Investigator Letter

To the Grant Review Committee:

Please accept the enclosed research proposal entitled, "**Prelinguistic vocal behaviors in infants at risk for cerebral palsy under 24 months**" in consideration for the American Speech-Language-Hearing Foundation New Investigators Research Grant. My research will examine prelinguistic vocal behaviors and their association with language performance in infants at high risk for cerebral palsy (hereafter, "CP-risk infants").

CP is a critically understudied population in the field of communication sciences and disorders. Speech and language developmental delays in CP are evident as young as two years of age, and communication in children with CP at two years is highly predictive of abilities at four years (Hustad et al., 2014, 2016). But *prelinguistic and early speech development in CP under two years of age has received almost no research attention*. Paradoxically, very early gross motor delays are often hallmark features of cerebral palsy and have received considerable attention in the research literature. The lack of empirical data on prelinguistic early vocal development presents a critical barrier for identifying communication delays and disorders, leading to missed opportunities to capitalize on early interventions for speech development that leverage early neuroplasticity. Early detection of communication impairment is particularly important in CP because of the extreme variation across severity profiles (Andersen et al., 2010). The study of prelinguistic vocal behavior has significant potential to offer prognostic information about speech or language development in CP. There exists a longstanding body of research on the stages of infant vocal development prior to the emergence of first words around 12 months (Oller, 2000; Stark, 1980). The timely emergence of these prelinguistic vocal stages is a robust predictor of typical speech development (Nathani et al., 2006; Oller, 1978) with delays observed in disorders such as autism, Down syndrome, and apraxia of speech (Lang et al., 2019; Lynch et al., 1995; Overby et al., 2020). Surprisingly few studies have directly studied CP beyond my own preliminary work (Levin, 1999; Long et al., under review). CP is a unique disorder because there is a greater need to discern motor speech impairments from intact language abilities. Earlier detection is critical to leverage plasticity for identifying appropriate interventions that foster language development if speech is not a viable option for individual children. Amazingly, this level of identification may be possible even before the typical onset of first words in infancy.

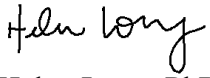
I am well suited to initiate an investigation of prelinguistic vocal development in CP because of my clinical and research experiences. My early professional career began as a speech-language pathologist (SLP) working with children who had complex communication needs secondary to autism and cerebral palsy which has guided the development of my clinical research questions. I completed my doctoral training with Dr. Kim Oller where I gained a foundational understanding of the evolutionary-developmental theory of typical and atypical prelinguistic vocal development. Immediately following my doctoral training, I was awarded a

highly competitive postdoctoral fellowship (T32) at the Waisman Center of the University of Wisconsin-Madison (100% salary support). In my postdoctoral training, I am currently studying speech and communication development in linguistic and older anarthric children with CP alongside the leading expert in this area, Dr. Katherine Hustad. Our work together has strengthened my foundational knowledge on longitudinal speech growth in school-age children with CP. From these experiences, I aim to bridge my understanding of infant vocal and early speech development in this population to continue building my programmatic line of clinical research in prelinguistic vocal biomarkers of communication impairment in infancy.

The New Investigators Research Grant will directly support foundational, clinical research needed to launch my career in this area. Specifically, it will support the coding and analysis of a preliminary study of infant vocal developmental patterns in 21 infants between 12-24 months of age at risk of CP. The **Specific Aims** of the project are to: **1) describe the complexity of prelinguistic vocal behaviors produced by CP-risk infants under 24 months,** and **2) examine the relationship between prelinguistic vocal developmental levels and language performance in CP-risk infants under 24 months.** For SA1, we will quantify prelinguistic vocal developmental levels of complexity to determine the extent to which vocal emergence may be delayed in CP. For SA2, we will compare these vocal developmental levels to these infants' receptive and expressive language performance to characterize the association between vocal and language performance. *My central hypothesis is that precursors to later speech impairment will be evident in early vocal behaviors; specifically, we anticipate measurable delays in the emergence of prelinguistic stages of development and unexpected patterns of vocal behavior because of the underlying neurological damage to the motor system.* Recordings of all 21 infants have already been collected as part of a prospective longitudinal project in my postdoctoral laboratory on early word production in CP. The proposed project presents a unique opportunity to gather data from these longitudinal samples that have not previously been examined and would not otherwise be possible to obtain in a one-year project. Funds for the proposed work are necessary to 1) purchase vocal coding analysis software and workstation equipment for real-time coding of infant vocalizations, and 2) support two research assistants for training, primary coding, and reliability coding of infant vocal behaviors. These materials are integral to support the ongoing study of prelinguistic biomarkers throughout my career.

Ultimately, the long-term goal of my research is to quantify early prelinguistic and speech developmental trajectories in CP-risk infants and to use these data to identify early indicators of dysarthria for individualized diagnostic and treatment. The proposed project is directly in line with ASHA's mission to support people with communication impairments through screening, diagnosis, and intervention for better communication and participation outcomes across life-long contexts. I am grateful for the opportunity to apply for this prestigious award and appreciate your consideration. Please contact me at the email address below if you have any questions.

Sincerely,



Helen Long, PhD, CCC-SLP
Postdoctoral Researcher
Waisman Center
University of Wisconsin-Madison
helen.long@wisc.edu

ABSTRACT

Communication problems affect the majority of children with cerebral palsy (CP). Speech and language delays are often present, which can affect communication effectiveness throughout the lifespan. There is a paucity of research that has sought to characterize early biomarkers of communication difficulties in this population. As a result, there is no prognostic information available to support an understanding of verbal trajectories in infants identified as being at risk of CP ("CP-risk infants"). There is a critical need to leverage prelinguistic vocal developmental behaviors for early identification of speech impairment in these infants under 24 months of age to support diagnostic and treatment planning as early as possible. Although many believe that speech begins with the onset of first words around 12 months of age, the emergence of prelinguistic vocal developmental stages at expected ages in infancy is a robust predictor of typical speech development. In a group of 21 CP-risk infants, the proposed research will examine the complexity of prelinguistic behaviors and examine their association with receptive and expressive language skills. Specifically, we will quantify infants' prelinguistic vocal developmental levels using the Stark Assessment of Early Vocal Development-Revised (SAEVD-R), a clinically translatable measure of prelinguistic vocal development. Vocalizations will be analyzed from recordings of the Communication and Symbolic Behavior Scales (CSBS) and compared to infants' performance on the Preschool Language Scales-5 (PLS-5). My central hypothesis is that precursors to later speech impairment will be evident in early vocal behaviors. Specifically, we hypothesize infants will demonstrate measurable delays in the emergence of prelinguistic stages of development and unexpected patterns of vocal behavior because of the underlying neurological damage to the motor system. Results from this project will provide preliminary data necessary to support ongoing research in the early detection of prelinguistic biomarkers of communication impairments in CP as well as the future development of clinically translatable tools to measure prelinguistic vocal developmental levels of children in medical settings. This work is critical to inform speech and language diagnostic and intervention planning to ultimately improve communication and participation outcomes in CP and other neurological disorders as young as infancy.

RESEARCH PLAN

A. Specific Aims

Over 50% of children with cerebral palsy (CP) present with communication impairments, which can greatly impact academic achievement, social participation, and emotional wellbeing across the lifespan (Parkes et al., 2010). It is well known that early speech and language intervention for children at risk leads to better communication outcomes later in life. However, CP is a condition known for its variability in severity profiles; speech impairments can range from complete anarthria to no perceptible impairment (Andersen et al., 2010). Our ability to predict speech outcomes in later childhood currently only begins at 3 years of age. There is potential to predict outcomes even earlier so that we can individualize interventions focused on speech and/or augmentative and alternative communication (AAC) modalities sooner in life to improve communication participation during critical periods in infancy. Early vocal development milestones and trajectories, prior to the age of 2 years, are one avenue to advance our ability to predict speech development beginning at earlier ages in children with CP, yet this avenue has received almost no research attention. The study of prelinguistic infant vocal development has great potential to inform even earlier detection of impairment in CP at even younger ages—possibly as early as 6 months of age. The investigation of infant vocal development is an untapped area of clinical research in CP-risk infants that can directly support the early detection of prelinguistic vocal biomarkers of communication impairment in this population.

The present research aims offers a preliminary investigation of prelinguistic vocal behaviors in CP-risk infants alongside early language abilities. I am well positioned to initiate this work because of my clinical experiences as a speech-language pathologist and research training in infant vocal development and communication in CP with Drs. Kim Oller and Katherine Hustad, respectively. *My central hypothesis is that precursors to later speech impairment will be evident in early vocal behaviors. Specifically, we hypothesize infants will demonstrate measurable delays in the emergence of prelinguistic stages of development and unexpected patterns of vocal behavior associated with the underlying neurological damage to the motor system.* The following Specific Aims will use a dataset of 21 infants between 12-24 months prospectively identified for risk of CP. We will quantify infant vocalizations using a gold-standard, validated coding protocol of 23 vocal types classified within five developmental levels.

Specific Aim 1: Describe the complexity of prelinguistic vocal behaviors produced by CP-risk infants under 24 months. We will calculate the 21 infants' vocal developmental levels from lab recordings. Overall rates of infant vocalizations across the five developmental levels in each infant will be observed. We hypothesize that infants will produce a larger number of less complex prelinguistic vocalization, which may be predictive of later communication delay or disorder.

Specific Aim 2: Examine the relationship between prelinguistic vocal developmental levels and language performance in CP-risk infants under 24 months. We will compare the 21 infants' vocal developmental levels to their receptive and expressive language performance. We predict that differences in the association between vocal developmental levels and language abilities may be predictive of later speech-language profile groups known to be observed in older children with CP. These findings would support the central hypothesis that anomalous prelinguistic vocal development in CP may be more highly associated with impairment in expected verbal expression performance than their overall language abilities.

B. Significance of Research

Background. As many as 50-90% of children with CP present with secondary communication, speech, or language impairments (Mei et al., 2014). The severity of speech impairments in CP can be highly variable across individual children and therefore difficult to predict at early ages (Hustad et al., 2015; Lee et al., 2014). Previous studies have shown clear differences in speech and language development in CP as young as two years of age, and that communication at age two years is highly predictive of abilities at four years of age (Hustad et al., 2014, 2016) However, there is limited evidence examining prelinguistic developmental patterns in this population under 24 months of age. This is a crucial barrier to progress in the early identification of speech impairment in CP because we are missing a critical period of time to capitalize on early interventions for speech development that leverage infant neuroplasticity.

There exists a longstanding body of research on prelinguistic vocal development and its stages prior to the emergence of first words around 12 months (Oller, 1978, 2000; Stark, 1980) Infant speech-like vocalizations or "protophones" (Oller, 2000) are considered infrastructural; in other words, mastery of early vocal stages is crucial to the development of more advanced stages, including the production of first words (Oller et al., 2016; Stark, 1981). These sounds become increasingly advanced across the first year of life in the phonatory, articulatory, and intonational domains, and transition through stages of cooing, marginal babbling of syllables with slow transitions between the consonant and vowel (CV), and eventually canonical babbling of mature, adult-like CV syllables. Age-expected prelinguistic vocal development is a robust indicator of typical speech development (Nathani et al., 2006; Oller et al., 1998). Delays in prelinguistic vocal development are commonly observed across neurological disorders with secondary communication impairment, including autism, Down syndrome, and childhood apraxia of speech (Lang et al., 2019; Lynch et al., 1995; Overby et al., 2020).

A limited number of cross-sectional studies have evaluated canonical babbling development in infants and young children with CP. Levin (1999) found delays in the onset of canonical babbling in six out of eight 11-12 month old infants. In a retrospective study using medical histories of 46 children with CP, Otapowicz et al. (2005) found delayed "cooing" in 50% of children, and 96% of those delayed were later diagnosed with dysarthria. Finally, Nyman & Lohmander (2018) observed canonical babbling delays in 28% of 10-24-month-olds in a "neurodevelopmental delay group" which consisted of infants with CP, Down syndrome, and other genetic syndromes. This work highlights the potential for early vocal development to inform our understanding of speech and language difficulties that become readily observable at older ages. However, these studies are limited in their methods, the number of infants studied, and the age ranges studied. Thus, there is a critical need for large scale, systematic study and comparison of prelinguistic vocal developmental behavior (and potential delays) to advance our understanding of how early vocal development can inform later speech and language outcomes.

I initiated the first two prospective, longitudinal studies of prelinguistic vocal development in CP. The first study compared canonical babbling emergence in two CP-risk infants to 10 typically developing (TD) infants between 5-16 months of age (Long et al., under review). Both infants at risk had low rates of canonical babbling relative to the TD infants. The infant who was later diagnosed with severe CP and anarthria presented with reduced canonical babbling consolidation over time (i.e., rates did not consistently increase across the ages studied), whereas the infant later diagnosed with only mild CP and mild speech impairments presented with an expected increasing consolidation that followed a delayed, but similar, trajectory to the TD group

average. Findings from this work highlight the need for additional study of longitudinal canonical babbling consolidation. My second study, conducted in my postdoctoral laboratory, examined the longitudinal emergence of marginal and canonical babbling (two prelinguistic vocal stages associated with the maturation of primitive articulation abilities) in 14 CP-risk infants at four age points under 24 months of age (Long et al., in preparation). Overall, the group showed consistent consolidation of marginal babbling but a protracted rate of growth of canonical babbling. Interestingly, the group did not reach mastery of the canonical babbling stage by 24 months, suggesting significantly delayed prelinguistic vocal development compared to typical expectations. There is a critical need for a more precise analysis of prelinguistic vocalizations across all stages of development in CP under 24 months of age, beyond just canonical babbling, to investigate the predictive value of different stages of development as biomarkers of later communication impairment.

Innovation. The proposed research is the first project of its kind to prospectively examine prelinguistic vocal developmental levels in a group of CP-risk infants. Furthermore, there are no known studies investigating the association between prelinguistic vocal developmental levels and language performance in this population. Many studies have utilized canonical babbling ratios (CBR) to assess vocal development in clinical populations; however, CBRs are used binarily to classify an infant as having mastered (or not yet mastered) the canonical babbling stage. A more fine-grained level of analysis is necessary to assess the prelinguistic emergence of verbal communication in a population at risk of different trajectories of impairment. This project will use a validated, clinically translatable coding protocol that classifies vocalizations across five levels of developmental complexity. Because of the variable nature of impairment and its severity in cerebral palsy, it is critical that we initiate a study evaluating prelinguistic vocal behaviors and the association with language development in infants at risk. My postdoctoral mentor, Dr. Hustad, has the only dataset in the world available to initiate this level of study. This dataset prospectively follows the communication, speech, and language development of ~50 CP-risk infants from the first few years of life. My unparalleled training in infant vocal development with Dr. Kim Oller and communication and speech development in CP with Dr. Katherine Hustad place me in a unique position to initiate this research project.

Impact. Research findings from this work will provide a critical set of pilot data that will support the development of a K23 application to receive additional training in the temporal relationship between infant vocal and motor development and in statistical modeling of longitudinal speech growth trajectories to begin to bridge our understanding of prelinguistic and early speech development in CP. The work completed during this award period is also foundational to my plans to develop and refine screening and diagnostic tools for measuring developmental progress in children at risk for CP across clinical settings. Presently, speech-language pathologists are often left out of the CP early detection team because medical providers often presume that communication performance is not immediately relevant to the diagnosis of CP. However, the proposed research and ongoing work based on our findings may support the argument that the emergence of the speech capacity begins in infancy, early intervention is critical, and clinical biomarkers of communication impairment have great potential to inform even the diagnosis of CP itself alongside multidisciplinary observations. Ultimately, the proposed work is a foundational step in establishing my programmatic line of clinical research supporting infants and young children at high risk of having complex communication needs.

C. Design Methods, Procedure, and Evaluation

Infant Dataset. For both Specific Aims, professional quality laboratory recordings and assessment data of 21 infants between 12-24 months are available for immediate coding and analysis (11 female, mean age: 18 mo., range: 13-24 mo.). All infants were recruited through local and regional medical centers through the Clinical Translational Core at the Waisman Center of the University of Wisconsin-Madison to participate in an ongoing longitudinal project on the Study of the Emergence and Acquisition of Language and Speech in ~50 infants and young children at risk for CP between 0-5 years (5R01DC009411, PI: *Hustad*). CP risk status was based on medical records and birth history. During participation in the longitudinal project, families brought their participating infant to the laboratory every 1-2 months for assessment starting at the point of recruitment (between 5-14 months). At these laboratory visits, infants participated in a variety of communication, language, and speech tasks. The 21 infants used in the present project were selected from the larger cohort of ~50 children for having completed the relevant assessment data required for analysis between the observed ages of study. The present research project will use data collected during administration of the Communication and Symbolic Behavior Scales (**CSBS**) and Preschool Language Scales-5 (**PLS-5**). My Specific Aims have not been previously studied with these data. All standardized assessment data is already scored. Infant vocal behaviors produced during the CSBS require coding classification by trained coders to analyze vocal developmental levels for this research project. My doctoral work provided me with a unique skillset to train new coders to classify prelinguistic vocal behaviors captured in these recordings. This award will provide necessary funding to purchase dedicated equipment and to fund two student research assistants for vocal coding.

Infant Vocal Coding Protocol. Two graduate student research assistants, hereafter, "**coders**," will be trained on classifying infant vocal behaviors according to the Stark Assessment of Early Vocal Development-Revised (SAEVD-R, Nathani et al., 2006). A thorough description of the coder training process is described below in *Coder Training.* The SAEVD-R is a coding scheme of 23 vocalization types used to classify infant vocalizations across five developmental levels of complexity. These five developmental levels correspond to the stages of prelinguistic vocal development originally described by Oller (2000) and Stark (1980), defined in **Table 1.** Although many studies of infant vocal development have analyzed canonical babbling ratios (CBR), the SAEVD-R is a more clinically translatable tool that supports a fine-grained level of classification for prelinguistic developmental complexity, more so than a simple binary criterion judgment. Thus, the greater precision of developmental classification from the SAEVD-R was deemed appropriate to examine prelinguistic vocal emergence in CP, a disorder well known for its wide variability of severity profiles. We have added "Words" as a sixth development level to the SAEVD-R classification scheme because the proposed research examines

TABLE 1. SAEVD-R vocal developmental levels and categories

Developmental Level	Expected onset age	Description (# vocal types included)
1: Reflexive	0-2 mo	Vegetative sounds (burp, cough, etc.), crying, fussing, phonation with closed vocal tract (4)
2: Phonation	1-4 mo	Primitive vowel-like sounds, raspberries, laughs (5)
3: Expansion	3-8 mo	Resonant full vowel sounds and glides, squeals, growls, primitive "marginal" syllables with immature consonants (6)
4: Canonical Syllables	5-10 mo	Single (CV) and repeated mature consonant-vowel syllables (CVCV, CVC), whispered productions (5)
5: Advanced Forms	9-18 mo	Complex, multisyllabic strings, varied intonation patterns, diphthongs (3)
6: Words	12-15 mo	Referential, clear semantic meaning (2)

an age range beyond the expected prelinguistic stages of emergence. Thus, if an infant in our sample is verbal, they will be classified appropriately as being in a linguistic developmental level. Although infant words tend to be immature in form compared to adult-like production of words, they can be reliably identified by parents and unfamiliar listeners based on environmental information including adult speech and infant eye gaze, body movements, and gestures (Vihman et al., 1985). During video observation, coders will classify each infant utterance in real-time using the 23 vocalization types of the SAEVD-R plus two Level 6 categories: Word and Multi-word, summing to a total of 25 vocalization categories. Coders will be allowed to stop the video to make a categorical judgment of each infant utterance and will have a copy of the description of each vocal type nearby to support coding fidelity. Vocal coding is estimated as ~40-60 minutes for each ~20-minute CSBS recording. Approximately 36 hours of vocal coding are therefore built into the *Management Plan* timeline for primary and reliability coding.

Coder Reliability: One primary coder will complete the vocal coding of all 21 recordings. The second coder will complete 20% of recordings (5 total). A two-way random (consistent raters, sample data) Interclass Correlation Coefficient (ICC, McGraw & Wong, 1996) will be conducted to assess inter-coder reliability across the 5 developmental levels.

Vocal Production during the Communication and Symbolic Behavior Scales (CSBS). As previously stated, infant vocal coding will be conducted using infant recordings during administration of the CSBS. All infants completed the CSBS with a research speech-language pathologist at a laboratory visit between 12-24 months of age. CSBS sessions were selected for vocal analysis because of the standardized nature of the assessment protocol to reduce potential parent-child interaction differences affecting infant vocal production, as may be the case in parent-child interaction sessions (McCathren et al., 1999). Recordings have been pre-screened to ensure that infants produced a minimum of 5 vocalizations for inclusion of analyses.

Coding software. Vocal coding will be conducted using the Action Analysis and Coding Training (**AACT**) software (Oller & Delgado, 1999). AACT is a general-purpose software package used for real-time and repeat-observation recording and analysis in behavioral research. AACT is unique from other behavioral coding software because it was specifically developed to support visualization of video and scrolling spectrographic information from the audio signal for coding infant vocalizations in real time. A laboratory license of AACT is required to support this project and ongoing coding of prelinguistic vocalization in my research. I am well acquainted and trained on the capabilities of this software; thus, I am immediately prepared to train coders on its use in the first two months of this project. Two workstations and accessories are also included in the proposal budget for AACT installation and dedicated coding.

Coder Training. I have created vocal training modules to support coder classification of infant vocalizations according to the SAEVD-R protocol. The use of training modules will ensure a standardized procedure for subsequent coding in future studies. These modules are adapted from the training protocol used in my doctoral laboratory with Dr. Oller at the University of Memphis (Oller et al., 2019). Training modules include 5-10-minute videos on the theoretical framework for understanding the stages of infant vocal development, explicit descriptions and examples of vocalization categories, and an introduction to coding in AACT. The primary training goals are to calibrate coders' perceptual judgment of 1) syllables versus utterances, 2) prelinguistic vocalizations versus words, and 2) vocal categories across the SAEVD-R levels. Following engagement with the training modules, coders will practice coding three 15-minute laboratory recordings of parent-child interactions. Their results will be compared with a "key" developed

from my coding as a gold standard. Discrepancies between student coding and the key will be highlighted and discussed with individual coders, and a second round of practice coding will ensue. This will continue until coders reach a criterion of not more than 10% discrepancy from the key on the targeted categories. Training is expected to last ~30 hours, split across 6 weeks of 4-hour increments. Each 4-hour training session will include direct instruction, module completion, coding practice, discussion of ambiguous information, and review of reliability training. Additional meetings will be scheduled to discuss discrepancies if coders do not reach the 10% criterion by the fourth week of training to maintain the proposed timeline. During the training period, coders will also be tasked with extracting the video and audio of CSBS recording sessions (~20 minutes) from the full laboratory recording (often ~1.5 hours in length). The segmented CSBS recordings will be used for formal vocal coding.

Specific Aim 1 (SA1): Describe the complexity of prelinguistic vocal behaviors produced by CP-risk infants under 24 months.

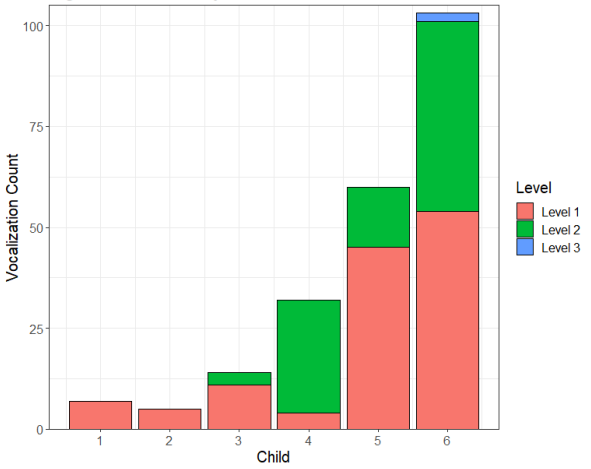
SA1: Rationale. The emergence of prelinguistic vocal developmental stages is well documented as being prerequisite to production of first words. Delays in the canonical babbling stage, for example, are commonly observed in children with developmental disorders (Lang et al., 2019; Oller et al., 1999). There is surprisingly little research examining prelinguistic vocal developmental patterns in CP (Levin, 1999; Long et al., under review). The first aim seeks to describe the prelinguistic vocal developmental levels of 21 children at risk of CP to develop a foundational understanding of prelinguistic development and the emergence of speech in a population at great risk of communication impairment. Importantly, the SAEVD-R was developed to support a comparison of vocal developmental levels to expected ages of emergence, thus, describing prelinguistic vocal complexity using this coding protocol supports a relative comparison to typical, developmental expectations.

SA1: Hypothesis. We predict that we will observe a larger number of vocalizations categorized into lower SAEVD-R developmental levels than higher developmental levels across all 21 CP-risk infants, indicative of vocal stage delays compared to typical ages of onset. This hypothesis is based in the theoretical, clinical assumption that delays in the emergence of prelinguistic vocal stages are indicative of later communication disorders.

SA1: Analysis Plan. Coded infant vocalizations will be aggregated within their SAEVD-R developmental level. For example, the four vocalization types of Level 1 (vegetative sounds, cries, singleton "quasivowel" utterances—i.e., phonation with closed vocal tract—and multisyllabic quasivowel utterances) will be combined into one "Level 1" category to represent all vocalizations that make up that developmental level. We will then compare the frequency of vocalizations in each developmental level. We will first assess the descriptive profiles of individual CP-risk infants. Specifically, we will examine rankings of the frequency of vocalizations within each developmental level and look for patterns of rankings across infants. A Friedman test will also be conducted as a non-parametric alternative to the one-way repeated measures ANOVA to determine if there are statistically significant differences of infants' vocalizations across all 6 Levels. Bonferroni corrections will be included to account for the 15 paired comparisons ($\alpha = .003$). Based on my hypothesis, we predict greater differences between lower and higher SAEVD developmental levels than sequential levels.

SA1: Preliminary Data. SAEVD-R levels of six anarthric children with CP between 19-58 months of age were previously coded in the WISC Lab using the methods described in this proposal (Erwin & Hustad, 2007). **Figure 1** shows that 5/6 infants produced more Level 1 vocalizations than Level 2 or 3 vocalizations. Interestingly, none of the infants produced any Levels 4-5 vocalizations (Level 6 not included). Because all infants were identified as anarthric, these preliminary data highlight a potential relationship between the production of more complex vocalizations and speech impairment severity levels.

Figure 1: Preliminary SAEVD-R levels in older children with CP

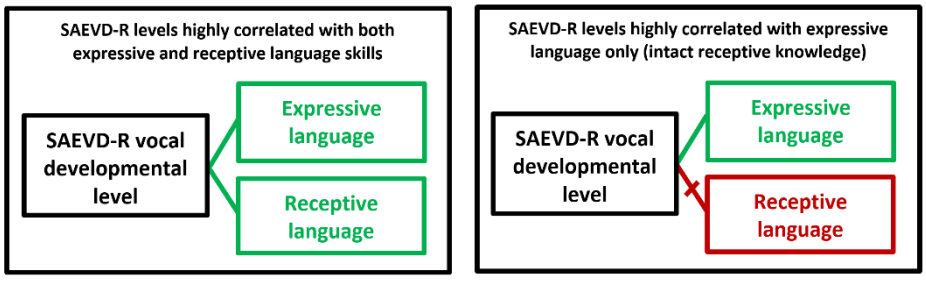


Specific Aim 2 (SA2): Examine the relationship between prelinguistic vocal developmental levels and language performance in CP-risk infants under 24 months.

SA2: Rationale. Prelinguistic vocal developmental delays are frequently observed in children later diagnosed with neurodevelopmental disorders with secondary language impairment (Lang et al., 2019; Lohmander et al., 2017; Oller et al., 1999). CP, although primarily a motor disorder, also frequently presents with co-occurring communication impairment because of the neurological impact on the motor speech system affecting verbal, expressive communication abilities (Hustad et al., 2010, 2016; Parkes et al., 2010). Because prelinguistic vocal development is prerequisite to speech and language emergence in typical infants, we aim to investigate potential differences across infants' vocal developmental level profiles and how these profiles may correspond to different constellations of language abilities.

SA 2: Hypothesis. We predict that the relationship between vocal developmental levels, expressive language, and receptive language will be variable across profiles of infants, and thus less predictable than for other populations with motor speech impairments (e.g., Down syndrome). Specifically, we presume that infants with CP will present with different constellations of language impairment. First, we hypothesize that the SAEVD-R developmental levels of some infants in our sample will be highly correlated with only expressive language—and *not* receptive language—abilities. This prediction is based in the assumption that for some children, CP-specific motor impairments impact expected verbal expression performance, more so than their overall language abilities. Conversely, other children with CP are known to have either global cognitive and language impairments, or imperceptible levels of impairment. Thus, we hypothesize that for some infants in our sample, SAEVD-R developmental levels will be highly correlated with their expressive communication *and* auditory comprehension scores, potentially indicative of more (or less) severe levels of impairment across all areas of development. Our hypotheses are illustrated in **Figure 2**.

Figure 2. Schematic for the predicted associations across different profiles of CP-risk infants.



SA2: Language Measure. The 21 infants' standard scores of the Expressive Communication and Auditory Comprehension subtests of the Preschool Language Scales, 5th edition (**PLS-5**) will be compared to the classification of infants' SAEVD-R developmental levels. All 21 infants completed the PLS-5 around the same age as the CSBS (mean age: 17 mo, range: 12-22). The PLS-5 was selected as the language measure because it is a validated, comprehensive infant and toddler language assessment. The PLS-5 was thus deemed an appropriate measure to examine the association of SAEVD-R developmental levels with the receptive and expressive language abilities of infants in our sample. Standard scores of the auditory and expressive subtests are already calculated for all 21 infants for analysis.

SA2: Vocal Developmental Level. Vocal developmental levels will be calculated from the same SAEVD-R coding conducted for SA1. For SA2, each infant will be classified for a single vocal developmental level based on the number of vocalizations produced in each category. **Table 2** demonstrates a schematic used to calculate two sample infant's prelinguistic vocal developmental level. Infant 1 would be identified as "Level 2" because they produced the highest number of vocalizations (n=26) within the Level 2 category. Infant 2 would be identified as "Level 5" because they produced more sounds in the Level 5 category (n=29) than any other level.

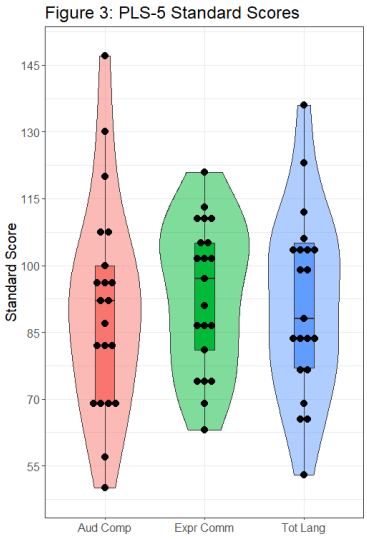
Table 2. Schematic for judging prelinguistic developmental level

Level	Infant 1 Count	Infant 2 Count
Level 1	23	10
Level 2	26*	15
Level 3	10	8
Level 4	2	24
Level 5	0	29*
Level 6	0	1

SA2: Analysis Plan. Two Spearman's rank-order correlations will be conducted to examine the association between infants' SAEVD-R developmental levels, PLS-5 expressive communication standard scores, and SAEVD-R levels with PLS-5 auditory comprehension standard scores. A non-parametric correlation analysis was selected because of the small sample size. In one profile of infants, we predict low SAEVD-R levels, low expressive communication scores, and high auditory comprehension scores. These results would preliminarily support the early detection of impaired expressive communication but typical language comprehension, which could ultimately support clinicians' decision to begin introducing advanced AAC modalities. In other infants, high correlations across all three measures may occur in two other profiles of infants: 1) low SAEVD-R level, low expressive communication, and low auditory comprehension, and 2) high SAEVD-R level, high expressive communication, and high auditory comprehension. These findings would preliminarily support the early identification of infants with 1) global cognitive and motor impairments (low SAEVD-R levels and low language scores), and infants with 2) overall mild or no perceptible impairment (high SAEVD-R levels and high language scores), respectively. Early identification of these profiles can support clinical decision-making for the introduction of low-tech AAC or continuing the treatment of speech as a viable option. If our hypotheses are correct, all three profiles would correspond to the speech-language profile groups in CP identified by Hustad at al. (2010): Speech motor impairment with typical language comprehension (LCT), SMI with language comprehension impaired (SMI-LCI), and no speech motor impairment (NSMI).

SA2: Preliminary Data. Descriptive statistics of the PLS-5 auditory and expressive language standard scores for all 21 infants are presented in **Figure 3**. Interestingly, our sample of 21 infants demonstrate a lower median of Auditory Comprehension standard scores than Expressive Communication. In this sample, we expect the SAEVD-R developmental levels to be more

highly correlated with the smaller range of Expressive Communication standard scores than the wider range of Auditory Comprehension scores.



Potential Problems and Alternative Outcomes. Data collection from all 21 infants that will be used to address the Aims of this project is complete. For this reason, few problems can be anticipated. In order to analyze the data as described, the project requires additional funds to support training and coding of infant vocalizations using the SAEVD-R levels. The timeline for these activities is described in *Management Plan and Budget.* For coder training, if coders do not reach the 10% discrepancy criterion (as described in *Coding Training*) by the fourth week of training, additional 1-hour meetings will be scheduled to maintain the proposed timeline. Previous research using similar coder training methods has demonstrated adequate coder calibration for coder agreement within a similar timeframe proposed in our project (Oller et al., 2019).

It is important to note that Aims 1 and 2 are independent of one another, thus, unexpected findings in one Aim will not directly impact the analysis of the other Aim. Because very little research has ever studied prelinguistic vocal behaviors in CP, even unexpected findings from our project can guide future research directions and inform our theoretical understanding of typical and atypical vocal development in human infants. For example, in Specific Aim 1, if infants are in fact observed to produce a larger number of vocalizations categorized into *higher* SAEVD-R developmental levels than *lower* levels, we may hypothesize that these findings support the evolutionary-developmental theory that prelinguistic vocal stages emerge robustly in humans, even in the face of neurological damage, in support of the capacity for advanced communication abilities in our species (Oller, 2019). Similarly, because of the comparable mean of receptive and expressive language scores used in SA2, we may observe no strong correlations among infants' SAEVD-R developmental levels and language performance. In this case, we may seek to conduct a principal component analysis across identified vocal developmental levels, cognition, language, and functional communication measures collected between the ages studied to determine which measures account for the most variance across all variables. This particular level of analysis is already planned for future studies examining data clustering of infants under 24 months that map onto established speech-language profile groups (Hustad et al., 2010).

Scientific Career Goals. In achieving each of the three Aims, scientific knowledge is advanced toward the identification of prelinguistic markers of communication impairments in children with CP, a disorder known for its wide variability of severity profiles and diagnostic uncertainty in infancy. This research not only has the potential to provide prognostic information to clinicians and families of children with CP in this population, but also the diagnosis of CP at even earlier ages. Findings from this research proposal will build the foundation for my programmatic line of research on the early detection of speech impairments in children with developmental disabilities. This work can greatly inform speech and language diagnostic and treatment planning and improve treatment outcomes for these children. Future immediate directions of this research include 1) developing a prelinguistic classification system of vocal development in CP based on the SAEVD-R for use as a clinical tool to identify estimated prelinguistic developmental age equivalencies, 2) studying the impact of infant-directed speech

on prelinguistic vocal performance in CP, and 3) developing early intervention treatments supporting parallel speech and AAC use. The research conducted under this grant is foundational to the development of future NIDCD K- and R- grants to conduct innovative research in this area. This research award will also support my ongoing success as I transition into independence as a clinical scientist in a tenure-track faculty position.

D. Facilities and Resources

Mentorship Team. Katherine Hustad, PhD, CCC-SLP will continue to serve as my primary postdoctoral mentor as PI of the WISC Lab. Dr. Hustad is dually appointed as a Full Professor in the Department of Communication Sciences and Disorders and the Waisman Center of the University of Wisconsin-Madison. Her research examines longitudinal speech, language, and communication development in children with cerebral palsy (CP) using observational and experimental research methods. Her work has been NIH funded since 2003 (5 R01 grants, 1 R03 grant; 1 K23 grant) with over 80 publications. Dr. Hustad and I will continue to meet for weekly laboratory meetings and weekly one-on-one meetings to discuss project status throughout the award period. Tristan Mahr, PhD, will serve as the statistical consultant for this project. Dr. Mahr has been a data scientist within the WISC Laboratory at the Waisman Center since 2018 with 17 publications. He has an extensive biostatistical background studying growth in child development using Bayesian statistics, functional programming, and mixed effects models required to support my ongoing research. Dr. Mahr and I will meet for weekly laboratory meetings and schedule weekly group meetings with Dr. Hustad during the data analysis period projected in *Management Plan.*

The **Waisman Center** of the University of Wisconsin-Madison (a Research I University) is a comprehensive multidisciplinary center with faculty from more than 25 affiliated departments conducting biological and behavioral research in human development, developmental disabilities, and neurodegenerative diseases. As an appointee of the Waisman Center T32 postdoctoral program, I regularly attend interdisciplinary research and training activities through the Waisman Center. These include a weekly John D. Wiley research seminar series featuring presentations from renowned behavioral and biological scientists in intellectual and developmental disabilities (IDD) and a twice monthly Ethics & Professional Development seminar. The T32 program will pay 100% of my salary with 100% dedicated research time throughout the project period. The T32 also includes travel funding for 1-2 conferences per year. State-wide recruitment for infant participation in the larger longitudinal project occurs through the Waisman Center's Clinical Translational Core registry.

The **Wisconsin Intelligibility, Speech, and Communication (WISC) Laboratory,** housed within the Waisman Center at UW-Madison, stores the dataset of laboratory recordings to be used in this project. I have been a postdoctoral researcher in the WISC Lab since April 2021. It is a well-equipped research laboratory space with a recording suite, recording studio, and four student and staff workspace rooms. The recording suite is fitted with state-of-the-art digital audio and video recording equipment, children's furniture, and toys. The laboratory staff includes the PI (*Hustad*), two research lab managers, a data scientist (*Mahr*), two postdocs (including me), and ~5 research assistants per year. I have a shared office space in the WISC Lab with a personal desk, laboratory computer, wired internet, remote and direct access to the secure WISC Lab server, and office supplies that will remain available to me throughout the course of the project. Laboratory meetings are held twice weekly to discuss ongoing research projects and logistics. Travel funding support is also available through Dr. Hustad's ongoing research grants.

REFERENCES

Andersen, G., Mjøen, T. R., & Vik, T. (2010). Prevalence of Speech Problems and the Use of Augmentative and Alternative Communication in Children With Cerebral Palsy: A Registry-Based Study in Norway. *Perspectives on Augmentative and Alternative Communication*, *19*(1), 12. doi: 10.1044/aac19.1.12.

Coleman, A., Weir, K. A., Ware, R. S., & Boyd, R. N. (2013). Relationship between communication skills and gross motor function in preschool-aged children with cerebral palsy. *Archives of Physical Medicine and Rehabilitation*, *94*(11), 2210–2217. doi: 10.1016/j.apmr.2013.03.025.

Erwin, E., & Hustad, K. (2007). *Vocal behaviors of prelinguistic children with cerebral palsy*. American Speech-Language Hearing Association Convention, Boston, MA.

Hidecker, M. J. C., Slaughter, J., Abeysekara, P., Ho, N. T., Dodge, N., Hurvitz, E. A., Workinger, M. S., Kent, R. D., Rosenbaum, P., Lenski, M., Vanderbeek, S. B., DeRoos, S., & Paneth, N. (2018). Early predictors and correlates of communication function in children with cerebral palsy. *Journal of Child Neurology*. doi: 10/gcvwfr.

Hustad, K. C., Allison, K. M., Sakash, A., McFadd, E., Broman, A. T., & Rathouz, P. J. (2016). Longitudinal development of communication in children with cerebral palsy between 24 and 53 months: Predicting speech outcomes. *Developmental Neurorehabilitation*, *20*(6), 1–8. doi: 10.1080/17518423.2016.1239135.

Hustad, K. C., Allison, K., McFadd, E., & Riehle, K. (2014). Speech and language development in 2-year-old children with cerebral palsy. *Developmental Neurorehabilitation*, *17*(3), 167–175. doi: 10.3109/17518423.2012.747009.

Hustad, K. C., Gorton, K., & Lee, J. (2010). Classification of speech and language profiles in 4-year-old children with cerebral palsy: A prospective preliminary study. *Journal of Speech, Language, and Hearing Research*, *53*(6), 1496–1513.

Hustad, K. C., Oakes, A., & Allison, K. (2015). Variability and diagnostic accuracy of speech intelligibility scores in children. *Journal of Speech, Language, and Hearing Research*.

Lang, S., Bartl-Pokorny, K. D., Pokorny, F. B., Garrido, D., Mani, N., Fox-Boyer, A. V., Zhang, D., & Marschik, P. B. (2019). Canonical babbling: A marker for earlier identification of late detected developmental disorders? *Current Developmental Disorders Reports*, *6*(3), 111–118.

Lee, J., Hustad, K. C., & Weismer, G. (2014). Predicting speech intelligibility with a multiple speech subsystems approach in children with cerebral palsy. *Journal of Speech, Language, and Hearing Research*, *57*(5), 1666–1678. doi: 10/f8k5q4.

Levin, K. (1999). Babbling in infants with cerebral palsy. *Clinical Linguistics & Phonetics*, *13*(4), 249–267. doi: 10.1080/026992099299077.

Lohmander, A., Holm, K., Eriksson, S., & Lieberman, M. (2017). Observation method identifies that a lack of canonical babbling can indicate future speech and language problems. *Acta Paediatrica, International Journal of Paediatrics*, *106*(6), 935–943. doi: 10/f939rh.

Long, H. L., Eichorn, N., & Oller, D. K. (under review). *A probe study on vocal development in two infants at risk of cerebral palsy*. doi: osf.io/ev397.

Long, H. L., Mahr, T. J., & Hustad, K. C. (in preparation). *Longitudinal development of marginal and canonical babbling in 14 infants at risk of cerebral palsy*.

Lynch, M. P., Oller, D. K., Steffens, M. L., Levine, S. L., Basinger, D. L., & Umbel, V. (1995). Onset of speech-like vocalizations in infants with Down syndrome. *American Journal of Mental Retardation*, *100*(1), 68–86.

McCathren, R. B., Yoder, P. J., & Warren, S. F. (1999). The relationship between prelinguistic vocalization and later expressive vocabulary in young children with developmental delay. *Journal of Speech, Language, and Hearing Research*, *42*(4), 915–924. doi: 10/gddc5k.

McGraw, K. O., & Wong, S. P. (1996). Forming inferences about some intraclass correlation coefficients. *Psychological Methods*, *1*(1), 30–46. doi: 10/br5ffs.

Mei, C., Reilly, S., Reddihough, D., Mensah, F., & Morgan, A. (2014). Motor speech impairment, activity, and participation in children with cerebral palsy. *International Journal of Speech-Language Pathology*, *16*(4), 427–435. doi: 10/gf6x3m.

Nathani, S., Ertmer, D. J., & Stark, R. E. (2006). Assessing vocal development in infants and toddlers. *Clinical Linguistics and Phonetics*, *20*(5), 351–369. doi: 10/dkbqjx.

Nyman, A., & Lohmander, A. (2018). Babbling in children with neurodevelopmental disability and validity of a simplified way of measuring canonical babbling ratio. *Clinical Linguistics and Phonetics*, *32*(2), 114–127. doi: 10/gbvt2q.

Oller, D. K. (1978). Infant vocalizations and the development of speech. *Allied Health and Behavioral Sciences*, *1*, 523–549.

Oller, D. K. (2000). *The emergence of the speech capacity*. Lawrence Erlbaum Associates.

Oller, D. K. (2019). Evolutionary-developmental modeling of neurodiversity and psychopathology. *Behavioral and Brain Sciences*, *42*, 1–4. doi: 10/gpxjv9.

Oller, D. K., Caskey, M., Yoo, H., Bene, E. R., Jhang, Y., Lee, C.-C., Bowman, D. D., Long, H. L., Buder, E. H., & Vohr, B. (2019). Preterm and full-term infant vocalization and the origin of language. *Scientific Reports*, *9*(1), 14734. doi: 10/gnzf8m.

Oller, D. K., & Delgado, R. E. (1999). *Logical International Phonetics Programs (Windows version) [Computer software]*. Intelligent Hearing Systems.

Oller, D. K., Eilers, R. E., Neal, A. R., & Cobo-Lewis, A. B. (1998). Late onset canonical babbling: A possible early marker of abnormal development. *American Journal on Mental Retardation*, *103*(3), 249.

Oller, D. K., Eilers, R. E., Neal, A. R., & Schwartz, H. K. (1999). Precursors to speech in infancy: The prediction of speech and language disorders. *Journal of Communication Disorders*, *32*(4), 223–245. doi: 10.1016/s0021-9924(99)00013-1.

Oller, D. K., Griebel, U., & Warlaumont, A. S. (2016). Vocal development as a guide to modeling the evolution of language. *Topics in Cognitive Science*, *8*(2), 382–392.

Otapowicz, D., Sobaniec, W., Kułak, W., & Okurowska-Zawada, B. (2005). Time of cooing appearance and further development of speech in children with cerebral palsy. *Roczniki Akademii Medycznej w Białymstoku (1995)*, *50 Suppl 1*(October), 78–81.

Overby, M. S., Belardi, K., & Schreiber, J. (2020). A retrospective video analysis of canonical babbling and volubility in infants later diagnosed with childhood apraxia of speech. *Clinical Linguistics and Phonetics*, *34*(7), 634–651. doi: 10/gpxr4r.

Parkes, J., Hill, N., Platt, M. J., & Donnelly, C. (2010). Oromotor dysfunction and communication impairments in children with cerebral palsy: A register study. *Developmental Medicine & Child Neurology*, *52*(12), 1113–1119. doi: 10/dqzbcp.

Stark, R. E. (1980). Stages of speech development in the first year of life. In *Child Phonology* (Vol. 1, pp. 73–90). Academic Press.

Stark, R. E. (1981). Infant vocalization: A comprehensive view. *Infant Mental Health Journal*, *2*(2), 118–128.

Vihman, M. M., Macken, M. A., Miller, R., Simmons, H., & Miller, J. (1985). From babbling to speech: A re-assessment of the continuity issue. *Language*, *61*(2), 397–445.

MANAGEMENT PLAN AND BUDGET

A. Management Plan

Research proposal timeline. Duration from December 2022 - December 2023.

Activity	Dec	Jan	Feb	Mar	Apr	May	Jun	Jul	Aug	Sep	Oct	Nov	Dec
Equipment purchase/set-up													
Coder interviews													
Coder training													
Video extraction													
Vocal coding*													
Data cleaning/wrangling													
Data analysis													
Manuscript writing													
Submit manuscript: JSLHR													
Submit abstract: ICIS talk													
Prepare NIDCD K23													
Submit NIRG final report													

** Includes primary and reliability vocal coding; ICIS= International Congress of Infant Studies; NIRG = New Investigators Research Grant*

IRB Approval. The ongoing IRB for the longitudinal research project under which this research project is subsumed was originally approved in 2018. It was most recently approved at a continuing review in Dec 21. Dr. Hustad and I will ensure that the IRB is resubmitted at the next continuing review prior to the expiration date of Dec 22.

Equipment Purchase and Set-Up. In Dec 22, we will purchase the AACT software, two workstations, and coding accessories. During this time, we will install the software onto the workstations to prepare for coder training and formal vocal coding.

Research Assistant Hiring, Training, and Vocal Coding. In Jan 22, Dr. Hustad and I will interview 3-4 graduate students in the speech-language pathology program at UW-Madison to fill the two student research assistant positions. Coders will be required to have had at least one course in phonetic transcription. From Feb – Jul 23, we will initiate coder training, video extraction, and vocal coding as described in the *Research Plan*. All coding data is expected to be complete and ready for analysis by Jul 23.

Manuscript Preparation. From Jul – Oct 23, I will consult with Dr. Mahr to ensure appropriate data cleaning and wrangling in preparation for formal analysis. Once complete, my mentorship team and I will initiate the formal statistical analyses for SA1 and SA2. This period will overlap with manuscript writing, between Sept – Dec 23. I will submit the final manuscript to ASHA's Journal of Speech, Language, and Hearing Research in Dec 23.

Additional Activities. I will meet weekly with my mentorship team in laboratory meetings to discuss the project. I will also meet with Dr. Hustad for weekly one-on-one meetings throughout the entire course of the project. Dr. Mahr will join our one-on-one meetings in Jul – Oct 23 to support data wrangling and analysis. In Sept 23, I will submit an abstract to present the study findings at the International Congress of Infant Studies (ICIS) in Glasgow, UK. The ICIS Congress is a bi-annual international conference that hosts world-renowned scientists presenting research across all infant developmental domains. From Aug – Dec 23, I will prepare and submit

a K23 award to the NIDCD to receive additional training with Dr. Hustad and other Waisman Center faculty to study the temporal relationship between early infant vocal and motor development in typical and CP-risk infants, and statistical modeling of longitudinal growth in children. Finally, I will write and submit the final report of activities and outcomes from this research project before the deadline of Dec 31, 2023.

B. Budget

The start-up funding required to engage in the proposed work (technology and training) will also support future studies examining prelinguistic vocal development using the same methods. In the case of leftover funds, we will use these resources for infant recruitment and additional research assistant vocal coding of new laboratory recordings for analysis of relevant data to be included in the proposed research.

Budget and justification.

Item	Cost	Justification
Coding workstation technology		
2 dedicated Dell OptiPlex 7780 All-in-One desktop computers	$2,190	*Primary computer for coding* (1 @ $1,095) Dedicated workstations to support current and future vocal coding of research projects examining prelinguistic development in CP.
2 Sony MDR ZX110NC Noise Cancelling Headphones	$160	*Headphones for focused coding* (1 @ $80) Noise-cancelling headphones required for vocal coding of specific vocal behaviors in a laboratory with 3-4 other laboratory assistants in the room.
Seagate Backup Plus Hub 8TB External Hard Drive	$150	*Backup data storage management* Required to store high-quality video, audio, and coded data to reduce the chances of data loss.
AACT software		
AACT coding software	$4,500	*Laboratory license of AACT software* AACT is required for infant vocal coding because it was specifically developed for coding and analyzing up to four levels of infant vocal behaviors in real-time.
Research assistant vocal coding		
2 research assistants at $15/hour, 10 hours per week	$3,000	Training, video extraction, and primary (100%) and reliability (20%) vocalization coding
Total	$10,000	

BIOGRAPHICAL SKETCH

Provide the following information for the Senior/key personnel and other significant contributors.
Follow this format for each person. **DO NOT EXCEED FIVE PAGES.**

NAME: Long, Helen Lauren

eRA COMMONS USER NAME (credential, e.g., agency login): hlong1

POSITION TITLE: Postdoctoral Trainee

EDUCATION/TRAINING *(Begin with baccalaureate or other initial professional education, such as nursing, include postdoctoral training and residency training if applicable. Add/delete rows as necessary.)*

INSTITUTION AND LOCATION	DEGREE *(if applicable)*	Completion Date MM/YYYY	FIELD OF STUDY
Indiana University, Bloomington, IN, USA	BA	05/2010	Speech & Hearing Sciences; Slavic & Eastern European Languages and Culture
Florida State University, Tallahassee, FL, USA	MS	08/2012	Speech-Language Pathology
University of Memphis, Memphis, TN, USA	PhD	12/2020	Communication Sciences and Disorders
University of Wisconsin, Madison, WI, USA	Postdoc	In Progress	Communication Sciences and Disorders

A. Personal Statement

My programmatic line of research aims to identify prelinguistic vocal biomarkers for communication impairment in children with cerebral palsy (CP) as young as infancy. A key component of this line of work necessitates foundational learning of prelinguistic vocal correlates of speech, language, and communication performance in this population. This work is critical to the ongoing study of the early detection of predictors for communication impairment in neurological disorders. My research questions are based in my clinical experience as a speech-language pathologist and doctoral and postdoctoral training with Dr. D. Kimbrough Oller and Dr. Katherine Hustad, leading experts in infant vocal development and speech development in CP, respectively. My research to date has used observational research methods to evaluate features of the vocal and speech signal that can signal information about developmental progress to parents in typical and atypical development. My dissertation with Dr. Oller innovatively offered perspective on the role of vocalization across social and nonsocial contexts as a signal of developmental progress. Following my doctoral training, I transitioned to a T32 postdoctoral training program at the UW-Madison Waisman Center to work with Dr. Hustad where I am currently. Our research longitudinally studies functional communication, school-based treatment goals, and changes in speech impairment classification in school-aged children with CP. These experiences provided invaluable training to support my programmatic line of research in vocal predictors of speech impairment in infants at risk for CP. The proposed research and subsequent studies will ground the future directions of this unexplored line of research, namely to support the development of diagnostic tools and early interventions for children with CP. The specific aims in this project will directly support my transition to an independent investigator, and

provide the critical preliminary information needed as I develop a K23 and future R01 applications.

1. **Long HL**, Mahr TJ, Natzke P, Rathouz PJ, Hustad KC. Longitudinal change in speech classification between 4 and 10 years in children with cerebral palsy. Dev Med Child Neurol. 2022 Mar 9. doi: 10.1111/dmcn.15198. Epub ahead of print. PMID: 35262181.
2. Koopmans C, Sakash A, Soriano J, **Long HL**, Hustad KC. Functional Communication Abilities in Youth With Cerebral Palsy: Association With Impairment Profiles and School-Based Therapy Goals. Lang Speech Hear Serv Sch. 2022 Jan 5;53(1):88-103. doi: 10.1044/2021_LSHSS-21-00064. Epub 2021 Nov 12. PMID: 34767477.
3. **Long HL,** Bowman DD, Yoo H, Burkhardt-Reed MM, Bene ER, Oller DK. Social and endogenous infant vocalizations. PLoS One. 2020 Aug 5;15(8):e0224956. doi: 10.1371/journal.pone.0224956. PMID: 32756591; PMCID: PMC7406057
4. **Long HL,** Oller DK, Bowman DA. Reliability of Listener Judgments of Infant Vocal Imitation. Front Psychol. 2019 Jun 11;10:1340. doi: 10.3389/fpsyg.2019.01340. PMID: 31244735; PMCID: PMC6579846.

B. Positions, Scientific Appointments, and Honors

Positions and Scientific Appointments

2021-	NICHD T32 Postdoctoral Trainee, Waisman Center, Univ. of Wisconsin-Madison
2015-2020	Research Assistant, Origin of Language Laboratory, Univ. of Memphis
2014-2021	Speech-Language Pathologist, Invo-Progressus Therapy
2012-2014	Speech-Language Pathologist, Easter Seals Massachusetts
2011-2012	Research Assistant, Experimental Child Phonetics Lab, Florida State Univ.
2009-2010	Research Assistant, Voice Physiology Lab, Indiana University-Bloomington

Other Experience and Professional Memberships

2021-	Scientific Review Committee, Am. Acad. for Cerebral Palsy and Dev. Medicine
2021-	Speech and Language Sciences Review Committee, ASHA Convention
2021-	Scholarship Review Committee, Am. Acad. for Cerebral Palsy and Dev. Medicine
2020-	Ad hoc reviewer, AJSLP, JSLHR, JCSD, etc.
2020-	Co-founding collaborator, CSDisseminate
2019-2021	Member, International Congress of Infant Studies
2018-	Membership Committee, Am. Acad. for Cerebral Palsy and Dev. Medicine
2013-	Member, American Speech-Language Hearing Association (CCC-SLP)
2009-2012	Member, National Student Speech-Language Hearing Association

Honors

2020	Graduate Student Association President Service Award, University of Memphis
2020	Celebrate Student Success Award, University of Memphis
2020	OrthoPediatrics™ Scholarship, Am. Acad. for Cerebral Palsy and Dev. Medicine
2019	S.P. Wong Award for Best Presentation in Statistical Application, U. of Memphis
2012	Red Apple Award for Outstanding School Personnel, Southborough Education Foundation
2012	Outstanding 2nd Year Master's Student Award, Florida State University
2011	Dr. Avery Vaughn Scholarship Fund for Excellence, Florida State University

2006-2010 H. Fullmer Faculty Scholarship Award, Indiana University

C. Contributions to Science

1. **Evolutionary origins of language and infant vocalizations as fitness signals.** This line of research is founded in the notion that the stages of prelinguistic vocal abilities follow a natural logic of development foundational to advanced linguistic skills in humans. My doctoral dissertation evaluated the role of social and endogenous factors influencing infant vocalizations as "fitness" signals, i.e., indicators of developmental wellbeing. Specifically, I investigated the reliability of listener judgments of the degree of infant vocal imitativeness as a measure of the saliency of potential vocal fitness signals. I found high intra- and inter-rater agreement among listeners, suggesting vocal imitation has the potential to be used as a signal of fitness in early development. I also quantified the extent to which infants produce social and endogenous (i.e., nonsocial) vocalizations across contexts. The findings highlight a potentially robust internal social motivation for vocalization, even in the presence of likely social-cognitive differences such as risk for autism. This line of research offers perspective on the ways in which social and endogenous factors reveal natural selection pressures on fitness signaling in the human infant.

    a. **Long HL,** Oller DK, Bowman DA. Reliability of Listener Judgments of Infant Vocal Imitation. Front Psychol. 2019 Jun 11;10:1340. doi: 10.3389/fpsyg.2019.01340. PMID: 31244735; PMCID: PMC6579846.
    b. **Long HL,** Bowman DD, Yoo H, Burkhardt-Reed MM, Bene ER, Oller DK. Social and endogenous infant vocalizations. PLoS One. 2020 Aug 5;15(8):e0224956. doi: 10.1371/journal.pone.0224956. PMID: 32756591; PMCID: PMC7406057.
    c. Oller DK, Griebel U, Bowman DD, Bene E, **Long HL**, Yoo H, Ramsay G. Infant boys are more vocal than infant girls. Curr Biol. 2020 May 18;30(10):R426-R427. doi: 10.1016/j.cub.2020.03.049. PMID: 32428468; PMCID: PMC8204662.
    d. Oller DK, Ramsay G, Bene E, **Long HL**, Griebel U. Protophones, the precursors to speech, dominate the human infant vocal landscape. Philos Trans R Soc Lond B Biol Sci. 2021 Oct 25;376(1836):20200255. doi: 10.1098/rstb.2020.0255. Epub 2021 Sep 6. PMID: 34482735; PMCID: PMC8419580.

2. **Prelinguistic vocal development in clinical populations.** During my doctoral work, I independently sought experiences supporting my research training of vocal development in disorders at risk of speech or language impairments. The last paper in my dissertation examined canonical babbling in infants at risk of autism. Our findings suggested a potential robust internal motivation to produce a high rate of canonical syllables in both social and nonsocial contexts, even in the likely presence of autism. I also collaborated on a project evaluating vocal development in tuberous sclerosis. We found delayed canonical babbling and a low rate of their canonical syllable production compared to typical expectations. Because of my interest in CP, I independently initiated two preliminary studies to study this population in my doctoral program. First, I completed a longitudinal observation of canonical babbling in two infants at risk for CP between 5-16 months of age. We observed delayed canonical babbling and unexpected variability in its emergence over time. Second, I coordinated a research collaboration with a local neuromuscular clinic to study speech and

language milestones in CP. We found that motor impairments significantly impact infants' ability to perform receptive and expressive language tasks, but the severity of impairment did not predict whether the child was receiving speech therapy. These findings indicate a critical need for further study of prelinguistic vocal patterns relative to later outcomes.

a. Gipson TT, Ramsay G, Ellison EE, Bene ER, **Long HL**, Oller DK. Early Vocal Development in Tuberous Sclerosis Complex. Pediatr Neurol. 2021 Dec;125:48-52. doi: 10.1016/j.pediatrneurol.2021.08.009. Epub 2021 Sep 10. PMID: 34628143; PMCID: PMC8557126.
b. **Long, HL,** Oller, DK, Romer, K, Friener, L, Warner, W, Spence, D, & Rhodes, LN. Pre-Speech and Early Speech Development of Young Children Diagnosed with Cerebral Palsy. [Abstract]. Dev Med Child Neurol, 2020 Oct;62,Suppl. 3:79-134. doi: 10.1111/dmcn.14662

Other Publications (under peer review)

c. **Long, HL,** Eichorn, N, & Oller, DK. A Probe Study on Vocal Development in Two Infants at Risk for Cerebral Palsy. [Preprint], osf.io/ev397/.
d. **Long, HL,** Ramsay, G, Bowman, D, Burkhardt-Reed, MM, & Oller, DK. Canonical Babbling in Vocal Turn Taking and Independent Vocal Play. [Preprint], doi: 10.1101/2020.10.09.333872

3. **Speech, language, and communication development in cerebral palsy**. My work as a NICHD T32 postdoctoral trainee has focused on the study of speech, language, and communication development in school-aged children with CP. In an investigation on school-based treatment goals, we found only the most severely impaired children had augmentative and alternative communication (AAC) goals, although children with lower levels of communication impairment also had reduced intelligibility. These findings suggest the need to integrate AAC into speech treatment, especially in early ages. I also led an examination of longitudinal speech changes in CP between 4-10 years. We found that speech impairments are likely to improve over time with ongoing speech development except in the most severe cases, suggesting an immediate need for AAC as early as age four. A recently submitted manuscript evaluating marginal and canonical babbling development in infants at risk for CP between 11-24 months found that the emergence of prelinguistic vocal stages is significantly delayed by at least one year. The clinical implications of this research supports the need for continued research in this area.

a. Hidecker, MJC & **Long, HL** When to refer: Early indicators for communication concerns in cerebral palsy. American Academy of CP Dev Med 74th Annual Meeting. 2020 Sept.
b. Koopmans C, Sakash A, Soriano J, **Long HL**, Hustad KC. Functional Communication Abilities in Youth With Cerebral Palsy: Association With Impairment Profiles and School-Based Therapy Goals. Lang Speech Hear Serv Sch. 2022 Jan 5;53(1):88-103. doi: 10.1044/2021_LSHSS-21-00064. Epub 2021 Nov 12. PMID: 34767477.
c. **Long HL**, Mahr TJ, Natzke P, Rathouz PJ, Hustad KC. Longitudinal change in speech classification between 4 and 10 years in children with cerebral palsy. Dev

Med Child Neurol. 2022 Mar 9. doi: 10.1111/dmcn.15198. Epub ahead of print. PMID: 35262181.

Other Publications (under peer review)

d. **Long, H. L.,** Mahr, T. J., & Hustad, K. C. Longitudinal Development of Marginal and Canonical Babbling in Infants at Risk for Cerebral Palsy.

Complete List of Published Work in MyBibliography:
https://www.ncbi.nlm.nih.gov/myncbi/1RMPzPxaXG8Qm/bibliography/public/

KEY PERSONNEL

1. **Primary Postdoctoral Mentor:** Dr. Katherine Hustad will continue to serve as my primary postdoctoral mentor throughout the course of the project. The proposed research utilizes a dataset collected within her laboratory. We will hold weekly laboratory meetings and weekly one-on-one meetings for ongoing mentorship to support the research proposal and to discuss logistical needs of the study throughout the entire course of the project (December 2022-December 2023). Additional one-on-one meetings will be scheduled as needed.

   Katherine C. Hustad, PhD, CCC-SLP
   Professor
   Department of Communication Sciences & Disorders
   University of Wisconsin-Madison
   Waisman Center, Room 463
   Wisconsin Intelligibility, Speech, and Communication (WISC) Laboratory
   1500 Highland Avenue
   Madison, WI 53705
   kchustad@wisc.edu
   (608) 265-9977

2. **Statistical consultant:** Dr. Tristan Mahr will serve as the statistical consultant for this project. We will meet at weekly laboratory meetings to discuss ongoing progress on the proposed research. Dr. Mahr will join my weekly one-on-one meetings with Dr. Hustad during the anticipated timeline of data management, wrangling, and analysis (May-August 2023) to discuss specific statistical analysis methods. Additional meetings will be scheduled as needed.

   Tristan J. Mahr, PhD
   Research Scientist II
   University of Wisconsin-Madison
   Waisman Center, Room 465
   Wisconsin Intelligibility, Speech, and Communication (WISC) Laboratory
   1500 Highland Avenue
   Madison, WI 53705
   mahr@wisc.edu
   (608) 263-5822