurious behavior in intellectual disabilities. New York, NY: Elsevier; 2008.
- Rojahn J, Schroeder SR, Mayo-Ortega L, Oyama-Ganiko R, LeBlanc J, Marquis J, Berke E. Multi-method assessment of behavior problems among infants and toddlers at risk for neurodevelopmental disorders. *Research in Developmental Disabilities*. 2013; 34:1804–1814. [PubMed: 23511345]
- Sameroff, AJ. The transactional model of development: How children and contexts shape each other. Washington, DC: American Psychological Association; 2009.
- Sandman CA, Kemp AS, Mabini G, Pincus D, Magnussen M. Self-injurious behavior: The role of self-injurious behaviour in the organisation of behaviour. *Journal of Intellectual Disability Research*. 2012; 56:516–526. [PubMed: 22452417]
- Schopler, E.; Reichler, RJ.; Renner, BR. The Child Autism Rating Scale. Los Angeles, CA: Western Psychological Services Corporation; 1988.
- Schroeder SR. Usage of stereotypy as a descriptive term. *Psychological Record*. 1970; 20:337–342.
- Schroeder SR, Courtemanche AB. Early prevention of severe behavior disorders: An integration. *Journal of Mental Health Research in Intellectual Disabilities*. 2012; 5:203–214. <http://dx.doi.org/10.1080/19315864.2011.593697>. [PubMed: 23139733]
- Schroeder, SR.; Kanoy, R.; Thios, S.; Mulick, JA.; Rojahn, J.; Stephens, M. Antecedent conditions affecting management and maintenance of programs for the chronically self-injurious. In: Hollis, J.; Meyers, CE., editors. Life threatening behavior. Washington, DC: AAMD Monograph Series, No. 5; 1982. p. 105-159.
- Schroeder, SR.; Loupe, PS.; Tessel, RE. Animal models of self-injurious behavior: Induction, prevention, and recovery. In: Glidden, LM., editor. International review of research in mental retardation. Vol. 36. New York, NY: Elsevier; 2008. p. 195-231.
- Schroeder SR, Mulick JA, Rojahn J. The definition, taxonomy, epidemiology, and ecology of self-injurious behavior. *Journal of Autism and Developmental Disorders*. 1980; 10:417–432. [PubMed: 6985454]
- Schroeder SR, Rojahn J, Xiaohu An, Mayo-Ortega L, Oyama-Ganiko R, LeBlanc JM. The Parental Concerns Questionnaire: A valid brief screening instrument for severe behavior problems among infants and toddlers at risk for developmental delays. *Journal of Developmental and Physical Disabilities*. 2013; 26:237–247. <http://dx.doi.org/10.1007/s10882-013-9359-8>.

- Schroeder SR, Richman DM, Abby L, Courtemanche AB, Oyama-Ganiko R. Functional analysis outcomes and comparison of direct observations and informant rating scales in the assessment of severe behavior problems of infants and toddlers at-risk for developmental delays. *Journal of Developmental and Physical Disabilities*. 26:325–334. 2014. <http://dx.doi.org/10.1007/s10882-014-9386-2>.
- Symons FJ, Sperry LA, Dropik PL, Bodfish JW. The early development of stereotypy and self-injury: a review of research methods. *Journal of Intellectual Disability Research*. 2005; 49:144–158. [PubMed: 15634323]
- Tate BG, Baroff GS. Aversive control of self-injurious behavior in a psychotic boy. *Behavior Research and Therapy*. 1968; 4:281–287.
- Taylor L, Oliver C, Murphy G. The chronicity of self-injurious behavior: A long term follow-up: A total population study. *Journal of Applied Research in Intellectual Disabilities*. 2011; 24:105–117.
- Thelen E. Rhythmical stereotypies in normal human infants. *Animal Behavior*. 1979; 27:699–715.
- Thelen, E. Normal infant stereotypies: A dynamic systems approach. In: Sprague, RL.; Newell, KM., editors. *Stereotyped movements: Brain and behavior relationships*. Washington, DC: American Psychological Association; 1996.
- Usrey KM, Manzardo AM, Roberts JL, Schroeder SR, Butler MG. Genetic analysis in a cohort of Peruvian children with atypical development and/or aberrant behavior. under review.
- Wacker D, Berg W, Harding J, Derby M, Asmus J, Healy A. Evaluation and long-term treatment of aberrant behavior displayed by young children with disabilities. *Developmental and Behavioral Pediatrics*. 1998; 19:260–266.
- Wacker D, Harding JW, Berg WK, Lee JF, Schieltz KM, Padilla YC, et al. Shahan TA. An evaluation of persistence of treatment effects during long-term treatment of destructive behavior. *Journal of the Experimental Analysis of Behavior*. 2011; 96:261–282. [PubMed: 21909168]
- Wacker DP, Harding JW, Morgan TA, Berg WK, Schieltz KM, Lee J. An evaluation of resurgence during Functional Communication Training. *Psychological Record*. 2013; 63:3–20.
- Wetherby, AM.; Prizant, BM. *Communication and Symbolic Behavior Scales Developmental Profile*. Baltimore, MD: Paul Brookes Publishing Co., Inc; 2002.

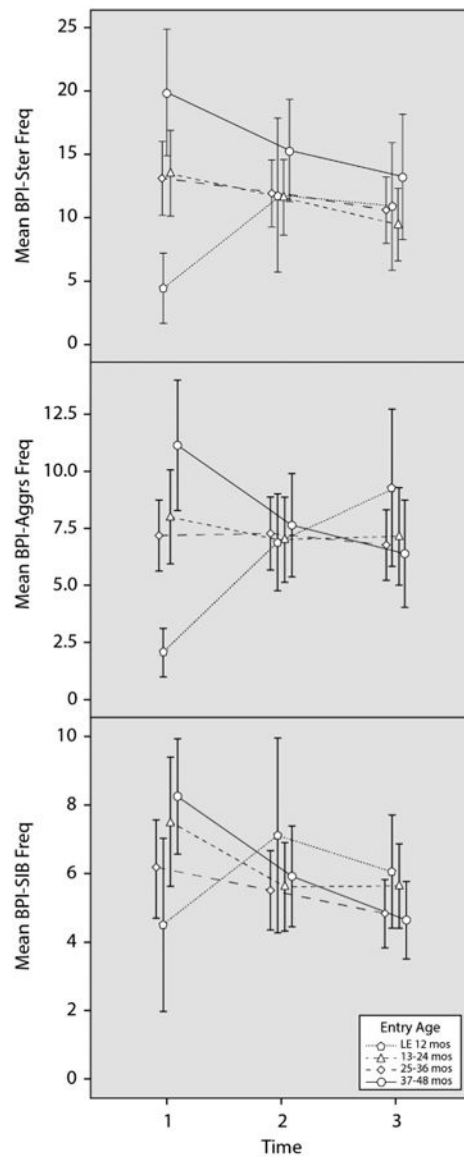


Figure 1.

Frequency of severe behavior disorders (SBDs) as a function of Age at Times 1, 2, 3 (birth, 6, 12 months). Top panel is mean frequency of stereotyped behavior; middle panel is mean frequency of aggression; bottom panel is mean frequency of self-injurious behavior (SIB). Means and standard deviations can be found in Table 8.

Note: BPI-01 = Behavior Problems Inventory.

Table 1
Item Topographies for Frequency of SIB, Stereotyped Behavior, and Aggression on the BPI-01

SIB Items (abbr.)	Mean	Stereotyped behavior	Mean	Aggression	Mean
Self-biting	.39	Rocking Back and forth	.73	Hitting others	1.22
Hits head	1.11	Sniffing objects	.41	Kicking others	.64
Hits body (not head)	.31	Spinning own body	.71	Pushing others	.61
Self-scratching	.31	Waving/shaking arms	.88	Biting others	.99
Vomits/ruminates	.42	Rolling head	.48	Grab/pull others	.90
Pinches self	.23	Whirling around	.56	Scratching others	.63
Pica	1.00	Repetitive body moves	.49	Pinching others	.82
Inserts objects in orifices	.19	Pacing	.58	Spitting on others	.18
Pulls finger or toe nails	.12	Twirling objects	.54	Verbal abuse	.03
Inserts fingers in orifices	.49	Repetitive hand moves	.76	Destroying things	.93
Air-swallowing	.24	Yelling and screaming	1.04	Being mean or cruel	.45
Hair-pulling	.75	Sniffing own body	.17		
Teeth-grinding	.12	Bouncing around	.66		
Extreme drinking	.98	Spinning objects	.46		
		Bursts of running	.66		
		Complex hand moves	.47		
		Manipulating objects	.79		
		Sustained finger moves	.38		
		Rubbing self	.22		
		Gazing at hands or objects	.95		
		Maintains bizarre postures	.31		
		Clapping hands	.36		
		Grimacing	.57		
		Waving hands	.12		

Note: SIB = self-injurious behavior; BPI-01 = Behavior Problems Inventory.

Table 2

BPI-01 Frequency Scores as a Function of Gender

Variable	Female (<i>n</i> = 68)		Male (<i>n</i> = 112)		<i>F</i> (1,177)	<i>p</i>	η^2
	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>			
SIB	5.99	5.94	7.25	6.25	1.80	.181	.010
Stereotypy	10.76	11.82	15.55	13.54	5.79	.017	.032
Aggression	6.63	7.27	8.33	7.43	2.25	.136	.013

Note: BPI-01 = Behavior Problems Inventory; SIB = self-injurious behavior.

Table 3
BPI-01 Frequency Scores as a Function of Age (months) at Entry

Variable	4-12 (n = 18)		13-24 (n = 49)		25-36 (n = 77)		37-48 (n = 36)		F(1,75)	p	η^2
	M	SD	M	SD	M	SD	M	SD			
SIB	4.50	5.36	7.51	6.60	6.13	6.31	8.25	5.05	2.08	.105	.034
Stereotypy	4.44	5.87	13.51	11.85	13.10	12.76	19.89	14.99	6.28	.000	.097
Aggression	2.06	2.24	8.00	7.22	7.18	6.84	11.14	8.57	6.90	.000	.105

Note: BPI-01 = Behavior Problems Inventory; SIB = self-injurious behavior.

Table 4
BPI -01 Frequency Scores as a Function of Primary Clinical Diagnosis

Variable	Down syndrome (<i>n</i> = 45)		At-risk for autism (<i>n</i> = 74)		Atypical development (<i>n</i> = 61)				
	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>	<i>F</i> (1,177)	<i>p</i>	η^2
SIB	4.56	4.35	8.77	6.94	5.97	5.51	7.98	.000	.083
Stereotypy	7.49	9.95	21.16	13.15	9.25	10.40	26.66	.000	.232
Aggression	5.47	5.96	9.92	8.27	6.61	6.47	6.45	.002	.068

Note: BPI-01 = Behavior Problems Inventory; SIB = self-injurious behavior.

Table 5
BPI-01 Frequency Scores as a Function of Intellectual Level (BSID III)

Variable	BSID Low (<i>n</i> = 100)		BSID High (<i>n</i> = 59)		<i>F</i> (1,117)	<i>p</i>	η^2
	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>			
SIB	6.24	5.22	8.00	7.43	3.06	.082	.019
Stereotypy	14.26	12.82	14.73	13.83	.047	.829	.000
Aggression	6.95	6.83	9.97	8.18	6.24	.013	.038

Note: BPI-01 = Behavior Problems Inventory; BSID = Bayley Scales of Infant Development; SIB = self-injurious behavior.

Table 6
BPI-01 Frequency Scores as a Function of Communication Level

Variable	CSBS Lowest quartile (<i>n</i> = 38)		CSBS Highest quartile (<i>n</i> = 39)		<i>F</i> (1,75)	<i>p</i>	η^2
	<i>M</i>	<i>SD</i>	<i>M</i>	<i>SD</i>			
SIB	6.11	5.32	6.28	6.72	.016	.899	.000
Stereotypy	12.84	13.20	12.10	13.48	.059	.808	.001
Aggression	5.03	5.85	10.38	8.61	10.10	.002	.110

Note: BPI-01 = Behavior Problems Inventory; CSBS = Communication, Symbolic, and Behavior Assessment Scale; SIB = self-injurious behavior.

Table 7
Multiple Regression Analyses of Risk Factors for SIB, Stereotyped Behavior, and Aggression

Factor	Unique contribution		
	SIB	Stereotypy	Aggression
CSBS	.003	.016	.007
Bayley	.000	.003	.006
Age	.000	.023	.006
Gender	.003	.015	.010
Vision	.010	.001	.017
Income	.030	.019	.008
Parent Education	.002	.004	.020
Diagnosis	.037	.118	.025
	$R^2 = .152$	$R^2 = .350$	$R^2 = .235$

Note: CSBS = Communication, Symbolic, and Behavior Assessment Scale; Bayley = Bayley Scales of Infant Development; SIB = self-injurious behavior.

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Table 8
Mean BPI-01 Subscale Frequency Scores as a Function of Age at Times 1, 2, 3

Entry age	SIB			Stereotypy			Aggression			
	Time 1	Time 2	Time 3	Time 1	Time 2	Time 3	Time 1	Time 2	Time 3	
0–12 months	<i>M</i>	4.50	7.11	6.06	4.44	11.78	10.89	2.06	6.89	9.26
	<i>SD</i>	5.36	6.03	3.51	5.87	12.89	10.67	2.24	4.51	7.32
13–24 months	<i>M</i>	7.51	5.61	5.63	13.51	11.61	9.45	8.00	7.00	7.14
	<i>SD</i>	6.60	4.53	4.31	11.85	10.40	9.98	7.22	6.53	7.49
25–36 months	<i>M</i>	6.13	5.51	4.83	13.10	11.92	10.60	7.18	7.27	6.77
	<i>SD</i>	6.31	5.09	4.35	12.76	11.54	11.45	6.80	7.03	6.79
37–48 months	<i>M</i>	8.25	5.92	4.64	19.89	15.31	13.25	11.14	7.64	6.39
	<i>SD</i>	5.05	4.42	3.37	14.99	12.10	14.86	8.57	6.79	7.02

Note: BPI-01 = Behavior Problems Inventory; SIB = self-injurious behavior.

Table 9
Mean BPI-01 Subscale Frequency Scores as a Function of Diagnosis at Times 1, 2, 3

Diagnosis	SIB			Stereotypy			Aggression		
	Time 1	Time 2	Time 3	Time 1	Time 2	Time 3	Time 1	Time 2	Time 3
At Risk for Autism	<i>M</i> 8.77	6.41	5.58	21.16	16.96	14.81	9.92	8.86	8.09
	<i>SD</i> 6.94	5.48	4.45	13.15	12.52	13.69	8.27	7.77	7.86
Down syndrome	<i>M</i> 4.56	4.38	4.82	7.49	9.27	8.73	5.47	4.93	6.18
	<i>SD</i> 4.35	3.61	3.94	9.95	10.26	11.79	5.96	3.88	6.74
Other	<i>M</i> 5.97	6.05	4.82	9.25	9.48	7.59	6.61	6.95	6.41
	<i>SD</i> 5.51	4.85	3.70	10.40	9.16	7.66	6.47	6.14	6.22

Note: BPI-01 = Behavior Problems Inventory; SIB = self-injurious behavior.