An Illustration of Using Multiple Imputation Versus Listwise Deletion Analyses: The Effect of Hanen's "More Than Words" on Parenting Stress

Rebecca G. Lieberman-Betz, Paul Yoder, Wendy L. Stone, Allison S. Nahmias, Alice S. Carter, Seniz Celimli-Aksoy, and Daniel S. Messinger

Abstract

This investigation illustrates the effects of using different missing data analysis techniques to analyze effects of a parent-implemented treatment on stress in parents of toddlers with autism symptomatology. The analysis approaches yielded similar results when analyzing main effects of the intervention, but different findings for moderation effects. Using listwise deletion, the data supported an iatrogenic effect of Hanen's "More Than Words" on stress in parents with high levels of pretreatment depressive symptoms. Using multiple imputation, a significant moderated treatment effect with uninterpretable regions of significance did not support an iatrogenic effect of treatment on parenting stress. Results highlight the need for caution in interpreting analyses that do not involve validated methods of handling missing data.

Key Words: missing data analysis; multiple imputation; autism spectrum disorder; early intervention; parent stress

Stories may be more persuasive than data for many people. In scientific communities, however, we need both. An effective "story" for many scientists interested in developmental disabilities might be a well-implemented treatment-efficacy study that focuses on a topic they care about. Such an illustration about missing data appears necessary because most treatment-efficacy studies in psychology and education do not yet handle missing data appropriately. The current investigation is an attempt to illustrate the value of missing data imputation using a study on the effects of teaching parents to use a treatment method with their very young children with autism, a difficult-to-test population. Difficult-to-test populations and highly stressed families are arguably more prone to producing missing data than other populations.

Randomized Controlled Trials, Intent to Treat, and Missing Data

Well-conducted randomized controlled trials (RCTs) and an assumption of nonsignificant

pretreatment differences between groups on all relevant variables allow for causal inferences of treatment effects (Shadish, Cook, & Campbell, 2002). In addition, RCTs provide opportunities to examine putative moderators of treatment effects. It is now widely accepted in the medical and epidemiology fields that the most internally valid tests of randomized clinical trials involve analyzing all participants assigned to treatment groups regardless of treatment-protocol compliance or presence of data at particular measurement periods (i.e., intent-to-treat [ITT], analyses) (Lachin, 2000; Newell, 1992). The rationale for this preference is that random assignment of participants to groups is more effective in creating groups that are comparable on all relevant variables when ITT principles are followed than when they are not (Lachin, 2000). Importantly, randomization has been found to maximize the probability of pretreatment comparability even on variables that are unmeasured. Comparability on *all* relevant (i.e., correlated with the outcome) variables, even those that are unmeasured, is needed for most threats to internal

validity to be controlled in between-group experiments (Shadish et al., 2002).

One challenge in the analysis of longitudinal data (including RCTs) in the behavioral sciences, however, is missing data (Enders, 2011). In fact, missing data in quantitative studies involving longitudinal measurement is ubiquitous and can occur for a variety of reasons, including (a) participants missing entire measurement periods, (b) participants partially completing measures during a specific measurement period (e.g., skipping items on a questionnaire, not completing all of the questionnaires), and (c) participants failing to complete the study (Schafer & Graham, 2002). Rubin and colleagues (Little & Rubin, 2002; Rubin, 1976) described three ways in which data can be missing: (a) missing completely at random (MCAR), (b) missing at random (MAR), and (c) missing not at random (MNAR). Patterns of missing data that are MCAR have no correlation with any other measured variables, or to the variable containing the missing data; data that are MAR are related to other measured variables, but not to the variable containing the missing values; and data that are MNAR are related to the variable containing the missing values (Baraldi & Enders, 2010; Enders, 2011). Analyzing data only from participants who complete testing (listwise deletion) has been shown to reduce the probability that randomization produces comparable treatment groups on all relevant pretreatment variables (Lachin, 2000). Thus, analyses that account for the missing data must be conducted for researchers to draw appropriate conclusions from RCT findings (Moher, Schulz, & Altman, 2001).

Multiple imputation (MI) is one of two recommended methods for analyzing data sets with missing data (Enders, 2010). Multiple imputation methods have been shown to provide unbiased parameter estimates with small sample sizes (Graham & Shafer, 1999; Schlomer, Bauman, & Card., 2010), making MI useful for research involving people with developmental disabilities, which often involves small participant sample sizes. For example, a simulation study by Barnes, Lindborg, and Seaman (2006) demonstrated little effect of small sample size on the standardized bias of mean change using several methods of imputation, including the Bayesian least-squares method, which serves as the foundation for the multiple imputation method described in the current article.

Despite consensus from methodologists regarding the importance of addressing missing data in RCTs, missing-data techniques such as MI are still not in widespread use by treatment researchers (Baraldi & Enders, 2010; Enders, 2011; Horton & Kleinman, 2007). For example, in a review of 71 RCTs published in four highly regarded medical journals, Wood, White, and Thompson (2004) found that of the 63 trials with missing data, 65% used listwise deletion methods, which involve removing participants with missing data from analyses. Listwise deletion has been found to bias parameter estimates (e.g., Buhi, Goodson, & Neilands, 2008; Newman, 2003). More recently, Schlomer et al. (2010) found that across 37 articles reporting quantitative data published in a single volume of the Journal of Counseling Psychology, only 14 articles reported the percentage of data missing, and 11 of them used listwise deletion methods in their analyses. Finally, previous criteria from the What Works Clearinghouse (WWC, 2010) for determining whether an intervention meets "evidence standards" for effectiveness made no mention of how to address missing data in experimental or quasi-experimental research designs. (WWC, of the Institute of Education Sciences, is a resource for educators to determine the evidence base supporting instructional practices. Teams of reviewers systematically review the education research using specific standards developed for evaluating group and single-subject design studies.) Current WWC criteria (2014) state a preference for conducting analyses on observed data (i.e., those cases where data are not missing) unless a study does not meet their standards for attrition. In this case, WWC provides listwise deletion and multiple imputation (among others) as equally acceptable means of handling missing data. Indeed, the criteria indicate listwise deletion is the "most straightforward" approach to handling missing data.

Simulation studies have been valuable in showing that missing data can have serious consequences on the interpretation of findings in group design studies (e.g., Ibrahim, Chen, Lipsitz, & Herring, 2005). It appears, however, these simulations have not convinced treatment researchers in psychology, education, and related sciences because widespread adoption of appropriate methods for handling missing data is not occurring. The current article provides an accessible, real-world example of why appropriate handling of missing data is integral to analysis

of RCT data and the building of a solid evidence base in treatment research. Therefore, the purpose of this article is to compare the results of two missing data analysis techniques, listwise deletion and multiple imputation, in addressing research questions regarding parental well-being and parent-implemented intervention for young children with autism spectrum disorders (ASD).

The Example Study

Parents Are Being Asked to Implement Treatments for Young Children With Autism Spectrum Disorder (ASD)

Recognizing the central role of caregiving relationships in early childhood (Sameroff & Emde, 1989), the majority of recently developed interventions for very young children with ASD have been parent-implemented (Dawson et al., 2010; Meadan, Ostrosky, Zaghlawan, & Yu, 2009; Vismara & Rogers, 2010). There is evidence that parents can be taught to implement early interventions with comparable fidelity to professionals (Ingersoll & Gergans, 2007). A recent report indicates that Hanen's "More Than Words" (HMTW) intervention benefits children's communication if the children begin treatment with low object interest (Carter et al., 2011). Yet, as parents are placed in roles in which they provide or support their children's early interventions, it becomes critical to assess the impact of these roles on parental well-being.

Parenting Stress and Parent-Implemented Treatments in Families of Children With ASD

Parents of children with ASD report significantly higher levels of parenting stress than parents of children with other developmental disorders and parents of typically developing children (Baker-Ericzén, Brookman-Frazee, & Stahmer, 2005; Dumas, Wolf, Fisman, & Culligan, 1991; Estes et al., 2013 Noh, Dumas, Wolf, & Fisman, 1989). Elevated levels of stress have been well documented in parents of older children with ASD (Bouma & Schweitzer, 1990; Dumas et al., 1991; Holroyd & McArthur, 1976; Rodrigue, Morgan, & Geffken, 1990; Sanders & Morgan, 1997), as well as in parents rearing very young children with ASD (Baker-Ericzén et al., 2005; Davis & Carter, 2008; Estes et al., 2013; Hastings & Johnson, 2001). Emerging evidence suggests that

participation in a parent skills training program can reduce parenting stress and improve parental mental health outcomes for parents rearing a child with ASD (Tonge et al., 2006; Whittingham, Sofronoff, Sheffield, & Sanders, 2009), including those with children who have been recently diagnosed (Tonge et al., 2006).

Most parent-implemented interventions for young children with ASD focus on providing parents with strategies for enhancing their children's social and communicative behaviors. Such treatments might influence parenting stress, in part, because the core social and communication deficits in ASD have been associated with parenting stress in parents of children with ASD (Baker-Ericzén et al., 2005; Davis & Carter, 2008; Donenberg & Baker, 1993; Herring et al., 2006; Lecavalier, Leone, & Wiltz, 2006; Tomanik, Harris, & Hawkins, 2004). Preliminary evidence suggests that even low-intensity parent-implemented interventions may be effective in reducing parenting stress (Wong & Kwan, 2010). HMTW is a parent-implemented intervention that teaches parents strategies for supporting their children's social-communication development within 11 group and individual sessions over the course of 12 weeks. Based on previous findings, it is possible that such a low-intensity treatment targeting a primary deficit area in children with ASD could affect the stress levels in parents implementing the treatment.

Research has also found that parent-implemented interventions can increase parenting stress (Brinker, Seifer, & Sameroff, 1994; Osborne, McHugh, Saunders, & Reed, 2008; Sameroff & Emde, 1989), suggesting that some parents are more vulnerable than others to experiencing participation in parent-implemented interventions as stressful. The possibility that some parents may experience *more stress* when asked to implement treatment does not address the question of which parent characteristics might be used to identify parents who are susceptible to such iatrogenic effects of parent-implemented treatments on parental stress.

Parents' Depressive Symptoms as a Potential Moderator of Parent-Implemented Treatment on Parenting Stress

Parents rearing children with ASD have also been found to be at increased risk for depressive

symptoms (Benson, 2006; Davis & Carter, 2008; Dumas et al., 1991). Moreover, initial levels of mothers' depressive symptoms appear to remain high throughout the preschool years (Carter, Martínez-Pedraza, & Gray, 2009). Given that feelings of hopelessness and self-critical cognitions are central to depression, one can imagine that asking parents who already experience depressive symptoms to take on the challenge of learning new parenting skills as part of their child's early intervention program could result in increasing their stress. In contrast, one can also imagine that if parents do not feel depressed, learning about new ways to foster their children's development may reduce parenting stress. Based on research and previous understanding of patterns of stress and depression in parents of children with ASD, parent depressive symptoms might function as a moderator of treatment effects.

Research Questions

In the current study, we used two data-analysis methods, listwise deletion and MI analyses, to examine (a) the extent to which Hanen's "More Than Words" (HMTW; Sussman, 2004), a parentimplemented intervention designed specifically for parents of young children with autism, causes a reduction in parenting stress, and (b) whether parents' pretreatment depressive symptoms moderate the effects of HMTW on changes in parenting stress. For parents with relatively low depressive symptoms before treatment, the prediction was that parents in the HMTW group would experience more decrease in parental stress compared to the control group. There was not an a priori prediction made for the parents with high depressive symptoms. This analysis is a follow-up to another article presenting the lack of main effects of HMTW on child outcomes but a moderated HMTW effect on child communication when children had low object interest (Carter et al., 2011).

Method

Participants

Participants were 62 children (51 boys and 11 girls) and their parents (91% mothers). The mean age of the children was 21.3 months (SD = 2.8 months; range = 15.5–25.0 months) when they were enrolled in the study. Inclusion criteria were as follows: (a) a predetermined *Screening Tool*

for Autism in Two-year-olds (STAT, Stone, Coonrod, & Ousley, 2000; Stone, Coonrod, Turner, & Pozdol, 2004; Stone, McMahon, & Henderson, 2008) risk score of 2.25 for children under 24 months and 2.0 for children 24 months of age, and (b) a clinical impression of symptoms consistent with ASD. Data from these 62 families were included in MI analyses. Analyses using listwise deletion included only those participants with pretreatment data and at least one follow-up data point on the primary outcome measures (*n* = 48) (see Figure 1). The participant sample included in both sets of analyses was educationally and ethnically diverse compared to many studies of children with ASD (see Table 1).

Importantly, the presence of missing data was not associated with treatment attendance in the HMTW group. Within the treatment group, 28% of participants were missing the follow-up (Time 3) stress outcome measure (n = 9). To determine the proportion of participants assigned to the treatment group with missing outcomes who "completed" treatment, treatment "goers" were defined as those attending over half of treatment sessions (i.e., those who attended a total of six or more sessions). It was determined that 33% of participants assigned to the treatment group with missing Time 3 outcome data did not complete the treatment (n = 3), and 17% of those participants assigned to treatment without missing Time 3 outcome data did not complete the treatment (n = 4). The presence of missing outcome data was not significantly associated with treatment completion ($\Phi = .173$, p = .327).

Procedures

After parents provided IRB-approved informed consent, each family participated in a Time 1 pretreatment visit, at which time children received the Mullen Scales of Early Learning Expressive and Receptive Language Scales (MSEL; Mullen, 1995); parents were interviewed with the Vineland Adaptive Behavior Scales II Communication and Socialization domains (Vineland; Sparrow, Cicchetti, & Balla, 2005); and parents completed questionnaires about family demographics and the child's participation in "business as usual" interventions, along with standardized measures assessing stress and depression (see "Measures" section). Parents completed the same psychological well-being measures at a Time 2 posttreatment visit and a Time 3 follow-up visit, which occurred

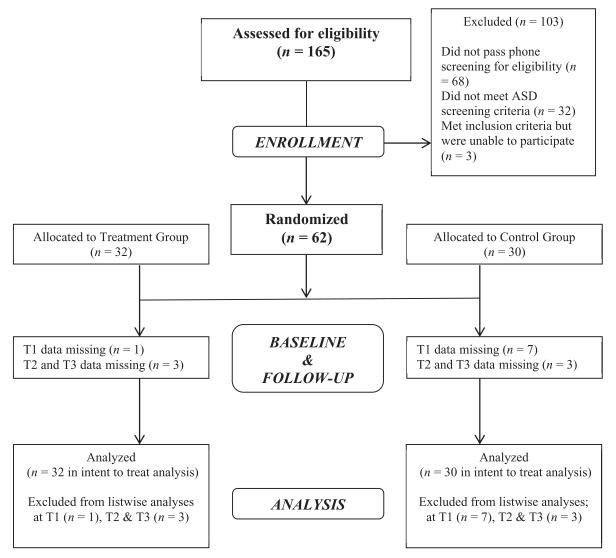


Figure 1. Flowchart of participants through various stages of the study for listwise deletion and multiply imputed study samples. *Note:* ASD = autism spectrum disorder; T1, T2, T3 = Time 1, Time 2, Time 3 (time points for treatment stages).

approximately five months (M = 5.3, SD = .47) and nine months (M = 9.3, SD = .56) after the Time 1 visit. Detailed information about the procedures and measures are provided in the first report of this study (Carter et al., 2011). Ninety-two percent of children who received Time 3 evaluations (46/50) met criteria for ASD based on both the *Autism Diagnostic Observation Schedule* (ADOS; Lord, Rutter, DiLavore, & Risi, 2002) and a clinical impression based on the *Diagnostic and Statistical Manual of Mental Disorders*, 4th Edition (DSM-IV). Two intervention and two control group participants did not meet ASD criteria at Time 3.

Hanen's "More Than Words" Intervention: Content and Fidelity Assessment

The aim of the HMTW intervention is to teach parents of young children with ASD strategies to enhance their children's communication. Strategies focus on improving two-way interactions, social skills, and understanding of language. The program involves eight group sessions (with parents only) and three in-home individualized parent-child sessions conducted by certified speech-language pathologists with specific training in HMTW. It is designed to be carried out in 12 weeks. During each session, parents are taught

Table 1
Time 1 Child and Parent Characteristics for MI Sample

	HMTW (n = 32)	Control $(n = 30)$
	Mean (SD)	Mean (SD)
CA (months)	21.1 (2.7)	21.5 (2.8)
Mullen Expressive Language Age (months)	8.2 (6.0)	7.3 (3.7)
Mullen Receptive Language Age (months)	8.4 (5.4)	8.2 (4.4)
Vineland Socialization Standard Score*	74.0 (6.5)	72.4 (6.7)
Vineland Communication Standard Score*	66.6 (12.9)	63.2 (9.1)
Parent Age*	33.7 (5.6)	35.5 (5.9)
Race* (%)		
White	64.5	57.7
Hispanic	29.0	34.6
Black	6.2	0
Asian	0	7.7
Education* (%)		
High school diploma	19.4	11.5
Some college/ 2 year or Associate's degree	32.3	34.6
College graduate	35.5	34.2
Graduate degree	16.1	19.2

Note. MI = Multiple imputation; HMTW = Hanen's "More Than Words" intervention; CA = chronological age; * As values for these variables were based on available data, ns vary.

to use everyday activities and play interactions as contexts for improving their child's communication and social skills. More information about HMTW, as implemented in the current study, is available in (Carter et al., 2011).

To assess the fidelity of treatment implementation, the speech-language pathologists who administered the intervention completed a set of checklists to assess specific content and process features expected for each HMTW session. Checklists were completed by speechlanguage pathologists for 97% of the group sessions and 78% of the individual sessions. To assess the reliability of their self-ratings, a random sample of the checklists from the group (23%) and individual (34.5%) sessions was rated by an observer who viewed videotapes of the sessions. Mean item-by-item agreement was 92% (SD = 10) and 92% (SD = 11) for group and individual sessions, respectively. The results of the speechlanguage pathologists' ratings indicated that 88% (SD = 4.7) of the intended elements of the group sessions and 89.9% (SD = 7.9) of intended elements in the individual sessions were implemented.

Measures

Parenting Stress Index/Short Form (PSI/ **SF**; **Abidin**, **1995**). The PSI/SF is a reliable and valid self-report measure designed to assess parenting stress focusing on the parent-child system. The PSI short form was administered at pretreatment (Time 1), posttreatment (Time 2), and 4-month follow-up (Time 3). It contains 36 items that are rated on a 5-point Likert-type scale ranging from 1 (strongly agree) to 5 (strongly disagree), with higher scores indicating higher levels of stress. In the current sample, internal consistency of the Total Stress raw score was .95, .94, and .93 at Times 1, 2, and 3, respectively. Raw change scores on the Total Stress score from Time 1 to Time 2 and from Time 1 to Time 3 were used in analyses as outcome variables.

Center for Epidemiologic Studies Depression Scale (CES-D; Radloff, 1977). The CES-D is a self-report measure designed to assess depressive symptomatology in community samples. The 20 items assess the frequency and occurrence of specific depressive feelings and behaviors, and are rated on a 4-point Likert-type scale ranging from zero (rarely or none of the time) to

3 (most or all of the time), with higher scores indicating greater risk for depression. In one of our ongoing investigations, the mean CES-D total score for primary caregivers of 14-24-month-olds in a representative Northeastern urban and suburban region was 8.6, SD = 7.7, N = 528. In the current sample, internal consistency of the total raw score at Time 1 was .92. The total raw score from this scale was used at Time 1 as a putative moderator of treatment effects on parenting stress.

Analysis Approach

Listwise deletion. In listwise deletion, cases with any missing data on analyzed variables are eliminated from analyses, reducing the sample size included in the analyses to only those participants with complete data for all time points required to derive the analyzed variable score (Baraldi & Enders, 2010). Such a reduction in sample size can lead to reduced power. In addition, listwise deletion assumes that the mechanism for the missing data is missing completely at random (MCAR), a stringent assumption (Enders, 2010). A common consequence of using listwise deletion is that the remaining groups are significantly different on pretreatment variables related to outcomes of interest. Some of these pretreatment variables may not be analyzed in a particular study. Hence, the use of listwise deletion can lead to inaccurate interpretation of between-group differences on posttreatment or gain scores and biased parameter estimates (Baraldi & Enders, 2010). Even when data are MCAR, simulation studies show that MI approaches to handling missing data produce more accurate analysis results than does listwise deletion (Ender, 2010).

Multiple imputation. MI involved three primary steps. First, using the MI procedure in Statistical Analysis System (SAS) software, multiple complete data sets were created (i.e., imputation step) using a Markov Chain Monte Carlo and expectation maximization (EM) method to impute missing values. This procedure uses variables with observed scores to estimate the values for variables with missing values using an iterative process that reduces the estimation error with each iteration. Following Enders (2010), all variables in the data set were used as sources of information to impute missing values. In addition, all transformations, change scores, and interaction terms were created from available data

before imputation, which reduces bias in pooled parameter estimates (von Hippel, 2009). As recommended by Graham (2009), 40 complete data sets were imputed. Many data sets were imputed because past research has indicated that estimating population parameters from this quantity of data sets yields more valid results than testing research questions with fewer imputed data sets (Enders, 2010).

Second, the substantive analyses were conducted on all imputed data sets (i.e., analysis step) using the regression and generalized linear model procedures in SAS (PROC REG and PROC GLM). During the analysis step, we used paired *t*-tests and multiple linear regressions as called for by the research questions. For example, to test the moderated treatment effect, a multiple regression was used to test the statistical interaction between the moderator and the treatment group predicting the change in stress in each imputed data set.

Third, using the multiple imputation analysis procedure in SAS (PROC MIANALYZE), the parameters (e.g., regression coefficients and standard errors) from the substantive analyses were estimated by pooling across the results of these analyses (the pooling step). One pools across the results of multiple imputed data sets because any one imputed data set will have more error than the pooled results from many imputed data sets (Enders, 2010). This data-based method for dealing with missing data has been found to be as accurate as the primary alternative, full information maximum likelihood (Enders, 2010; Graham, 2009), and is thought to be more easily understood by psychologists and educators.

Results

Preliminary Analyses

Preliminary analyses included univariate inspection of the data, $\log 10$ or square root transformations when appropriate (Tabachnick & Fidell, 2007), and *t*-tests and chi square analyses to test for differences on Time 1 measures (i.e., primary outcome variables, putative moderator, demographic characteristics, and clinical characteristics) that relate theoretically or empirically to the outcomes. No between-group differences were found for any Time 1 variable (all ps > .10). Chi square analyses comparing the percentage of parents with analyzable data in the treatment and control groups were conducted to determine

whether differential attrition threatened the interpretation of any observed group differences for any analyzed outcome variable. Differential attrition did not occur (all ps > .10). Analyses of variance and regression models were conducted to examine the potential influence of site and site-by-treatment interactions on the outcome variables. Main effects of site and site-by-treatment interactions for both study outcomes were non-significant (all ps > .20).

Attendance to nonproject treatments was not significantly different between groups for any measurement period (all ps > .05). To provide a context for this experiment, we describe participation in nonproject treatment. Across treatment and control groups, participants received an average of 9.8 hr (SD = 19.6), 26.1 hr (SD = 37.0), and 31.9 hr (SD = 33.5) of all therapies combined (i.e., speech therapy, occupational therapy, and applied behavior analysis) per week at Times 1, 2, and 3 respectively. With regard to speech therapy alone, across both groups, participants received an average of 2.2 hr (SD = 3.7), 4.4 hr (SD = 4.8), and 5.1 hr (SD = 3.9) per week at Times 1, 2, and 3 respectively.

Means and standard deviations. Means and standard deviations of the raw scores on the PSI-SF were 87.0 (21.8) for the treatment group and 76.2 (26.7) for the control group at Time 1, 80.2 (20.5) for the treatment group and 80.9 (28.5) for the control group at Time 2, and 78.2 (21.7) for the treatment group and 72.6 (23.8) for the control group at Time 3. Across the primary variables analyzed in this report, data were missing for 13% of the CES-D at Time 1 and 15%, 26%, and 26% of the PSI at Times 1, 2, and 3, respectively. In total, 20% of primary variables for the 62 participants were missing. Results of Little's test indicated the data were not significantly different from that expected from MCAR data ($\chi^2 = 1404.66$, df = 1585, p = 1.000). Graphical diagnostics indicated nonsignificant autocorrelation among successive imputations.

Time effects on primary dependent variables. For both treatment groups combined, parenting stress decreased from Time 1 to Time 2 based on listwise deletion and MI methods, t(40) = 3.6, p = .001, and t(61) = 2.4, p < .01. Parenting stress also decreased from Time 1 to Time 3 based on listwise deletion and MI, t(41) = 3.29, p = .002, and t(61) = 2.66, p < .01, respectively (see Table 2).

Table 2
Effect Sizes for Effects on Stress for Listwise Deletion
and Multiple Imputation (MI) Analyses

	Listwise deletion	MI	
	Time main effect; d		
T1 to T2	29*	61*	
T1 to T3	30*	68*	
	Treatment main	Treatment main effect; d	
T1 to T2	.37	.23	
T1 to T3	.22	.022	
	Depression-moderated treatmer		
	effect**; R squared change		
T1 to T2	.04	.01	
T1 to T3	.19*	.13*	

Note. T1, T2, T3 = time points for treatment stages. * = Significant at .05 level; ** = analyses controlled for initial depression and treatment main effects.

Primary Tests of the Research Questions

Hierarchical Linear Regression was used to test the main effect of the HMTW intervention on parenting stress and to determine whether Time 1 parental depression moderated the effect of the HMTW intervention on change in parenting stress. When the product term was significant, regions of significance were identified (Preacher, Curran, & Bauer, 2006). Regions of significance specify the upper and lower values of parental depression at which the intervention and control groups are significantly different on the outcome variable. For the multiply-imputed data, interpretability of regions of significance (i.e., whether participant scores across treatment and control groups fell within the upper or lower regions of significance) was determined using (a) observed scores of participants and (b) averaged withinparticipant depression scores pooled across the 40 imputed datasets. There was no evidence that there was undue influence of any data points as the maximum observed Cook's D was .28.

The between-group effect sizes for analyses using listwise deletion and MI approaches are presented in Table 2. Contrary to expectations, there was no main effect of HMTW intervention on change in stress for either approach and either change interval. For change in stress from Time 1 to Time 2, the statistics for the main effects of treatment were t(39) = 1.1, p = .26 (listwise deletion) and t = -0.67, p = .51 (MI). A similar pattern was seen when comparing the change in stress from Time 1 to Time 3 between the

treatment and control groups t(40) = .70, p = .50 (listwise deletion), t = .07, p = .94 (MI). Also contrary to expectations, initial level of depressive symptoms did not moderate HMTW's effects on changes in parenting stress between Times 1 and 2 t(40) = 1.35, p = .19 (listwise deletion) and t = .99; p = .33 (MI). As predicted, pretreatment depressive symptoms did moderate treatment effects on changes in parental stress from Time 1 to Time 3 using listwise deletion, t(41) = 3.04, p = .004. This moderated treatment effect was also significant using MI, t = 2.79, p = .0095.

Listwise deletion and MI, however, produced substantively different interpretations of the meaning of this result. This difference is particularly important given the potential implications of interpreting these apparently similar findings. As shown in Figure 2a (listwise deletion results), parents in the HMTW group with slightly lower than average depression scores evidenced greater reduction in parenting stress than those in the control group. In contrast, parents in the HMTW group with highly elevated depression scores reported an increase in stress when compared to those assigned to the control group. The latter finding is consistent with iatrogenic effects of HMTW on parenting stress. Conversely, although MI results (Figure 2b) indicate that the interaction was significant, neither the upper nor the lower region of significance was interpretable (i.e., no participant scores were contained within either region of significance). This was the case using both observed and imputed values of the moderator. Importantly, there is no basis for predicting an iatrogenic effect of HMTW on parenting stress in parents with initially high depressive symptoms.

Discussion

The goal of this article was to illustrate the importance of selecting appropriate methods for handling missing data by comparing the use of two methods of missing data analysis in examining whether Hanen's "More than Words" (HMTW) intervention resulted in greater reductions in parenting stress compared to a "business as usual" control group. Multiple imputation is a state-of-the-art technique that provides unbiased parameter estimates when data are MNAR and MAR, outperforming listwise deletion, which has been found to produce biased parameter estimates when these patterns of missing data are present. In

addition, MI provides better estimates than listwise deletion even when data are MCAR (Enders, 2010). Thus, when results differ between methods for addressing missing data, the field is better served by placing greater weight on MI results than on listwise deletion results.

First, the results of the two missing data analysis approaches showed two similar findings, which suggest the robustness of these effects. In both analyses, there was no main effect of the HMTW intervention in reducing parenting stress when analyses were conducted. In addition, improvements in the mean level of parenting stress were observed over time across the two groups.

The cut-off scores for the regions of significance and interpretation of the statistical interaction between depressive symptoms at Time 1 and group assignment on stress, however, depended on whether listwise deletion or multiple imputation (i.e., intent to treat) analyses were conducted. Unlike interactions between nonexperimental variables, when one of the variables in the statistical interaction is an experimental manipulation (e.g., presence or absence of HMTW), the interpretation regarding the between-group difference is the more important one because it potentially allows more confidence in the causal influence of the manipulated variable on the dependent variable than does the association involving the nonexperimental variable.

The use of listwise deletion revealed a significant moderated treatment effect on stress with interpretable regions of significance at both ends of the depressive symptom continuum. If the less-robust listwise deletion results are interpreted, one finds confirmation of the prediction that for parents with initially lower depressive symptoms prior to treatment, parental stress was reduced more in the HMTW group than in the control group. The less-robust analysis approach, however, also incorrectly supported an interpretation that for parents with initially high depressive symptoms before treatment, parental stress was reduced less in the HMTW group than the control group. The latter results are evidence of an iatrogenic effect.

In contrast, the findings from the more robust MI analysis indicate a significant depression-moderated effect of HMTW on stress, but with regions of significance that are not interpretable because they are outside of the range of depression scores obtained in our sample. Consistency across

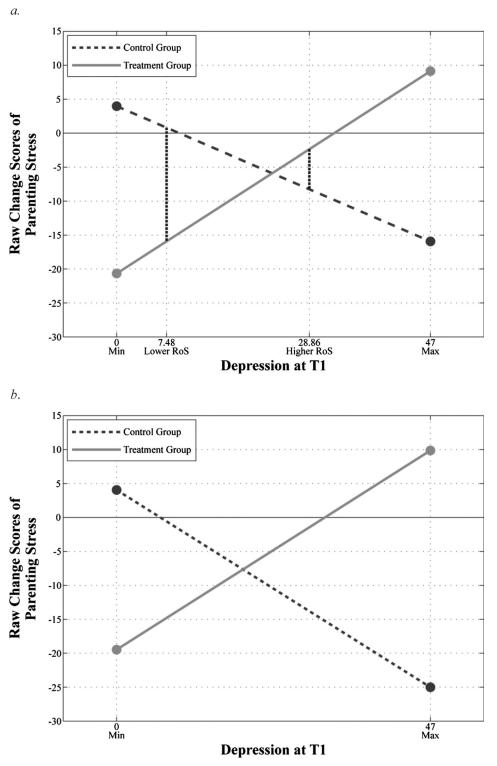


Figure 2. Moderation of Treatment by Time 1 Depression Predicting Time 1 to Time 3 Parenting Stress Change Scores (a) in Listwise Deletion Analysis and (b) in Multiple Imputation (MI) Analysis. Note: For Parenting Stress, more negative change scores are optimal. T1 = 1st time point; Max = maximum value of T1 moderator variable; Min = minimum value of T1 moderator variable; Higher RoS = higher region of significance; Lower RoS = lower region of significance. There are no regions of significance in (b) because they lie outside of the range of depressive symptom scores in the study sample.

two reasonable methods for assessing interpretability of the regions of significance (i.e., observed scores and imputed scores) further strengthens the conclusion that the significant interaction cannot be interpreted with regard to the effect of HMTW on stress. Using the validated MI approach, there was no suggestion that HMTW may increase stress for parents beginning the treatment with relatively high depressive symptomatology. Thus, we can only interpret the less interesting difference in association between depression symptoms and stress as a function of the treatment groups. The explanation for this difference in association is unclear. Regardless, the difference in interpretation afforded by the two methods of handling missing data is salient.

Strengths and Limitations of the Study

The strengths and limitations of the HMTW study have been noted elsewhere (Carter et al., 2011). In sum, strengths were that the initial randomization resulted in groups that were comparable at Time 1 on variables that were theoretically or empirically related to parenting stress; that differential attrition did not occur across the HMTW intervention and "business as usual" control groups; and that completion of treatment was not associated with the presence of missing data. Limitations were the smaller-than-recommended size of the parent groups and the reliance on a single method and informant (i.e., the parent) for measuring the primary outcome and moderator variable.

Integration With Extant Literature

Regardless of treatment group, parents in both the HMTW and "business as usual" groups reported reductions in parenting stress between initial enrollment and the second and third testing sessions (approximately five and nine months later). Few studies have examined longitudinal patterns of parenting stress over time in families of young children with ASD. The current findings of improvements in parental well-being may reflect the specific timing of the first assessment. Many parents had learned of their child's diagnosis immediately before completing the Time 1 parenting stress measure. Thus, the higher initial scores may reflect, in part, the inherent stress of receiving the diagnosis of ASD, and associated negative cognitions and beliefs about what the diagnosis might mean for their child's

future (Goin-Kochel, Mackintosh, & Myers, 2006; Wachtel & Carter, 2008; Wolf, Noh, Fisman, & Speechley, 1989). Accordingly, it appears that as a group, parents may experience declines in stress in the year that follows receipt of an early diagnosis of ASD. We could not find another reporting of this type of reduction in parenting stress in the ASD literature.

Conclusion

The results of the current study point to the effect that decisions about how to manage missing data can have on the findings of treatment studies, and tell a cautionary tale regarding interpretation of findings when state-of-the-art missing data methods are not used by researchers. This study serves as a good exemplar for illustrating the divergent results obtained using different methods for handling missing data. Because of the small sample size and presence of attrition, this study represents a typical intervention study in treatment research involving people with developmental disabilities. It is likely that researchers across fields (e.g., clinical psychology, special education, communication sciences) will be able to identify similar logistical and methodological concerns in their own work, making the current illustration for why it is important to analyze data using the most appropriate and state-of-the-art methods particularly salient to treatment researchers.

References

Abidin, R. R. (1995). Parenting Stress Index: Professional Manual (3rd ed.). Odessa, FL: Psychological Assessment Resources, Inc.

Baker-Ericzén, M. J., Brookman-Frazee, L., & Stahmer, A. (2005). Stress levels and adaptability in parents of toddlers with and without autism spectrum disorders. *Research and Practice for Persons with Severe Disabilities, 30*, 194–204. http://dx.doi.org/http://dx.doi.org/10. 2511/rpsd.30.4.194

Baraldi, A. N., & Enders, C. K. (2010). An introduction to modern missing data analyses. *Journal of School Psychology*, 48, 5–37. http://dx.doi.org/10.1016/j.jsp.2009.10.001

Barnes, S. A., Lindborg, S. R., & Seaman, J. W. (2006). Multiple impuation techniques in small sample clinical trials. *Statistics in Medicine*, 25, 233–245. http://dx.doi.org/10.1002/sim.2231

- Benson, P. R. (2006). The impact of symptom severity of depressed mood among parents of children with ASD. *Journal of Autism and Developmental Disorders*, 36, 685–695. http://dx.doi.org/10.1007/s10803-006-0112-3
- Bouma, R., & Schweitzer, R. (1990). The impact of chronic childhood illness on family stress: A comparison between autism and cystic fibrosis. *Journal of Clinical Psychology*, 46, 722–730. http://dx.doi.org/10.1002/1097-4679 (199011)46:6<722::aid-jclp2270460605>3.0. co;2-6
- Brinker, R. P., Seifer, R., & Sameroff, A. J. (1994). Relations among maternal stress, cognitive development, and early intervention in middle- and low-SES infants with developmental disabilities. *American Journal on Mental Retardation*, 98, 463–480.
- Buhi, E. R., Goodson, P., & Neilands, T. B. (2008). Out of sight, not out of mind: Strategies for handling missing data. *Amercian Journal of Health Behavior*, 32, 83–92. http://dx.doi.org/10.5993/AJHB.32.1.8
- Carter, A. S., de L. Martínez-Pedraza, F., & Gray, S. A. (2009). Stability and individual change in depressive symptoms among mothers raising young children with ASD: Maternal and child correlates. *Journal of Clinical Psychology*, 65, 1270–1280. http://dx.doi.org/10. 1002/jclp.20634
- Carter, A. S., Messinger, D. S., Stone, W. L., Celimli, S., Nahmias, A. S., & Yoder, P. (2011). A randomzied controlled trial of Hanen's 'More Than Words' in toddlers with early autism symptoms. *Journal of Child Psychology and Psychiatry*, *52*, 741–752. http://dx.doi.org/10.1111/j.1469-7610.2011.02395.x
- Davis, N. O., & Carter, A. S. (2008). Parenting stress in mothers and fathers of toddlers with autism spectrum disorders: Associations with child characteristics. *Journal of Autism and Developmental Disorders*, 38, 1278–1291. http://dx.doi.org/10.1007/s10803-007-0512-z
- Dawson, G., Rogers, S., Munson, J., Smith, M., Winter, J., Greenson, J. ... Varley, J. (2010). Randomized, controlled trial of an intervention for toddlers with autism: the Early Start Denver Model. *Pediatrics*, 125, e17–e23. http://dx.doi.org/10.1542/peds.2009-0958
- Donenberg, G., & Baker, B. L. (1993). The impact of young children with externalizing behaviors on their families. *Journal of Abnormal Child Psychology: An official publication of the*

- International Society for Research in Child and Adolescent Psychopathology, 21, 179–198. http://dx.doi.org/10.1007/bf00911315
- Dumas, J. E., Wolf, L. C., Fisman, S. N., & Culligan, A. (1991). Parenting stress, child behavior problems, and dysphoria in parents of children with autism, Down syndrome, behavior disorders, and normal development. *Exceptionality*, *2*, 97–110. http://dx.doi.org/10.1080/09362839109524770
- Enders, C. K. (2010). *Applied missing data analysis*. New York, NY: Guilford.
- Enders, C. K. (2011). Missing not at random models for latent growth curve analyses. *Psychological Methods*, 16, 1–16. http://dx.doi. org/10.1037/a0022640
- Estes, A., Olson, E., Sullivan, K., Greenson, J., Winter, J., Dawson, G., & Munson, J. (2013). Parenting-related stress and psychological distress in mothers of toddlers with autism spectrum disorders. *Brain & Development*, 35, 133–138. http://dx.doi.org/10.1016/j.braindev. 2012.10.004.
- Goin-Kochel, R. P., Mackintosh, V. H., & Myers, B. J. (2006). How many doctors does it take to make an autism spectrum diagnosis? *Autism*, 10, 439–451. http://dx.doi.org/10. 1177/1362361306066601
- Graham, J. W. (2009). Missing data analysis: Making it work in the real world. *Annual Review of Psychology*, 60, 549–576. http://dx.doi.org/10.1146/annurev.psych.58.110405. 085530
- Graham, J. W., Cumsille, P. E., & Elek-Fisk, E. (2003). Methods for handling missing data. In J. A. Schinka & W. F. Velicer (Eds.), Research methods in psychology (pp. 87–114). In I. B. Weiner (Editor-in-Chief), Handbook of psychology, vol 2. New York, NY: John Wiley & Sons.
- Graham, J. W., & Schafer, J. L. (1999). On the performance of multiple imputation for multivariate data with small size. In R. H. Hoyle (Ed.), *Statistical strategies for small sample research* (pp. 1–29). Thousand Oaks, CA: SAGE Publications.
- Hastings, R. P., & Johnson, E. (2001). Stress in UK families conducting intensive home-based behavioral intervention for their young child with autism. *Journal of Autism and Developmental Disorders*, 31, 327–336. http://dx.doi.org/10.1023/a:1010799320795
- Herring, S., Gray, K., Taffe, J., Tonge, B., Sweeney, D., & Einfeld, S. (2006). Behaviour

- and emotional problems in toddlers with pervasive developmental disorders and developmental delay: Associations with parental mental health and family functioning. *Journal of Intellectual Disability Research*, 50, 874–882. http://dx.doi.org/10.1111/j.1365-2788.2006. 00904.x
- Holroyd, J., & McArthur, D. (1976). Mental retardation and stress on the parents: A contrast between Down's syndrome and childhood autism. *American Journal of Mental Deficiency*, 80, 431–436.
- Horton, N. J., & Kleinman, K. P. (2007). Much ado about nothing: A comparison of missing data methods and software to fit incomplete data regression models. *The American Statistician*, 61, 79–90. http://dx.doi.org/10.1198/000313007X172556
- Ibrahim, J. G., Chen, M-H., Lipsitz, S. R., & Herring, A. H. (2005). Missing-data methods for generalized linear models: A comparative review. *Journal of the American Statistical Association*, 100, 332–346. http://dx.doi.org/10.1198/016214504000001844
- Ingersoll, B., & Gergans, S. (2007). The effect of a parent-implemented imitation intervention on spontaneous imitation skills in young children with autism. *Research in Developmental Disabilities*, 28, 163–175. http://dx.doi.org/10.1016/j.ridd.2006.02.004
- Lachin, J. M. (2000). Statistical considerations in the intent-to-treat principle. *Controlled Clinical Trials*, *21*, 167–189. http://dx.doi.org/10.1016/S0197-2456(00)00046-5
- Lecavalier, L., Leone, S., & Wiltz, J. (2006). The impact of behaviour problems on caregiver stress in young people with autism spectrum disorders. *Journal of Intellectual Disability Research*, 50, 172–183. http://dx.doi.org/10.1111/j.1365-2788.2005.00732.x
- Little, R. J. A., & Rubin, D. B. (2002). Statistical analysis with missing data. New York, NY: Wiley.
- Lord, C., Rutter, M., DiLavore, P., & Risi, S. (2002). Autism Diagnostic Observation Schedule manual. Los Angeles, CA: Western Psychological Services.
- Meadan, H., Ostrosky, M. M., Zaghlawan, H. Y., & Yu, S. (2009). Promoting the social and communicative behavior of young children with autism spectrum disorders: A review of parent-implemented intervention studies. *Topics in Early Childhood Special Education*, 29, 90–104. http://dx.doi.org/10.1177/0271121409337950

- Moher, D., Schulz, K., Altman, D.; CONSORT GROUP (Consolidated Standards of Reporting Trials). (2001). The CONSORT statement: revised recommendations for improving the quality of reports of parallel-group randomized trials. *Annals of Internal Medicine*, 134, 657–662. http://dx.doi.org/10.7326/0003-4819-134-8-200104170-00011
- Mullen, E. M. (1995). *Mullen Scales of Early Learning*. Circle Pines, MN: American Guidance Service Inc.
- Newell, D. J. (1992). Intention-to-treat analysis: Implications for quanitatitive and qualitative research. *International Journal of Epidemiology*, 21, 837–841. http://dx.doi.org/10.1093/ije/21.5.837
- Newman, D. A. (2003). Longitudinal modeling with randomly and systematically missing data: A simulation of ad hoc, maximum likelihood, and multiple imputation techniques. *Organizational Research Methods*, 6, 328–362. http://dx.doi.org/10.1177/1094428103254673
- Noh, S., Dumas, J. E., Wolf, L. C., & Fisman, S. N. (1989). Delineating sources of stress in parents of exceptional children. *Family Relations*, 38, 456–461. http://dx.doi.org/10.2307/585753
- Osborne, L. A., McHugh, L., Saunders, J., & Reed, P. (2008). Parenting stress reduces the effectiveness of early teaching interventions for autistic spectrum disorders. *Journal of Autism & Developmental Disorders*, 38, 1092–1103. http://dx.doi.org/10.1007/s10803-007-0497-7
- Preacher, K. J., Curran, P. J., & Bauer, D. J. (2006). Computational tools for probing interactions in multiple linear regression, multilevel modeling, and latent curve analysis. *Journal of Educational and Behavioral Statistics*, 31, 437–448. http://dx.doi.org/10.3102/10769986031004437
- Radloff, L. S. (1977). The CES-D Scale: A self-report depression scale for research in the general population. *Applied Psychological Measurement*, 1, 385–401. http://dx.doi.org/10. 1177/014662167700100306
- Rodrigue, J. R., Morgan, S. B., & Geffken, G. (1990). Families of autistic children: Psychological functioning of mothers. *Journal of Clinical Child Psychology*, 19, 371–379. http://dx.doi.org/10.1207/s15374424jccp1904_9
- Rubin, D. B. (1976). Inference and missing data. *Biometrika*, 63, 581–592. http://dx.doi.org/10. 1093/biomet/63.3.581

- Sameroff, A. J., & Emde, R. N. (1989). Relationship disturbances in early childhood: A developmental approach. New York, NY: Basic Books.
- Sanders, J. L., & Morgan, S. B. (1997). Family stress and adjustment as perceived by parents of children with autism or Down syndrome: Implications for intervention. *Child & Family Behavior Therapy*, 19(4), 15–32. http://dx.doi.org/10.1300/J019v19n04_02
- Schafer, J. L., & Graham, J. W. (2002). Missing data: Our view of the state of the art. *Psychological Methods*, 7, 147–177. http://dx.doi.org/10.1037/1082-989X.7.2.147
- Schlomer, G. L., Bauman, S., & Card, N. A. (2010). Best practices for missing data management in counseling psychology. *Journal of Counseling Psychology*, 57, 1–10. http://dx.doi.org/10.1037/a0018082
- Shadish, W., Cook, T., & Campbell, D. (2002). Experimental and quasi-experimental designs for generalized causal inference. Boston, MA: Houghton-Mifflin.
- Sparrow, S., Cicchetti, D., & Balla, D. (2005). Vineland-II: Vineland Adaptive Behavior Scales: Survey forms manual. Circle Pines, MN: American Guidance Service.
- Stone, W. L., Coonrod, E., & Ousley, O. (2000). Screening Tool for Autism in Two-year-olds (STAT): Development and preliminary data. *Journal of Autism & Developmental Disorders*, 30, 607–612. http://dx.doi.org/10.1023/A: 1005647629002
- Stone, W. L., Coonrod, E. E., Turner, L. M., & Pozdol, S. L. (2004). Psychometric properties of the STAT for early autism screening. *Journal of Autism and Developmental Disorders*, 34, 691–701. http://dx.doi.org/10. 1007/s10803-004-5289-8
- Stone, W. L., McMahon, C. R., & Henderson, L. M. (2008). Use of the Screening Tool for Autism in Two-year-olds (STAT) for children under 24 months: An exploratory study. *Autism*, 12, 557–573. http://dx.doi.org/10. 1177/1362361308096403
- Sussman, F. (2004). More Than Words: Helping parents promote communication and social skills in children with autism spectrum disorder. Toronto, Canada: The Hanen Centre.
- Tabachnick, B. G., & Fidell, L. S. (2007). *Using multivariate statistics* (5th ed.). Boston, MA: Allyn & Bacon/Pearson Education.
- Tomanik, S., Harris, G. E., & Hawkins, J. (2004). The relationship between behaviours exhibited

- by children with autism and maternal stress. *Journal of Intellectual and Developmental Disability*, 29, 16–26. http://dx.doi.org/10.1080/13668250410001662892
- Tonge, B., Brereton, A., Kiomall, M., Mac-Kinnon, A., King, N., & Rinehart, N. (2006). Effects on parental mental health of an education and skills training program for parents of young children with autism: A randomized controlled trial. *Journal of the American Academy of Child & Adolescent Psychiatry*, 45, 561–569. http://dx.doi.org/10.1097/01.chi.0000205701.48324.26
- Vismara, L. A., & Rogers, S. J. (2010). Behavioral treatments in autism spectrum disorder: What do we know? *Annual Review of Clinical Psychology*, 6, 447–468. http://dx.doi.org/10. 1146/annurev.clinpsy.121208.131151
- Von Hippel, P. T. (2009). How to impute interactions, squares, and other transformed variables. *Sociological Methodology*, *39*, 265–291. http://dx.doi.org/10.1111/j.1467-9531. 2009.01215.x
- Wachtel, K., & Carter, A. S. (2008). Reaction to diagnosis and parenting styles among mothers of young children with ASDs. *Autism: The International Journal of Research and Practice*, 12, 575–594. http://dx.doi.org/10.1177/1362361308094505
- What Works Clearinghouse. (2010). Procedures and standards handbook, version 2.1. Retrieved from http://ies.ed.gov/ncee/wwc/pdf/reference_resources/wwc_ procedures_v2_1_standards_handbook.pdf
- What Works Clearinghouse. (2014). Procedures and standards handbook, verson 3.0. Retrieved from http://ies.ed.gov/ncee/wwc/pdf/reference_resources/wwc_procedures_v3_0_standards_handbook.pdf
- Whittingham, K., Sofronoff, K., Sheffield, J., & Sanders, M. R. (2009). Stepping Stones Triple P: An RCT of a parenting program with parents of a child diagnosed with an autism spectrum disorder. *Journal of Abnormal Child Psychology*, *37*, 469–480. http://dx.doi.org/10. 1007/s10802-008-9285-x
- Wolf, L. C., Noh, S., Fisman, S. N., & Speechley, M. (1989). Brief report: Psychological effects of parenting stress on parents of autistic children. *Journal of Autism and Developmental Disorders*, 19, 157–166. http://dx.doi.org/10. 1007/bf02212727

Wong, V. C. N., & Kwan, Q. K. (2010). Randomized controlled trial for early intervention for autism: A pilot study of the Autism 1-2-3 project. *Journal of Autism and Developmental Disorders*, 40, 677–688. http://dx.doi.org/10.1007/s10803-009-0916-z

Wood, A. M., White, I. R., & Thompson, S. G. (2004). Are missing outcome data adequately handled? A review of published randomized controlled trials in major medical journals. *Clinical Trials*, 1, 368–376. http://dx.doi.org/10.1191/1740774504cn0320a

Received 01/19/2013, accepted 11/05/2013.

The research reported here was supported in part by the Institute of Education Sciences, U.S. Department of Education, through Grant #R305B090015 to the University of Pennsylvania. The opinions expressed are those of the authors and do not represent the views of the Institute or the U.S. Department of Education.

Authors:

Rebecca G. Lieberman-Betz, Assistant Professor of Special Education; **Paul Yoder**, Professor of Special Education, Department of Special Education,

Peabody College, Vanderbilt University; **Wendy L. Stone**, Professor of Psychology, Department of Pediatrics, Vanderbilt University; **Allison S. Nahmias**, Doctoral Candidate in Clinical Psychology, Vanderbilt University; **Alice S. Carter**, Professor of Psychology, Department of Psychology, University of Massachusetts-Boston; **Seniz Celimli-Aksoy**, Research Assistant; and **Daniel S. Messinger**, Professor of Psychology, Department of Psychology, University of Miami.

Rebecca G. Lieberman-Betz is now with the Department of Communication Sciences and Special Education, University of Georgia; Wendy L. Stone is now with the Department of Psychology, University of Washington; Allison S. Nahmias is now with the Department of Psychology, University of Pennsylvania; Seniz Celimli-Aksoy is now with the Department of Teaching and Learning, University of Miami.

Address correspondence concerning this article to Rebecca G. Lieberman-Betz, Department of Communication Sciences and Special Education, 565 Aderhold Hall, University of Georgia, Athens, GA 30602 (e-mail: rglb@uga.edu).