Effects of a Naturalistic Intervention on the Speech Outcomes of Young Children with Cleft

Palate

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Abstract

Objective: The purpose of this study was to investigate the extent to which a naturalistic communication intervention, Enhanced Milieu Teaching with Phonological Emphasis (EMT+PE) improved the speech outcomes of toddlers with CP±CL.

Design: This study was a stratified randomized controlled trial.

Setting: Treatment was delivered in a university clinic by a trained speech language pathologist.

Participants: Thirty children between 15 and 36 months (M = 25) with nonsyndromic CP±CL and typical cognitive development were randomly assigned to a treatment (EMT +PE) or business as usual (BAU) comparison condition.

Interventions: Participants in the EMT+PE treatment group received 48, 30-minute sessions, over a six-month period. Fidelity of treatment was high across participants.

Main outcome measures: The primary outcome measures were percent consonants correct (PCC), consonant inventory, compensatory articulation errors, and nasal emission.

Results: Regression analyses controlling for pre-intervention child characteristics were conducted for PCC and consonant inventory. Intervention was not a significant predictor of post-intervention outcome. Words per minute (WPM) differentiated the children who benefited from the intervention from those who did not. Reduction in compensatory errors and nasal emission occurred in both groups but to a greater degree in the EMT+PE group.

Conclusion: EMT + PE is a promising early speech intervention for young children with CP±CL, especially for children with higher rates of word use.

Key words: nonsyndromic clefting, speech development, speech disorders

Early speech and language development in children with cleft palate with or without cleft lip (CP±L) is characterised by delays in the onset and complexity of canonical babbling and slow early development of vocabulary and speech sound development (Chapman, Hardin-Jones & Halter, 2003; Chapman, Hardin-Jones, Schulte & Halter, 2001). Some children appear to make progress on their speech and language milestones following palate repair without intervention, while others continue to show delays or differences in their speech and/or language performance. Research to date has provided possible factors that affect early speech and language development for children with CP±L (Chapman et al., 2001; Scherer et al., 1999) and identified skills predictive of communication outcomes.

Limitations in consonant inventory that are characteristic of children with CP±L may limit the differentiation of vocabulary acquisition due to the restrictions in the sounds available for early words (Scherer, Williams, Stoel-Gammon & Kaiser, 2012). Studies suggest that young children with CP±L show slower early vocabulary development and increased lexical selectivity; they produce more words beginning with nasals, glides, and vowels than words with high-pressure consonants (Chapman et al., 2003; Scherer, 1999; Scherer, Williams & Proctor-Williams, 2008). These speech sound limitations reduce intelligibility, which, in turn, may reduce children's communicative attempts using words (Scherer, Boyce, & Martin, 2013). When children make fewer communicative attempts using words, they have fewer opportunities to practice sound production and to receive feedback from their communicative partners (Frey, Kaiser & Scherer, 2018).

In previous research, when language intervention was used to bootstrap speech sound production, young children with CP±L made gains in both vocabulary and phonologic acquisition; however, these few studies have limitations in the description of the intervention,

fidelity measurement, and/or absence of a comparison group (Ha, 2015; Pamplona, Ysunza, Ramirez, 2004; Scherer, 1999; Scherer, D'Antonio, & McGahey, 2008). The limitations of these published studies restrict the interpretation of early intervention effects and decision-making regarding recommendations for intervention timing, dosage, and approach.

The effects of EMT have been documented over a wide range of language targets, including vocabulary and word combinations, across populations of children with language, behavior, or cognitive challenges (Kaiser & Roberts, 2013; Kaiser, Scherer, Frey, & Roberts, 2017; Peredo, Zeyala & Kaiser, 2017). The impact of EMT on speech development, however, has been less studied but is of interest for a number of clinical populations receiving early intervention. Camarata (2010) used naturalistic early intervention augmented with speech recasting to improve both language and speech in children with speech sound disorders, Down syndrome, and autism. The proximity of the adult model to the child production within a functional context provides an opportunity for the child to compare the model to his/her production. Scherer (1999) assessed the effects of EMT on a set of broad speech recasts for three children with cleft palate. No specific sounds were targeted within the target vocabulary. This study shows the impact of vocabulary expansion on speech production even without targeting specific sounds.

EMT +PE is a specific modification of traditional EMT that was adapted to address the specific needs of young children with both speech and language delays (Scherer & Kaiser, 2010). The phonological emphasis (PE) integrates speech recasting strategies within the EMT language modeling and prompting strategies. Speech production criteria are included in the procedures for selecting speech and language intervention targets. Speech recasting has been an effective naturalistic strategy for improving young children's speech accuracy and intelligibility and may

address the particular speech production deficits common in children with clefts (Camarata, 2010).

EMT+PE includes four components: (a) Environmental Arrangement which involves arranging the physical and social context to maintain child interest and engagement and optimize opportunities to prompt language; (b) Responsive Interaction strategies that balance turns, mirror child nonverbal play and communicative behaviors as an opportunity for modeling, respond to child communicative initiations with models, and expand children's utterances; (c) Milieu Teaching episodes that prompt children to use target vocabulary and increase intelligibility with elicited modeling, time delay, and incidental teaching; and (d) Speech Recasting to provide feedback and models for correct production of children's target sounds. EMT+PE attempts to increase consonant inventory and vocabulary development concurrently by selecting words that incorporate target speech sounds, increasing child rate of talking, modeling correct phonological and semantic forms, and providing contingent semantic and phonological feedback from an adult.

Kaiser et al. (2017) reported the language and broad speech outcomes of a pilot study comparing EMT+PE intervention to a "business-as-usual" (BAU) comparison for children under three years of age with nonsyndromic CP±L. The children receiving the EMT+PE intervention showed significant pre to post intervention gains in receptive language, expressive vocabulary skills, and percent consonants correct (PCC) in comparison to the gains by the comparison group. Significant differences between groups were not found for number of different words (NDW) used per minute, complexity of language use as measured by mean length of utterance in morphemes (MLUm), rate of communication, as measured by the number of vocalizations per minute, or expressive language scores on the *Preschool Language Scales* –

4th edition (PLS-4). Effect sizes, however, for all language and speech measures were positive and ranged from .04 to .65, indicating that children in the EMT+PE group performed better at the end of the study than children in the BAU group on vocabulary and speech accuracy measures. The current study extended the findings from Kaiser et al. (2017) through conducting a more indepth analysis of the treatment effects on speech skills.

The purpose of the current study was to examine the specific speech outcomes for toddlers with CP±L who received the EMT+ PE early intervention compared to a group of children assigned to the comparison group. The following research questions were addressed:

- 1. Does intervention condition predict post intervention percent consonants correct (PCC) and consonant inventories after controlling for pre-intervention child characteristics?
- 2. Are pre-intervention child characteristics (i.e. speech, word rate, and/or vocabulary) correlated with post- intervention speech outcomes (i.e. PCC and consonant inventory)?
- 3. What are the observed changes in place, manner of articulation features, audible nasal emission and compensatory articulation for children in the EMT+PE and BAU groups following intervention?

Method

A small, stratified, randomized group design study was conducted to evaluate the effects of EMT+PE on the speech of young children with nonsyndromic, repaired cleft palate with or without cleft lip.

Participants

Thirty children with CP±L were enrolled in this study; 15 were randomly assigned to the EMT+PE intervention (Age M = 23.13 months) and 15 to the "business as usual" (BAU) comparison group (Age M = 24.07 months). BAU comparison groups are those in which

children may attend the same or similar interventions available in the community.

Children were recruited for participation in the study at two sites in the southeastern United States continuously between December 2009 and June 2012. Children were included in this study if they (a) were between 15 and 36 months old; (b) had a cognitive scale composite score of 80 or above on the Bayley Scales of Infant and Toddler Development-III (Bayley-III; Bayley, 2006); (c) could produce at least five different words per parent report on the MacArthur-Bates Communicative Development Inventory (MCDI; Fenson et al., 2007); and (d) were considered at-risk for speech and language delay based on their errors on the *Profiles of* Early Expressive Phonology (PEEPS; Stoel-Gammon & Williams, 2013). Children were excluded from the sample if they (a) had a sensorineural hearing loss or sound field hearing threshold over 30dB HL, as measured by an audiologist or confirmed by the medical record; (b) were multilingual or non-English speaking based on parent report; (c) had a syndrome diagnosis from a geneticist; and/or (d) had more than three additional dysmorphic features in addition to the cleft palate (Jones, 1988). Table I. shows participant characteristics for the EMT+PE intervention and BAU comparison group. After the initial screening, children who met the inclusion criteria were assigned to the EMT+PE or BAU comparison group using a random number generator for assignment.

----Insert Table I. here---

Procedures

Pre-post assessment. All children received a comprehensive assessment of speech and language skills prior to and following intervention. The *Bayley Scales of Infant Development III-Cognitive subtest* (Bayley, 2006) was administered to all participants during the pre-intervention

assessment only. The *Preschool Language Scale - Fourth Edition* (PLS-4; Zimmerman et al., 2002), a standardized, norm referenced assessment, was individually administered to assess children's receptive and expressive language skills before and after intervention. Children's language skills were also assessed through 30-minute, naturalistic, play-based language samples (LS) with a trained clinician and parent-child interaction (PCX) sessions in the clinic. In addition to the standardised, norm-referenced and observational measures, parents completed the MCDI (Fenson et al., 2007) as a measure of expressive vocabulary before and after intervention. The PEEPS (Stoel-Gammon & Williams, 2013) was the primary speech measure used throughout the study. The phonetic transcriptions of the PEEPS were used to calculate percent consonants correct, consonant inventory, and audible nasal emission. Toys representing each word are available in the Supplementary Material, Table 1.

During the assessment, children wore a digital recorder inserted in a vest to ensure high-quality recordings of their speech. The session also was video and audio recorded. After the session, each child response on the PEEPS was phonetically transcribed using the international phonetic alphabet while listening to the digital recording and viewing the video recording.

BAU comparison group. The children in the comparison group did not participate in any of the experimental intervention (EMT+PE) sessions but did participate in routine follow-up through their local cleft palate team. Participants' cleft palate teams and community intervention providers received copies of the assessments performed in this study. Six children in the comparison group received community-based early intervention services, per parent report, during the study period. Six of the 15 children received community-based services in their homes once monthly for 4-6 months during the study.

EMT+PE Group. Children in the EMT+PE treatment group received intervention during

individual, 30-min play sessions, twice weekly, in a clinic room. Children participated in a total of 48 intervention sessions over the course of about six months. The intervention was provided with two licensed SLPs, one at each of the collaborating sites. Parents were permitted to be in the room during the assessments and intervention or in an observation room during the session; however, the parents were not specifically trained to administer the intervention. The children received routine follow-up by their craniofacial teams but no other speech-language or early intervention during the EMT+PE intervention. Procedures for implementing EMT+PE included are described in detail in Kaiser, Scherer and Frey (2017). A description of the strategies and fidelity criterion is included in Supplementary Table 2.

Intervention target selection. Five speech targets were identified from the PEEPS pretest results for each participant. These were embedded in single and multiple word target language levels. Speech targets were identified after reviewing the PEEPS analyses and were selected based on the guidelines included in Peterson-Falzone, Hardin-Jones, and Karnell (2010). Nasals, stops, and fricatives were given priority as target sounds, anterior place of articulation was targeted before posterior place of articulation, and pre-vocalic positions were addressed before post-vocalic position. The target words were chosen based on their syllable structure in the PEEPS assessment (i.e. CV, VC, CVC, CVCV, CVCVC), recommendations from the parent regarding use in the home environment and ease with which they could be incorporated into age appropriate play. Typical word targets included names for objects, actions, or locations that began with stop and fricative consonants.

Treatment fidelity. Before beginning the study, clinicians received extensive training on EMT+PE intervention through practice in the clinic with children with CP+/-L and/or other language impairments not enrolled in the study, review of written materials (research articles,

chapters describing the intervention, handouts summarising intervention strategies), review of video examples of the intervention implemented by other therapists, and practice with coaching and feedback from senior therapists experienced in the components of the intervention. Data were summarized for each component category and reviewed in relation to established fidelity criterion in EMT research literature (see Kaiser & Hampton, 2016).

Reliability

The SLP was knowledgeable of group assignment. To control for potential bias, child responses on the PEEPS were scored independently a second time by the first author, who was blind to group assignment. Point-to-point agreement was calculated for all of the items on each PEEPS assessment for each child. Disagreements in consonant transcription were resolved by consensus. The first coder also re-transcribed 50% of each sample to assess intraobserver agreement. Intra- and inter-judge reliability was assessed for 50% of the total PEEPS transcriptions; reliability for coding place and manner agreements, compensatory articulation, and nasal emission was calculated separately. Intra-judge PEEPS transcription reliability was 92-95% agreement, and inter-judge rating was 87-90% across the two time points for total transcriptions. Both place and manner features showed intra-judge reliability of 97-98% agreement and inter-judge reliability of 95-98% across the two time points. Reliability of compensatory errors and audible nasal emission are reported separately since they are often problematic. Intra and inter-judge reliability for compensatory articulation agreement was 90-92% and 85-89%, respectively. Intra and inter-judge reliability for ratings of nasal emission were 86-88% and 78-85%.

All intervention and interaction assessment sessions were video recorded, transcribed, and then coded. Average percentage of agreement for each coding category was as follows: 80%

(SD = 9.3) for child language targets, 97% (SD = 3.98) for matched/unmatched turns, 86% (SD = 5.41) for recasts/expansions, and 88% (SD = 6.60) for therapist language level. If agreement was below 85% on any coding category within any session, the two coders met to review and discuss each coding discrepancy before coding any additional intervention sessions.

Data Analysis

Assessment data were double entered independently by two research assistants into a database, and any disagreements in data entry were resolved by consensus. Demographic information and assessment data were summarized using appropriate descriptive statistics, and group differences at pre-intervention were examined. Data analyses were conducted in R (R Core Team, 2018) using ggplot2 (3.1.0; Wickham, 2016) and ez (4.4-0; Lawrence, 2016). For Research Question 1, we conducted a general linear model (GLM) regression analysis, which is a statistical procedure to examine the predictive relationship between one or more independent variables and a dependent variable (e.g. post-intervention PCC). We examined the predictive power of experimental condition, number of different words (NDW), and words per minute (WPM), controlling for number of words produced on pre-test and age, for postintervention PCC and consonant inventory. All predictors were entered into the model simultaneously, as there were no predictions about the unique or increasing contribution of a single predictor. NDW and WPM were based on transcripts of the 20-minute parent-child interaction (PCX) observation at pre-test. These predictors were selected a priori based on previous studies using a portion of the data presented here (Frey, et al., 2018; Kaiser, et al., 2017). NDW, WPM, and pre-intervention consonant inventory were all highly correlated with one another suggesting the presence of multicollinearity; however, all predictors were included in the model as they represented unique theoretical constructs.

To address Research Question 2, we examined the correlations among pre-intervention characteristics and post-intervention PCC and consonant inventories. For Research Questions 3 and 4, we completed a descriptive analysis of change from pre to post intervention for audible nasal emission (Research Question 3) and for changes in place and manner of articulation features and percentage of compensatory articulation (Research Question 4) for children in the EMT+PE and comparison groups.

Result

Descriptive statistics of pre-intervention characteristics for Enhanced Milieu Teaching with Phonological Emphasis group (EMT+PE) and Business as Usual group (BAU) are presented in Supplementary Table 3. Linear regression statistics, examining predictors of treatment response, are presented in Table II. Results of the correlation analyses, examining the relationship between child characteristics and post-intervention speech outcomes, are provided in Supplementary Table 4. Table III and Supplementary Table 5 present data associated with our post-hoc descriptive analyses.

Pre-intervention Comparison

No significant differences between groups were observed for age (t(26.66) = -0.38, p = 0.7054, d = -0.14), NDW (t(26.89) = -0.85, p = 0.4051, d = -0.31), WPM (t(27.89) = -0.02, p = 0.9857, d = -0.01), CDI (t(21.46) = -1.08, p = 0.2884, d = 0), or consonant inventory (t(25.23) = -1.37, p = 0.1822, d = -0.50); however, there was a difference for pre-intervention PCC (t(27.99) = -2.17, p = 0.0382, d = -0.79) which indicated that children in the EMT+PE group had lower speech accuracy as measured by PCC at pre-intervention. Supplementary Material Table 3 presents pre-intervention characteristics by treatment group.

Factors Predicting Post-Intervention Outcomes

EMT+PE and the BAU groups were compared on post-intervention performance on speech measures using age and pre-intervention performance as control variables. Pre-intervention NDW and WPM were examined as predictors for the PCC and consonant inventory (see Table II). Both regressions were significant (PCC: F (5, 24) = 9.31, p < .001; consonant inventory: F (5, 24) = 7.95, p < .001). Treatment condition was not a significant predictor for PCC (t = 1.13, p = 0.2696) or consonant inventory (t = 0.74, p = 0.4643). Models could be simplified by removing pre-intervention NDW and WPM as neither of these predicted post-intervention PCC or consonant inventory and for consonant inventory were highly correlated with pre-intervention scores (r = 0.90 and r = 0.82 respectively). Although for post-intervention consonant inventory, the assumption of independent predictors was violated, the model does provide us with initial information about which pre-intervention characteristics might predict post-intervention performance.

---Insert Table II---

Relationships Between Child Characteristics and Speech Outcomes

All pre-intervention variables were significantly correlated with post-intervention PCC and consonant inventories. Pre-intervention PLS scores, PCC, and WPM showed large effect sizes (i.e. $r^2 > 0.68$, Supplementary Material Table 4). The pre-intervention MCDI raw score and NDW had moderate effect sizes (r^2 between 0.60 and 0.65). Small effect sizes differences between groups in age were observed, indicating that age was a relatively poor predictor of later speech outcomes.

Examination of Audible Nasal Emissions

Audible nasal emission (ANE) was observed in the speech of three children in the EMT+PE group and two children in the comparison group prior to intervention affecting

between 7-15% of responses per child. Percent ANE decreased for three children (two in EMT/PE and one in comparison group) to 5-10% at post-intervention; two children (one in EMT/PE and one in comparison group) showed no change in ANE from pre- to post-intervention, with 7% and 11%, respectively. We do not have definitive information on velopharyngeal function for these children. To date, two of the five children with ANE have had secondary palatal management. One of the children was in the comparison group (11% ANE post-intervention), and one was in the EMT/PE group (5% ANE post-intervention).

Changes in Articulation

Treatment group findings. The mean and standard deviation of the change scores for place and manner of articulation was compared for EMT+PE and BAU comparison groups, and no differences were observed between the groups. (The complete data set for the change scores is in Supplementary Table 5). Given the importance of rate of word use as demonstrated by WPM's high correlation with intervention outcome, we examined the descriptive statistics for WPM in the whole sample (M = 6.81, SD = 5.94, Range = [0, 17.10], Median = 6.10). Inspection of the data showed that there was a bi-modal distribution, with no scores between 8.20 and 11.20 WPM at pre-intervention. Therefore, we split the treatment and controls samples at 10 WPM and completed a series of post-hoc analyses comparing the changes from pre to post intervention for place, manner, and compensatory articulation using the resulting four groups. Means and standard deviations for age, NDW, vocabulary size (as reported by parents on the MCDI), WPM, and pre-intervention speech scores for participants divided into four groups based on number of words at pretest (EMT+PE: High-rate and Low- rate; BAU: High-rate and Low-rate) are provided in Table III.

Place and manner. Place and manner of articulation performance for children in the

four groups, based on performance on the PEEPS, are presented in Figure 1 and Supplementary Table 5. The complete data set for the change scores is in Supplementary Materials. Effect sizes were calculated for the low rate and high-rate EMT+PE and comparison groups. There were clinically meaningful benefits (i.e. d > 0.5) for the low-rate EMT+PE group for changes for liquids (d = 0.58) and glides (d = 0.94) only; all other effect sizes for the low-rate groups were not clinically meaningful and ranged from 0 (dentals) to -0.47 (affricates). Effects sizes indicated that children with a speaking rate of less than 10 words per minute had limited benefits of EMT+PE. However, for the high-rate children, EMT+PE resulted in greater change, which was also clinically meaningful. The high-rate intervention group exceeded the comparison group in overall consonants (d = 1.91), and stop (d = 0.79), fricative (d = 1.43), and liquid (d = 2.88) consonants. The greatest gains were observed in the high-pressure consonant categories of stop and fricatives, which were speech targets in this study. Additionally, there was an advantage for high-rate children in the EMT+PE intervention for alveolar consonants (d = 2.48) with the EMT+PE group gaining approximately six alveolar consonants while the comparison group gained one from pre- to post-intervention. Effect sizes also revealed clinically meaningful differences in favour of the high-rate EMT+PE group for dental (d = 0.72) and labial (d = 1.73) consonants; other effect sizes ranged from -1.04 (affricates) to zero (velars, glottals, and nasals). The effect sizes for the high-rate group indicate that participation in EMT+PE results in clinically meaningful changes in both manner and place of articulation.

Compensatory articulation. The percent of compensatory substitutions produced preand post-intervention by children in each group is presented in Figure 2. The data suggest that children in the EMT+PE group who were low-rate talkers had a greater decrease in the use of compensatory errors from pre- intervention to post-intervention. Children who were high-rate talkers in both EMT+PE and comparison groups showed a reduction in compensatory errors from pre to post intervention. For all participants, compensatory errors occurred more often in word final position (86.67% in final position collapsed across time). Examination of the change in use of compensatory errors (glottal stops) was examined in two ways: first, we looked at children who had glottal stops at T0 and what changes occurred at T1. Of the 15 children in the intervention group, 8 of them had some glottal stops at T0 and, although they still were using glottals at T1, they decreased their use of glottals particularly in word final position at T1 by replacing glottals with oral consonants. Additionally, 3 children used some glottals at T1 when they had not used them in T0. These children were those that used very few consonants at T0. We also examined the 6 children in the BAU group who used glottal stops at T0. As with the TX group, these children continued to have some glottal stop use although they were used less at T1. Additionally, 3 more children used glottal stops at T1 who had not used them at T0. This data suggests that glottal stops were emerging to mark prevocalic, or more often, postvocalic syllables and then declined as oral consonants were added.

- --Insert Figure 1 here--
- --Insert Table III here--
- --Insert Figure 2 here--

Discussion

The purpose of this study was to analyze the speech outcomes of children with CP±L who received an early intervention (EMT+PE) compared with a group of children who were in a BAU comparison group. Overall, there were no significant effects of the intervention on PCC or consonant inventory for the two groups. However, secondary analysis suggests that the children in the EMT+PE group who had higher-rates of word use at pre-intervention (greater than 7-10

WPM) showed more change in their consonant inventories, speech accuracy, and compensatory articulation use from pre- to post-intervention than children in the EMT+PE low-rate group and both the high and low- rate BAU comparison groups.

Predictors of Response to Treatment

We explored predictors of response to treatment in several different ways, including regression, correlation, and descriptive analysis. The first research question examined the predictive power of pre-intervention treatment outcome, language, and age for post-intervention speech outcomes. The only significant predictor for the regressions was pre-intervention speech performance; however, the small sample size, variability in pre-intervention performance, and possible multicollinearity may have masked important information about predictors of post-intervention outcomes. The second research question correlated pre-intervention language, speech, rate, and vocabulary variables with post-intervention PCC and consonant inventories. The correlational analysis identified several pre-intervention child characteristics that could provide guidance for the application of EMT+PE to young children with CP±L to maximize speech outcomes. The third research question addressed a descriptive analysis of change in place and manner of articulation following the intervention. The descriptive analysis provided further evidence that speaking rate at pre-intervention was related to who benefitted from EMT+PE intervention.

Speaking rate (words per minute), number of different words, and overall vocabulary size showed moderate to large effect sizes as predictors of consonant inventory and speech accuracy following intervention. Studies have shown that consonant inventory and PPC (particularly for high-pressure consonants) for children from 18-20 months of age is correlated with later speech outcomes (Chapman et al., 2003). It appears that these same variables are still correlated to

intervention outcomes for young children. Of particular interest was the importance of rate before intervention as a predictor of speech outcome. Children who made the largest gains in speech production spoke at a rate of at least 7-10 words per minute, had at least 20 total words in the PCX, had 50 words reported by parents on the MCDI, and were 23-33 months of age. These findings suggest that children who do not yet meet these rate and vocabulary size criteria might benefit more from EMT only, focusing on increasing the rate and diversity of spoken language before introducing the speech recasting component. Alternatively, including the PE component did not appear to inhibit speech production. The optimal sequencing of intervention components in naturalistic interventions for this population is an important area for future research.

Children in the EMT+PE group who had a higher rate of words spoken per minute during the PCX showed a greater change in speech sound production from pre- to post-intervention compared to children in the EMT+PE group who had lower WPM and all children in the comparison group. The children in this study responded to the EMT+PE intervention differentially based on pre-intervention WPM used in the parent-child interaction sample (high-rate vs low-rate). The low-rate children performed generally the same pre and post intervention. Both groups gained consonant inventory and accuracy but to a lesser degree than the high-rate children.

The rate at which children use words in conversation is a critical factor affecting children's opportunities to learn from adult responses and feedback. The dosage of an early naturalistic speech-language intervention depends to a certain extent on the frequency and quality of the opportunities to respond to child utterances with meaningful input that presents expanded language models and provides corrective feedback for speech production. Frey et al. (2018) examined the rate of caregivers' responses to intelligible and unintelligible speech from

young children with and without CP±L. Although all caregivers were highly responsive, they found significant differences in child intelligibility and rate of spoken language (as measured by WPM) between toddlers with and without CP±L. More specifically, children with CP±L produced 50 words less and received 90 words less of adult input than noncleft children in a 10minute sample of caregiver-child interaction (because children with CP±L provided fewer opportunities for caregivers to respond). Over time, reduced frequency of meaningful, semantic input from the adult communicative partner may provide children with CP±L fewer opportunities to learn new language compared to noncleft children (Frey et al., 2018). Rate of word production is particularly important when providing feedback for speech production. In the current study, it likely that the children who gained the most in speech production were those high rate talkers who received phonological feedback and models of correct production via the recasting component of the EMT + PE intervention. For low-rate talkers, it may be more beneficial to focus first on increasing the rate of and diversity of spoken words before focusing on speech production. It may be important to build a language and communication foundation to support the use of naturalistic strategies that promote speech production, such as speech recasting. The specific speech performance outcomes for the children in the high-rate EMT+PE group included greater gains in consonants with alveolar, labial, and dental placements than observed for the children in the high-rate comparison group. Alveolar place of articulation has been described as problematic for young children with CP±L (Klinto, Salameh, Olsson, Flynn, Svensson & Lohmander, 2014). Thus, this exploratory evidence of gains on acquisition of consonants with alveolar place of articulation is a potentially important outcome of the intervention. The target sounds in the EMT+PE intervention contained predominantly stop and fricative consonants; however, six of the eight target stop or fricative consonants had labial and

alveolar placements. In addition to place of articulation gains, the children in the high-rate EMT+PE group made gains greater than the comparison group for speech accuracy, particularly in the stop and fricative manners of articulation. These two manners of articulation require production of high oral air pressure necessitating adequate velopharyngeal function. Children in the comparison group showed better growth in affricates; however, few children in either the EMT+PE or the comparison groups used affricates. Overall, the data suggest improvements in speech accuracy and increases in number of consonants following EMT+PE; however, changes in speech accuracy were not necessarily linear. In other words, children added or substituted new nontarget sounds before they acquired the targeted sound production. This pattern reflects the early stage of phonological acquisition in which children are adding and eliminating phonological processes (Sosa & Stoel-Gammon, 2012).

Limitations and Future Research

Sample size was a limitation of this study. A larger clinical trial with multiple sites is needed to address the dosage and timing of strategies in the intervention. Additionally, pre-intervention PCC influenced the treatment response and, therefore, future studies should use random assignment to groups stratified by PCC level to minimize the effect of speech disorder severity. Additionally, we used a new phonological assessment measure (PEEPS) because there are few standardized measures available for this age range. PEEPS has a small normative sample and limited validity and reliability data.

Additionally, it is likely that active coaching of parents to implement the intervention would also improve outcomes by increasing the dosage of the intervention in contexts where children are motivated to talk with familiar communication partners (Roberts & Kaiser, 2011). While the current study did not train parents as intervention agents, the parents in the

intervention group did have the opportunity to observe the intervention with their child and the clinician. Passive observation of intervention may have an impact on parent interaction with their child; however, parental behaviors were not measured in this study. However, a combined clinician plus parent model, similar to Roberts and Kaiser (2013), might provide greater benefits for the child and support parents in interacting with their children.

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Figure Legends

Figure 1. The gain in number of consonants by place of articulation (Top Panels) and Percent Consonants Correct for manner of production (Lower Panels) between pre and post intervention on the PEEPS is presented for the EMT+PE groups (black bars) and the business-as –usual (BAU) groups (gray bars). A positive gain score indicates that children had a higher score at post-test than pre-test on the PEEPS measure. For example, on average the high rate children in the EMT+PE group produced six more alveolar consonants at post-test than pre-test while the BAU group produced one more alveolar consonant. Another example is that on average high rate children in the EMT+PE group gained approximately 40% Stop PCC from pre- to post-test while the BAU group gained 20% on stop production, a 20% advantage for the EMT+PE group. The data for the Low Rate children in each group is presented in the panels on the left and the High Rate children in panels on the right.

Figure 2. Percent of compensatory substitutions used for pre and post intervention for EMT+PE (black bars) and BAU (gray bars). The data for the low-rate children in each group is presented in the left panel and the high-rate children in right panel.

Table I.

Demographic characteristics of the groups at pre-intervention.

| | EMT+PE | BAU |
|-----------------------------|--------------|--------------|
| Sample size | 15 | 15 |
| Gender | | |
| Male | 8 (53.33%) | 10 (66.67%) |
| Female | 7 (46.67%) | 5 (33.33%) |
| Race/Ethnicity ^a | | |
| Caucasian | 12 (80%) | 11 (73.33%) |
| African American | 0 | 1 (6.67%) |
| Hispanic | 0 | 1 (6.67%) |
| Asian | 3 (20%) | 1 (6.675) |
| Cleft Palate Type | | |
| Cleft Palate Only | 4 (26.67%) | 4 (26.67%) |
| Unilateral CL/P | 8 (53.33%) | 8 (53.33%) |
| Bilateral CL/P | 3 (20%) | 3 (20%) |
| Age of palate repair a, b | 12.47 (4.10) | 10.54 (1.73) |
| Range | [9, 25] | [7, 14] |
| Mother's Education | | |
| Level ^a | | |
| GED | 1 (6.67%) | 0 |
| High School | 3 (20%) | 0 |

| Some College or 2yr | 4 (26.67%) | 4 (26.67%) |
|----------------------------------|------------|-------------|
| degree | | |
| 4 Year Degree or more | 7 (46.67%) | 10 (66.67%) |
| Annual Gross Income ^a | | |
| < \$25,000 | 0 | 2 (13.33%) |
| \$30,000 - \$74,999 | 7 (46.67%) | 6 (40%) |
| ≥ \$75,000 | 8 (53.33%) | 6 (40%) |

Notes. ^a One record was not completed for participant in BAU group. ^b Mean (standard deviation) [minimum, maximum]. Count (percentage).

Table II.

General linear regression model statistics for post-intervention speech outcomes.

| Percent Consonants Correct | | | | | | |
|------------------------------|------------------|--------------|-------|-------|-------|--|
| Variable | В | SE B | β | t | p | |
| Constant | 17.76 | 13.72 | | 1.29 | 0.21 | |
| Pre-test score | 0.93 | 0.26 | 0.71 | 3.58 | 0.001 | |
| Age | -0.19 | 0.63 | -0.05 | -0.29 | 0.77 | |
| Treatment condition (EMT+PE) | 7.85 | 6.94 | 0.16 | 1.13 | 0.26 | |
| Number of different words | -0.46 | 0.50 | -0.29 | -0.92 | 0.37 | |
| Rate of speaking | 2.42 | 1.37 | 0.52 | 1.76 | 0.37 | |
| Adj. R ² | 0.59 | | | | | |
| F(5, 24) | 9.31 ($p = 0$. | 0000) | | | | |
| | Consc | nant Invento | ry | | | |
| Constant | 10.68 | 4.47 | | 2.39 | 0.03 | |
| Pre-test score | 1.02 | 0.36 | 0.90 | 2.80 | 0.009 | |
| Age | -0.09 | 0.23 | -0.08 | -0.42 | 0.68 | |
| Treatment condition (EMT+PE) | 1.71 | 2.29 | 0.10 | 0.74 | 0.46 | |
| Number of different words | -0.18 | 0.19 | -0.34 | -0.88 | 0.38 | |
| Rate of speaking | 0.46 | 0.47 | 0.30 | 0.98 | 0.33 | |
| Adj. R ² | 0.56 | | | | | |
| F(5, 24) | 7.95 (p = 0. | 0001) | | | | |

Notes. B = unstandardized beta estimate. SE B = standard error of unstandardized beta. $\beta = \text{standardized beta estimate}$. t = T - statistic estimate. p = p-value estimate

Table III.

Pre-intervention characteristics by treatment condition and rate of speaking grouping.

| | | Treatment Condition | | | |
|--|-------------------------------|-------------------------------|-------------------------------|--------------------------------|--|
| Variable | EMT+PE | | BAU | | |
| | Low-rate (< 10 words per min) | High-rate (>10 words per min) | Low-rate (< 10 words per min) | High-rate (> 10 words per min) | |
| Age (months) | 20.50 (4.93) [13, 30] | 28.40 (3.85) [25, 33] | 21.80 (7.84) [15, 35] | 28.60 (3.85) [24, 34] | |
| Parent report vocabulary (CDI raw) | 54.60 (62.72) | 314.80 (179.26) | 163.40 (251.34) | 405.25 (260.27) | |
| Number different words (PCX) | 5.80 (4.12) | 28.60 (16.10) | 8.20 (8.93) | 38.80 (10.40) | |
| Words per minute (PCX) | 3.54 (3.06) | 13.30 (1.45) | 3.36 (2.94) | 13.77 (2.56) | |
| Pre-intervention total PCC (PEEPS) | 18.70 (12.92) | 41.60 (19.11) | 33.8 (16.69) | 55.2 (13.33) | |
| Pre-intervention consonant inventory (PEEPS) | 4.40 (4.27) | 12.60 (5.31) | 6.40 (6.22) | 19.60 (4.16) | |

Notes. Mean (Standard Deviation). [Minimum, Maximum]. CDI = MacArthur-Bates Communicative Development Inventory (Fenson et al., 1993). PCX = Parent-child interaction. PCC = Total percent consonants correct. PEEPS = Profiles of Early Expressive Phonology (Stoel-Gammon & Williams, 2013).

Supplementary Table 1.

PEEPS Word List

| Words |
|------------------|
| Belly button |
| Chin |
| Ear |
| Eye |
| Finger |
| Foot * |
| Hand |
| Knee |
| Leg * |
| Mouth * |
| Nose * |
| Toe * |
| Tongue |
| Tooth |
| Tummy |
| Bib |
| Diaper (nappies) |
| Baby * |
| Hair |
| Doll |
| Peek-a-boo |
| Girl |
| Hat * |
| Sock * |
| Off |
| Ball * |
| Bed |
| Cookie (biscuit) |
| Cup * |
| Duck |
| Pig |
| Puppy (doggy) * |
| Shoe * |
| Book |
| Kitty (cat) |
| Meow |
| Cow |
| Moo |
| Mouse |
| Dog * |
| Moon |

Supplementary Table 2.

Fidelity Criteria and Therapist Use of EMT+PE Strategies

| Fidelity Measure | Description | % Criterion | % Mean (SD) |
|-------------------------------------|---|-------------|-------------|
| Matched turns | Percentage of therapist's utterances that were in response to a child's communicative attempt or utterance | >75 | 97 (2.9) |
| Talk at child's level | Percentage of therapist's utterances that were at child's language level | >50 | 88(7.7) |
| Recasted incorrect child utterances | Percentage of child words containing speech errors that were immediately followed by a therapist's recast of the word(s) containing the speech error(s) | >40 | 78(17.5) |
| Expanded child utterances | Percentage of child utterances to which the therapist responded by repeating child's utterance and adding one or more words | >40 | 52(18.1) |

| Time delay strategies | Percentage of correct implementation of time delay episodes | >80 | 98(8.0) |
|---------------------------------|---|-----|----------|
| Prompting strategies | Percentage of prompting episodes that were delivered in response to a child request, followed a system of least to most prompts, and ended with therapist providing the desired action or object to the child | >80 | 98(12.1) |
| Speech recasting | Repeating a word the child said with emphasis of a target consonant in the word. | >40 | 45(10.2) |
| Words containing speech targets | Percentage of words therapist used during the session that contained at least one of the child's speech targets | >25 | 34(16.0) |

Supplementary Table 3.

Pre-Intervention comparison by treatment group.

| Pre-Intervention characteristics | | | | |
|--|--------------------------|--------------------------|--|--|
| | Treatment Condition | | | |
| Variable | EMT+PE | BAU | | |
| Age (months) | 23.13 (5.89) [13, 33] | 24.07 (7.40) [15, 35] | | |
| Parent report vocabulary (CDI raw) | 141.33 (166.82) | 232.50 (268.74) | | |
| Number different words (PCX) | 13.4 (14.45) | 18.4 (17.76) | | |
| Words per minute (PCX) | 6.79 (5.14) | 6.83 (5.76) | | |
| Pre-intervention total PCC (PEEPS) * | 26.33 (18.34) | 40.93 (18.41) | | |
| Pre-intervention consonant inventory (PEEPS) | 7.13 (5.99) | 10.80 (8.55) | | |

Notes. N = 30. Mean (Standard Deviation). [Minimum, Maximum]. CDI = MacArthur-Bates Communicative Development Inventory (Fenson et al., 1993). PCX = Parent-child interaction. PCC = Total percent consonants correct. PEEPS = Profiles of Early Expressive Phonology (Stoel-Gammon & Williams, 2013). CI = confidence interval.

^{*} *p* < .05. ** *p* < .01. *** *p* < .001.

Supplementary Table 4.

Pearson correlations between child characteristics at pre-intervention and dependent at post-intervention.

| Pre-intervention characteristics | Post intervention PCC | Post intervention Consonant inventory |
|---|-----------------------|--|
| Age | 0.490 *** (0.24) | 0.531 *** (0.28) |
| PLS - Total (raw score) ^a | 0.684 *** (0.47) | 0.717 *** (0.51) |
| Parent reported vocabulary (CDI raw) ^a | 0.606 *** (0.37) | 0.638 *** (0.41) |
| Number different words (PCX) | 0.656 *** (0.43) | 0.682 *** (0.47) |
| WPM (PCX) | 0.684 *** (0.47) | 0.686 *** (0.47) |
| Pre-intervention PCC (PEEPS) | 0.742 *** (0.55) | 0.723 *** (0.52) |
| Pre-intervention consonant inventory (PEEPS) | 0.613 *** (0.38) | 0.767 *** (0.59) |

Notes. ^a N = 29. r (effect size r²). PLS = Preschool Language Scales (Zimmerman & Steiner, 2002). CDI = MacArthur-Bates Communicative Development Inventory (Fenson et al., 1993). PCX = Parent-child interaction. PCC = Total percent consonants correct. WPM = words per minute. PCC = percent consonant correct by manner. PEEPS = Profiles of Early Expressive Phonology (Stoel-Gammon & Williams, 2013). P-values were adjusted for multiple test using false-discovery rate.

^{*} p < 0.05; ** p < 0.01; *** p < 0.001

Supplementary Table 5.

Full summary data of change scores for PCC by manner and consonant inventory by place by group.

| Variable | | EMT+PE | | | BAU | |
|-----------|--------------|--------------|---------------|--------------|--------------|---------------|
| v arrable | Low-rate | High-rate | Whole group | Low-rate | High-rate | Whole group |
| | | | Place | | | |
| Labial | 2.40 (1.65) | 3.20 (1.92) | 2.67 (1.72) | 2.2 (2.53) | 0.6 (0.89) | 1.67 (2.23) |
| Dental | 0.50(0.85) | 1.20 (0.45) | 0.73 (0.79) | 0.5 (0.85) | 0.4 (1.51) | 0.47 (1.06) |
| Alveolar | 4.30 (1.77) | 6.40 (1.14) | 5.00 (1.85) | 3.3 (2.95) | 1.4 (2.61) | 2.67 (2.89) |
| Palatal | 0.90 (0.88) | 1.00 (1.22) | 0.93 (0.96) | 0.8 (0.79) | 1.6 (2.41) | 1.07 (1.49) |
| Velar | 1.60 (1.71) | 1.60 (1.14) | 1.60 (1.50) | 2.5 (2.22) | 1.6 (0.89) | 2.20 (1.89) |
| Glottal | 0.30 (0.95) | -0.20 (0.45) | 0.13 (0.83) | 0.7 (1.33) | -0.2 (0.45) | 0.40 (1.18) |
| | | | Manner | | | |
| Total | 25.8 (16.98) | 37.8 (9.42) | 29.80 (15.65) | 17.9 (20.53) | 20.0 (9.25) | 18.60 (17.22) |
| Stop | 42.1 (20.81) | 37.4 (19.50) | 40.53 (19.81) | 43.3 (38.91) | 25.0 (10.22) | 37.20 (32.91) |
| Fricative | 28.4 (30.99) | 48.0 (30.99) | 34.93 (27.52) | 39.4 (31.57) | 18.4 (31.57) | 32.40 (30.71) |
| Affricate | 5.0 (15.81) | -5.0 (27.39) | 1.67 (19.97) | 14.4 (23.24) | 40.0 (23.24) | 22.93 (36.88) |
| Nasal | 50.8 (40.20) | 19.6 (20.01) | 40.40 (37.22) | 59.9 (40.94) | 19.2 (29.36) | 46.33 (41.45) |
| Liquid | 22.5 (21.38) | 52.6 (19.77) | 32.53 (24.93) | 8.4 (26.92) | -7.8 (22.09) | 3.00 (25.84) |
| Glide | 60.0 (51.64) | 36.6 (58.29) | 52.20 (53.06) | 8.6 (57.21) | 60.0 (54.77) | 25.73 (59.92) |

Notes. Mean (SD). EMT+PE = Enhanced Milieu Teaching plus Phonological Emphasis; BAU = Business as usual. Manner is change in percent correct.



