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Why Dose Frequency Affects Spoken Vocabulary in Preschoolers With Down Syndrome

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Abstract

In an earlier randomized clinical trial, daily communication and language therapy resulted in more favorable spoken vocabulary outcomes than weekly therapy sessions in a subgroup of initially nonverbal preschoolers with intellectual disabilities that included only children with Down syndrome (DS). In this reanalysis of the dataset involving only the participants with DS, we found that more therapy led to larger spoken vocabularies at posttreatment because it increased children's canonical syllabic communication and receptive vocabulary growth early in the treatment phase.

Keywords

Down syndrome; dose frequency; early intervention; Milieu Teaching; spoken vocabulary; canonical syllables; receptive vocabulary

The majority of reports on vocabulary development of children with Down syndrome (DS) suggest a pattern of slow early lexical development that yields later spoken vocabulary deficits that are more severe than expected, given the children's global cognitive impairment (Cardoso-Martins, Mervis, & Mervis, 1985; Caselli, Monaco, Trasciani, & Vicari, 2008; Miller, 1992, 1999; Warren et al., 2008; c.f., Caselli et al., 1998; Galeote, Soto, Checa, Gomez, & Lamela, 2008; Vicari, Caselli, & Tonucci, 2000). This difficulty in spoken vocabulary acquisition is observed even when chronological age, mental age, and level of cognitive impairment are all controlled (Warren et al., 2008; Yoder & Warren, 2004; Yoder,

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Woynaroski, Fey, & Warren, 2014). Thus, the literature strongly suggests that children with DS struggle significantly with spoken vocabulary development.

In an earlier report involving a subgroup of toddlers (18 – 27 months) with intellectual disabilities (ID) due to DS, children who received daily 1-hr sessions (i.e., a higher dose frequency) of a naturalistic, play-based early communication intervention called Milieu Communication Teaching (MCT) had superior spoken vocabulary outcomes after 9 months of treatment relative to children who received only weekly 1-hr sessions (i.e., a lower dose frequency) of the same therapy method, controlling for initial level of cognitive impairment (Yoder et al., 2014). We interpret these findings to mean that level of cognitive impairment accounts for much variance in spoken vocabulary development, and that, once level of cognitive impairment is controlled, effects of dose frequency on spoken vocabulary outcomes in children with DS were apparent. However, we do not yet understand the mechanisms through which more frequent MCT sessions per week translated to gains on spoken vocabulary outcomes of toddlers with DS.

Rationale for the Proposed Mediators of the MCT Effect on Spoken Vocabulary

More frequent MCT might affect spoken vocabulary outcomes of toddlers with DS by increasing the complexity of their prelinguistic productions. In a secondary analysis of our dataset including subgroups of children with ID with DS and without DS, we recently discovered that dose frequency facilitated increases in the proportion of intentional communication acts with canonical syllables that children produced (i.e., canonical syllabic communication; Woynaroski, Yoder, Fey, & Warren, 2014). Canonical syllables are vocalizations that include at least one consonant and vowel combination produced with adult-like speech timing (Oller, 1978). However, we did not test whether dose frequency affected canonical syllabic communication in just the subgroup of children with ID due to DS. Therefore, it is unclear whether the effect of increased dose frequency on spoken vocabulary was, in fact, preceded by an effect of dose frequency on canonical syllabic communication in the subgroup of children with DS.

Although we have not tested this in our earlier studies, more frequent MCT might also affect spoken vocabulary outcomes of children with DS by boosting receptive vocabulary in children with DS. One previous study found that the component of MCT employed for children in the prelinguistic stage, Prelinguistic Milieu Teaching, was found to facilitate receptive language for a subgroup of children with ID who had parents with relatively high formal education or initially high responsivity to children's communication (Yoder & Warren, 2001). Additionally, we found that an increased dose frequency of our present version of MCT produced an effect on receptive vocabulary for the subgroup of our broader sample of children with ID who played functionally with a variety of objects at study outset (Fey, Yoder, Warren, & Bredin-Oja, 2013). These findings provide empirical support for our hypothesis that more frequent MCT sessions might have enhanced receptive vocabulary growth in our sample of children with DS before we detected a dose frequency effect on spoken vocabulary outcomes.

We have presented our rationale for proposing that canonical syllabic communication and receptive vocabulary may be affected by increased dose frequency in children with DS. However, for canonical syllabic communication and receptive language to explain (i.e., mediate) the effect of dose frequency on spoken vocabulary in our sample of children with DS, these variables must also be linked with later spoken vocabulary outcomes. Various metrics of canonical syllabic vocalization or communication during the prelinguistic period have been shown to predict later spoken language in young children with ID (Yoder & Warren, 2004; Yoder, Warren, & McCathren, 1998), as well as children with specific expressive language delays (Wetherby, Yonclas, & Bryan, 1989; Whitehurst, Smith, Fischel, Arnold, & Lonigan, 1991) and typically developing children (Menyuk, Liebergott, & Schultz, 1986; Stoel-Gammon, 1989; Watt, Wetherby, & Shumway, 2006; Wetherby, Cain, Yonclas, & Walker, 1988). Similarly, early receptive vocabulary predicts later spoken vocabulary in children with ID (Chapman, Seung, Schwartz, & Kay-Raining Bird, 2000; Vandereet, Maes, Lembrechts, & Zink, 2010). Therefore, we anticipated that canonical syllabic communication and receptive vocabulary during the treatment phase would predict spoken vocabulary outcomes at the end of the treatment phase in our sample of children with DS.

Thus, there is reason to believe early effects of dose frequency manipulations of MCT will occur on both canonical syllabic communication and receptive vocabulary. Additionally, these dose frequency effects might explain the previously discovered effect of dose frequency on spoken vocabulary outcomes in children with DS at the posttreatment period when level of cognitive impairment was controlled (Yoder et al., 2014).

Research Questions

We had two research questions:

1. Controlling for level of cognitive impairment, does daily MCT have a greater effect than weekly MCT on canonical syllabic communication and receptive vocabulary of children with DS early in the treatment phase?
2. Do early dose frequency effects on canonical syllabic communication and receptive vocabulary explain our previously reported dose frequency effects on spoken vocabulary outcomes of children with DS (controlling for cognitive impairment)?

Method

Design

In this between-groups experiment, children were randomly assigned to either (a) one 1- hr MCT session per week (i.e., weekly treatment), or (b) five 1-hr MCT sessions per week (i.e., daily treatment). Select communication and spoken language abilities were assessed at four measurement periods: (a) pre-treatment (Time 1), (b) three months after treatment onset (Time 2), (c) 6 months after treatment onset (Time 3), and (d) immediately following completion of the full nine month treatment protocol (Time 4). In our dataset, canonical syllabic communication was measured only at Times 1 and 2. Receptive vocabulary and spoken vocabulary were measured at all time periods. For this re-analysis, we included measures of canonical syllabic communication from Time 2, receptive vocabulary from Time 1 to Time 3, and spoken vocabulary from Time 4.

Participants

In recruiting for our larger study we advertised for 18– 27-month-old children from English-speaking homes with general developmental delay, significant delays in the acquisition of words, and no diagnosis of autism. Inclusion criteria were (a) 20 or fewer spoken or signed words in the expressive lexicon, as reported by the primary caregiver on the MacArthur-Bates Communicative Development Inventories: Words and Gestures vocabulary checklist (MB-CDI, Jackson-Maldonado et al., 2003).); (b) Bayley Scales of Infant and Toddler Development, Third Edition (Bayley III; Bayley, 2006) Cognitive Composite (CC) standard score between 55 and 75; (c) a score of under 2.75 for children 18–23 months old and a score of under 2 for children 24 –27 months old on the Screening Tool for Autism in Two-Year-Olds (STAT) (Stone & Ousley, 1997, indicating low risk of autism spectrum disorder; (d) normal hearing in at least one ear, as determined by a hearing screening; (e) normal, or corrected-to-normal, vision per parent report; and (f) motor skills sufficient to sit unsupported and to engage in play with an interventionist. Diagnosis of DS was confirmed by the child's parents, who had learned of the diagnosis either at birth or earlier, through physician identification or genetic testing. We do not know whether any of these children had a mosaic variant of DS rather than the full trisomy 21, but no parent reported such variation.

Thirty-five preschoolers or toddlers with DS and their parents were included for analysis in this report. All of the children had fewer than 20 words as indicated by parent vocabulary checklist. Within our sample of children with DS, 16 children were randomly assigned to receive weekly MCT, and 19 children were assigned to receive daily MCT. Table 1 provides additional detail on sample characteristics at Time 1. Importantly, treatment groups were nonsignificantly different on all variables of interest at entry to the study.

Treatment Dose Frequency

All children received MCT comprising Prelinguistic Milieu Teaching (Yoder & Warren, 1998), Milieu Language Teaching (Hancock & Kaiser, 2006), and Responsivity Education (Yoder & Warren, 2002). A detailed description of MCT is provided in Fey et al. (2013) and Warren et al. (2008). Briefly, MCT begins with prelinguistic targets. At first, children are encouraged to combine unconventional gestures (e.g., tapping an object with fingers or giving an object to the adult) or nonword vocalizations with attention to object and person. Later, goals shift to combining conventional gestures (e.g., a head nod or shake, a distal point) with attention to adult. When children show readiness for word production, targets shift to using words to communicate about child-selected foci and activities. Concurrent with MCT, parents were taught to follow their children's attentional and communicative leads and to talk about the children's play actions and foci of attention. Interventionists were college graduates with some training in child development who were supervised by licensed and certified speech-language pathologists. They provided MCT in homes or child care centers according to parent preference. Details of interventionist training have been described in Fey et al. (2013). The contrast between one and five 1-hr sessions per week represents our test of MCT dose frequency. We refer to the between-group mean differences on spoken vocabulary, canonical syllabic communication, or receptive vocabulary as *dose frequency* effects.

Measuring fidelity to the dose frequency protocol: The interventionists recorded attendance to, and duration of, each session for each participant. Additionally, 30-min segments of one 60-min treatment session per month were coded to quantify the rate of correct MCT teaching episodes per minute. The estimated cumulative number of correct MCT teaching episodes was computed as the product of (a) average duration of sessions, (b) total number of sessions attended, and (c) rate of correct teaching episodes per minute. Mean cumulative exposure to correct teaching episodes was more than four times greater for daily ($M = 9718$, $SD = 3417$) versus weekly ($M = 2242$, $SD = 519$) MCT groups, $t(33) = 8.6$, $p < .001$, $d = 2.7$. Further details about fidelity of treatment are available in previous reports (Fey et al., 2013; Yoder et al., 2014).

Procedures and Metrics

Level of cognitive impairment—At Time 1, the Bayley III CC (Bayley, 2006) standard score was used to quantify children's *level of cognitive impairment* (i.e., the degree to which cognitive ability was delayed relative to chronological age expectations). This measure is among the most widely used instruments for evaluating intellectual abilities of infants and toddlers (Anderson, De Luca, Hutchinson, Roberts, & Doyle, 2010). Level of cognitive impairment was used as a covariate in analyses related to both research questions.

Canonical syllabic communication—Canonical syllabic communication at Time 2 was summed across three sampling contexts that varied in the level of structure provided and in the familiarity of the adult interaction partner to increase the likelihood of obtaining a stable, and thus valid, estimate of the construct of interest in our sample of young children with DS (Sandbank & Yoder, 2014). Adult interaction partners were blind to treatment group assignment in these contexts with the exception of the parents in the parent-child interaction session.

Communication and symbolic behavior scales—Of the three contexts included in the Time 2 evaluations, the Communication and Symbolic Behavior Scales (CSBS; Wetherby & Prizant, 1993) was the most structured. The adult interaction partner was unfamiliar to the child. Only the Communicative Temptations and the Book Sharing components were coded for canonical syllabic communication.

Examiner-child semi-structured free play—The examiner-child semi-structured free play (ECSS) served as a less structured sampling context relative to the CSBS. The adult interaction partner was unfamiliar to the child. Adult interaction partners were asked to follow the child's lead and to respond to child communications, but to provide limited scaffolding for communication and play behaviors. The total duration of the ECSS was 15 min.

Parent-child free play—The parent-child free play (PCFP) sampling context was the least structured of the communication samples, but the adult interaction partner (the parent or primary caregiver) was familiar to the child and aware of the child's prescribed dose frequency assignment. The PCFP involved 10 minutes of free play and 5 min of unstructured book sharing. Parents were instructed to offer their child a choice of two play sets and to play as they would typically play at home. They were also told that they could switch to the second toy set if their child became disinterested in the first set. For the book sharing segment of this sample, parents were told that they should "look at" the books with their child for 5 min, but they were not directly instructed to read the books. Parents were not told to respond to communication acts in any particular way. Additional details on the three communication samples are provided in Woynaroski et al. (2014).

Communication sample coding—Media files of the three communication samples were coded for intentional child communication acts and for the production of canonical syllables by trained research staffers who were blind to treatment assignment. Each communication sample was coded by a primary coder, and 14% of the samples were randomly selected for independent transcription and analysis by a secondary coder. A coding supervisor reviewed primary and secondary transcripts to discuss coding discrepancies in an attempt to minimize observer drift.

Intentional child communication acts were defined as (a) nonword vocalizations or unconventional gestures combined with coordinated attention to object and person, (b) conventional gestures combined with attention to adult, or (c) symbolic forms (i.e., words, sign language). Canonical syllables were defined as vocal communication acts in which a rapid transition occurred between vowel-like and consonant-like sounds. The metric employed in analyses of canonical syllabic communication was the proportion of child communication acts in which canonical syllables were used across the three Time 2 communication samples (i.e., number of child communication acts including a canonical syllable/total number of child communication acts). This variable, *canonical syllabic communication*, was a dependent variable in analyses addressing the first research question and a putative mediator in analyses addressing the second research question. The intra-class correlation coefficient (ICC) estimate of the inter-observer reliability, calculated in a way that included errors on presence of a communication act and presence of a canonical syllable, was .92. This ICC indicates that there was high consistency between raters in coding and computing proportion of communication acts with canonical syllables.

Receptive and spoken vocabulary—Receptive and spoken vocabulary size were estimated using an adaptation of the MB-CDI (Jackson-Maldonado et al., 2003). We excluded sound effects, animal sounds, generic caregiver names, and words associated with routinized games and activities because these items are less likely to be used to build grammatical utterances. Additionally, we added a response column for "signs and understands" and altered the production column to read "says and understands (may also sign)." *Spoken vocabulary* was the sum of words marked under "says and understands (may also sign)." Words that were signed only were not included in the spoken vocabulary count. The Time 4 estimate of spoken vocabulary was the dependent variable for the second research question. *Receptive vocabulary* was the sum of "understands only" + "signs and understands" + "says and understands (may also sign)."

We used growth curve modeling (i.e., mixed level modeling) to capture children's receptive vocabulary growth across our three measurement periods (Times 1–3). Growth curves derived from three or more measurement periods have been shown to yield estimates of constructs with less measurement error than scores estimated at only one or two measurement periods (Singer & Willett, 2003). With extant data available for three measurement periods for receptive vocabulary, we were able to fit the data to a simple linear growth model, which included intercept and slope parameters. To control for initial levels of receptive vocabulary, we centered children's time in study at Time 1, which enables interpreting the intercept as estimated initial receptive vocabulary level (Singer & Willett, 2003). The slope parameter in this simple linear model indexed growth per month in receptive vocabulary across the first 6 months in treatment. Thus, we used the ordinary least squares estimate of the simple linear slope for receptive vocabulary growth from Time 1 to Time 3 as a dependent variable for the first research question and as a putative mediator for the second research question.

Statistical Plan

The first research question regarding early effects of MCT dose frequency on canonical syllabic communication and receptive vocabulary was addressed through analysis of covariance (ANCOVA) with dose frequency of MCT as the independent variable. Contemporary mediation analysis methods were subsequently used to evaluate whether early effects of MCT dose frequency on canonical syllabic communication and receptive vocabulary could account for the previously observed effect of dose frequency on spoken vocabulary in children with DS (Preacher & Hayes, 2004). Current recommendations for mediation analysis require directly testing the significance of the mediated effect using bias-corrected, bootstrapped confidence intervals when sample sizes are small (Preacher & Hayes, 2004).

Statistically controlling the level of cognitive impairment was necessary to detect the dose frequency effect on spoken vocabulary in preschoolers with DS in our previous report (Yoder et al., 2014). Our intent was to explain the effect of dose frequency on vocabulary using mediation analyses. Doing so required level of cognitive impairment to be statistically controlled in all analyses in this report. Further details regarding mediation analyses are provided in the "Results" section.

Results

Preliminary Results

Documentation that dose frequency was successfully manipulated through group assignment and adherence to the research protocol—As noted earlier, the daily dose frequency provided more than 4 times the teaching episodes as the weekly dose. In some trials, between-group differences in attrition and attendance to non-project treatment can explain between-group differences after randomization occurs, but this is not so for the present study. Nonproject treatment attendance during the treatment phase was similar (about 2.5 hr per month in each group) and nonsignificantly different ($p = .56$) between dose frequency groups. Attrition was low (5%).

Tests of statistical assumptions—There were no violations of homogeneity of slopes in analyses of the effect of dose frequency using level of cognitive impairment as a covariate (p values for these tests ranged from .36 to .88). Tests of the assumption of homoscedasticity indicated a need to transform the two vocabulary measures. After log10 ($x + 1$) transformation, analyses involving the vocabulary measures no longer violated this assumption. Therefore, all analyses using the vocabulary measures used the log10 transformed metric. There was no evidence of undue influence of any given data point in any analysis, max Cook's D values ranged from .2 to .23. Thus, the analyzed data met important statistical assumptions.

Growth model for receptive vocabulary—Mixed level modeling indicated significant fixed effects for both the intercept, $t(34) = 27.86$, $p < .001$, and the slope, $t(34) = 6.09$, $p < .001$. These results indicate that the average receptive vocabulary at Time 1 and the mean growth rate of receptive vocabulary from Time 1 to Time 3 were significantly greater than 0. Significant random effects for the intercept, $\chi^2(34) = 237$, $p < .001$, and the slope, $\chi^2(34) = 142$, $p < .001$, were also observed. These results indicate that receptive vocabulary level at Time 1 and growth rate of receptive vocabulary from Time 1 to Time 3 varied significantly across participants. The average initial receptive vocabulary level (i.e., the mean number of words that children were reported to understand) in the transformed metric (i.e., log10) was 1.54. The average growth rate of receptive vocabulary from Time 1 to Time 3 in the log-transformed units was .065. Using the untransformed receptive vocabulary scores for interpretative purposes, the average initial receptive vocabulary was 44.8 words understood, and the average growth rate of receptive vocabulary was 11 words per month. The reliability of the slope, the measure of growth of receptive vocabulary per month, was .76.

Primary Results

Dose frequency effects on canonical syllabic communication and receptive vocabulary—Controlling for level of cognitive impairment, children in the daily MCT group had significantly higher canonical syllabic communication than children in the weekly MCT group after 3 months of treatment, $F(1,34) = 5.86$, $p = .02$ ($d = .77$). Table 2 provides descriptive statistics by group. Also controlling for level of cognitive impairment, children in the daily MCT group had significantly greater growth in receptive vocabulary than children in the weekly MCT group over the first 6 months of treatment, $F(1,34) = 6.8$, $p = .01$ ($d = .76$). Table 2 provides transformed descriptive statistics, which represent the metric of the variables that was statistically analyzed. The untransformed values are easier to understand: The untransformed IQ-adjusted means were 7 words ($SD = 7$) and 15 words gained per month ($SD = 15$) for weekly and daily MCT groups, respectively. From Time 1 to Time 3, the average increase in untransformed number of words understood in the weekly versus daily MCT groups were 46 and 86, respectively.

Mediation of dose frequency effects—Mediation models can be used to determine whether overall (i.e., total) effects, such as our previous observation of an effect of higher dose frequency on the spoken words of our participants with DS, can be explained by effects that occur earlier in the treatment phase, such as the effect of dose frequency on canonical communication acts and receptive vocabulary. This evaluation requires attention to multiple effects (i.e., paths). Figures 1 and 2 will aid understanding the results of our analyses. The coefficients on these figures are standardized (i.e., vary from –1.0 to 1.0) and, thus, provide readers with a better understanding of the magnitude of the various paths for these effects than the unstandardized coefficients, which are reported in the text and tested for significance.

We define some terms here for readers who are unfamiliar with contemporary mediation analysis (Preacher & Hayes, 2004). The effect we want to explain is the “total effect.” In our case, the total effect is the previously reported effect of dose frequency on spoken vocabulary in DS *without* controlling for mediators, such as canonical syllabic communication or growth of receptive vocabulary (Yoder et al., 2014). This total effect is equal to the sum of the “direct” and “indirect” effects. The direct effect, in this case, is the dose frequency effect on spoken vocabulary, *controlling* for a mediator, such as canonical syllabic communication or receptive vocabulary. The indirect effect, in this case, is the dose frequency effect on spoken vocabulary *through* either canonical syllabic communication or receptive vocabulary. The indirect effects are composed of two paths: A and B. The “A path” represents the effect of dose frequency on the putative mediator. The “B path” represents the effect of the putative mediator on spoken vocabulary, controlling for dose frequency. The product of the coefficients for the A and B paths (AB) quantifies the magnitude of the indirect effect. When the confidence interval for the product AB does not include zero, it is significant. This indicates that the direct effect is significantly reduced or nonsignificant relative to the total effect and that the total effect is explained, at least in part, by the indirect effect. That is, a “mediated” treatment effect is shown when the indirect effect is significant.

Canonical syllabic communication as mediator—As we previously reported, the total effect of dose frequency on spoken vocabulary for our participants with DS was positive and significant, d = .67, standardized coefficient = .38; 95% CI [.03, .73] (Yoder et al., 2014). The average untransformed gains for the number of words said for the weekly vs. daily MCT groups were 54 and 70, respectively. The indirect effect of dose frequency on spoken vocabulary through canonical syllabic communication was also significant, AB = .18; 95% CI [.03, .50]. The direct effect of dose frequency on spoken vocabulary, controlling for canonical syllabic communication, was not significant, .20; 95% CI [–.15, .57]. Thus, the effect of dose frequency on canonical syllabic communication mediates the effect of dose frequency on spoken vocabulary. See Figure 1 for a depiction of this mediated treatment effect. When the mediator was controlled, the effect size for the dose frequency effect on spoken vocabulary was reduced from a Cohen’s d of .77 to .35.

Receptive vocabulary as mediator—The indirect effect of dose frequency on spoken vocabulary through growth rate of receptive vocabulary was also significant, AB = .19; 95% CI [.05, .38]. The direct effect of dose frequency on spoken vocabulary, controlling for receptive vocabulary, was not significant, .20; 95% CI [–.18, .57]. Thus, the effect of dose frequency on receptive vocabulary growth also mediates the effect of dose frequency on spoken vocabulary. See Figure 2 for an illustration of this mediated treatment effect. When the mediator was controlled, the effect size for the dose frequency effect on spoken vocabulary reduced from a Cohen's *d* of .77 to .35. It is coincidental that the reduction in effect size of dose frequency on spoken vocabulary when the mediator is controlled is the same across mediational models.

Exploratory Analysis

Because canonical syllabic communication and receptive vocabulary growth were both found to be significant mediators, one might wonder which is most explanatory of the dose frequency effect on spoken vocabulary in DS. Due to a small sample size, we cannot answer this important question directly. However, it may be useful to know that canonical syllabic communication and growth rate of receptive vocabulary were positively correlated ($r = .40$, $p = .01$).

Discussion

We previously reported that increasing the dose frequency of MCT led to improved spoken vocabulary outcomes in preschoolers with DS (Yoder et al., 2014). Because spoken vocabulary is a particularly difficult aspect of language development for children with DS, we wanted to understand *why* more MCT would enhance spoken vocabulary in this population. Our re-examination of the extant dataset from our prior report suggests that the effect of increased dose frequency on spoken vocabulary in children with DS can be explained by (i.e., was mediated by) more immediate effects of MCT on children's canonical syllabic communication and receptive vocabulary growth.

As indicated previously, the extant literature provides piecemeal evidence and a theoretical rationale for the predicted indirect effects. Our previous research demonstrated that both of our putative mediators were sensitive to environmental manipulations (i.e., treatment effects). We previously found effects for earlier versions of MCT on receptive vocabulary (Fey et al., 2013; Yoder & Warren, 2001) and recently reported an effect for increased dose frequency of MCT on canonical syllabic communication (Woynaroski et al., 2014). However, these prior effects were found in children with ID due to multiple etiologies, not just DS. The current results extend the aforementioned findings to show that increased MCT dose frequency boosts canonical syllabic communication and receptive vocabulary in a subset of preschoolers with ID who tend to show disproportionate deficits in spoken language development—preschoolers with DS.

Previous research had additionally demonstrated that these key skills were linked with later spoken language skill in young children with ID due to multiple etiologies (Chapman et al., 2000; Vandereet et al., 2010; Yoder & Warren, 2004; Yoder et al., 1998). Our results confirm that canonical syllabic communication and receptive vocabulary specifically predict later spoken vocabulary size in children with ID due to DS. Most important, the present findings demonstrate that the early effects of MCT dose frequency on canonical syllabic communication and receptive vocabulary account for our previously observed effect of dose frequency on spoken vocabulary in this sample of children with DS (Yoder et al., 2014).

Clinical Importance of the Effect Sizes

As we have discussed in previous articles, we lack the information necessary to definitively interpret the clinical importance of the "moderate" or "medium" effect sizes (between .67 and .77) that we have observed for increased MCT dose frequency on generalized communication and language outcomes in children with DS. To do so, we would need to know what effect sizes tend to be observed for children with a similar etiology and level of ID (e.g., children with DS) on highly generalized measures of communication and spoken language in well-designed studies of early communication interventions. Currently, we lack this information. Thus, the field would greatly benefit from a meta-analytic summary across many well-conducted language intervention studies in young children with DS. If such a meta-analysis were available, we could compare the effect sizes achieved here with the mean and standard deviation of effect sizes observed across available language interventions for children with DS. We wish to highlight that the average effect sizes for highly generalized dependent variables (i.e., outcomes measured in contexts that are different from the treatment context on multiple dimensions), such as those involved in the present investigation, are likely to be lower than the effect sizes for potentially context-bound dependent variables (i.e., outcomes measured in treatment sessions or in context similar to treatment sessions) (Yoder, Bottema-Beutel, Woynaroski, Chandrasekhar, & Sandbank, 2014). Such a research synthesis will provide us with the needed benchmarks for assessing whether the effect sizes reported here represent an improvement over the effect sizes that have been observed in other internally valid studies examining effects of treatment on generalized characteristics in children with DS.

Additionally, the clinical importance of the size of *indirect* effects is also difficult to judge. There is no currently available and widely accepted way of calculating an effect size for indirect effects that involve covariates (Preacher & Kelley, 2011). The current study uses a covariate, IQ, in both models. However, it is noteworthy that the dose frequency effect on spoken vocabulary was lessened from medium-to-large to small-to-medium in each of our mediation models.

Strengths of the Study

One strength of the current study was the measure of canonical syllabic communication, which aggregated across three sampling contexts to afford a more stable estimate than is afforded by estimating from a single communication sample (Sandbank & Yoder, 2014). Additionally, direct observation measures of canonical syllabic communication enabled the use of blind coders and, with the exception of the parent-child interaction session, blind adult examiners, thus reducing the likelihood of correlated measurement error (Yoder & Symons, 2010). Aggregating scores from parent-child sessions with scores from two other communication samples, which were administered by blind examiners, reduced the probability that the nonblind status of parents could have influenced the effects involving canonical syllabic communication.

To our knowledge, this is the first study to explain why more frequent treatment facilitates spoken vocabulary acquisition in children with DS. For those unfamiliar with mediational analysis in which the independent variable is manipulated in the context of randomized controlled trials, it might be useful to discuss why we can claim that our mediated effects "explain" the total effect of dose frequency on spoken vocabulary in children with DS. In many instances, mediation analyses are conducted in a correlational research design. In those instances, it would not be appropriate to interpret a mediated effect as an "explanation" for the total effect. However, several elements of our experimental design allow relatively strong causal inferences. For example, we randomly assigned children to weekly versus daily MCT and closely monitored the interventionists to ensure that the two groups received quite different doses of MCT. Our fidelity of treatment and attendance data indicated the experimental manipulation was successful in achieving more than a four-fold difference in cumulative treatment intensity between groups. Additional analyses ruled out several possible alternative explanations for be-tween-treatment group differences on our outcome measure, spoken vocabulary, and on our putative mediators, canonical syllabic communication and receptive vocabulary.

Thus, we have a strong basis for inferring causality for two of the three effects comprising each of our mediation models. That is, we can have as much confidence as possible from a single, unreplicated study that the between-group differences on the mediators and the outcome were due to dose frequency. However, an association between the mediator and the outcome is also necessary to state that a mediator "explains" a total effect. Our ability to infer that the mediator influenced the outcome is weaker than the other effects in the mediation model because the research design used to test the effect between the mediator and the outcome is correlational, not experimental. However, the present combination of effects is as close as we can get in clinical science to understanding how or why a treatment has an effect on a major outcome such as spoken vocabulary.

Weaknesses of the Study

In addition to the weaknesses discussed in Fey et al. (2013), the current analysis is limited by two issues. First, we have a reduced sample size (as this reanalysis involved only the subset of our ID sample that was diagnosed with DS). Having a small sample size increases the likelihood that effects of interest will not be detected (i.e., elevates type II error rate) and potentially reduces the stability of effect size estimates. Fortunately, the small sample size did not prevent detection of the primary effects of interest in the present report. Unfortunately, we did not have sufficient sample size to test which of the mediators better accounts for the effect of dose frequency on spoken vocabulary in children with DS. Therefore, at this time it is best to consider both mediators important explanatory variables for the dose frequency effect on spoken vocabulary.

A second weakness is that one of our mediators (receptive vocabulary) and our outcome (spoken vocabulary) were measured only by parent report. Scores from parent reports in general, and the MB-CDI in particular, have been shown to have relatively strong reliability and validity (Jackson-Maldonado et al., 2003). However, parents could not be blind to the dose frequency level their children received, and lack of blindness can inflate the size of treatment effects (Yoder & Symons, 2010).

Implications for Theory

It has been proposed that children with DS may struggle with spoken language acquisition due to difficulty with representing, planning, or executing motor programs to produce the speech sounds that compose words (i.e., the motor deficit hypothesis; Miller & Leddy, 1999). Alternatively or additionally, children with DS may need more models to learn new words because they are slow to process phonemes and other information (i.e., slow speech processing hypothesis; Yoder, Camarata, Camarata, & Williams, 2006) and have auditory memory limitations (Chapman & Hesketh, 2001).

Although the current findings do not definitively confirm or refute any of these hypotheses, they are compatible with all three theories. The mediation model involving canonical syllabic communication is compatible with the concept that more MCT therapy may aid practice and feedback in a way that enables sufficient vocal control and accuracy to enhance spoken vocabulary despite possible motor dysfunction. The mediation model involving receptive vocabulary is compatible with the concept that children with DS may be able to overcome slower information, including speech, processing and weaker memory abilities to acquire spoken words when more models of the words are provided in early communication intervention offered with greater frequency. These possibilities provide encouraging messages for maximizing the potential of children with DS.

Implications for Clinical Practice and Conclusion

Our prior report suggested that providing more frequent sessions of an early communication intervention, MCT, boosts spoken vocabulary outcomes in children with DS after an extended duration of treatment (i.e., 9 months). This is good news for children with DS, who typically struggle to learn to talk. The present results extend our previous finding by demonstrating that effects of more frequent treatment on canonical syllabic communication and receptive vocabulary preceded and mediated the effect of dose frequency on spoken vocabulary. Our findings suggest that gains on measures of canonical syllabic communication and receptive vocabulary are appropriate intermediate goals when the overarching goal is for young children with DS to learn to communicate with spoken words. Children with DS are likely to exhibit significant progress on these measures before consistent gains in word production are observed. Future work is needed to aid policymakers, educators, and insurance companies in deciding whether the effect sizes that we have observed in this efficacy study justify paying for an increased dose frequency of early communication intervention for children with DS when implemented by community service providers.

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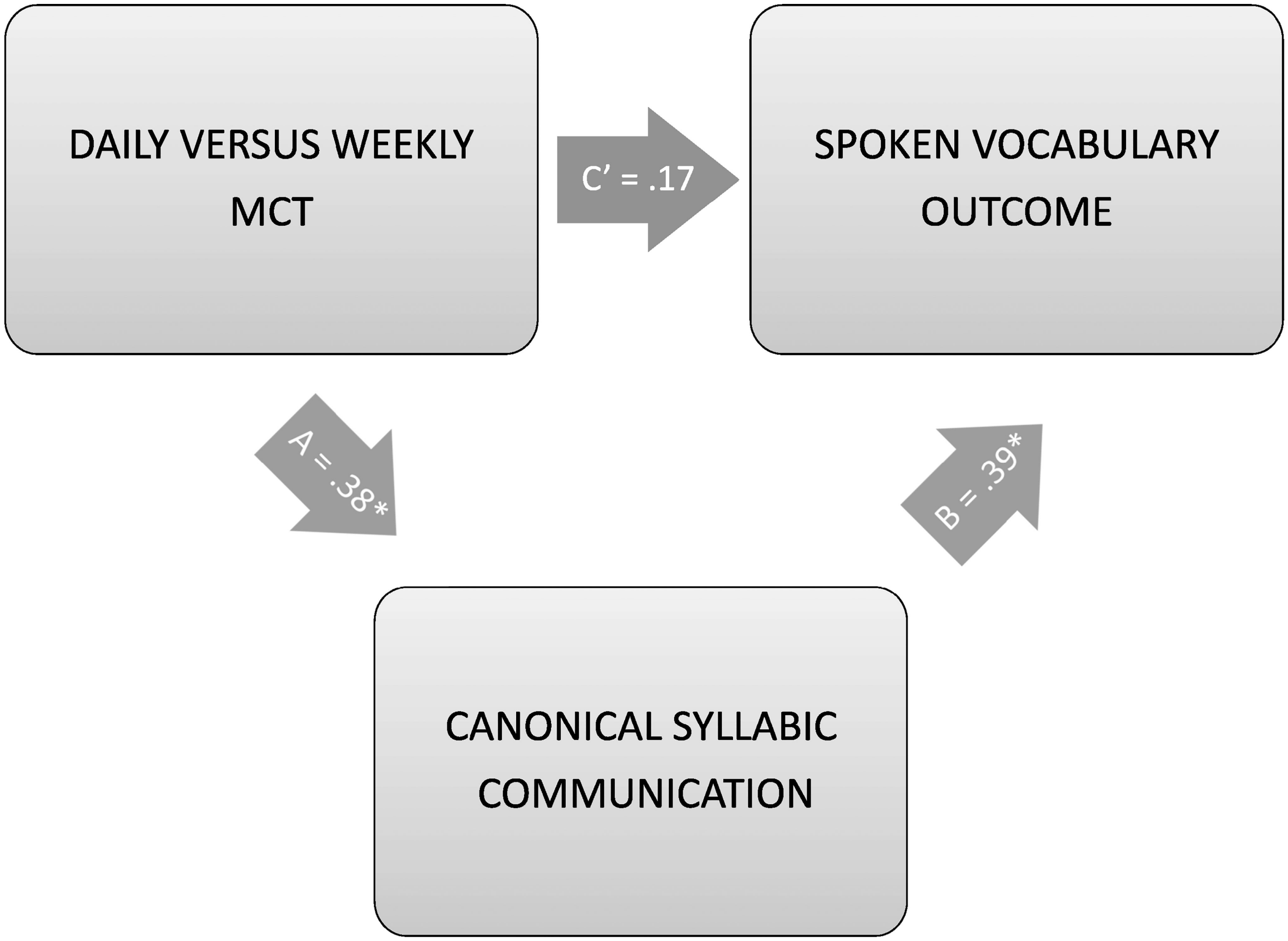

Figure 1.
Canonical syllabic communication mediates the effect of Milieu Communication Teaching (MCT) dose frequency on spoken vocabulary outcomes in preschoolers with Down syndrome. Coefficients in the figure are standardized regression coefficients. * indicates that $p < .05$. Note that each component of this mediation model controls for level of cognitive impairment as indexed by Bayley III Cognitive Composite standard score (not depicted).

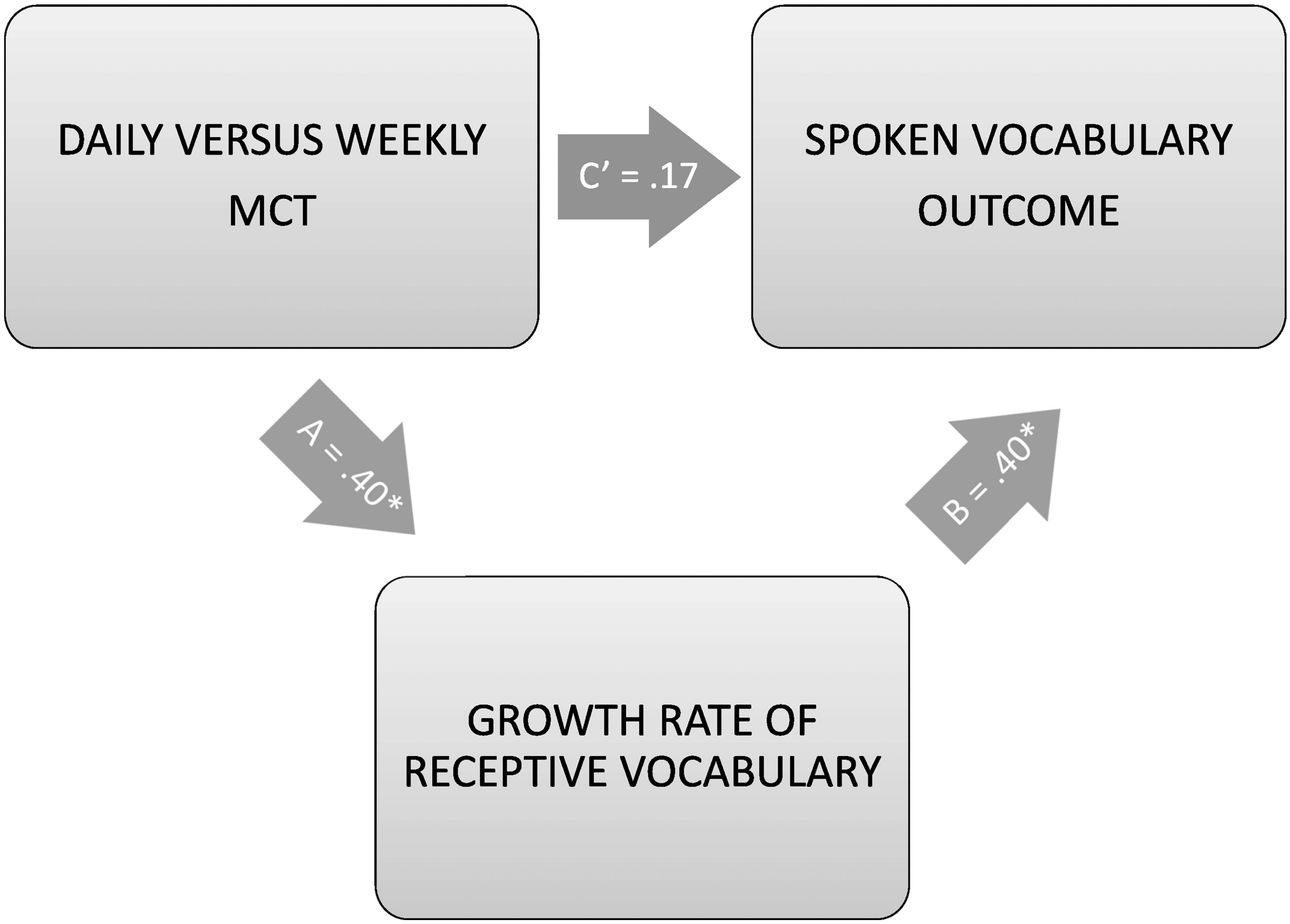

Figure 2.

Growth rate of receptive vocabulary mediates the effect of Milieu Communication Teaching (MCT) dose frequency on spoken vocabulary outcomes in preschoolers with Down syndrome. * indicates that $p < .05$. Note that each component of this mediation model controls for level of cognitive impairment as indexed by Bayley III Cognitive Composite standard score (not depicted).

Table 1

Pre-Experimental Child Characteristics by Dose Frequency Group

Pretreatment characteristic	Group	*M(SD)*
Chronological age[a]		
	Weekly	22.50(3.09)
	Daily	21.62(2.61)
Bayley III mental age		
	Weekly	12.69(1.66)
	Daily	12.05(2.09)
Bayley III CC		
	Weekly	65.94(6.38)
	Daily	63.95(7.18)
Number of words spoken[b]		
	Weekly	.94(1.34)
	Daily	1.37(1.61)
Number of words understood[b]		
	Weekly	53(55)
	Daily	36(23)
Canonical syllabic communication[c]		
	Weekly	.14(.17)
	Daily	.24(.21)

CC = Cognitive Composite standard score.

[a]in months.

[b]from MacArthur-Bates Communicative Development Inventories, Words and Gestures vocabulary checklist.

[c]Proportion of communication acts with canonical syllables summed across the Communication and Symbolic Behavior Scales, a semi-structured communication sample with an examiner, and a parent-child free play session. *Note*. No Time 1 means were significantly different between groups.

Table 2

Effects of Dose Frequency on Putative Mediators in Preschoolers With Down Syndrome

Variable	Daily *M* (*SD*)	Weekly *M* (*SD*)
Canonical syllabic communication	.42 (.23)	.25 (.22)
Growth rate of receptive vocabulary per month[a]	.09 (.06)	.04 (.04)

All values represent adjusted means controlling for level of cognitive impairment as indexed by Bayley III Cognitive Composite Standard Score.

[a]Values are log10 transformed.