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A Meta-Analysis of the Association Between Vocalizations and Expressive Language in Children with Autism Spectrum Disorder

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Abstract

Background—Targeting the frequency or complexity of prelinguistic vocalizations might improve the language trajectory of children with autism spectrum disorder (ASD) who exhibit continued expressive language deficits.

Aims—This meta-analysis evaluates the strength of the association between various measures of vocalizations and expressive language in young children with ASD and five putative moderators of that association to inform prelinguistic intervention development: consonant-centricity, communicativeness, concurrent versus longitudinal research design, risk for correlated measurement error, and publication status.

Methods and Procedures—We systematically searched databases and other sources for correlations between vocalizations and expressive language in children with ASD less than 9 years old. Using robust variance estimation, we calculated the weighted mean effect size and conducted moderator analyses.

Outcomes and Results—Nine studies (19 reports), which included 362 participants and 109 unique effect sizes, met inclusion criteria. The weighted mean effect size between vocalizations and expressive language was significant ($r = .50$, 95% CI [.23, .76]). As predicted, concurrent correlations were significantly stronger than longitudinal correlations. Other moderator effects were not detected.

Conclusions and Implications—Young children with ASD demonstrate a strong association between vocalizations and expressive language skills. Future experimental studies should investigate causal relations to guide intervention development.

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Keywords

autism spectrum disorder; vocalizations; language; meta-analysis

1. Introduction

Many children with autism spectrum disorder (ASD) continue to demonstrate language skills below those of their peers and a substantial portion do not obtain useful speech, which is defined as “expressive language that may be used frequently, communicatively, referentially, and in a semantically diverse manner” (Yoder, Watson, & Lambert, 2015, p. 1254). Addressing expressive language deficits is particularly important in light of the strong predictive relation between expressive language skills and social and adaptive outcomes for children with ASD (Billstedt, Gillberg, & Gillberg, 2005; Howlin, 2000). Researchers must continue to optimize expressive language interventions (e.g., Tager-Flusberg, Paul, & Lord, 2005). Early deficits in vocal development may dramatically influence spoken language development in children with ASD (e.g., Patten et al., 2014; Paul, Chawarska, Cicchetti, & Valkmar, 2008; Plumb & Wetherby, 2013). In this meta-analysis, we aim to synthesize the current literature on the association between vocalizations and expressive language in children with ASD to inform vocalization measurement and language interventions targeting prelinguistic vocalizations.

1.1. Continuity of Babbling and Spoken Words in Typical Development

Infraphonological theory (Oller, 2000) offers a framework for vocal development, in which vocalizations become more speech-like across development. For example, early infant vocalizations exhibit limited speech-likeness but show capabilities necessary for speech (e.g., phonation). The speech-likeness of vocalizations becomes particularly prominent when canonical syllables emerge. The phonological similarities between infant vocalizations and early words, differentiated acoustic characteristics of babbling across languages, and correlations between early vocalizations and later language skills in children with typical development strongly suggest that early vocalizations are continuous with language development (McCune & Vihman, 2001; McGillion et al., 2017; Oller, 2000; Rvachew, Mattock, Polka, and Ménard, 2006; Vihman, 2017). These contemporary reports effectively counter prior theories positing discontinuity between babbling and spoken words (e.g., Jakobson, 1968). Vocalization frequency and consonant use in prelinguistic vocalizations predict later expressive language in children with typical development (Camp, Burgess, Morgan, & Zerbe, 1987; Stoel-Gammon, 1991; Watt, Wetherby, & Shumway, 2006). Intervening at the prelinguistic stage may alter a child’s trajectory for producing spoken words.

1.2. Areas of Need in Vocal Development in Children with ASD

Differences in the total frequency of vocalizations between children with and without ASD or those at high versus low risk for ASD have been mixed (Patten et al., 2014; Paul, Fuerst, Ramsay, Chawarska, & Klin, 2011; Sheinkopf, Mundy, Oller, & Steffens, 2000). Yet, replicated deficits in the use of canonical syllables and speech-like vocalizations have been

shown (Patten et al., 2014; Paul et al., 2011; Plumb & Wetherby, 2013; Werner, Dawson, Munson, & Osterling, 2005). Children with or at high risk for ASD also present with less diverse vocalizations, including smaller consonant inventories, than children with typical development (Paul et al., 2008; Paul et al., 2011; Wetherby, Watt, Morgan, & Shumway, 2007). In addition, children with ASD are more likely to produce vocalizations without a communicative purpose compared with typically developing, same-age peers (Plumb & Wetherby, 2013; Shumway & Wetherby, 2009).

1.3. Defining and Measuring Vocalizations in Children with ASD

Vocalization definitions vary across studies. Authors of studies may or may not distinguish between communicative versus noncommunicative vocalizations, non-word vocalizations versus spoken words, and distress signals (e.g., crying) versus non-distress signals. Various vocalization measures differ in emphasis on particular vocalization features (e.g., acoustic qualities, communicative function, and phonetic content) and collection methods (e.g., human coding or automated vocal analyses). The heterogeneity in vocalization definitions and measurement techniques may at least partially explain variations in associations between vocalizations and expressive language.

2. The Current Literature Synthesis

Despite multiple reports of associations between vocalizations and expressive language in children with ASD, to our knowledge, no systematic reviews or meta-analyses have been completed on this association. The current meta-analysis was conducted to inform intervention by providing a more accurate population estimate of the association between vocalizations and expressive language for children with ASD than point estimates from single studies. Further, the meta-analytic approach permits consideration of factors that may explain differences in reported findings to inform (a) the measurement of vocalizations and (b) the development of language interventions targeting prelinguistic vocalizations.

2.1. Factors that May Influence the Association Between Vocalizations and Expressive Language

2.1.1. Consonant-centricity—Consonant-centric measures characterize vocalizations based on the presence of consonants (e.g., consonant inventory and proportion of vocalizations with a canonical syllable). Non-consonant-centric vocalization measures focus on vowels or do not emphasize consonants over vowels or vice versa (e.g., they simply report number of vocalizations or communicative vocalizations). Vocalizations that exclude consonants are more likely to be atypical or less speech-like than vocalizations with consonants (Oller et al., 2010). Nonetheless, “consonant-centric” and “speech-like” are not synonymous. Some vocalizations may contain consonants, but lack “speech-like rhythmic organization” due to excessively long or excessively short duration, for example (Oller et al., 2010, SI Appendix, p. 24).

Consonant use in prelinguistic vocalizations correlates positively with later expressive language skills, including vocabulary and phonological skills, in typical development (e.g., Vihman & Greenlee, 1987; Watt et al., 2006). The onset of canonical babbling, which

includes consonants by definition, is a critical language development step. Canonical babbling not only demonstrates important child skills, but it also influences adult use of language facilitation strategies. Caregivers respond to child vocalizations with consonants more consistently and with more complex language than vocalizations without consonants or non-speech productions (Gros-Louis, West, Goldstein, & King, 2006; Warlaumont, Richards, Gilkerson, & Oller, 2014). In addition, maternal responsiveness positively correlates with child language (Haebig, McDuffie, & Weismer, 2013; Wu & Gros-Louis, 2014). Child vocalizations with consonants may elicit enhanced caregiver input, which in turn, increases the semantic basis for expressive language development. Thus, consonants may be associated more closely with expressive language than vocalizations without consonants for children with ASD.

2.1.2. Communicativeness—Communicative vocalizations measured at 18 to 24 months accounted for unique variance in language skills at three years of age over and above noncommunicative vocalizations (Plumb & Wetherby, 2013). Similarly, inventories of consonants used in communication acts of initially preverbal children with ASD accounted for unique variance in expressive language growth above and beyond eight other theoretically-motivated predictors and two background variables (Yoder et al., 2015). We predicted that communicative vocalizations would be correlated more strongly with expressive language than noncommunicative vocalizations.

2.1.3. Concurrent versus longitudinal correlations—Both concurrent and longitudinal associations between vocalizations and expressive language are reported in the literature (e.g., Paul et al., 2008; Plumb & Wetherby, 2013; Sheinkopf et al., 2000; Wetherby et al., 2007; Yoder et al., 2015). We included both types to maximize comprehensiveness. Concurrent and longitudinal correlations provide different levels of evidence for making causal inferences. Evidence for causal inferences must meet three criteria: (a) association between two variables of interest, (b) temporal precedence of the occurrence of the putative cause prior to the occurrence of putative effect, and (c) exclude all other possible explanations for the association. Concurrent correlations only meet the first criterion. Longitudinal correlations meet the first two, and, therefore, provide more convincing evidence. Neither can exclude all other possible explanations. We predicted that the weighted mean effect size for concurrent correlations would exceed that for longitudinal correlations.

2.1.4. Risk for correlated measurement error—For this meta-analysis, risk for correlated measurement error occurs when a vocalization measure is correlated with a language measure that was derived from the same context (e.g., a parent-child free play session) or the same source (e.g., parent report). When this type of correlated measurement error occurs, the apparent correlation between two variables of interest can be elevated artificially (Yoder & Symons, 2010). We predicted a higher mean effect size for effect sizes at risk for correlated measurement error than those not at risk.

2.1.5. Publication status—Publication bias, a known risk to the validity of meta-analyses, occurs “when published research on a topic is systematically unrepresentative of

the population of completed studies on that topic” (Rothstein, 2008, p. 61). We examine publication status as a possible indicator of publication bias. Additional means are described under Methods. We did not anticipate detecting a significant effect of publication status, but examination of such is an attribute of well-conducted research syntheses.

2.2. Research Questions

We addressed two research questions: (1) What is the average strength of the association between vocalizations and expressive language in young children with ASD? (2) Does the strength of the association between vocalizations and expressive language vary by consonant-centricity, communicativeness, research design, risk for correlated measurement error, or publication status? Summary-level effect sizes and their confidence intervals are weighted due to variation in sample size and number of relevant within-study effect sizes.

3. Methods

3.1. Inclusion Criteria

3.1.1. Population—Consistent with definitions of “young children,” the mean or median age for participants for each effect size must be no greater than 8 years, 11 months at the time of the vocalization measure (National Association for the Education of Young Children, n.d.). Participants must be diagnosed with ASD. Due to the anticipated publication dates of studies included in this meta-analysis and changing criteria and terminology for what is now termed autism spectrum disorder, studies including participants with autism spectrum disorder(s), autistic disorder, pervasive developmental disorder—not otherwise specified, autism, high-functioning autism, and Asperger’s disorder/syndrome were included if they met other inclusion criteria. Groups of children at “high-risk” for ASD were included only if they were later diagnosed with ASD.

3.1.2. Measures—Each included report must include at least one correlation between a child vocalization measure and a child expressive language measure. Parent report measures were permitted.

In this meta-analysis, “vocalizations” are defined as non-vegetative sounds produced by the child that are not distinct word approximations or words (e.g., coos, squeals, open vowels, marginal babbling, and consonant-vowel combinations). In an effort to include all relevant effect sizes, vocalization measures could include cries and/or laughs. However, measures exclusively describing cries were excluded. Vocalization measures at or above the word level (e.g., number of words or syllable shape diversity) were excluded. Children use vocalizations and early words at the preverbal stage of development. To guard against vocalization measures assessing language rather than non-word vocalizations, vocalization measures at risk of including words were excluded if the group’s mean developmental level exceeded 24 months. All automated vocal analyses variables (e.g., Language ENvironment Analysis [LENA] system) were considered at risk for including words.

“Language” is defined as a “socially shared code or conventional system for representing concepts through the use of arbitrary symbols and rule-governed combinations of those symbols” (Owens, 2008, p. 4). Each included report must include a measure of expressive

language (e.g., expressive vocabulary, mean length of utterance, morphosyntactic skills and speech accuracy) or total (i.e., receptive and expressive) language measured at the same time as the vocalization measure or a later time point. Total language measures were permitted due to the anticipated low prevalence of expressive language measures reported independently in the literature.

3.1.3. Study design elements—Reports included a Pearson's r , partial r , or convertible statistic with necessary assumptions met (e.g., beta coefficient, tau, and Spearman's rho) for a group of participants meeting the population criteria. The sample size on which effect size was estimated must include five or more participants. Correlations from groups with fewer participants are not sufficiently powered even for strong associations and are difficult to assess for meeting assumptions of correlations.

A variety of study designs, including mixed methods, were included as long as a concurrent or longitudinal correlation between the necessary measures was provided. Intervention studies were permitted if associations were reported prior to intervention or if the study reported no interaction between group membership and vocalizations predicting the language measure. Studies were not excluded by specific quality indicators. Instead, we conducted moderator analyses.

3.2. Exclusion Criteria

Single case research designs and qualitative designs were excluded, regardless of the number of participants due to the lack of correlations available. Review articles and other non-primary reports were excluded at the full text level. Due to limited resources to translate studies written in other languages, reports unavailable in English were excluded. No exclusions were made based on publication date.

Effect sizes were excluded if more than 33% of the group was (a) diagnosed with a sensory and/or physical limitations impacting language acquisition (i.e., cerebral palsy, motor speech disorders, cleft lip and/or palate, hearing loss, and history of a tracheostomy) or (b) bilingual or spoke a primary language other than English. Languages vary in phonetic and acoustic properties that may influence the association between vocalizations and expressive language. Identifying these influences was beyond the scope of this meta-analysis.

3.3. Search Strategy

Multiple study methods were used to locate all relevant effect sizes. The main search was completed through electronic databases. We searched the Education Database, ERIC, Health & Medical Collection, Linguistics and Language Behavior Abstracts, Linguistics Database, ProQuest Dissertations & Theses Global, Psychology Database, PsycINFO, and Social Science Database in ProQuest on September 30, 2016, and PubMed and Cumulative Index of Nursing and Allied Health Literature (CINAHL) on October 1, 2016. See Appendix for an example search. Additionally, the first author hand searched tables of contents from the past year for journals yielding at least five studies for the full text screening, conducted forward searches of all included articles, scanned included articles' reference lists, reviewed proceedings from the 2015 and 2016 International Meeting for Autism Research, and

contacted experts for unpublished findings. Supplementary searches were completed on November 22, 2016.

The second author independently screened 25% of reports at the title and abstract level and the full text level. The primary coder was blind to which reports would be selected for reliability. Discrepancy discussions occurred regularly. Point-by-point agreement (i.e., number of agreements divided by total number of agreements plus disagreements, multiplied by 100) was 93% at the title and abstract level. The primary coder was more conservative. At the full text level, point-by-point agreement was 99% for inclusion/exclusion of reports. The primary coder's judgments were used.

3.4. Coding the Studies

The first and second authors independently extracted data from every included report. The detailed coding manual may be requested from the first author. Point-by-point agreement across all variables was 83%. All disagreements were resolved by consensus. Consensus coding was used for data analysis.

Report level features included publication status and whether the report shared participants with other included reports. Effect size level features included sample size, number of participants with ASD, mean chronological age and developmental level at vocalization and language measures, consonant-centricity, vocalization communicativeness, vocalization and language measure reliability level, effect size value and type, risk for correlated measurement error, and handling of missing data. Fisher's z and its variance and standard error were computed.

3.5. Analytic Strategies

3.5.1. Effect size—Consistent with current meta-analytic techniques, Pearson's r correlations were converted to Fisher's z prior to analysis to account for each effect size's sample size (Borenstein, Hedges, Higgins, & Rothstein, 2009) using the Campbell Collaboration online calculator (http://campbellcollaboration.org/resources/effect_size_input.php; Wilson, n.d.). Effect sizes were transformed back to Pearson's r for reporting.

3.5.2. Robust variance estimation—Traditional meta-analytic techniques assume all effect sizes are independent. Effect sizes for the current analysis violate that assumption because individual reports present multiple effect sizes and use shared samples. We used robust variance estimation via the `robumeta.ado` file downloaded from the Stata Statistical Software Components archive to address the dependent effect sizes (Hedges, Tipton, & Johnson, 2010; Tanner-Smith & Tipton, 2014). We used a random effects model with approximately inverse variance weights because the primary cause for dependency is multiple effect sizes per study.

3.5.3. Moderator analyses—We conducted the five planned moderator analyses using meta-regression with robust variance estimation: consonant-centricity, communicativeness, research design, risk for correlated measurement error, and publication status. All

moderators were tested independently after examining intercorrelations among putative moderators.

4. Results

4.1. Study Selection

See Figure 1 for Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flow diagram. Database searches yielded a total of 1248 records. Other sources yielded 173 additional records. After eliminating duplicates and screening titles and abstracts, 301 records remained. At the full text level, coders independently evaluated each criterion in the order listed in Figure 1. The search yielded 109 unique effect sizes across nine studies described in 19 reports.

4.2. Study Characteristics

Table 1 displays study characteristics. The effect sizes included at least 362 unique participants with mean age of 46.16 months ($SD = 23.35$ months; range = 6–114). We used the largest sample size of a single report in the FIRST WORDS® Project as a conservative estimate of this study's sample size.

A wide variety of vocalization measures were used (see Table 2). Although still diverse, fewer language measures were used and the vast majority were standardized assessments. Authors more consistently reported vocalization measure reliability. They rarely reported language measure reliability. This difference may be due to the tendency to use observational measurement for vocalization measures and standardized assessments for the language measures. Only reports using participants from the Yoder et al. (2015) study explicitly described the handling of missing data.

The overall risk of correlated measurement error is judged to be small and a positive indicator for the quality of included studies (Sandbank & Yoder, 2014). Only three reports, two of which were from the same study, showed any risk of correlated measurement error (Kim, 2008; Yoder et al., 2015; Yoder, Woynaroski, Keceli-Kaysili, & Watson, 2016). Two of these three reports used aggregate measures, which tend to increase measures' stability, with only some of the measures being at risk for correlated measurement error (Yoder et al., 2015; Yoder et al., 2016). Consequently, concern that correlated measurement error may have caused the association between vocalization and language measures is minimized.

4.3. Preliminary Analyses

Robust variance estimation requires users to input an assumed common correlation between the effect sizes. We examined the impact of assumed values of ρ in Stata (Tanner-Smith & Tipton, 2014). No variation in weighted mean effect sizes was observed for $\rho = 0.0$ to 0.9 in 0.1 increments. We used $\rho = 0.6$ for all analyses based on extant literature (Condouris, Meyer, & Tager-Flusberg, 2003; Wetherby et al., 2007).

Because partial r values control for at least one other variable, they are expected to be smaller than zero-order correlations. A moderator analysis using robust variance estimation indicated that the mean weighted effect sizes using partial r and r values were not

statistically different at a .01 alpha level (partial $r = .43$; $r = .52$; $p = .41$; 95% CI [-0.51, 0.86]). We used an alpha level of .01 for analyses with degrees of freedom less than four because we have weak confidence in the specific interpretation of the coefficients (Tanner-Smith & Tipton, 2014). All effect sizes were combined for remaining analyses.

4.4. Synthesis of Results

The overall mean effect size for the association between vocalizations and expressive language was significant and strong ($r = .50$, 95% CI [.23, .76]). We rejected the null hypothesis that vocalizations and expressive language are unrelated in children with ASD. Sensitivity analyses in which each report was excluded one at a time did not substantively alter the results, suggesting that no single report unduly influenced the overall mean effect size.

4.5. Heterogeneity

The Galbraith plot and τ^2 provide means for evaluating heterogeneity, which refers to variation in estimated 'true effects' (Borenstein et al., 2009). The Galbraith plot (Figure 2) provides an alternative graphing method for summarizing meta-analyses with too many effect sizes for an interpretable forest plot. 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). Numerous effect sizes fall outside the confidence interval indicating substantial heterogeneity. τ^2 uses the same scale as the effect size (i.e., -1 to 1 for Pearson's r ; Borenstein et al., 2009). Consistent with visual examination of the Galbraith plot, the τ^2 value of 0.14 is substantial. The substantial variability in effect size fulfills one of the necessary conditions for conducting moderator analyses.

4.6. Moderator Analyses

We proceeded with the planned moderator analyses for consonant-centricity, communicativeness, research designs, and publication status. We did not conduct the planned moderator analysis for risk for correlated measurement error, because only two studies exhibited risk for it. As predicted, the research design characteristic of concurrent versus longitudinal designs moderated the association between vocalizations and expressive language ($b = -.466$, $p = .04$). Concurrent correlations ($r = .77$, 95% CI [.45, 1.0]) were significantly stronger than longitudinal correlations ($r = .33$; 95% CI [.05, .60]). None of the other moderator analyses detected an effect. All p values for other moderator analyses exceeded .50 with large confidence intervals.

We used subgroup analyses to explore which moderators may warrant further investigation (see Table 3). Only three effect sizes specifically measured noncommunicative vocalizations. A weighted mean effect size could not be calculated for these three effect sizes, which came from a single study ($r = .01$, $r = .36$, and $r = .37$; FIRST WORDS® Project). Similarly, because 25 of the 26 effect sizes at risk for correlated measurement error were from the same study, the degrees of freedom for mean weighted effect size were too low.

We examined the correlations between the putative moderators to evaluate the degree to which each captured distinct characteristics of studies or dependent variables that might explain variation among effect sizes. Only three putative moderator pairs had correlations that exceeded $r = .3$: communicativeness and risk for correlated measurement error ($r = .48$), consonant-centricity and risk for correlated measurement error ($r = .53$), and consonant-centricity of vocalization measures and communicativeness ($r = .61$). Although there were a limited number of effect sizes at risk for correlated measurement error, most of the effect sizes used consonant-centric or communicative vocalization measures.

We evaluated risk for publication bias via funnel plots and the Egger's test. The funnel plot (see Figure 3) appears symmetrical with only one small study near the weighted mean effect size. The funnel plot should be interpreted with caution due to the small number of studies. The Egger's test provides a statistical test for a "small-study effect" wherein small studies have larger effect sizes than larger studies (i.e., evidence of publication bias; Egger, Smith, Schneider, & Minder, 1997). Using the Egger's test, we found little evidence of small study bias ($p = .86$).

5. Discussion

5.1. Summary of Evidence

Based on 109 effect sizes from nine studies with a total of at least 362 participants, the weighted mean effect size of the association between vocalizations and expressive language in children with ASD is strong ($r = .50$). Various measures of vocalizations account for 25% of the variance in expressive language in young children with ASD. However, as predicted, the mean effect size for concurrent correlations was significantly larger than that of longitudinal correlations. Nonetheless, the mean effect size for longitudinal correlations is notable. These weighted mean effect sizes provide more accurate estimates of the population estimate for the associations of interest than any single study. The strength of the association between vocalizations and expressive language is substantial, particularly for concurrent associations. Vocalizations correlate with expressive language in children with ASD to a similar or larger degree than other early communicative behaviors (Bottema-Beutel, 2016; Harbison, McDaniel, & Yoder, 2017). These findings support progressing toward testing the causal relation between vocalizations and expressive language in children with ASD.

Other tested moderators did not detect effects. Power for identifying moderators may be reduced by the magnitude of the true relation and/or the precision of the estimate, which is influenced by the number of subjects across studies and number of studies. Because only nine studies met inclusion criteria and many had relatively small sample sizes (mean $n = 46$), low power is likely due to limited precision. Omitted or imprecise reporting of key study characteristics, including vocalization communicativeness and reliability, further reduced power for specific analyses. A meta-analysis with more studies and a larger number of total participants might reveal significant moderator effects.

Consistent with other evaluations of publication bias, publication status was not a significant moderator of the association between vocalizations and expressive language. The large

number of unpublished findings included and the use of grey literature and supplementary searches further reduces the likelihood that the results are a function of publication bias.

5.2. Limitations

For the current meta-analysis, the most prominent limitation at the primary-study level was imprecision or ambiguity in defining and reporting key constructs and reliability, particularly for vocalizations. Surprisingly, eight reports did not define vocalizations explicitly. Differentiating vocalizations from language and language from prelinguistic communication is necessary for accurately identifying a child's current skills, capturing progress, and predicting growth, particularly for children with ASD who may remain in the preverbal or early stages of word learning for an extended time. Second, a number of screened reports were excluded despite using acceptable vocalization and expressive language measures because they did not report an association between them. Authors are encouraged to report such correlations even if they are not primary findings to advance the literature base. Third, nearly half of the included reports shared participants with another report, often without sufficient information as to which participants overlapped. Using a limited participant pool increases the risk of finding sample specific results that will not replicate. These weaknesses in reporting are not unique to this set of studies; instead, they permeate child language research and related areas.

Robust variance estimation works best with at least 40 studies and focuses on mean effect sizes and meta-regression coefficients, not heterogeneity parameters (Tanner-Smith & Tipton, 2014). This synthesis found far fewer than 40 relevant studies. As with any systematic review or meta-analysis, we may have failed to include relevant effect sizes. To guard against this concern, we completed supplementary searches and reliability checks at every screening level. We cannot draw causal conclusions from these correlational data. Other untested variables could moderate the association between vocalizations and language. Finally, we only included participants with a primary language of English.

5.3. Strengths

At least four strengths should be noted. First, we used robust variance estimation to prevent loss of potentially important effect sizes. Second, we located a large number of unpublished effect sizes, adding to the knowledge base and increasing the robustness of the findings. Third, we considered the effect of research design. Fourth, we conducted independent interrater reliability for screening at all levels and two coders independently extracted data from all included reports.

5.4. Future Research

At the primary-study level, we need additional studies that permit evaluation of this study's putative moderators. Low power may have limited our ability to detect moderator effects in the current study. At the meta-analytic level, our field would benefit from evaluating the association between vocalizations and expressive language in other populations (e.g., children with other disabilities, children with typical development, and children who speak languages other than English). The relation between dyadic variables focusing on the reciprocal nature of vocal exchanges and later spoken language should be evaluated as well

because addressing child and adult variables that influence one another and vocal development may be necessary for effective intervention (Gratier et al., 2015; Jaffe et al., 2001; Pickles et al., 2016). In addition, to progress toward developing an effective language intervention targeting prelinguistic vocalizations for preverbal children with ASD, a systematic review and meta-analysis of vocalization intervention strategies, such as the frequency, complexity, and communicative use of vocalizations including integration of vocalizations with other means of communication, is warranted.

5.5. Conclusions

The strong association between vocalizations and expressive language supports progressing toward evaluations of causal relations between vocalizations and expressive language. Due to the continuity of vocalizations and language, such interventions will need to consider the forms and functions of vocalizations and language. Viewing vocalizations and language as related, but not synonymous, may lead to intervention strategies that can improve the language development trajectory of children with ASD. Despite substantial heterogeneity between studies, only one moderator analysis was significant. Future attempts to identify heterogeneity, with the goal of informing intervention development, will benefit from primary studies clearly defining and reporting key constructs (e.g., vocalizations and language).

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Appendix

Search Strategy

ProQuest: Education Database, ERIC, Health & Medical Collection, Linguistics and Language Behavior Abstracts, Linguistics Database, ProQuest Dissertations & Theses Global, Psychology Database, PsycINFO, Social Science Database

Search: ti,ab,su(Autis* OR Asperger OR Aspergers OR PDD OR PDD-NOS OR “pervasive developmental disorder*”) AND ti,ab,su(infants OR infant OR children OR preschool OR preschoolers OR toddlers) AND ti,ab,su(vocalization OR vocalizations OR babble OR babbling OR babbles OR cooing OR coo OR consonant OR consonants OR vowel OR vowels OR “automated vocal analysis” OR LENA OR profile OR profiles) AND (correlat* OR longitudinal OR concurrent OR “related to” OR predict*) AND (language OR vocabulary OR speech OR articulation OR intelligibility OR expressive)

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What this paper adds?

- This study is the first known meta-analysis on the association between vocalizations and expressive language in young children with autism spectrum disorder (ASD).
- Nine studies (19 reports), which included at least 362 participants and 109 unique effect sizes, met inclusion criteria.
- The results indicate a significant weighted mean effect size of $r = .50$, 95% CI [.23, .76].
- The weighted mean effect size is particularly strong for concurrent correlations ($r = .77$, 95% CI [.45, 1.0]).
- The findings provide support for investigating causal relations between prelinguistic vocalizations and expressive language in young children with ASD to inform language intervention, particularly at the early stages of language development.

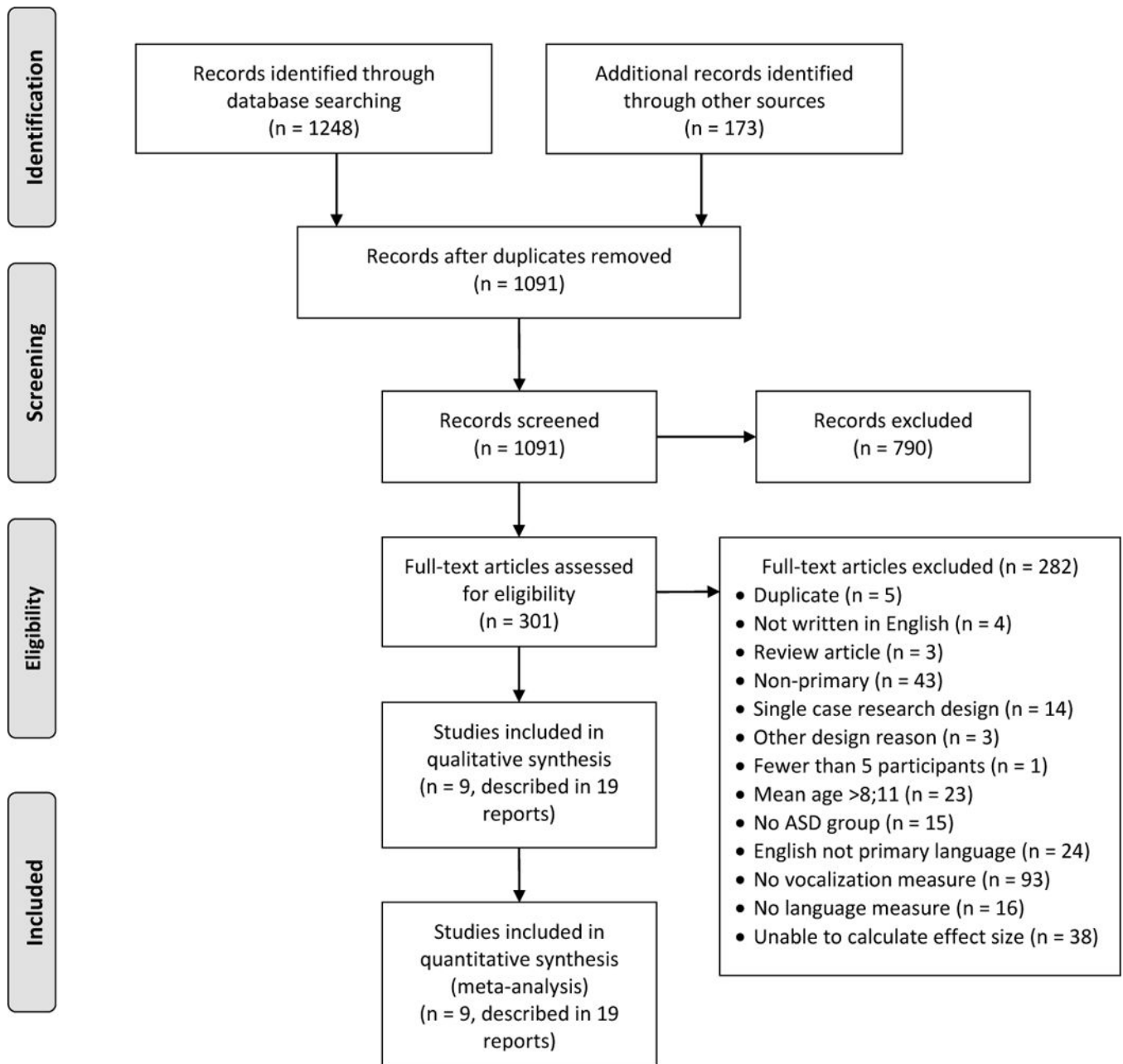


Figure 1. Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) flow diagram.

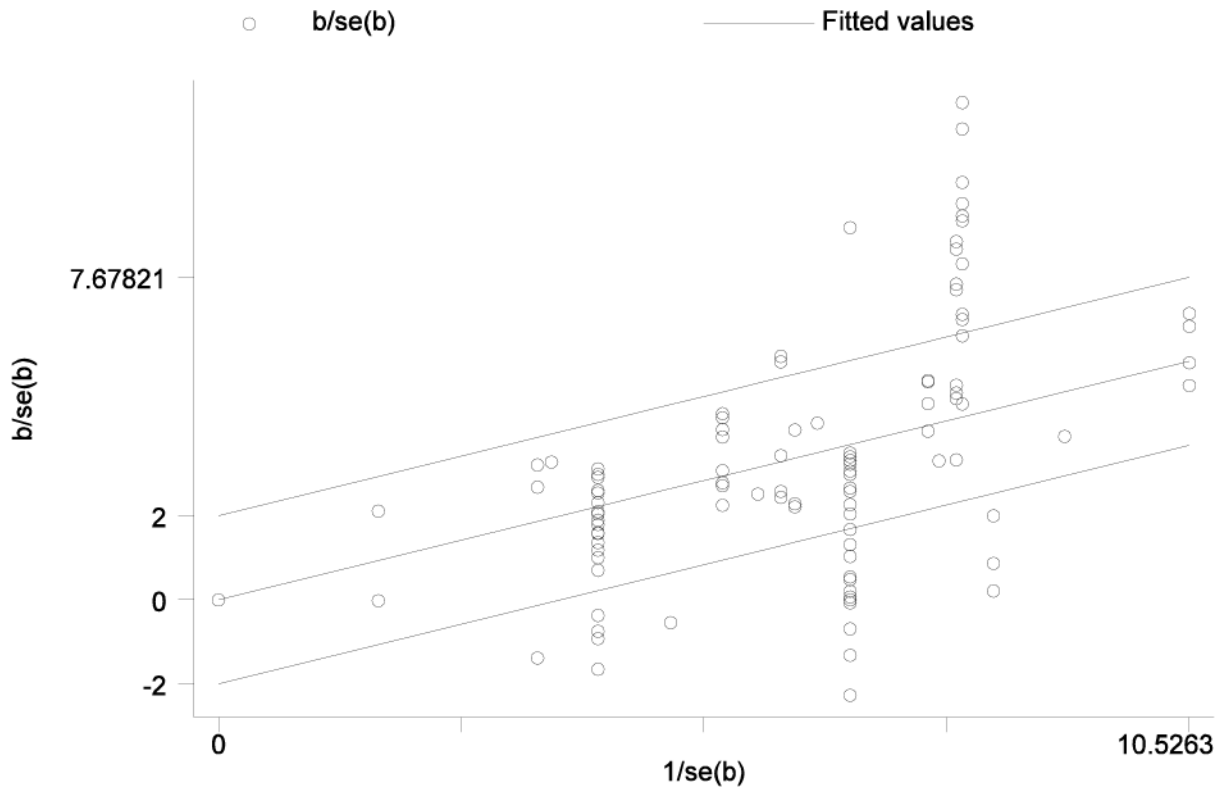


Figure 2. Galbraith plot for correlations between vocalization and language measures.

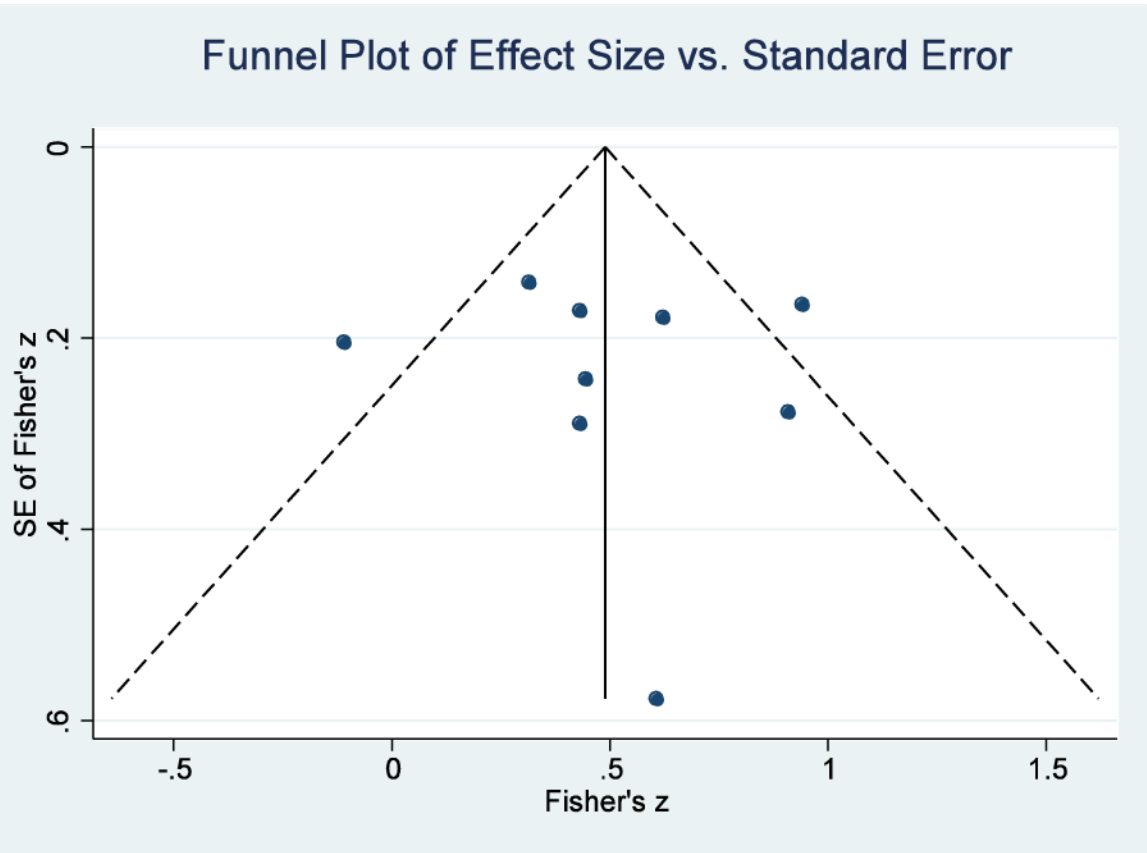


Figure 3. Funnel plot of effect size (Fisher's z) versus standard error for all included studies.

Table 1

Characteristics of Included Reports

Reference	Pub	Mean age at TI (m)	n	Total	Effect Sizes				ES
					Long (Time)	Conc	Cons	Com/Non	
Independent Samples									
Cardona (2004)	No	28.60	20	4	4(17)	0	4	4	0.42
Kim (2008)	Yes	48.00	27	1	1(N/A)	0	0	0	-0.11
Paul et al. (2008)	Yes	21.80	37	1	1(25)	0	1	1	$\neq^*0.41$
Sheinkopf et al. (2000)	Yes	44.67	15	3	0	3	1	0	0.41
Sigman & Ungerer (1984)	Yes	51.70	16	1	0	1	0	0	0.72
Talbott (2014)	No	8.93	6	2	2(3)	0	1	0	0.54
Yoder et al., 2013	Yes	38.60	40	2	0	2	0	0	0.74
Shared Samples									
FIRST WORDS® Project									
Book (2009)	No	21.98	45	1	0	1	1	1	0.30
McCoy (2013)	No	19.83	114	4	2(12)	2	4	4	0.52
Plumb (2008)	No	21.29	50	22	22(18)	0	1	2/3	$\neq^*0.19$
Plumb & Wetherby (2013)	Yes	21.29	50	4	4(18)	0	0	4	$\neq^*0.26$
Shumway & Wetherby (2009)	Yes	21.29	40	3	3(18)	0	0	3	$\neq^*0.43$
Swineford (2011)	No	19.80	74	3	1(16)	2	0	3	0.12
Wetherby et al. (2007)	Yes	21.36	42-64	5	4(16)	1	5	5	$\neq^*0.55$
Sample from Yoder, Watson, et al. (2015)									
Woynaroski (2014)	No	39.67	33	8	4(4)	4	4	4	$\neq^*0.56$
Woynaroski et al. (2017)	Yes	37.90	20	5	5(4)	0	1	1	0.45
Yoder, Watson, et al. (2015)	Yes	34.70	87	1	1(16)	0	1	1	$\neq^*0.40$
Yoder, Woynaroski, et al. (2015)	No	25.00	20	15	15(4)	0	1	0	0.26
Yoder et al. (2016)	No	42.13	62-68	24	16(4-12)	8	24	24	0.70
Total		46.16	362	109	85(11)	24	49	57/3	

Note.

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= For at least one effect size, the sign has been changed to reflect increased frequency of vocalizations being associated with higher language skills to be consistent with other effect sizes;

\hat{r} = Partial r for at least one effect size.

* = At least one effect size originally not reported as r or partial r . Com/Non = Communicative vocalization effects sizes/noncommunicative effect sizes; Conc = Concurrent; Cons = Consonant-centric; ES = unweighted mean effect size (r); Long = Longitudinal; m = Months; n = Average number of participants per effect size; N/A = Not applicable; Pub = Publication status; T1 = Time 1/time of vocalization measure; Time = Average time between vocalization and language measures in months.

Table 2

Vocalization and Language Measures of Included Reports

Reference	Vocalization measures	Language measures
Independent Samples		
Cardona (2004)	CSBS: Sounds Subscale CSBS: Speech Composite	Reynell Expressive ^E & Vineland Expressive ^E
Kim (2008)	Onset of Babbling	Onset of First Words ^E
Paul et al. (2008)	CSBS-DP: Consonant Inventory	Vineland Expressive (“good” or “poor”) ^E
Sheinkopf et al. (2000)	Canonical babbling, atypical vocalizations, and utterance count during ESCS	Reynell Expressive ^E
Sigman & Ungerer (1984)	Uzgiris-Hunt Ordinal Scales of Psychological Development: Vocal imitation	Clinical assessment ^E
Talbott (2014)	Home video diary: Vowel and consonant-vowel vocalizations	Mullen Expressive ^E
Yoder et al. (2013)	LENA Vocal Development Age Equivalency	LDS ^E & CDI ^E
Shared Samples		
FIRST WORDS® Project		
Book (2009)	SORF-Home: Communicative vocalizations with consonants	Mullen Verbal DQ ^T
McCoy (2013)	SORF-Clinic and SORF-Home: Lack of vocalizations with consonants	CSBS Words ^E and Mullen Verbal DQ ^T
Plumb (2008)	Transcribable and non-transcribable vocalizations during CSBS	Mullen Verbal DQ ^T
Plumb & Wetherby (2013)	Total and communicative vocalizations during CSBS-DP	Mullen Verbal DQ ^T
Shumway & Wetherby (2009)	CSBS-DP: Proportion of behavior regulation and joint attention acts with a vocalization	Mullen Verbal DQ ^T
Swineford (2011)	SORF-Home: Communicative vocalizations with consonants	CSBS-DP Words ^E & Mullen Verbal DQ ^T
Wetherby et al. (2007)	CSBS-DP: Consonant inventory	ADOS Communication ^T , CSBS-DP Words ^E & Mullen Verbal DQ ^T
Sample from Yoder, Watson, et al. (2015)		
Woynaroski (2014)	Canonical syllables and consonants during CSBS-DP and SSCS Multiple LENA variables	MB-CDI Expressive ^E
Woynaroski et al. (2017)	Canonical syllables and consonants during CSBS-DP and SSCS Multiple LENA variables	MB-CDI Expressive ^E
Yoder, Watson, et al. (2015)	CSBS-DP: Consonant inventory	MB-CDI Expressive ^E , CSBS-DP Words ^E & UCS NDW ^E
Yoder, Woynaroski, et al. (2015)	Multiple LENA variables	MB-CDI Expressive ^E
Yoder et al. (2016)	Various metrics for canonical syllables and consonant inventory	MB-CDI Expressive ^E , CSBS-DP Words ^E & UCS NDW ^E

Note. ADOS = Autism Diagnostic Observation Schedule (Lord et al., 2000); CDI = Child Development Inventory (Ireton, 1992); CSBS = Communication and Symbolic Behavior Scales (Wetherby & Prizant, 1993); CSBS-DP = Communication and Symbolic Behavior Scales–Developmental Profile (Wetherby & Prizant, 2002); Mullen Verbal DQ = Mullen Verbal Developmental Quotient (Mullen, 1995);

E = Expressive language only measure; ESCS = Early Social Communication Scales (Mundy et al., 2003); LENA = Language ENvironment Analysis; LDS = Language Development Survey (Rescorla, 1989); MB-CDI = MacArthur-Bates Communicative Development Inventories–Words and Gestures Form (Fenson et al., 2007); NDW = Number of different words; Reynell = Reynell Developmental Language Scales (Reynell & Gruber, 1990); SORF = Systematic Observation of Red Flags (Dow, Guthrie, Stronach, & Wetherby, 2016); SCS = Semi-structured communication sample;

T = Total language measure; UCS = Unstructured communication sample; Vineland = Vineland Adaptive Behavior Scales–Survey Edition (Sparrow, Balla, & Cicchetti, 1984).

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Table 3

Moderator Analysis Subgroup Results

Subgroup	<i>n</i>	<i>df</i>	<i>r</i>	95% CI
Overall weighted mean effect size	109	7.54	.50	[0.23, 0.76]
Consonant-centric	54	4.29	.62	[0.38, 0.87]
Non-consonant-centric	55	5.69	.39	[-0.02, 0.79]
Communicative	57	2.95	.54	[0.19, 0.90]
Undifferentiated	47	4.96	.44	[-0.01, 0.89]
Concurrent	24	3.94	.77	[0.45, 1.0]
Longitudinal	85	4.42	.33	[0.05, 0.60]
Published	24	5.92	.50	[0.18, 0.83]
Unpublished	83	2.63	.46	[0.10, 0.82]
At risk for correlated measurement error	26	1.00	–	–
Not at risk for correlated measurement error	83	6.43	.55	[.32, 0.78]

Note. When *df* is less than 4, results should be interpreted with caution. *df* = degrees of freedom; *n* = number of effect sizes; *r* = weighted mean effect size.