Title: Effectiveness of Responsivity Intervention Strategies on Prelinguistic and Language Skills of Children with ASD: A Meta-Analysis of Randomized Controlled Trials

Authors: Jena McDaniel[1], Madeline Friedrichs[2], Jordan Rangel[2], Mackenna Murphy[1], & Lily Black[1]

Introduction: Investigations of the effectiveness of communication and language interventions vary across multiple facets and have yielded widely varying results (Hampton & Kaiser, 2016; Reichow et al., 2018; Sandbank et al., 2020). One organizational approach to synthesizing prior results is to evaluate the effectiveness of interventions that include specific intervention components to identify active ingredients. Interventionists can then implement these essential strategies. The purpose of this systematic review and meta-analysis is to describe the current state of the literature for the role of responsivity intervention strategies to influence the direction of future research studies and clinical practice. We define responsivity intervention strategies as strategies designed to support turn-taking conversations through responding systematically to children's vocalizations and communicative attempts, following the child's lead, and providing targeted input. Responsivity intervention strategies are often used within an intervention package (e.g., naturalistic developmental behavioral interventions; Schreibman et al., 2015). They contrast adult-driven strategies that emphasize discrete training of specific behaviors using structured prompting. Multiple converging theories (e.g., social feedback theory [Goldstein & Schwade, 2008], social feedback loop theory [Warlaumont et al., 2014], and transactional theory of spoken language development [McLean & Synder-McLean, 1978]) provide theoretical support for the use of responsivity intervention strategies for supporting language development. We ask three research questions: (1) Is the mean effect size for interventions that use responsivity strategies on prelinguistic and/or language skills in children with ASD greater than zero for randomized controlled trials (RCTs)? (2) Does the mean effect size vary by interventionist, length of intervention, proximal versus distal outcome, context-bound versus generalized characteristic outcome, risk for correlated measurement error (CME), or publication status? (3) As an analysis of study quality, what percentage of included studies exhibit low, moderate, and high risk of bias?

Method: We conducted a systematic review (via primary and supplementary searches) to identify records of randomized controlled trials of children with ASD who were randomly assigned to an intervention that used responsivity intervention techniques or one that did not. Reports had to include prelinguistic and/or language outcomes (e.g., communication acts, gestures, joint attention, and receptive and expressive language). An independent coder completed reliability at the title/abstract and full text screening levels. Two independent coders extracted data and evaluated risk of bias (revised Cochrane risk-of-bias tool for randomized trials; Higgins et al., 2019) for all included articles. We calculated the mean effect sizes with robust variance estimation to include multiple effect sizes per study and conducted moderator analyses to explain heterogeneity.

Results: We identified 5671 reports for title and abstract screening (after removing duplicates) and 779 reports for full text review. Point-by-point agreement was 89% at the title and abstract level and 87% at the full text level for inclusion/exclusion. Point-by-point agreement was 94% for data extraction and 90% for bias coding. All disagreements for data extraction and bias coding were resolved by consensus. Consensus coding was used for data analysis. Forty-five reports from 33 RCTs met inclusion criteria (single case research design studies [n = 93] will be presented at a later date) and included 293 relevant effect sizes. The mean treatment group sample size and age by effect size are 26.7 (range: 5 - 82) and 42.1 months (range: 20.6 - 110.7 months).

For research question 1, the mean standardized group difference using robust variance estimation with random effects (Tanner-Smith & Tipton, 2014) is g = .37, 95% CI [0.22, 0.52]. The Galbraith plot provides an alternative graphing method for summarizing meta-analyses with too many effect sizes for an interpretable forest plot and for examining heterogeneity (Borenstein et al., 2009). More precise estimates lie further from the origin. As heterogeneity increases, more effect sizes fall outside of the 95% confidence interval (two parallel outer lines; Anzures-Cabrera & Higgins, 2010). Visual examination of the Galbraith plot and the τ^2 value of 0.18 indicate substantial heterogeneity and fulfills one of the necessary conditions for conducting moderator analyses.

For research question 2, context-bound outcomes exhibited a larger mean effect size (n = 68; mean ES = .47) than generalized or possibly context-bound behaviors combined (n = 225; mean ES = .25). None of the other moderator analyses yielded significant results. Of note, the mean effect size was greater than zero for effect sizes at-risk for CME (n = 145; mean ES = 0.39), but not for those free from CME risk (n = 148; mean ES = 0.14). Although a publication bias was not detected via the moderator analysis, the Egger's test was significant (p = .005; Figure 2), which suggests publication bias against small studies with negative results.

For research question 3, 25 studies were coded at overall high risk versus seven at moderate risk and one at low risk. A primary reason for studies being classified as high risk was the presence of risk for CME for the included outcome measure.

Discussion: We identified a significant, positive mean effect size for interventions that include responsivity strategies increasing prelinguistic and language skills in children with ASD compared with a randomly assigned control condition. Aside from context-bound outcomes exhibiting a larger mean effect size than generalized or possibly context-bound behaviors, the planned moderator analyses were non-significant. The majority of studies exhibited overall high risk for bias, often due to risk for CME. We discuss how to apply these findings to clinical practice and future research. Future directions for this meta-analysis include (a) testing of additional putative moderators to explain the observed heterogeneity in results and (b) synthesizing single case research design studies from this systematic search.

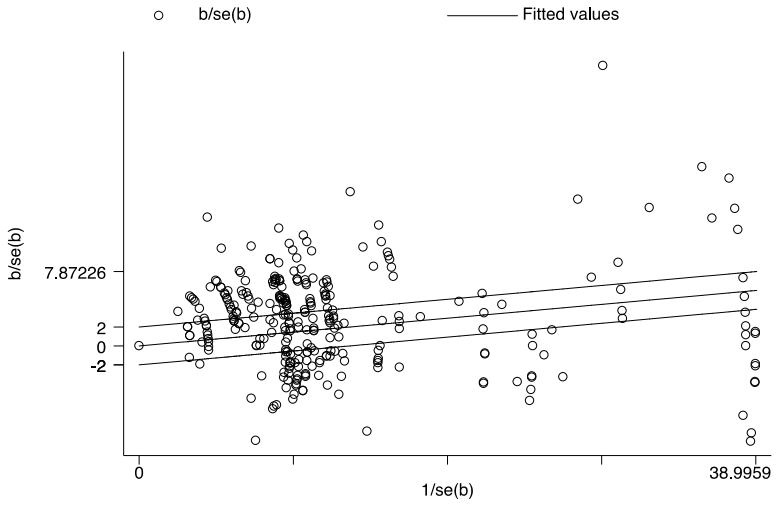

Figure 1. Galbraith plot for standardized mean difference between the responsivity intervention group and control group.

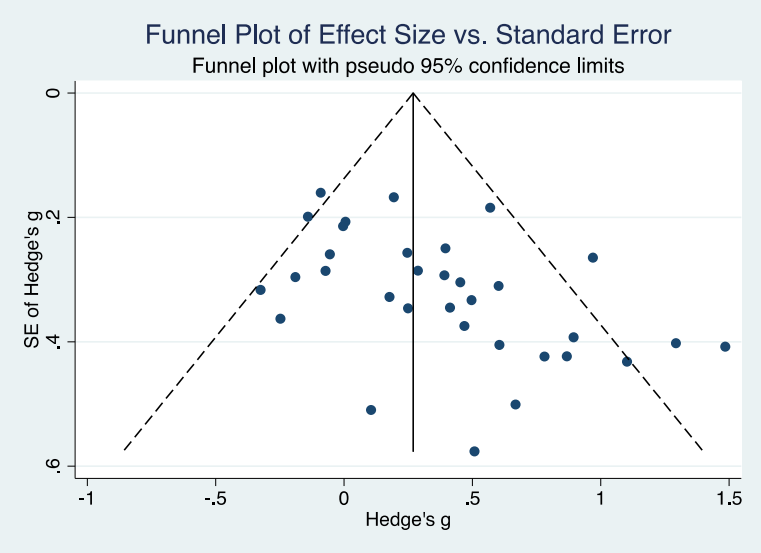

Figure 2. Funnel plot of effect size (Hedge's *g*) versus standard error for all included studies.

References:

Anzures-Cabrera, J., & Higgins, J. (2010). Graphical displays for meta-analysis: An overview with suggestions for practice. Research Synthesis Methods, 1, 66–80. http://dx.doi.org/10.1002/jrsm.6.

Borenstein, M., Hedges, L. V., Higgins, J. P. T., & Rothstein, H. R. (2009). Introduction to meta-analysis. Wiley: West Sussex, United Kingdom.

Goldstein, M., & Schwade, J. (2008). Social feedback to infants' babbling facilitates rapid phonological learning. *Psychological Science, 19*, 515-523.

Hampton, L., & Kaiser, A. (2016). Intervention effects on spoken-language outcomes for children with autism: A systematic review and meta-analysis. *Journal of Intellectual Disability Research, 60*, 444-463.

Higgins, J., Savovic, J., Page, M., & Sterne, J. (2019) Revised Cochrane risk-of-bias tool for randomized trials (RoB 2) SHORT VERSION (CRIBSHEET). https://colloquium2019.cochrane.org/sites/2019.colloquium.cochrane.org/files/uploads/users/u%5Buid%5D/20190814_RoB_2.0_cribsheet_parallel_trial%20.pdf

McLean, J., & Snyder-McLean, L. (1978). *A transactional approach to early language training*. Columbus, OH: Charles E. Merrill.

Reichow, B., Hume, K., Barton, E., et al. (2018). Early intensive behavioral intervention (EIBI) for young children with autism spectrum disorders (ASD). *Cochrane Database of Systematic Reviews*(5).

Sandbank, M., Bottema-Beutel, K., Crowley, S., et al. (2020). Project AIM: Autism intervention meta-analysis for studies of young children. *Psychological Bulletin, 146* [advanced online release].

Schreibman, L., Dawson, G., Stahmer, A. et al. (2015). Naturalistic developmental behavioral interventions: Empirically validated treatments for autism spectrum disorder. *Journal of Autism and Developmental Disorders, 45*, 2411-2428.

Tanner-Smith, E. E., & Tipton, E. (2014). Robust variance estimation with dependent effect sizes: Practical considerations including a software tutorial in Stata and SPSS. *Research Synthesis Methods, 5,* 13–30.

Warlaumont, A., Richards, J., Gilkerson, J., et al. (2014). A social feedback loop for speech development and its reduction in autism. *Psychological Science, 25*, 1314-1324.

[1] University of Kansas
[2] University of Kansas Medical Center