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## Language outcomes in children who are deaf and hard of hearing: The role of language ability before hearing aid intervention

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Language Outcomes in Children who are Deaf and Hard of Hearing: The Role of Language  
Ability Before Hearing Aid Intervention

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## Abstract

*Purpose:* Early auditory experiences are fundamental in infant language acquisition. Research consistently demonstrates the benefits of early intervention (i.e., hearing aids) to language outcomes in children who are deaf and hard of hearing. The nature of these benefits, and their relation with pre-fitting development are, however, not well understood.

*Methods:* This study examined Ontario Infant Hearing Program birth cohorts to explore predictors of performance on the Preschool Language Scale-4 (PLS-4) at the time of ( $N=47$ ) and after ( $N=19$ ) initial hearing aid intervention.

*Results:* Regression analyses revealed that before the hearing aid fitting, severity of hearing loss negatively predicted 19% and 10% of the variance in Auditory Comprehension and Expressive Communication, respectively. Post-hearing aid fitting, children's standard scores on language measures remained stable but they made significant improvement in their Progress Values, which represent individual skills acquired on the test, rather than standing relative to same-age peers. Magnitude of change in Progress Values was predicted by a negative interaction of pre-fitting language ability and severity of hearing loss for the Auditory Comprehension scale.

*Conclusions:* These findings highlight the importance of considering a child's pre-fitting language ability in interpreting eventual language outcomes. Possible mechanisms of hearing aid benefit are discussed.

**Keywords:** Children; hearing loss; language outcome; hearing aids

Language Outcomes in Children who are Deaf and Hard of Hearing: The Role of Language Ability Before Hearing Aid Intervention

It is well documented that early auditory experience dramatically impacts and shapes the language development of typically hearing infants and children (Kuhl et al., 2008; Maye, Weiss & Aslin, 2008; Maye, Werker & Gerken, 2002; Werker & Tees, 1984). In cases where access to auditory information is compromised by permanent childhood hearing loss, the importance of early access to auditory information has motivated the early identification of hearing loss via Universal Newborn Hearing Screening. Initial recommendations for Universal Newborn Hearing Screening were met with criticism based on both the absence of comprehensive follow-up assessment and intervention programs to support children who are deaf and hard of hearing (CD/HH) after screening, as well as a lack of evidence supporting benefits of such programs (Bess & Paradise, 1994). These concerns, along with the known importance of early access to auditory information, motivated the development of comprehensive Early Hearing Detection and Intervention (EHDI) programs to provide appropriate follow-up after initial detection of hearing loss. Furthermore, these criticisms and concerns influenced research evaluating the effectiveness of comprehensive EHDI programs. When considered as an overall predictor, the benefits of early hearing detection and intervention have been demonstrated (Wake et al., 2016; Vohr et al., 2008; Kennedy et al., 2006; Yoshinaga-Itano, Coulter & Thomson, 2001; Moeller 2000; Yoshinaga-Itano et al., 1998; see Yoshinaga-Itano 2003 for a review) and some work has revealed age-appropriate language achievement on certain measures of language ability in CD/HH who received early intervention (Fulcher, Purcell, Baker & Munro, 2012; Moeller 2000; Stika et al., 2014).

Recent work has moved beyond considering EHDI as a single factor to begin studying how specific aspects of early detection and intervention relate to language outcomes. Outcomes for Childhood Hearing Loss (OCHL), a well-controlled accelerated longitudinal study, recently explored various factors that predict outcomes in a sample of 317 CD/HH, most of whom ( $n = 308$ ) were fitted with hearing aids. The OCHL project explored language achievement using composite measures of language ability from 2 to 6 years of age and identified that while earlier hearing aid fitting was associated with improved language outcomes, it was not the only factor that predicted performance. Caregiver's quantity (amount) and quality (e.g., varied vocabulary, eliciting utterances that were longer and higher level) of speech contributed to both language performance and growth over time. Additionally, audiological characteristics such as hearing aid dosage (the residual difference between unamplified and amplified hearing levels, or rSII), amount of hearing aid use (both hours per day, and months with the hearing aid), and severity of hearing loss all contributed to both language performance and growth over time as well (Ambrose et al., 2015; Ambrose et al., 2014; Tomblin et al., 2015). Previous work has also explored the contributions of some of these factors to the outcomes of CD/HH who are fitted with hearing aids (Walker et al., 2015; Ambrose et al., 2014; Ching & Dillon, 2013; Ching et al., 2010), but has not considered all of them in such a large, single sample.

This recent research underscores the role that child-specific factors, beyond the presence/absence of EHDI intervention, may play in a child's language development and overall outcome. However, research has yet to consider the child's communicative development *prior to* hearing aid fitting as a predictive factor, despite recognition of the importance of early communicative experiences to eventual language outcome and its use as a rationale for the provision of early auditory access through EHDI programs. It is currently unknown how CD/HH

are able to perceive or process suboptimal speech signals that have not been amplified with hearing aids and use them to develop oral language. In regions with EHDI programs, the goal is for CD/HH whose families elect for amplification to be fitted with hearing aids promptly following confirmation of permanent hearing loss and parent readiness to proceed with fitting (Bagatto, Scollie, Hyde & Seewald, 2010; JCIH, 2007; SAC, 2010). Although CD/HH are identified and provided with hearing aids as early as possible, they are still faced with forming the perceptual and cognitive basis for their linguistic system for a number of months prior to personal amplification. This early auditory deprivation may have lasting, and cascading, impacts on a child's ability to learn spoken language later in development. For instance, infants' learning of phonemic categories at the age of 7.5 months has been shown to predict their vocabulary development at 30 months (Kuhl et al., 2008) and has been interpreted as a reflection of *neural commitment* to their native language. In the case of CD/HH, difficulty in learning information that is acquired earlier in development may result in cascading difficulties in language learning and therefore influence the degree of benefit hearing aids are able to provide. Evaluating the role of language and communication ability prior to receipt of hearing aids in eventual language outcome is crucial for understanding how CD/HH are able to use the amplified input provided by the hearing aids, and for developing theories regarding the mechanism by which intervention with hearing aids impacts the language outcomes of CD/HH.

EHDI programming was developed in the province of Ontario in 2002. The Ontario Infant Hearing Program (OIHP) serves as a branch of the Ontario Ministry of Children and Youth Services, with the goal to "facilitate the affected child's development of communication skills and readiness for school" (Bagatto et al., 2010, p. S71). As described in further detail below, data regarding the clinical management and assessment of all children enrolled in the OIHP are

maintained in a de-identified province-wide database. Access to this database was provided to the authors to facilitate assessment of audiological and language outcomes. As such, this database represented a serendipitous opportunity to explore our research questions in an ecologically valid context. However, due to the clinical nature of this database, many aspects of participant intervention and demographics were unavailable to the authors, which impacts the generalizability of this study to the broader deaf/hard of hearing pediatric population. For instance, while all children in our sample received hearing aids, it is unknown whether or not they were additionally learning signed language, or were considered candidates for cochlear implantation at later ages (although all children wore hearing aids for the duration of the study). In this regard, our data reflect the decision making practices of clinicians serving the OIHP, which are guided by policies and procedures designed in accordance with JCIH recommendations (Bagatto, Scollie, Hyde & Seewald, 2010; JCIH, 2007; SAC 2010). Although these limitations necessitate the application of additional caution in our interpretations, we present these data as preliminary evidence regarding the utility of considering pre-fitting language abilities in exploring eventual language outcomes for CD/HH, and consider potential mechanisms of hearing aid benefit to be evaluated in prospective studies.

### **Purpose**

The present work examined the role that the time prior to intervention with hearing aids plays in the language outcomes of CD/HH by examining their language performance around the time of hearing aid fitting and its relation with language performance after hearing aid fitting. Three main questions were addressed: 1) To what extent does hearing loss impact language performance prior to hearing aid intervention, 2) Can hearing aids ameliorate these effects, and if

so, 3) What factors predict the magnitude of change in language performance after exposure to hearing aids?

### **Methods**

This retrospective cohort analysis examined CD/HH serviced by the OIHP who were born in 2008 and 2011. Following recommendations set out by the Joint Committee on Infant Hearing and Speech Audiology Canada (Bagatto, Scollie, Hyde & Seewald, 2010; JCIH, 2007; SAC 2010), newborns in Ontario are universally screened for hearing loss and provided with appropriate family-centered follow-up services to confirm the presence of permanent hearing loss and provide intervention (e.g., hearing aids, communication development) according to the family's choices. The screening, audiological assessment, hearing aid fitting, and audiological outcome measurement components of the OIHP are based on evidence-based protocols that are implemented province-wide. Due to the varied nature of oral communication development service options, specifics regarding a child's intervention are not mandated by the OIHP, however, children identified with permanent hearing loss through the OIHP are routinely assessed using the *Preschool Language Scale, 4<sup>th</sup> edition* (PLS-4; Zimmerman, Steiner & Pond, 2009) to track their progress. Those data are entered into a provincial database.

### **Data collection**

Data were extracted from the OIHP clinical management database for the 2008 and 2011 birth cohorts as part of a larger evaluation of the program. Data extraction from the broader database proceeds through extraction requests to the Ministry. Thus, our use of these two birth cohorts was based on data availability. OIHP protocol mandates the collection of basic demographic data (sex, birth date, gestational age), audiological information (hearing thresholds, hearing aid fitting date, audibility provided by the hearing aid), outcome assessment information



(test results and scores), as well as information regarding appointment dates and complicating factors, for entry into a de-identified database. All information included in the database was obtained over the course of the child's care by their clinical service providers (e.g., audiologist, speech-language pathologist) who follow the relevant provincial protocols.

### **Participants**

Children within the database from the 2008 and 2011 birth cohorts had been identified as having permanent childhood hearing loss in at least one ear and had been fitted with hearing aids at some point during their care within the OIHP. Both children who were typically developing and children who had other comorbidities were included in the sample. Data from children were included in the current analyses if they had a PLS-4 language assessment conducted before or no more than two months after their first hearing aid fitting. On average, children had their language assessed within 1 month prior to fitting, however this time frame ranged from 13 months prior to fitting to 2 months after ( $M = -1.52$ ,  $SD = 4.23$ ). Forty-seven children were included in our final analyses of language ability around the time of hearing aid fitting. A subset of these ( $N = 19$ ) had data available for a second analysis of language ability both around the time of hearing aid fitting and at some time after the initial hearing aid fitting; these 19 children were additionally analyzed for language ability over time. The time between first and second PLS-4 assessment for children in this longitudinal group ranged between 4.63 and 11.27 months ( $M = 6.89$ ,  $SD = 2.25$ ). Due to the fact that pre-fitting language assessments were included any number of months before fitting, and up to two months after fitting, it is important to consider the proportion of time between the two assessments that the child had been fitted with their hearing aids. Of the 19 children in the longitudinal group, 13 of them had been fitted before their first assessment. Therefore, these children had access to their hearing aids during the time between assessments. Of the remaining

6 children, the pre-fitting assessment was, on average, 1 month before fitting ( $SD = 1.08$ ), ranging from 26 days to 3 months. The percentage of time between assessments that these 6 children had access to hearing aids ranged from 51% to 97% ( $M = 83\%$ ,  $SD = 0.17$ ). It is important to bear in mind, however, that proportion of time with access to hearing aids is not the number of hours per day that a child wears her hearing aid.

These broad inclusion criteria resulted in an extremely variable sample (see Table 1 for details), however, these facilitated our preliminary analyses. As a sample, our children were identified with hearing loss ( $M = 13.54$ ,  $SD = 15.56$ ) and fitted with hearing aids ( $M = 22.62$ ,  $SD = 16.63$ ) at older ages than the broader OIHP population, with an average hearing loss severity of 47.46 dB HL better-ear four pure tone average (BE-4PTA; described in further detail below). Of the children who had their language assessed before hearing aid fitting ( $N = 47$ ), 5 had unilateral losses. Of the children who had their language assessed both at the time of fitting and after ( $N = 19$ ), 2 had unilateral losses. It is important to note that the mean age of hearing aid fitting in the sample is older than JCIH benchmarks for children identified in the first few months of life (i.e., fitted by 6 months). Thus, as a group, our sample represents children who are identified with a hearing loss and fitted with hearing aids at older ages. This is possibly representative of our inclusion of children with comorbidities who, for a variety of reasons, may have received their hearing aids later, not of a failure in implementation of the OIHP. Children with comorbidity information entered in the database were included in our sample, as were children for whom the comorbidity field was left blank (i.e., it is unknown where or not they had a comorbidity). In the OIHP database, a comorbidity may be entered for the presence of medical issues (e.g., cerebral palsy or Down syndrome) or a complex factor (e.g., family or psychosocial challenges, inconsistent hearing aid use, Children's Aid involvement). Similarly, our sample

included children with unilateral hearing loss (represented by a BE-4PTA of less than 25 dB HL) who wear hearing aids, as well as children with profound bilateral hearing loss (BE-4PTA greater than 90 dB HL). Two children were excluded from our analyses because of discrepancies between the raw scores and the standard scores reported in the database that appeared to be due to data entry error and could not be resolved. One child was included but was missing scores for the PLS-4 Auditory Comprehension scale.

In accordance with the OIHP Protocol for the Provision of Amplification procedures (Bagatto, Scollie, Hyde & Seewald, 2010), infants and children are considered candidates for amplification if the hearing loss is permanent and hearing thresholds for either ear are 30 dB HL or greater at any frequency between 500 and 4000 Hz. Amplification is provided based on ear-specific threshold estimates at 500 and 2000 Hz using the Desired Sensation Level Method (Scollie et al., 2005) and real-ear-to-coupler difference measurements (Bagatto, Scollie, Hyde & Seewald, 2010; Bagatto et al., 2005). Given limitations in sample size, we were unable to statistically consider the unique effects of quality of the hearing aid fitting in the analysis. However, using normative values developed by Bagatto et al. (2011) to evaluate the audibility of speech provided by the hearing aids (e.g., speech intelligibility index, or SII) demonstrated that the audibility provided by hearing aids to the children in our sample fell within expected ranges in all but five cases (see Supplemental Figure 1). Four children had SIIs that fell below, and one child had values that fell above, the expected norms. SIIs were calculated using an Average Input (65 dB SPL) level.

### **Outcome measures**

Children's language ability was evaluated using the PLS-4, an omnibus language measure containing scales for Auditory Comprehension and Expressive Communication that is suitable

for children ranging from birth to 6 years, 11 months. Our analyses considered both standard scores and Progress Values (Zimmerman, Steiner & Pond, 2006) as outcome measures. Standard scores represent the performance of a child in standard deviation units on a distribution normed using the PLS-4 norming sample ( $M = 100$ ,  $SD = 15$ ), thus providing an estimate of the child's *relative* language abilities in comparison to age expectations. Progress Values (also known as growth scores or growth scale values) capture a child's *absolute* language abilities. They are similar to raw scores in this regard, but have the advantage over raw scores of being placed along an equal-interval scale, allowing for more accurate measurement of change over time. That is, Progress Values, unlike standard scores, do not consider a child's ability in relation to the PLS-4 norming sample, but rather provide an index of progress on the test specific to the individual child. For the purposes of our analyses, we chose to analyze both standard scores and Progress Values, which allowed us to consider children's performance and subsequent growth relative to both the normative sample as well as themselves. Considering a child's growth relative to the test norming sample using standard scores, as well as relative to themselves using Progress Values, enabled a richer interpretation than considering either score in isolation. Progress values indicate whether there has been improvement in language skills, while standard scores indicate whether the rate of improvement has been above or below the average rate for the child's same-age peers, both of which are important questions to examine about CD/HH.

Language ability around the time of fitting was operationalized using either the standard scores or Progress Values from a PLS-4 assessment either before or within two months of a child's first hearing aid fitting. Similarly, language ability after fitting was considered to be either the standard score or Progress Values from a PLS-4 assessment conducted sometime greater than two months after a child was first fitted with hearing aids. As previously noted, the

period between first and second assessments was not the same for all children in our sample. Time between assessment periods was statistically controlled by creating standardized residuals for the change scores, which was used as the outcome measure in all analyses of language growth.

### **Data Extraction**

We selected age at assessment and severity of hearing loss as relevant predictors for language ability around the time of hearing aid fitting. Since the PLS-4 standard scores are calculated using age, only severity of hearing loss was included as a predictor of language ability around the time of fitting for analysis of PLS-4 standard scores.

Severity of hearing loss was operationalized as the BE-4PTA calculated during the audiological assessment closest in date (but not following) the child's first language assessment. The BE-4PTA is the average of a child's dB HL thresholds across 500, 1000, 2000 and 4000 Hz. Pure tone averages are calculated for each ear individually and the BE-4PTA is the pure tone average of the ear with the lowest dB HL threshold, that is, the better hearing ear or the ear that has the least amount of hearing loss.

As previously outlined, the time between the two language assessments was not equal for all children, reflecting the variability in clinical assessment practices at the time of data collection. To control for these inequalities in our analyses of growth, the difference scores used for our growth analyses were residualized to account for the variance due to differences in time between assessments.

## **Results**

### **Language Ability Around the Time of Hearing Aid Fitting**

Hierarchical linear regression analyses were conducted on the Progress Values for the Auditory Comprehension and Expressive Communication scales, wherein predictors were entered into the analysis in the following order: Age at assessment, BE-4PTA, and the interaction between Age at assessment and BE-4PTA. The interaction between BE-4PTA and Age at assessment was included in our analyses because we predicted that cumulative experience with degraded auditory input will impact the growth in language ability associated with increasing age. R<sup>2</sup> change was evaluated for each model, and the most parsimonious model was considered to be the last model in which the additional predictors significantly contributed to the unique variance of the model, thus significantly improving explained variance. Details of these regression models are presented in Table 2.

The most parsimonious model for the prediction of Auditory Comprehension using Progress Values was the model that included Age at assessment, BE-4PTA, and the interaction between Age and BE-4PTA,  $R^2(\text{adj}) = 0.84$ ,  $F(3, 43) = 83.96$ ,  $p < 0.001$ . Within this model, both age and BE-4PTA were statistically significant predictors, but the interaction between these predictors did not significantly contribute to prediction.

Similarly, the most parsimonious model for the prediction of Expressive Communication using Progress Values was the model that included Age at assessment, BE-4PTA, and the interaction between Age at assessment, and BE-4PTA,  $R^2(\text{adj}) = 0.85$ ,  $F(3, 43) = 88.79$ ,  $p < 0.001$ . In this model, however, only age was found to be a statistically significant predictor of Expressive Communication.

A second set of hierarchical linear regression analyses was conducted for the standard scores, wherein BE-4PTA was entered. BE-4PTA was a significant predictor for both Auditory

Comprehension,  $R^2(\text{adj}) = 0.31$ ,  $F(1,45)=21.85$ ,  $p<0.001$ , and Expressive Communication scales,  $R^2(\text{adj}) = 0.15$ ,  $F(1,45)=9.3$ ,  $p=0.04$ . Details of these regression models are presented in Table 3.

### **Change in Language Ability**

There was no significant change in Auditory Comprehension and Expressive Communication standard score after hearing aid fitting,  $t(17)=-1.46$ ,  $p=0.16$  and  $t(18)=-0.76$ ,  $p=0.45$ , although Progress Values did show significant growth,  $t(17)=6.46$ ,  $p<0.001$  and  $t(18)=8.23$ ,  $p<0.001$ . Changes in standard score performance were also evaluated for individual children to identify whether or not there were subgroups of individual child performance that were masked by the group data, which would indicate that our null finding represented regression to the mean (see Supplemental Figure 2 depicting graphs of each child's performance over time with 90% confidence intervals). Using non-overlapping confidence intervals as a rough metric for determining significant change in relative standing, 14 children showed no significant change on either scale, 4 children (Child 3, 5, 6 & 12) showed significant change on one (but not both) scales and only one child (Child 8) showed significant change on both scales. Of the children who showed significant change, there was no consistency in the direction of change: some children's performance improved (Child 5 & 8), while others worsened (Child 3, 6 & 12). Thus, the null changes in children's standard score performance did not appear to be due to subgroups of growth patterns being masked in whole group analyses, and our subsequent analyses were conducted on our entire sample.

Hierarchical linear regression analyses were conducted on the residualized Progress Value difference scores, using predictors entered in the following order: Progress Values from the first assessment, BE-4PTA, and the interaction between Progress Values from the first assessment and BE-4PTA. Specifically, the dependent variables in these analyses were the Progress Value

difference scores where the variance due to unequal time between assessments across children was removed. The interaction between Progress Values and BE-4PTA was included in our analyses to investigate the degree to which the extent of the two risk factors (low initial language and more severe hearing losses) impacted the gain in language ability related to hearing aid fitting. As was the case in evaluating initial language ability, the most parsimonious model was considered to be the last model that produced significant  $R^2$  change. The most parsimonious model for residualized Auditory Comprehension Progress Value difference scores was the model that included first assessment Progress Values, BE-4PTA and the interaction between them, suggesting that the largest gains in language ability were made by those at the greatest initial risk: those with the weakest initial language abilities and the most severe hearing loss. In this model, BE-4PTA and the interaction between Progress Value at first assessment and BE-4PTA were significant predictors,  $F(3,14)=16.42$ ,  $p>0.001$ , explaining 70% of the variance. Our analyses did not produce a significant model of residualized Expressive Communication Progress Value change,  $F(1,17)=2.201$ ,  $p=0.1562$  (see Table 4 for a summary).

### **Language Ability after Fitting**

Final hierarchical regression analyses predicted Progress Value performance and standard scores after hearing aid fitting using BE-4PTA and Progress Value performance (or standard scores) from first assessments as predictors. Unlike previous analyses, the most parsimonious model was considered to be the model that significantly accounted for the most variance. We were not interested in  $R^2$  change for these regressions, as we were interested in evaluating how the relation between BE4PTA and language ability after hearing aid fitting changed when we accounted for the variance contributed by initial language ability. In all cases, the most parsimonious model was the model that included both BE-4PTA and pre-fitting Progress Value



(Auditory Comprehension:  $F(2,15)=90.01$ ,  $p<0.001$ , Expressive Communication:  $F(2,16)=49.87$ ,  $p<0.001$ ) or standard score (Auditory Comprehension:  $F(2,15)=12.51$ ,  $p<0.001$ , Expressive Communication:  $F(2,16)=5.298$ ,  $p=0.017$ ) as predictors. Unlike the regression models evaluating language ability prior to fitting, BE-4PTA was not a significant predictor for Auditory Comprehension Progress Value, Expressive Communication Progress Value, Auditory Comprehension standard score or Expressive Communication standard score in these models, whereas Progress Values and standard scores from initial language assessments were a significant predictor for all models except the model of Auditory Comprehension Progress Value post hearing aid fitting. Details of these regression models are included in Tables 5 and 6.

### **Discussion**

Our results indicate that severity of hearing loss impacted language ability prior to hearing aid fitting and that this had lasting effects on language outcomes after hearing aid fitting in our sample. Although children continued to acquire language skills after they are fit with hearing aids (as indicated by significant Progress Value change for both language scales), they maintained the same standing relative to same-age peers that they had before receiving hearing aids. The amount of Progress Value growth on the Auditory Comprehension scale was significantly predicted by an interaction of severity of hearing loss and Progress Values around the time of fitting, such that children with greater severities of loss experienced the greatest amount of Progress Value growth, but high levels of initial auditory comprehension abilities attenuated this growth. This suggests that the greatest benefits of hearing aids were delivered to the children who were at greatest initial risk: those with more severe hearing losses and the worst initial language comprehension ability. Furthermore, our analyses demonstrated that severity of hearing loss did not uniquely predict language ability after fitting. In our sample, the relation of

hearing loss severity to language ability after hearing aid fitting was driven by its relationship with language around the time of fitting, rather than further effects.

Considered together, our analyses highlight the importance of considering language outcomes of CD/HH in the context of their initial language abilities, rather than audiological characteristics in isolation. Studies that examine the language outcomes of children involved in EHDI programs have focused on providing evidence for the benefit of early hearing aid fitting supported by these programs as well as identifying factors that may improve language outcomes beyond amplification (Tomblin et al., 2015; Ambrose, VanDam, & Moeller 2014; Tomblin et al., 2014; Ching et al., 2010). However, these studies have not considered how language ability prior to hearing aid fitting may impact eventual language outcomes. Our results suggest that language ability prior to hearing aid fitting is another factor that predicts eventual language outcomes. With the increasing prevalence of EHDI programs, research examining language outcomes in CD/HH should increase attention to the role of unamplified development.

Exploring the language development of CD/HH before fitting will inform our understanding of *how* CD/HH are able to learn spoken language using hearing aids as well as improve early identification and remediation of language impairments. There are three possible mechanisms by which hearing aids may benefit the language development of CD/HH: catch-up, preservation/protection, or a combination of the two wherein the benefits change at different points in development. A catch-up hypothesis proposes that CD/HH acquire language skills at an increased rate after hearing aid fitting to acquire skills comparable to same-aged peers, whereas a preservation/protection hypothesis would argue that hearing aids benefit CD/HH by protecting their developing linguistic system from further declines associated with hearing loss. A preservation/protection hypothesis predicts that the language trajectory of CD/HH is in initial

decline before hearing aid fitting, and that their language abilities stabilize relative to same aged peers after fitting. It is beyond the scope of our data to differentiate between these two hypotheses. As a group, children in our sample acquired individual skills (as measured by significant Progress Value change) at a rate sufficient to maintain their standing relative to test norms (as indicated by null standard score growth). However, as we are restricted to language assessment at only two time points, it is unclear if the acquisition of skills would continue at a rate sufficient to increase standard score performance with time beyond the data points presented here.

Similarly, work by Tomblin et al. (2015) demonstrated that children who received intervention later than 6 months appeared to “catch up” to their earlier intervened peers, and documented that children receiving intervention before 6 months of age demonstrated stable language performance without significant change in their language performance relative to test norms. The authors posited that either a period of rapid catch-up or protection from effects of severity might explain stable language performance across ages in children receiving intervention before 6 months of age. However, their work examined data collected after hearing aid fitting, thus they were unable to measure language ability prior to fitting. In both cases, examining change in language outcome with respect to either *only* after fitting (Tomblin et al., 2015), or *only* immediately before and after (our data), limits our ability to propose a complete mechanism of hearing aid benefit. Indeed, it may be the case that *both* preservation and catch-up play a role at different points in development. For instance, hearing aid fitting may initially protect the child from further declines in language ability relative to same-aged peers, but the addition of speech-language interventions may facilitate catch-up. These possibilities highlight the importance of additional prospective research: developing a complete understanding of how

language develops prior to hearing aid fitting is necessary for improving identification and remediation of language impairments. Researchers and clinicians need to be able to form realistic expectations of language outcomes for CD/HH in order to identify when they are veering off course.

Although the importance of understanding the role of pre-fitting language in eventual outcomes is most pronounced for CD/HH who are later identified, as is the case for children in our sample, this also necessary to consider for infants born with hearing losses who are rapidly learning about their native language (Kuhl et al., 2008; Maye, Weiss & Aslin, 2008; Maye, Werker, Gerken, 2002; Werker & Tees, 1984). Amplification with hearing aids in infancy is increasingly common within the context of universal newborn hearing screening and EHDI programs. A caveat, however, is that appropriate methodology for the assessment of pre-fitting language abilities in pre-verbal infants is currently clinically challenging. The PLS-4 is currently the only standardized oral language assessment tool that is suitable for assessing children from birth through to 6 years (thus covering the entire age-range of children serviced by the OIHP), and this facilitated our analyses. However, the PLS-4 may not be especially sensitive to subtle differences in pre-linguistic performance in the first year of life, nor is it clear whether the PLS-4 is sufficiently sensitive to detect subtle changes in the developing child's linguistic system in a short time-span. There is some recent work using electroencephalography to identify speech processing differences in infancy related to later vocabulary development (e.g., Kuhl et al., 2008), however these tools are not normed, standardized, or clinically feasible. While we argue that consideration of pre-fitting abilities is important in understanding language outcomes even in infants fit early with hearing aids, we acknowledge that considerable work in assessment tools needs to be done before addressing these issues is possible. In the absence of these tools,

exploration of pre-fitting language ability is best considered in relation to children who are fit at older ages.

A full understanding of hearing aid benefit depends upon understanding how CD/HH acquire language without intervention with hearing aids and how that changes with the introduction of hearing aids at various ages. It is currently unknown what cognitive strategies, if any, CD/HH use to compensate for their sensory deficits prior to being fitted with hearing aids. The ability to adopt compensatory strategies may differentially predict better, or worse, language outcomes for CD/HH and enable early identification of persistent language delays. Similarly, understanding the way in which CD/HH use auditory information prior to fitting may expose malleable factors early in development for these children that can maximize hearing aid benefit. In the case of CD/HH who are fit later, this may include earlier hearing aid fittings, and caregiver training to facilitate language development. For infants who are deaf/hard-of-hearing and are being fit according to JCIH guidelines, earlier hearing aid fittings may not be clinically feasible. In these cases, maximizing language learning may involve caregiver training to provide optimal learning of auditory stimuli to facilitate language development prior to hearing aid fitting. If, as demonstrated here, hearing aids preserve pre-fitting language ability, then maximizing pre-fitting language ability may optimize outcomes. Research into the cognitive processes of CD/HH would provide stakeholders (speech-language pathologists, audiologists, educators, and caregivers) a starting point from which to begin intervening to maximize pre-fitting ability.

Our study also demonstrates the utility of using Progress or Growth Scale Values. Progress Values provided an index of language ability that allowed us to examine language growth that facilitated interpretation of our findings of null standard score changes. These scaling scores, first called W scores, were developed in the 1970s (Woodcock & Dahl, 1971) but are only

recently becoming available in tests of oral language development in the early years. Children in our study did not demonstrate significant standard score change. This lack of change is not evidence that growth did not occur; it is only evidence that the growth was not sufficient to alter children's standing relative to the norming sample (rather than relative to their own performance). In other words, they demonstrated a typical rate of growth in their language skills between assessments. Progress Values are sufficiently sensitive to capture a child's change in her own performance, which, when considered in concert with standard score, is especially informative.

Despite the benefits of using Progress Values, their use in CD/HH language outcome studies has not been adopted; standard scores are currently used for reporting results on standardized language assessments (Tomblin et al., 2015; Tomblin et al., 2014). This is, perhaps, due to the difficulty associated with calculating Progress, or Growth Scale, Values. The charts required to calculate the Progress Values for the PLS-4 were not included in the PLS-4 materials, but rather were later sold separately. However, due to difficulty in sales, the charts were never reprinted and are no longer available. Fortunately, Progress Values, renamed as Growth Scale Values are now being included in publications of child language measures such as the PLS-5 (Zimmerman, Steiner & Pond, 2011). With their increasing availability, we encourage their adoption, in conjunction with measures of relative performance, in the study of language growth and outcomes based on the aforementioned benefits.

As a retrospective study, our data included a number of limitations. First, known predictors of language (e.g., socio-economic status, characteristics of caregiver input, multilingual language environment) were not included in the database and thus were not available for consideration. Similarly, details about each child's communication development intervention (e.g.,

communication mode, type of intervention, frequency) were unknown and may have impacted our findings. Although the decision making for communication modality is multifactorial, surveys of communication modality choices suggest that between 87% and 96% of parents choose speech for either the sole communication modality or as a complement to signed input (e.g., sign language or signed English; Crowe, Fordham, McLeod & Ching 2014; Crowe, McLeod, McKinnon & Ching, 2014; Li, Bain & Steinberg, 2003). In addition, the PLS-4 was not administered to children in the OIHP who were using or being taught signed language as their primary mode of communication. Thus, it is likely that the children in our sample were receiving at least some degree of consistent spoken language input.

Additionally, the children in our sample are not representative of the entire OIHP population. Of the 155 children in the OIHP database who had PLS-4 assessment data entered, only 48 had a PLS-4 assessment prior to their hearing aid fitting. As described in the Methods, the children in our sample were identified and fitted at older ages than the broader OIHP population. Due to the nature of the data contained within the OIHP database, it is unclear whether or not our population differs in additional systematic ways from the broader population. The inclusion of children with various comorbidities in our sample also impacts the generalizability of our results. The language development and response to hearing aid fitting of children in our sample may have been influenced by their comorbidities in addition to their hearing loss. In particular, the growth rates of some children in our sample may have been slowed due to the presence of additional comorbidities, and our data might therefore underestimate the amount of growth that would be observed in a sample of children without comorbidities. Due to the sample specific nature of our work, prospective replication is needed in order to explicate the mechanism by which hearing aids benefit CD/HH.

We had an insufficient sample size to consider the relationship of our predictors across different levels of hearing loss severity, the impact of unilateral versus bilateral loss, as well as indicators of amplification dosage (eg., residual SII) and amount of hearing aid use per day, which have been demonstrated to impact language outcomes and trajectories after fitting (Tomblin et al., 2015). Our finding that severity of hearing loss do not significantly predict language ability post-fitting demonstrates the importance of considering metrics of hearing aid quality (SII) and amplification dosage (rSII) in analyses of language growth trajectories, as well as ability after hearing aid fitting, since BE4PTA did not appear to appropriately capture functional hearing post-fitting. Given the limitations of our study, we are unable to draw definitive conclusions. However, our analyses demonstrate the importance and feasibility of considering pre-fitting language abilities in research in language outcomes for CD/HH. Furthermore, this work contributes a demonstration of the utility of well-maintained EHDI program databases: despite our limitations, our work was conducted in an ecologically valid context. In future iterations of OIHP data management protocols, careful attention to the limitations impacting this study will provide researchers with access to data capable of addressing important theoretical questions in the CD/HH literature.

### **Conclusions**

Our retrospective cohort analysis represents a first attempt at studying the language outcomes of CD/HH in relation to their language ability prior to fitting. Despite limitations that hinder our ability to identify the mechanism of hearing aid benefit, our work illustrates the role of initial language ability as a child-specific factor influencing outcome, and calls for the consideration of initial language ability in future explorations of the outcomes of CD/HH to



clarify how CD/HH use unamplified and amplified input in the development of their linguistic systems.

## Tables

Table 1: *Summary of Participant Characteristics*

Variable	Group 1 (N = 47)			Group 2 (N = 19)		
	<i>n</i>	<i>M</i> (range)	<i>SD</i>	<i>n</i>	<i>M</i> (range)	<i>SD</i>
Comorbidity						
Specified	11			4		
Not-Specified	36			15		
BE4PTA (dB)		47.46 (11.24- 100.00)	22.93		47.45 (11.25 – 100.00)	22.93
Age at Fit (months)		22.62 (3.73 – 60.40)	16.63		15.5 (3.73 – 30.13)	10.48
Age at Pre-Fitting Assessment		21.22 (3.43- 55.10)	15.27		15.9 (4.3- 40.87)	10.58
Age at Post-Fitting Assessment					22.8 (10.43- 46.87)	12.08
Time Between Pre- and Post - Assessment (months)					6.89 (4.63 - 11.27)	2.25
PLS-4 standard score Pre-Fitting						
Auditory Comprehension Scale		89 <sup>a</sup> (50.00- 133.00)	21.62		89.04 (50.00- 133.00) <sup>b</sup>	21.61
Expressive Communication Scale		93.83 (50.00- 131.00)	17.77		93.83 (50.00- 131.00)	17.76
PLS-4 standard score Post-Fitting						
Auditory Comprehension Scale					93(50.00- 124.00) <sup>b</sup>	20.29
Expressive Communication Scale					96.74 (60.00- 125.00)	18.59

## PLS-4 Progress Value Pre-Fitting

Auditory Comprehension Scale	413.87 (212.00-583.00)	87.31	391.63 (212.00-511.00)	80.25
Expressive Communication Scale	401.05 (313.00-513.00)	54.29	401 (313.00-513.00)	54.29

## PLS-4 Progress Value Post-Fitting

Auditory Comprehension Scale			437.06 (348.00-561.00)	60.9
Expressive Communication Scale			439 (362.00-563.00)	52.87

Note. BE4PTA= Better Ear 4-Pure Tone Average. PLS-4 = Preschool Language Scale, 4<sup>th</sup> edition.

<sup>a</sup>  $N = 46$

<sup>b</sup>  $N = 18$

Table 2: Hierarchical Regression Model of Language Progress Value Prior to Hearing Aid Fitting

Predictor	PLS-4 Progress Value Pre-Fitting					
	Auditory Comprehension			Expressive Communication		
	$R^2$ (adj)	$\Delta R^2$	b	$R^2$ (adj)	$\Delta R^2$	b
Model 1	0.62***			0.69***		
Age at assessment			4.54***			4.33
Model 2	0.84*	0.22***		0.85***	0.16**	
Age at assessment			6.81**			5.62**
BE4PTA			-1.66*			-0.66
Age*BE4PTA			-0.02			-0.03

Note. PLS-4 = Preschool Language Scale, 4<sup>th</sup> edition. Auditory Comprehension Scale,  $N = 46$ . Expressive Communication Scale,  $N = 47$ . BE4PTA= Better Ear 4-Pure Tone Average.

\* $p < .05$ . \*\* $p < .01$ . \*\*\* $p < .001$ .

Table 3: *Hierarchical Regression Model of Language standard score Prior to Hearing Aid Fitting*

Predictor	PLS-4 standard score Pre-Fitting			
	Auditory Comprehension		Expressive Communication	
	$R^2$ (adj)	b	$R^2$ (adj)	b
Model 1	0.31***		0.15**	
BE4PTA		-0.54***		-0.32**

Note. PLS-4 = Preschool Language Scale, 4<sup>th</sup> edition. Auditory Comprehension Scale,  $N = 46$ . Expressive Communication Scale,  $N = 47$ . BE4PTA= Better Ear 4-Pure Tone Average.

\* $p < .05$ . \*\* $p < .01$ . \*\*\* $p < .001$ .

Table 4: *Hierarchical Regression Model of Change in Language After Hearing Aid Fitting*

Predictor	PLS-4 Progress Value Difference Score					
	Auditory Comprehension			Expressive Communication		
	$R^2$ (adj)	$\Delta R^2$	b	$R^2$ (adj)	$\Delta R^2$	b
Model 1	0.57***			0.06		
PLS-4 Progress Value Pre-Fitting			-0.29			-0.13
Model 2	0.73***	0.16*				
PLS-4 Progress Value Pre-Fitting			0.14			
BE4PTA			2.56**			
Progress Value*BE4PTA			-0.01*			

Note. PLS-4 = Preschool Language Scale, 4<sup>th</sup> edition. Auditory Comprehension Scale,  $N = 19$ . Expressive Communication Scale,  $N = 20$ . BE4PTA= Better Ear 4-Pure Tone Average. Outcome scores were standardized residuals of PLS-4 Progress Values removing the variance due to time between the pre-fitting and post-fitting assessments.

\* $p < .05$ . \*\* $p < .01$ . \*\*\* $p < .001$ .

Table 5: *Hierarchical Regression Model of Language Progress Value After Hearing Aid Fitting*

Predictor	PLS-4 Progress Value Post-Fitting					
	Auditory Comprehension			Expressive Communication		
	$R^2$ (adj)	$\Delta R^2$	b	$R^2$ (adj)	$\Delta R^2$	b

Model 1	0.16			0.11	
BE4PTA			-1.29		-1
Model 2	0.91***	0.75***		0.84***	0.73***
BE4PTA			0.46		0.12
PLS-4 Progress Value Pre-Fitting			0.78***		0.93***

Note. PLS-4 = Preschool Language Scale, 4<sup>th</sup> edition. Auditory Comprehension Scale, N = 19. Expressive Communication Scale, N = 20. BE4PTA= Better Ear 4-Pure Tone Average. \*\*\*p < .001.

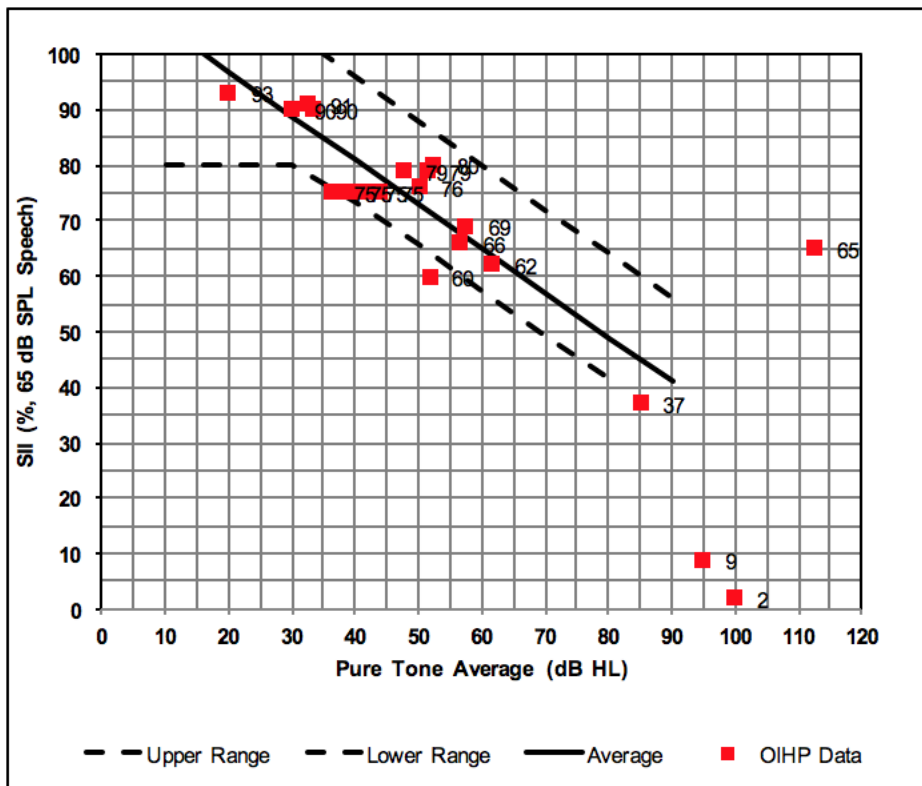
Table 6: Hierarchical Regression Model of Language standard score After Hearing Aid Fitting

Predictor	PLS-4 standard score Post-Fitting					
	Auditory Comprehension			Expressive Communication		
	R <sup>2</sup> (adj)	ΔR <sup>2</sup>	b	R <sup>2</sup> (adj)	ΔR <sup>2</sup>	b
Model 1	0.44***			0.14		
BE4PTA			-0.64**			-0.38
Model 2	0.57**	0.13*		0.32*	0.18*	
BE4PTA			-0.24			-0.11
PLS-4 standard score Pre-Fitting			0.55*			0.65*

Note. PLS-4 = Preschool Language Scale, 4<sup>th</sup> edition. Auditory Comprehension Scale, N = 17. Expressive Communication Scale, N = 18. BE4PTA= Better Ear 4-Pure Tone Average. \*p < .05. \*\*p < .01. \*\*\*p < .001.

Supplemental Materials

Supplemental Figure 1: Hearing Aid Speech Intelligibility Index (SII) Values (N=19) compared to Bagatto et al., 2011 normative data



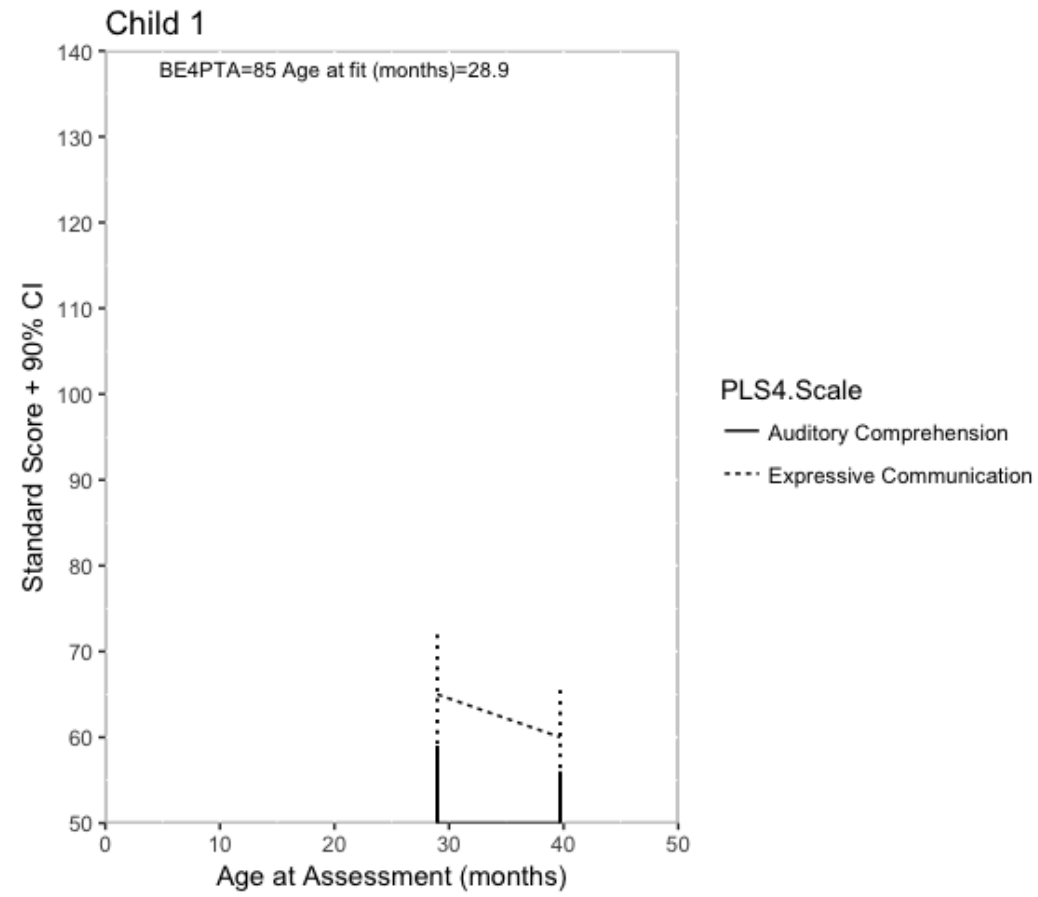
Supplemental Figure 2: Summary of Supplemental Figures

Subject Number	Audiological Characteristics		First Assessment Data		Second Assessment Data	
	BE4PTA	Age at Fit (months)	Auditory Comprehension (90% CI)	Expressive Communication (90% CI)	Auditory Comprehension (90% CI)	Expressive Communication (90% CI)
Child 1	85	28.9	50 (50 – 59)	65 (58 – 72)	50 (50 – 56)	60 (54 – 66)
Child 2	25	26.96	102 (94 – 110)	91 (84 – 98)	85 (79 – 91)	88 (82 – 94)
Child 3	38.33	20.07	120 (112 – 128)	96 (89 – 103)	123 (115 – 131)	123 (116 – 130)
Child 4	50	22.73	84 (76 – 92)	87 (80 – 94)	85 (78 – 92)	89 (82 – 96)
Child 5	56.67	20.97	99 (86 – 112)	108 (99 – 117)	87 (49 – 95)	79 (72 – 86)
Child 6	52.5	39.13	89 (83 – 95)	71 (65 – 77)	83 (76 – 90)	74 (68 – 80)

Child 7	41.25	19.73	81	96 (89 – 103)	/	79 (72 – 86)
Child 8	48.75	29.67	98 (91 – 105)	102 (95 – 109)	119 (113 – 125)	125 (119 – 131)
Child 9	51.25	6.73	101 (90 – 112)	99 (91 – 107)	90 (77 – 103)	87 (78 – 96)
Child 10	33.33	6.1	115 (104 – 126)	118 (110 – 126)	117 (104 – 130)	111 (102 – 120)
Child 11	51.67	13.13	73 (60 – 86)	83 (74 – 92)	99 (86 – 112)	97 (88 – 106)
Child 12	25	11.27	109 (79 – 107)	131 (118 – 144)	109 (96 – 122)	108 (99 – 117)
Child 13	42.5	4.33	119 (108 – 130)	106 (94 – 118)	97 (83 – 111)	118 (105 – 131)
Child 14	56.67	3.73	63 (52 – 74)	106 (94 – 118)	69 (55 – 83)	98 (85 – 111)
Child 15	36.25	4.87	101 (90 – 112)	107 (99 – 115)	103 (89 – 117)	118 (105 – 131)
Child 16	32.5	11.43	99 (86 – 112)	97 (88 – 106)	124 (111 – 137)	115 (106 – 124)
Child 17	43.33	13.7	71 (58 - 84)	87 (78 – 96)	86 (78 – 94)	86 (79 – 93)
Child 18	95	5.7	63 (52 – 74)	77 (69 – 85)	76 (62 – 90)	80 (67 – 93)
Child 19	100	5.3	59 (50 – 70)	91 (79 – 103)	72 (58 – 86)	103 (90 – 116)

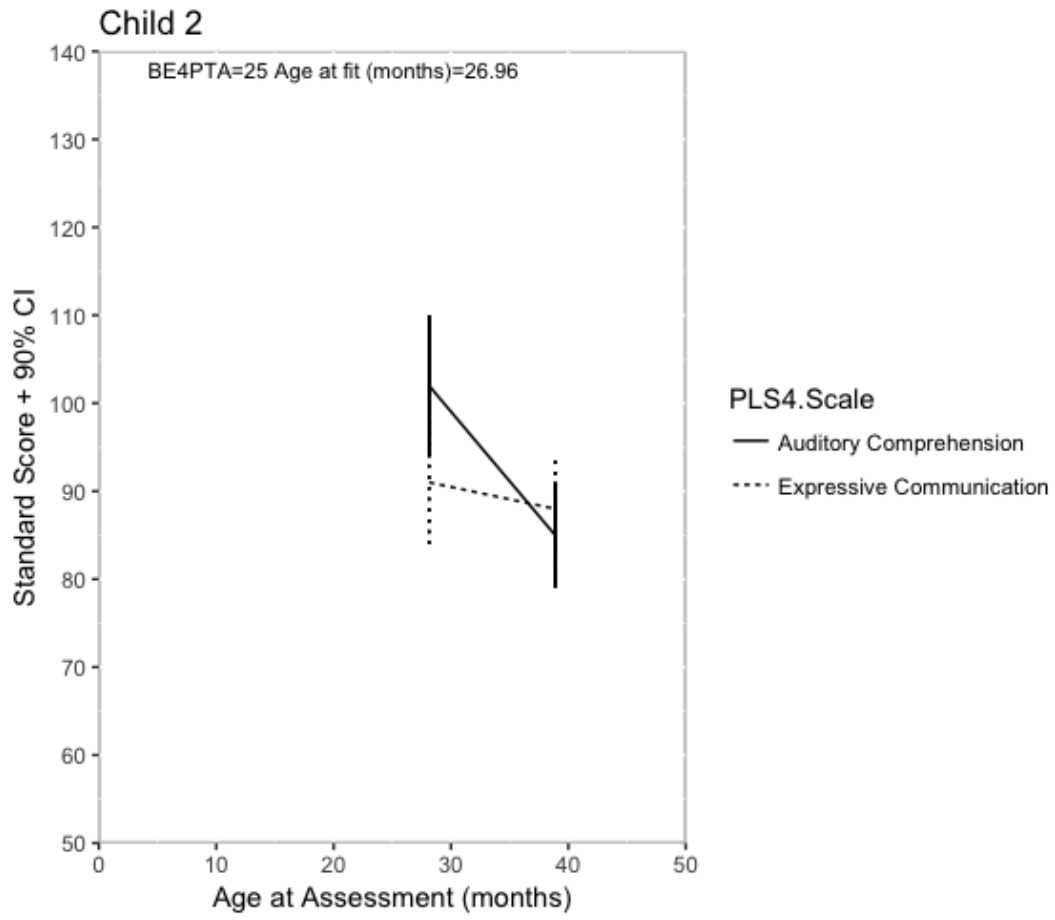
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Supplemental Figure 3

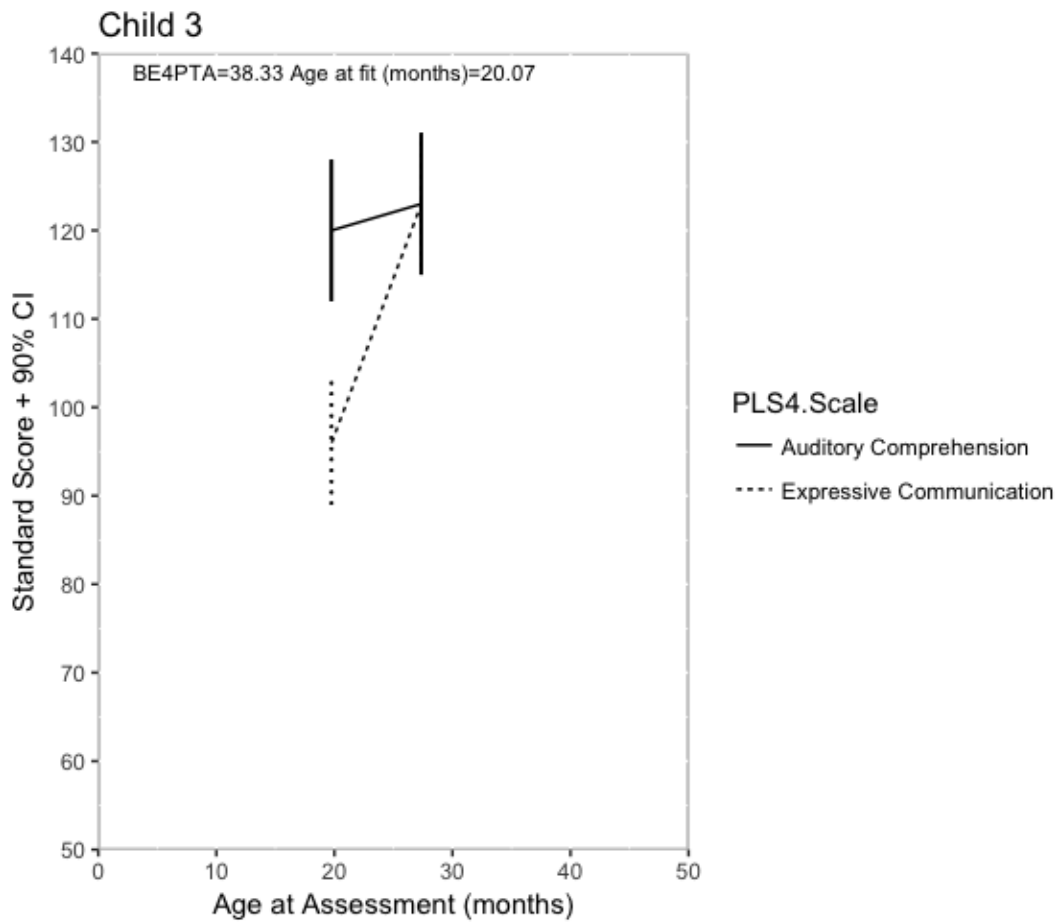




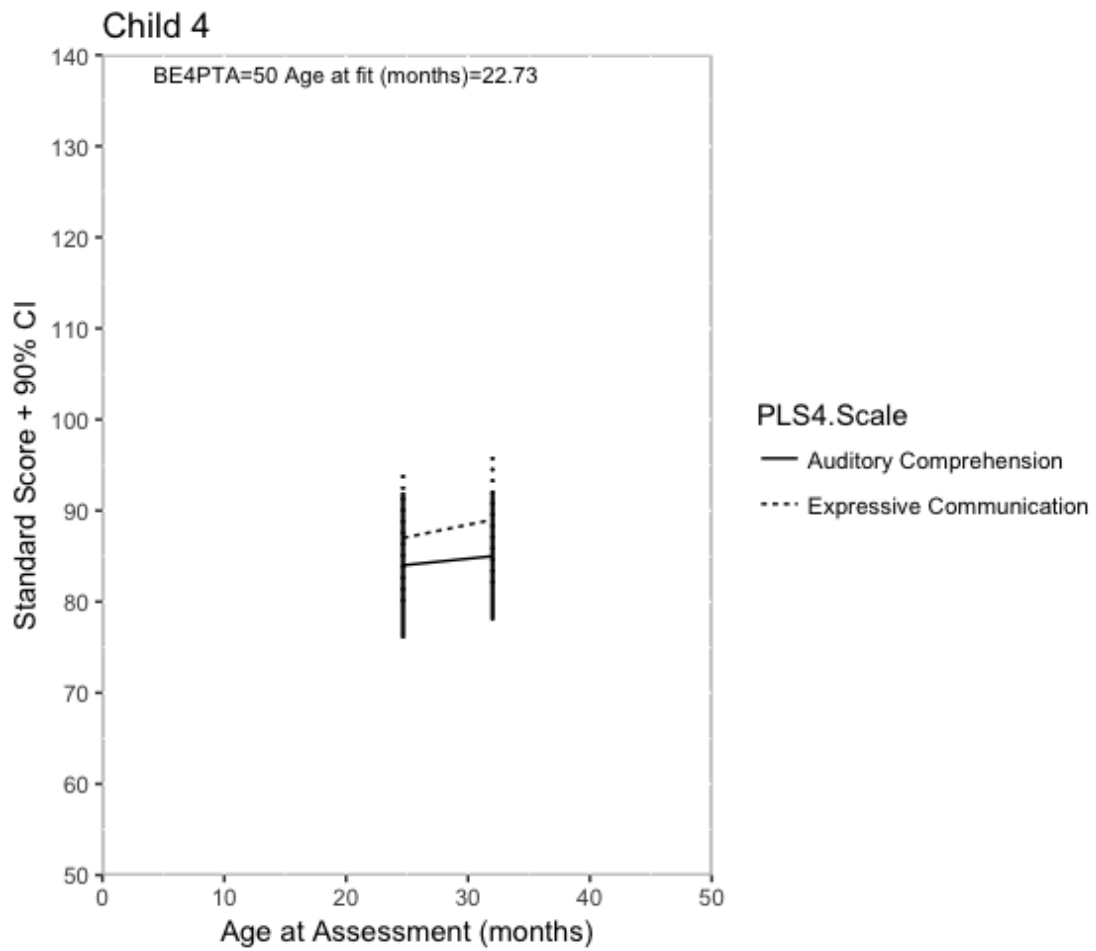
Supplemental Figure 4



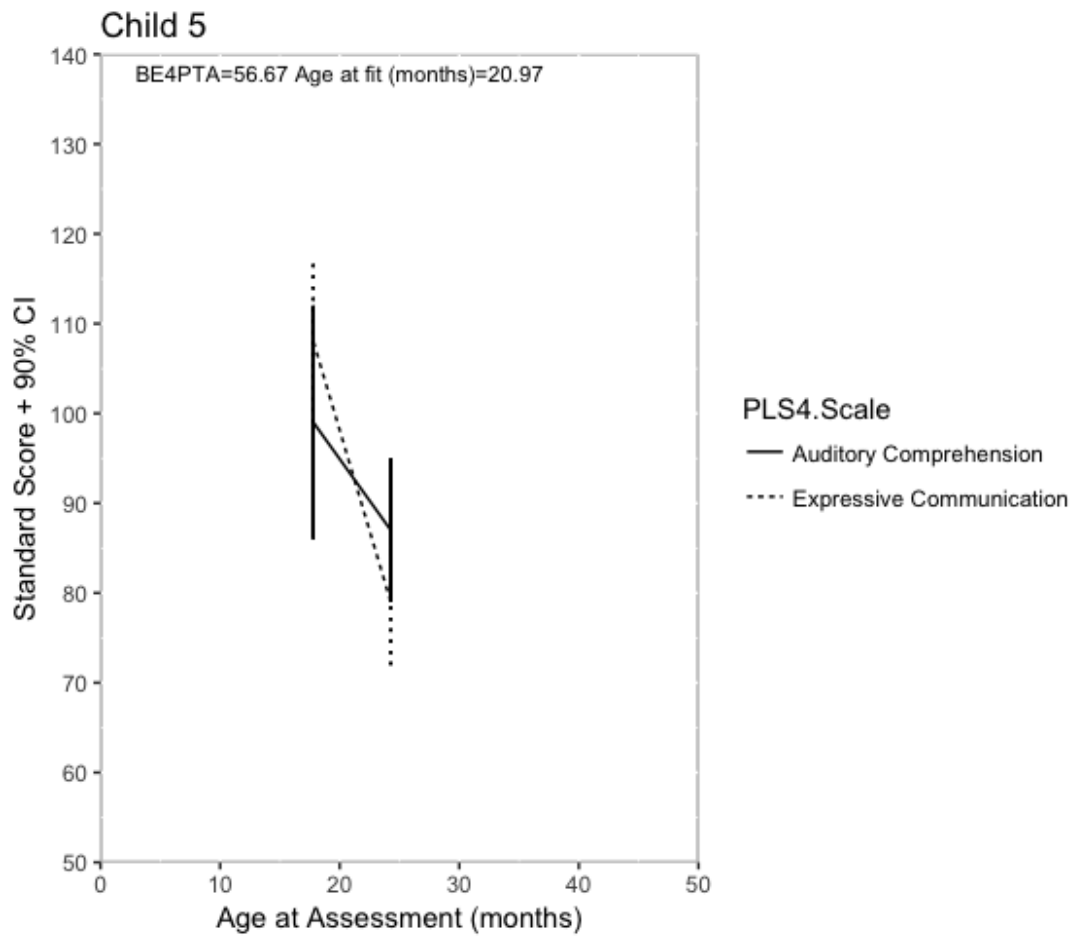
Supplemental Figure 5



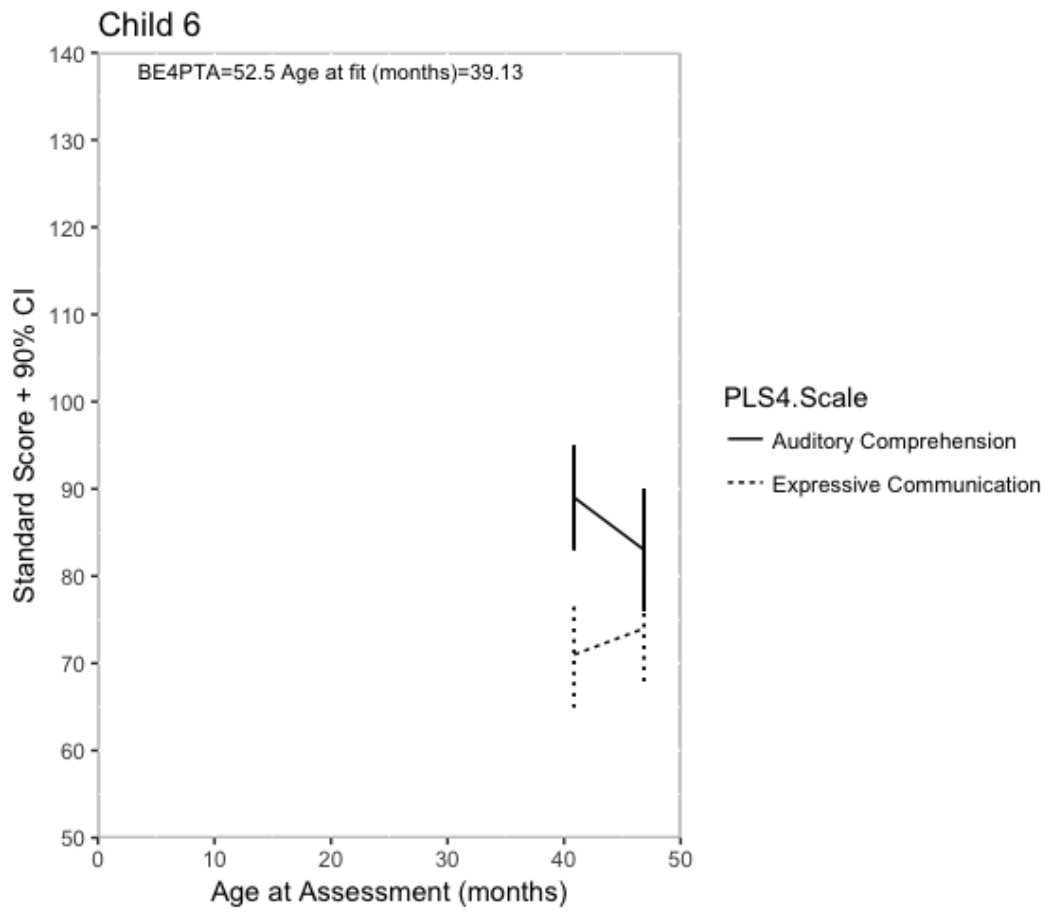
Supplemental Figure 6



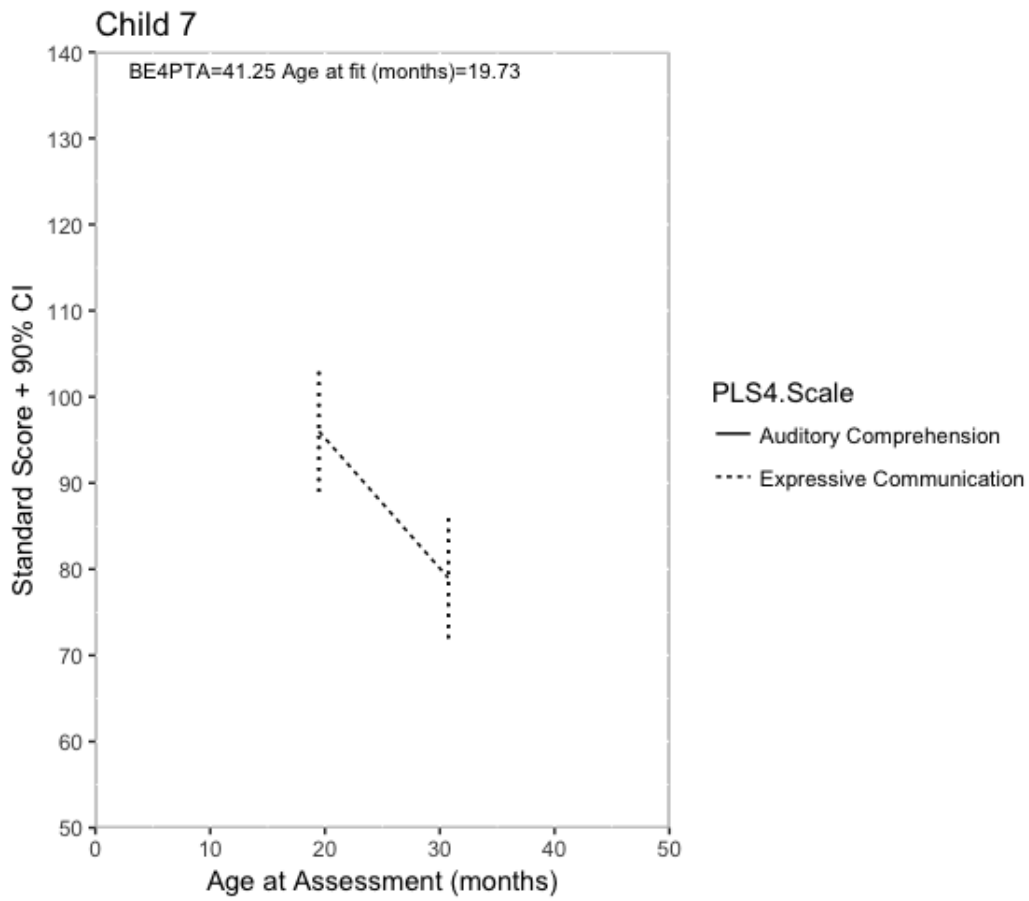
Supplemental Figure 7



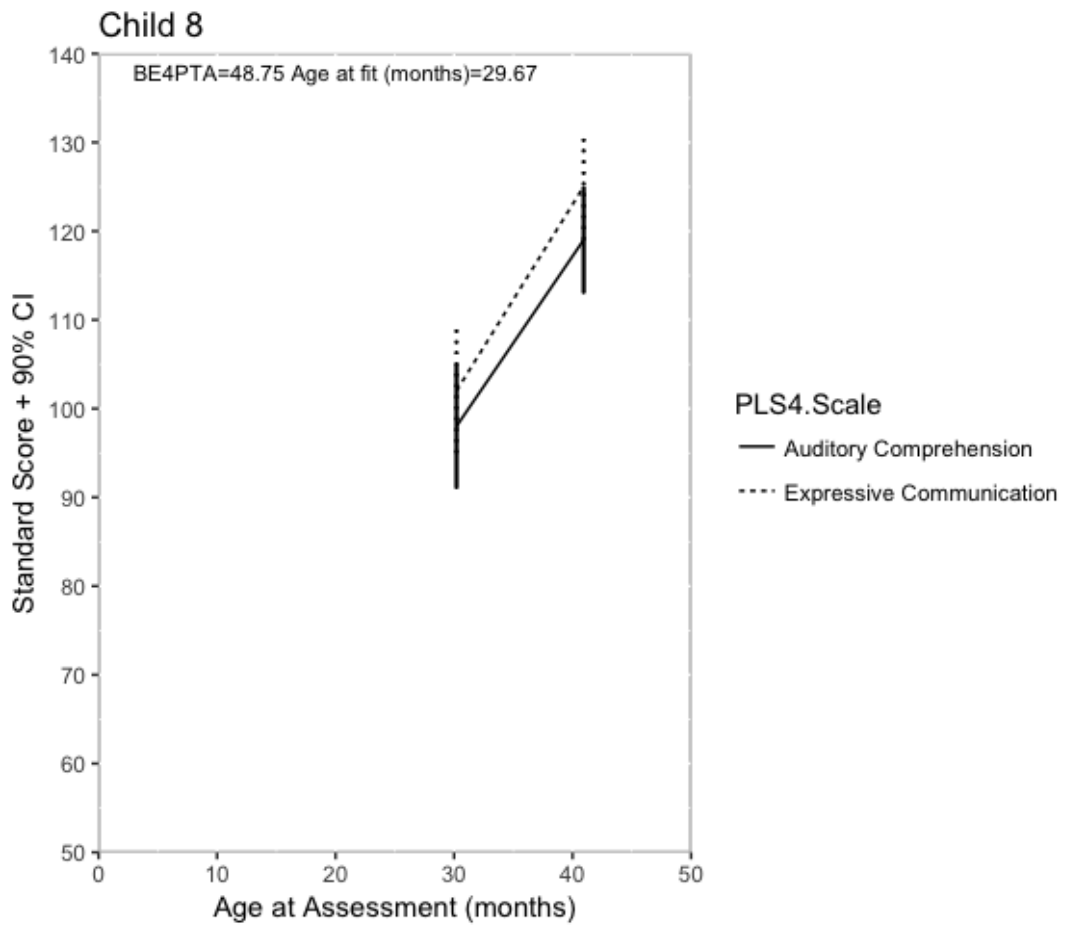
Supplemental Figure 8



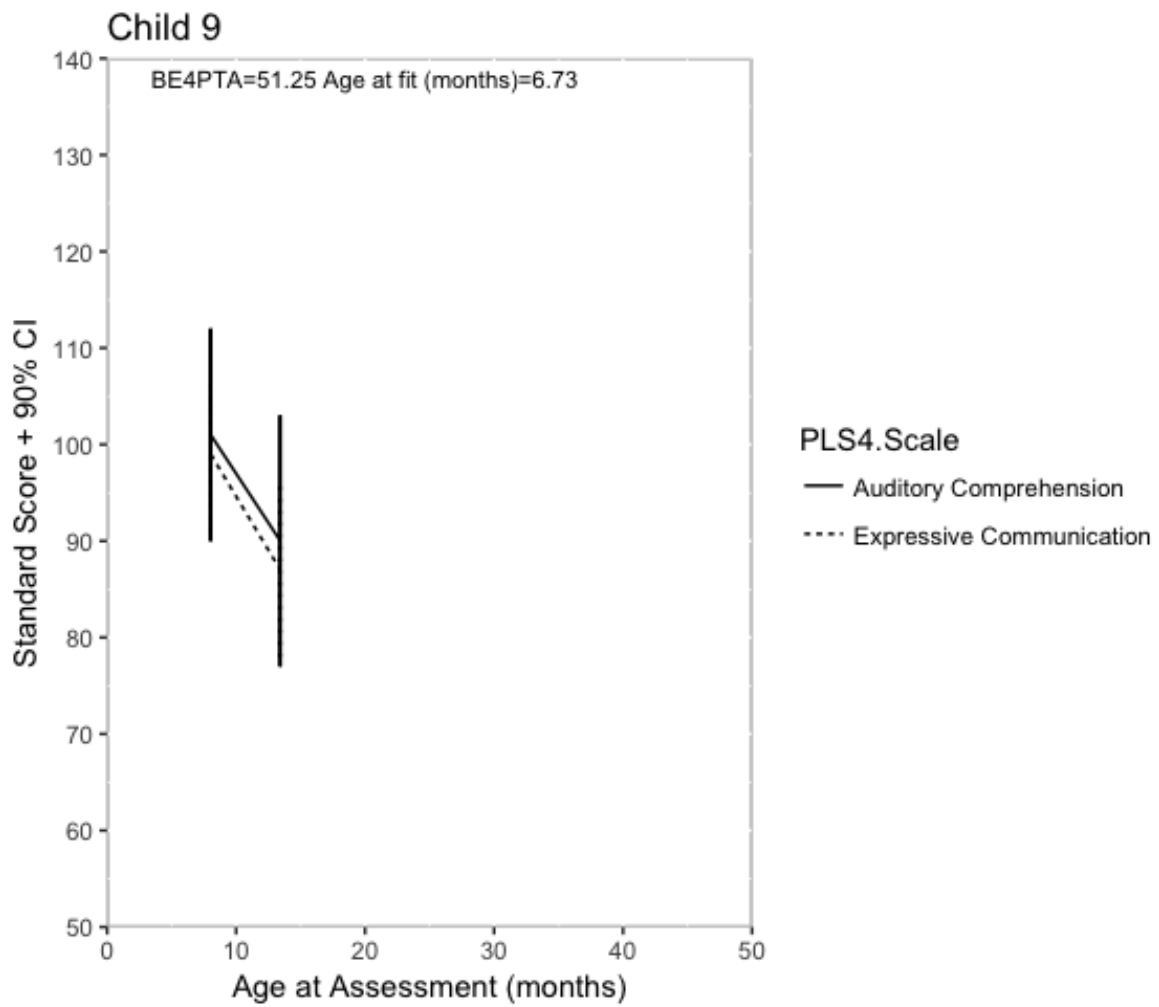
Supplemental Figure 9



Supplemental Figure 10

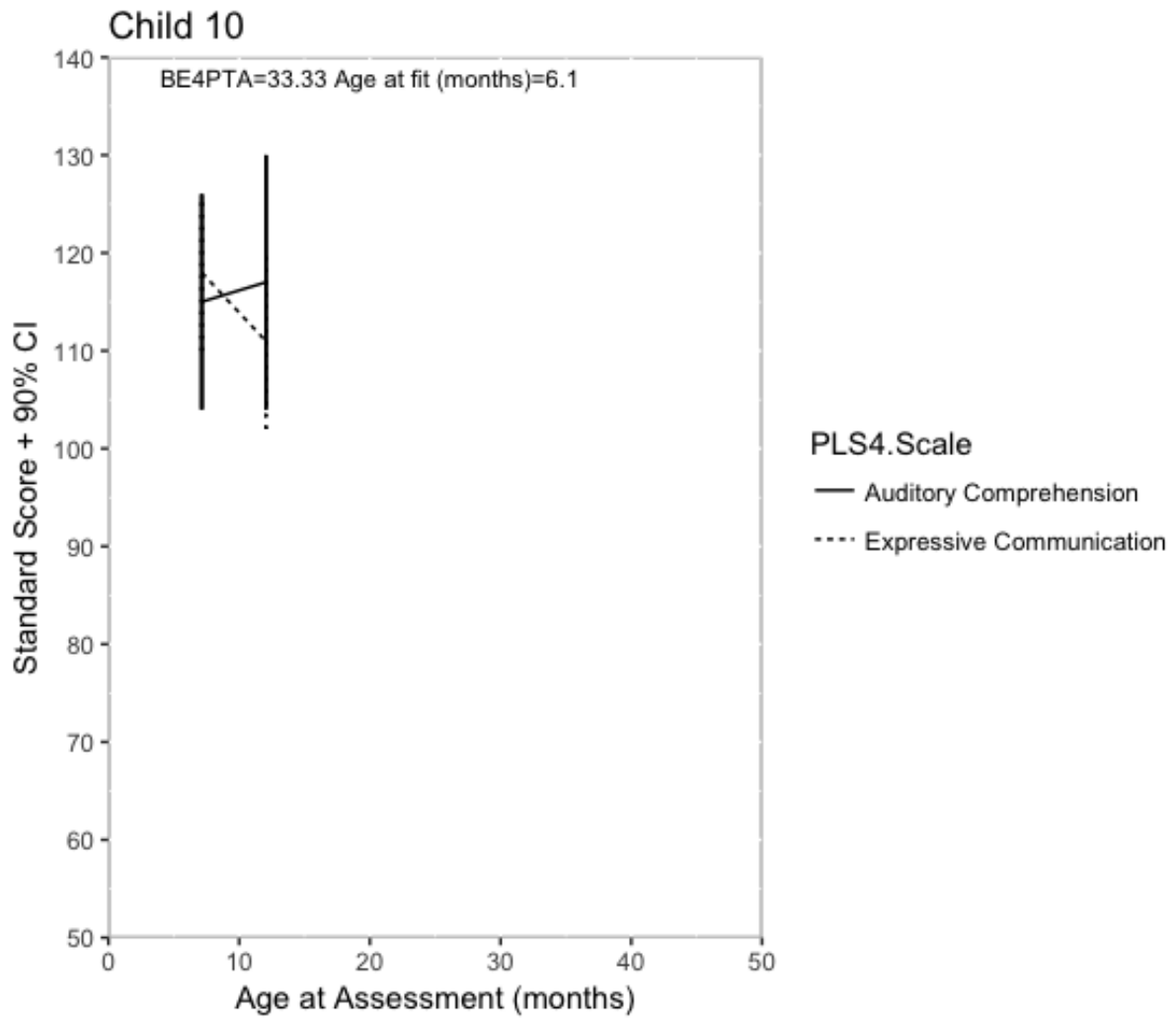


Supplemental Figure 11

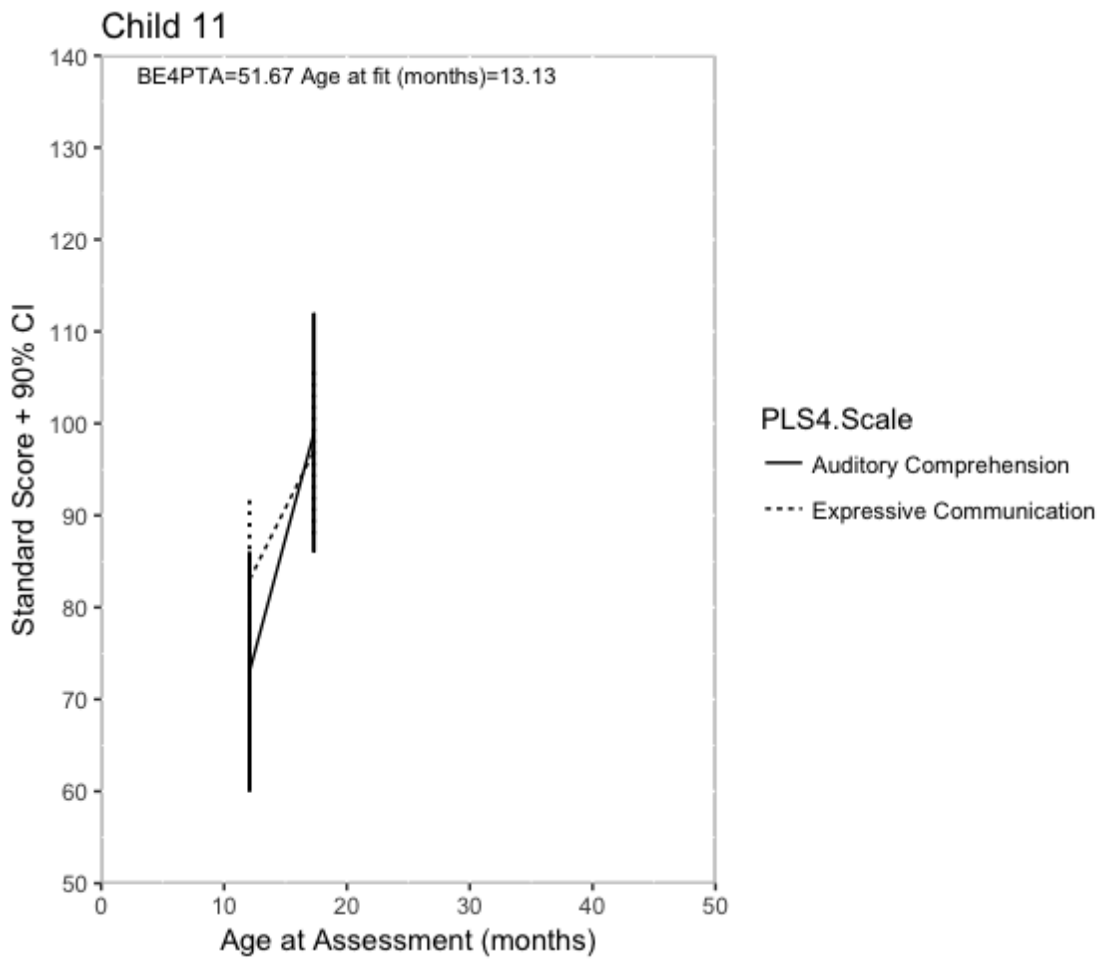




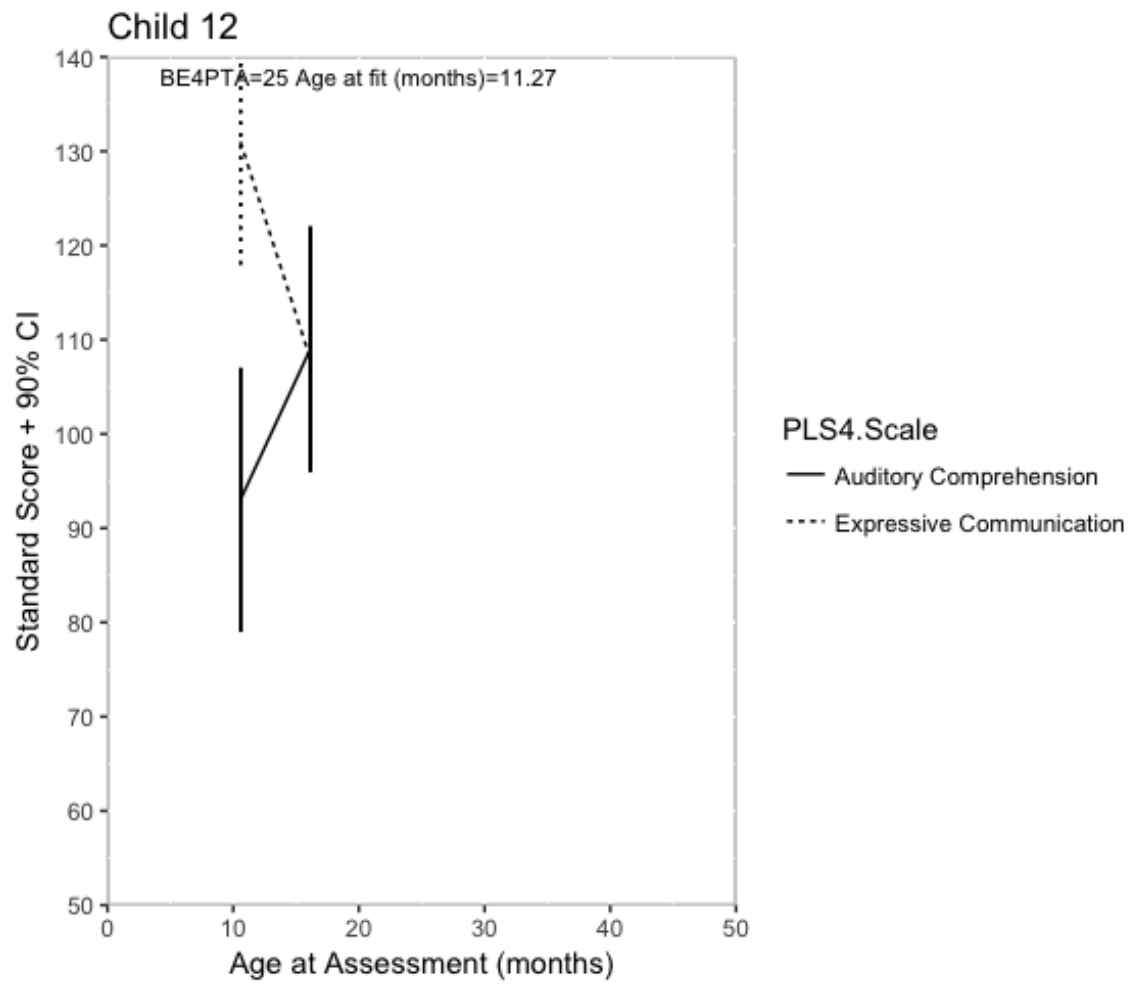
Supplemental Figure 12



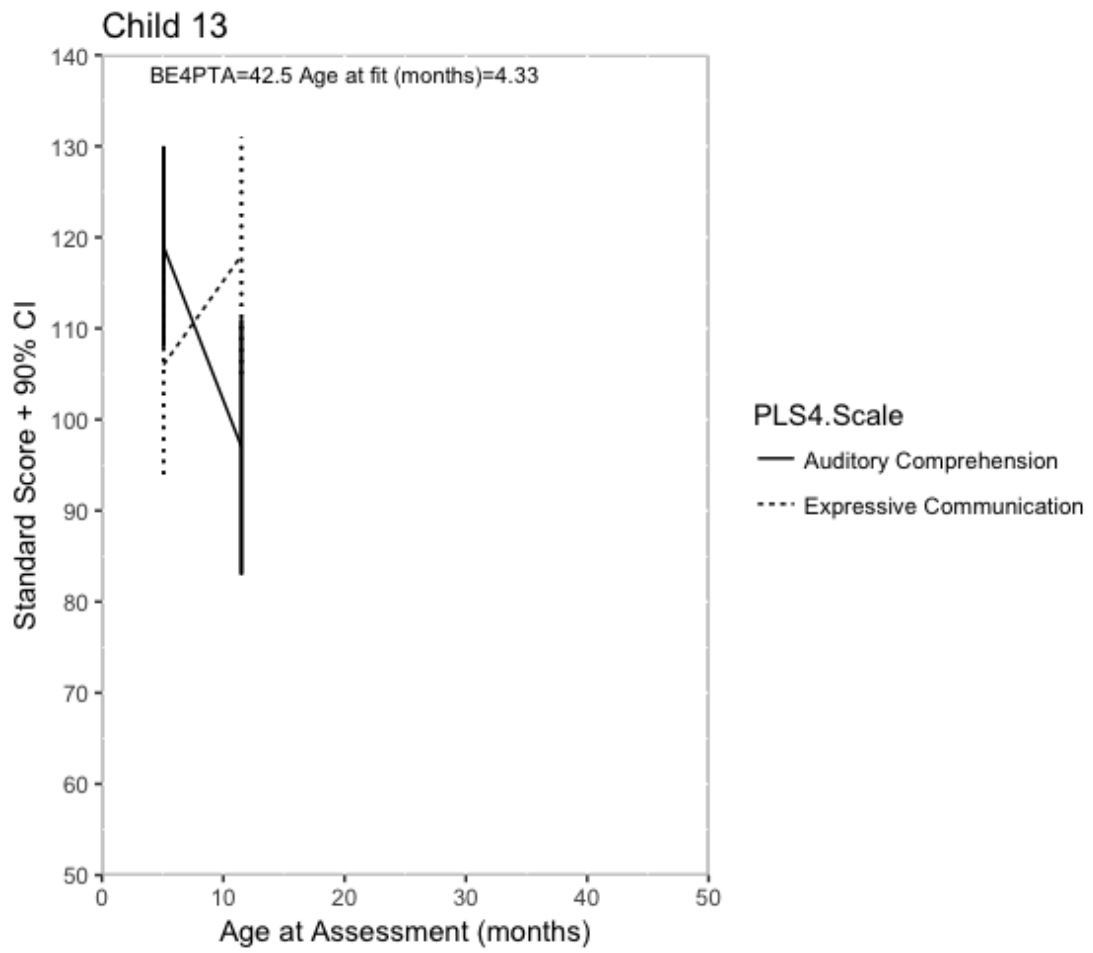
Supplemental Figure 13



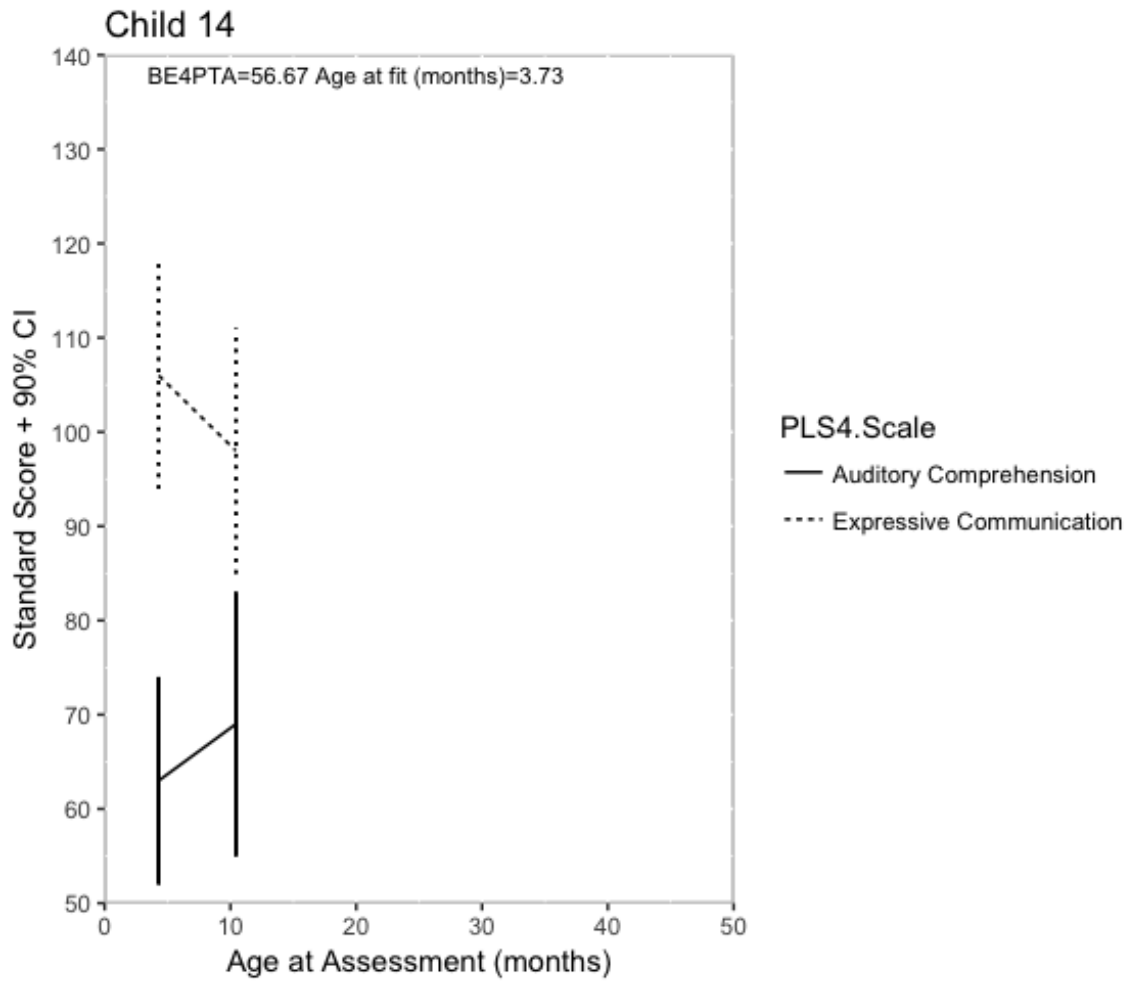
Supplemental Figure 14



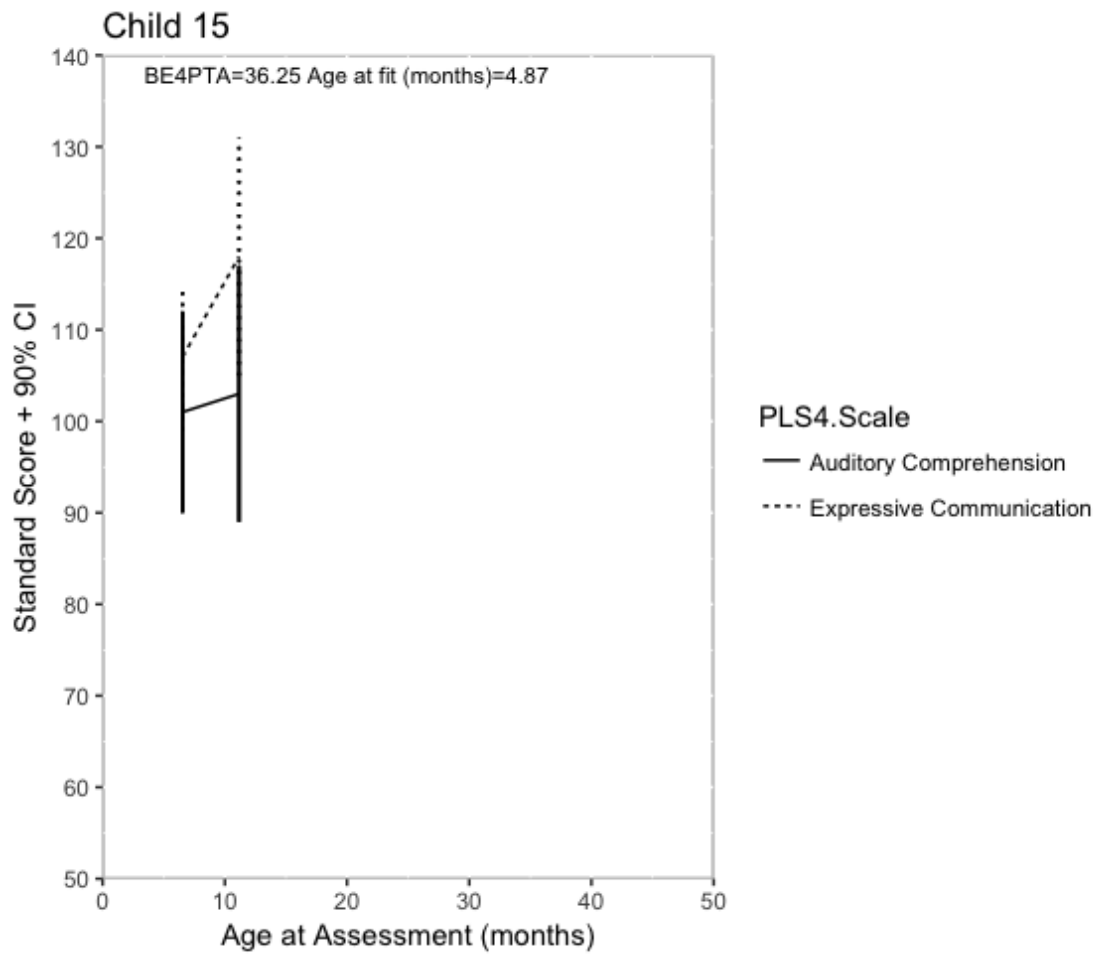
Supplemental Figure 15



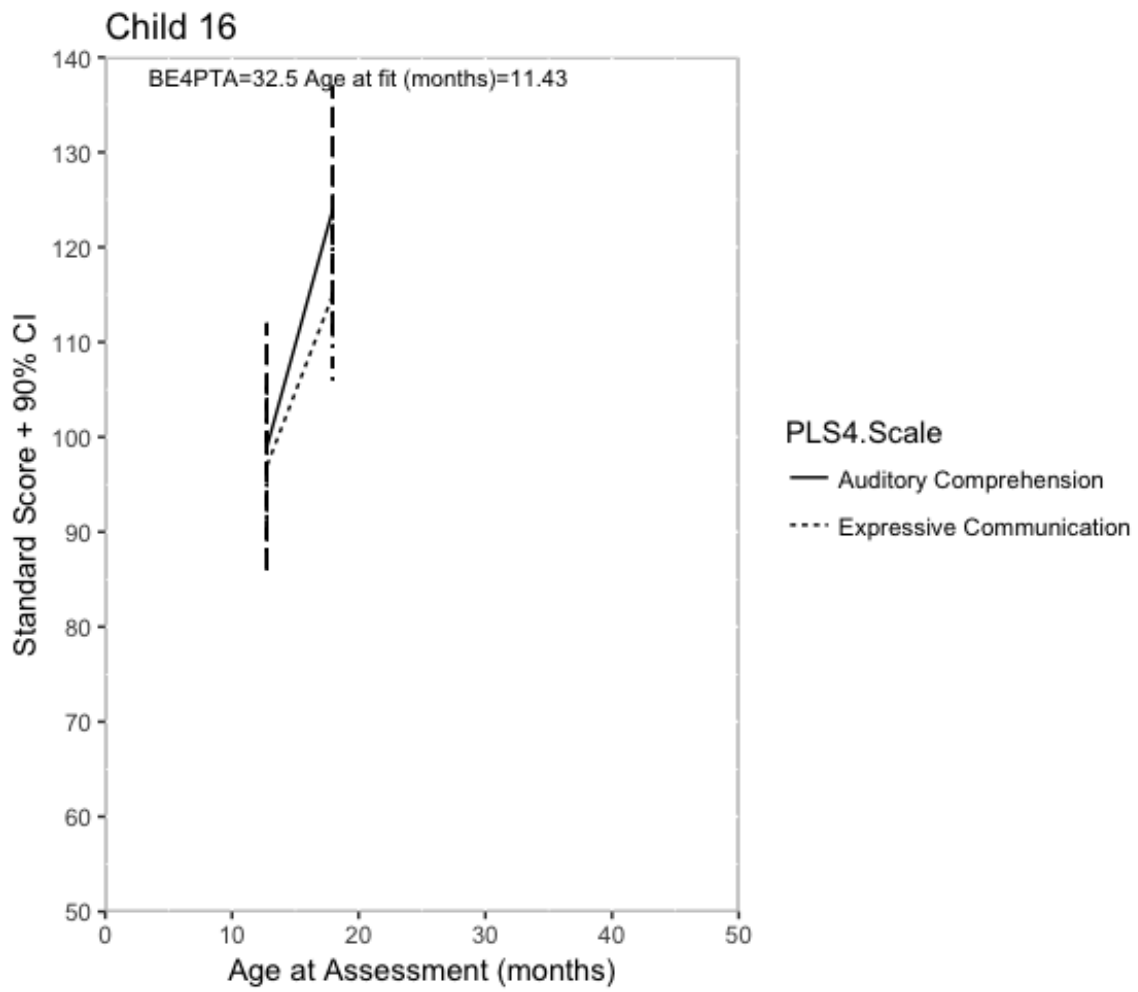
Supplemental Figure 16



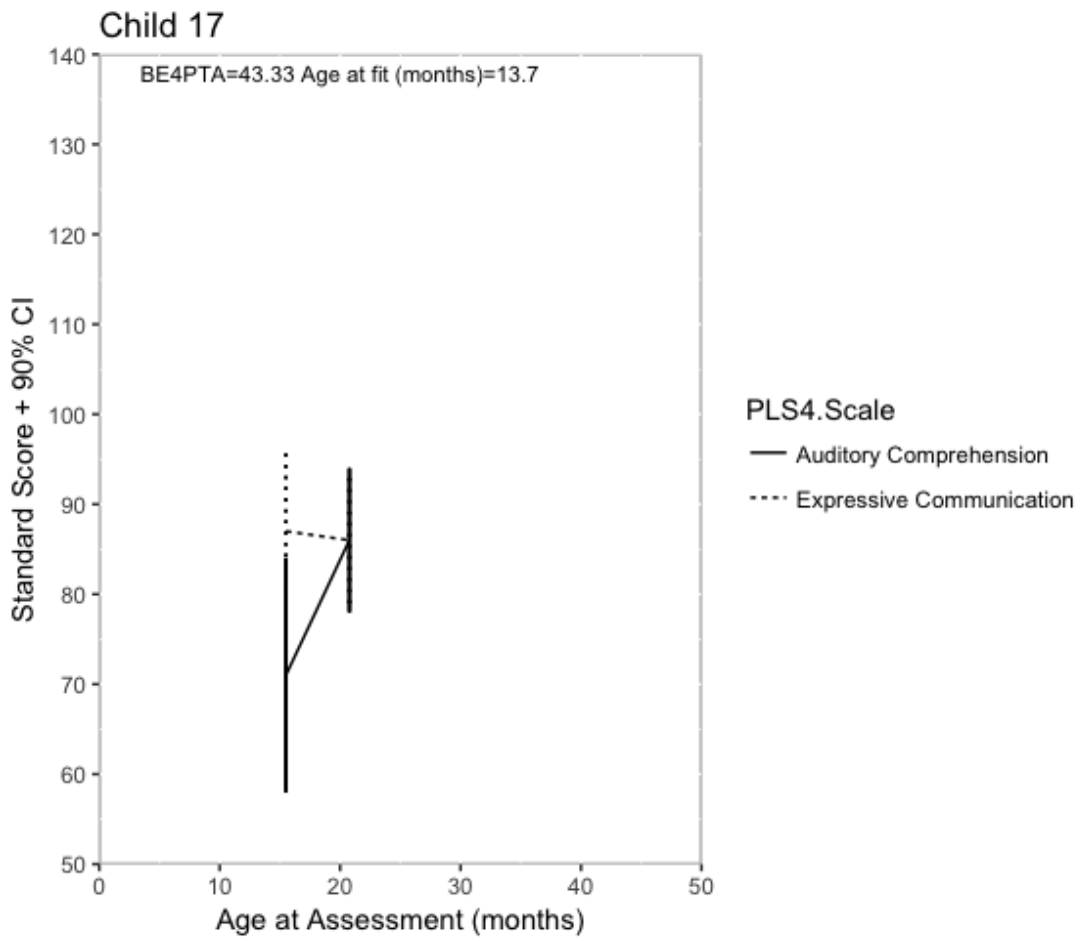
Supplemental Figure 17



Supplemental Figure 18

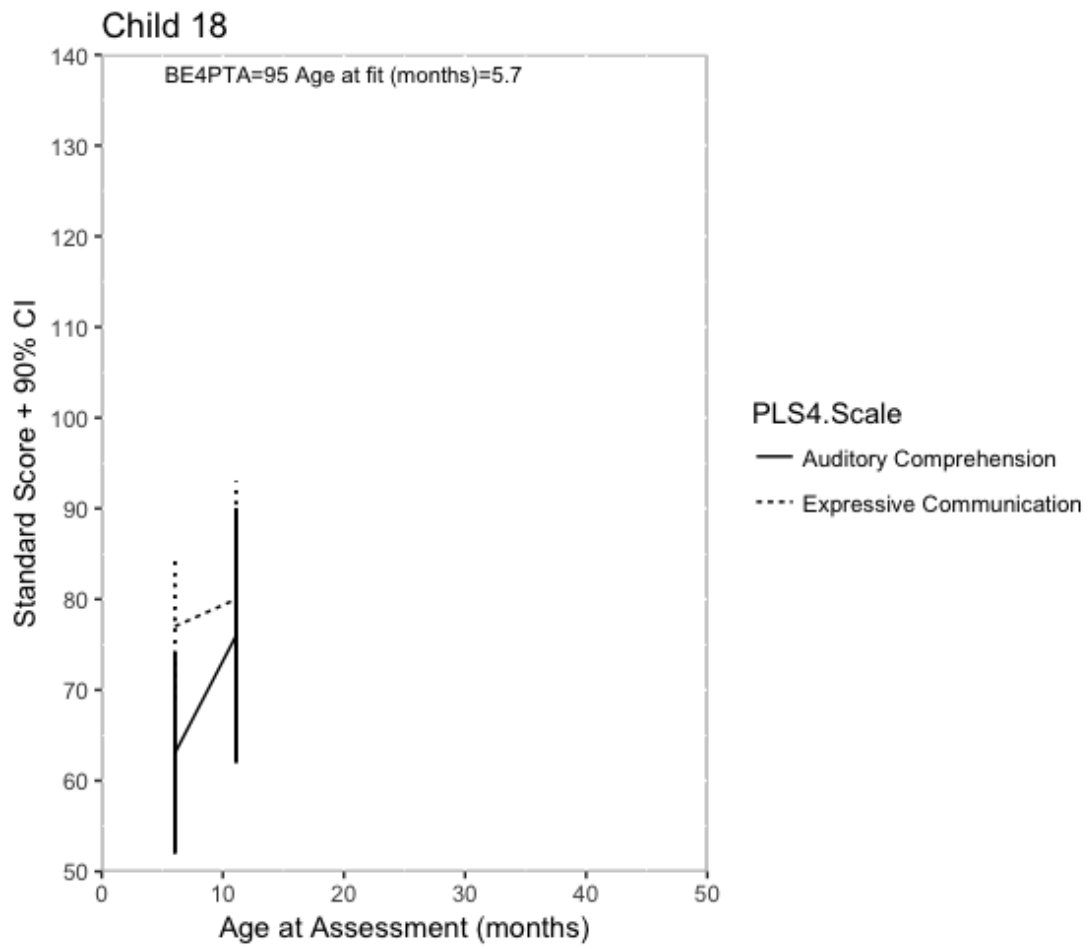


Supplemental Figure 19

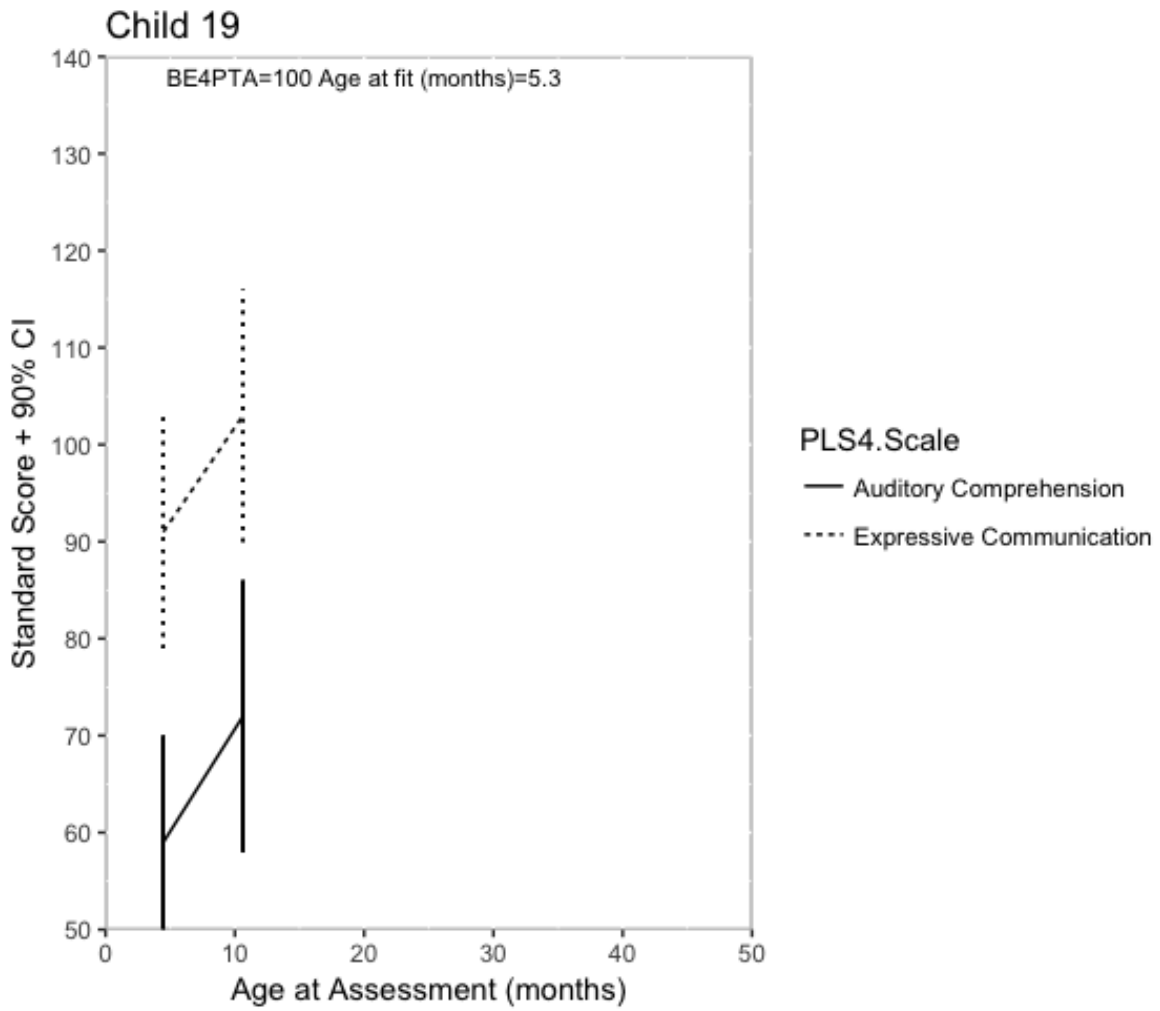




Supplemental Figure 20



Supplemental Figure 21



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