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Comparison of Developmental Trajectories for Auditory Cortex Development and Language

Development in Children with Hearing Loss With and Without Additional Disabilities

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Comparison of Developmental Trajectories for Auditory Cortex Development and Language Development in Children with Hearing Loss With and Without Additional Disabilities.

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Abstract

Objectives: The goal of this study was to explore if there was a difference in the developmental trajectories for auditory cortical development and language development for children who have hearing loss and additional disabilities compared to children with only hearing loss.

Methods: The Preschool Language Scales 5th Edition (PLS-5) was used to assess language skills and the P1 CAEP biomarker was recorded to assess cortical auditory development in sixty-six children with hearing loss who were enrolled in a study of language therapy delivered over a period of six months. Twenty-six children with only hearing loss constituted the control group and forty children formed the group with hearing loss and additional disabilities.

Results: There were no significant differences in P1 latencies between the groups at baseline or follow-up. In addition, PLS-5 Total Language Score, PLS-5 Auditory Comprehension Score, or PLS-5 Expressive Communication Score at baseline or follow-up were not significantly different between the two groups.

Conclusions: Our main finding is that there are no significant differences in auditory cortical development (as measured by P1 latencies) or language development (as measured by PLS-5 scores) between the control group of children with hearing loss and the group of children with hearing loss and additional disabilities. We also found both groups to be equivalent in terms of the proportions of children who showed a change from abnormal to normal cortical development (as measured by P1 latency) from baseline to follow-up.

Introduction

Children with hearing loss are likely to have additional disabilities, in fact, 20-40% of deaf or hard of hearing children have an additional disability (Cupples et al., 2018; Gallaudet Research Institute, 2011; Picard, 2004, Sharma, et al., 2013; Eisenberg, 2013; Birman, Elliott, & Gibson, 2012). However, these children were not always seen as good candidates for cochlear implants (CIs), and thus, have not always been included in research studies (Sharma et al., 2013). At present, there is a significant increase in children with hearing loss and additional disabilities receiving CIs warranting the need to study this population (Sharma et al., 2013). Studies have found children with hearing loss and additional disabilities can benefit from CIs but do so at a slower pace and to a lesser degree compared with children with hearing loss and no additional disabilities (Cupples et al., 2018; Mesallam, Yousef, & Almasaad, 2019).

Cupples et al. (2018) explored language and speech outcomes of children with hearing loss and additional disabilities. Children aged 5 years with hearing loss and multiple types of additional disabilities were evaluated on receptive and expressive language skills using direct assessment and caregiver report. Better language outcomes were associated with milder hearing loss, use of oral communication, higher levels of cognitive ability and maternal education, and earlier device fitting. This finding emphasizes the importance of early device fitting for children with hearing loss and additional disabilities, (Cupples et al., 2018).

Cortical auditory evoked potential (CAEP) P1 latency biomarker responses can be used to infer the maturity of the central auditory pathways and auditory cortex in deaf children by comparing their P1 latencies to age-normative data (Sharma et al., 2013; Eggermont, 1988; Wunderlich, Cone-Wesson, & Sheppard, 2006; Sharma, Dorman, & Spahr, 2002; Pang & Taylor, 2000; Ponton et al., 2000; Gilley et al., 2005). Thus, allowing analysis of normal versus abnormal auditory cortical maturation (Sharma, Dorman, and Spahr, 2002). In a study of 245 congenitally deaf children with CIs fit after various periods of auditory deprivation, it was shown that with less than 3.5 years of auditory deprivation before implantation P1 latencies matched age appropriate normal P1 latencies after 3-6 months of electrical stimulation from cochlear implants. However, children who have 7 or more years of auditory deprivation before implantation, never developed normal P1 latencies (Dorman et al., 2007).

Sharma et al. (2013) explored the clinical relevance of the P1 biomarker for evaluating cortical maturation in hearing impaired children with additional disabilities. They recorded CAEPs in five children with hearing loss and who had received CIs. Three of these children had additional disabilities which included CHARGE (coloboma of the eye, heart defects, atresia of the choanae, restriction of growth and development, and ear abnormalities and deafness), ANSD (Auditory Neuropathy Spectrum Disorder), and Pallister-Killian Syndrome. They found that for some children with multiple disabilities that received early intervention, central auditory development can proceed in a manner comparable to that seen for hearing-impaired children without multiple disabilities. A main finding was that the P1 biomarker is useful in assessing central auditory maturation in patients with additional disabilities. The authors concluded that the combination of hearing loss and additional disabilities poses unique challenges for deciding on appropriate intervention for these children. However, the P1 biomarker was able to provide valuable information regarding the effectiveness of different intervention strategies for children with hearing loss and additional disabilities. (Sharma et al., 2013). However, a limitation of the Sharma et al., (2013) study was that it did not have a large enough sample or examine language development in the children which is considered the gold standard for making intervention decisions. It is important for parents of these children to receive informed advice regarding development of the central auditory system and language acquisition to make decisions regarding early intervention and management using devices such as hearing aids or CIs (Cupples et al., 2018; Sharma et al., 2002; Sharma et al., 2002; Sharma et al., 2007; Dorman et al., 2007; Eggermont & Ponton, 2003).

The goal of this study was to explore if there was a difference in the developmental trajectories for auditory cortical development and language development for children who have hearing loss and additional disabilities compared to children with only hearing loss. To this end, We compared P1 biomarker latencies and performance on tests of language development in children with multiple disabilities and hearing loss to those with only hearing loss.

Methods

Participants

A power analysis with power ($1 - \beta$) set at 0.80 and α equal to 0.05 (two-tailed) indicated a sample size of 39 children would be needed to detect a small effect size ($d=0.2$). A total of 66 English speaking children between the ages of 13 months and 78 months were included in this study. All the children presented with moderately-severe to profound sensorineural hearing loss which was treated with either hearing aids or CIs. The children were divided into two groups based on the presence of another reported disability in addition to hearing loss (see table 1 some additional disabilities that were captured in this study). 40 children were classified as having multiple disabilities, while 26 children were found to have only hearing loss and were used as our control group. Independent samples t-tests were conducted to confirm the groups were not significantly different with reference to age ($t(63)= 1.14, p=0.258$), with the average age in the control group being 47 months old and the average age in the multiple disabilities group being 42 months old. Two proportion z-tests were completed to confirm groups did not significantly differ in terms of gender ($z(1)=0.03, p=0.859$), proportion of hearing aid users ($z(1)=0.61, p=0.435$), or proportion of CI users ($z(1)=0.00, p=1$). All of the children included in this analysis received listening and spoken language therapy services either in person or via telehealth above the services obtained through early intervention programs; however, there were no differences between the groups on the mode of therapy delivery ($z(1)=0.00, p=1$). The average length of therapy for the duration of the study for these children was 7.16 months. There was no significant difference in the length of therapy when comparing the groups ($t(63)=-1.23, p=0.229$). The average length of therapy for the control group was 7.58 months, while the average length of therapy for the multiple disability group was 6.89 months.

Measures

The Preschool Language Scales 5th Edition (PLS-5) was completed to assess language skills. The PLS-5 measures included scores for Auditory Comprehension (AC), Expressive Communication (EC), and a Total Language Score. All raw scores were converted to age equivalent scores in months (Pearson, et al., 2011; Sahli & Belgin, 2017). Data was collected at two timepoints by speech language pathologists and teachers of the Deaf/Hard of Hearing as part of a larger study. The baseline appointment occurred around the time of enrollment in the study. Follow up appointments occurred roughly 6-7 months after the baseline appointment.

The P1 CAEP biomarker was recorded to assess auditory development. Data was collected at two time points. The baseline appointment occurred around the time of enrollment in the study and follow up appointments occurred roughly 6-7 months after the baseline appointment. CAEPs were recorded in response to the speech syllable /ba/. The stimulus was delivered via a loudspeaker and at a comfortable loudness level. The responses were recorded using 5-8 electrodes depending on if the child had a hearing aid or CI. The CI artifact was minimized using a previously published method (Gilley et al., 2006). About 300 sweeps were collected for each participant using a 100 ms pre stimulus and a 600 ms post stimulus time window. An analog filter was set to 0.1 to 1000 Hz. Testing for subjects occurred in an electromagnetically shielded sound booth where the child was seated on a parent's lap in a comfortable chair. The child was offered a movie of their choice played on a TV monitor in front of them with no sound. The test

session lasted approximately an hour. For all subjects' testing, their hearing aids or CIs were set at their normal settings. (Sharma, et al., 2013; Gilley et al, 2006; Sharma et al., 2002; Sharma et al., 1997; Sharma, Dorman, & Spahr, 2002).

Statistical Analysis

All data was first visualized to identify outliers. The Shapiro-Wilk Test was completed to assess assumptions of normality. Due to a lack of normality across all variables, transformations were performed such that analyses were conducted with the inverse of the raw P1 latencies, the square root of the PLS-5 age equivalent scores at baseline, and log of the PLS-5 age equivalent scores at follow-up. However, the P1 baseline latencies in the disability group ($p=0.039$), as well as P1 latencies at baseline collapsing across the groups ($p=0.047$) continued to demonstrate evidence of a non-normal distribution. A non-normal distribution was also indicated for the transformed PLS-5 EC Score ($p=0.045$). Therefore, both parametric (t-tests) and non-parametric (Mann-Whitney tests) were performed. Since the results were not different between the parametric and non-parametric tests, only the parametric test results are reported here. Two-tailed independent sample t-tests were used to assess group differences between those with multiple disabilities and those with hearing loss in cortical auditory and language measures. T-tests were completed for baseline measures, follow-up measures, and the change in outcomes between these two time points. Two proportion z-tests were used to examine the proportion of children with abnormal P1 latencies in comparison to age matched normative values for each group. Finally, Pearson correlations were examined between P1 latencies and all PLS-5 scores at baseline and follow-up.

Results

Group Differences in Cortical Auditory Evoked Potentials

At baseline and follow-up, three of the children could not complete CAEP testing, or their results contained excessive noise resulting in measures from 63 children being used in this analysis. In comparing a group of children with multiple disabilities to a control group of children with only hearing loss, there were no significant differences in P1 latencies at baseline ($t(32)=-0.03$, $p=0.974$) or follow-up ($t(43)=1.03$, $p=0.200$), as can be seen in Figure 1. Furthermore, in examining the change in P1 latencies from baseline to follow-up there were no significant differences between the groups ($t(36)=-1.50$, $p=0.142$). Specifically, the average change in P1 latency for children with only hearing loss was 34.59 ms, while the average change in P1 latency for children with multiple disabilities and hearing loss was 24.75 ms (see Figure 2).

P1 latencies were classified as normal or abnormal in comparison to age matched normative values for each child (Sharma, Dorman, & Spahr, 2002). The proportion of children with abnormal P1 latencies was calculated, and in a two proportion z-test it was found that there were no significant differences in the proportions of children with abnormal P1 latencies between the groups ($z(1)=0.00$, $p=0.944$). Additionally, we examined group differences in the proportion of children who had abnormal P1 latencies at baseline and normal P1 latencies at follow-up visits. There were no significant differences between the control and multiple disability groups in the proportion of children who went from having abnormal P1 latencies to normal P1 latencies through the course of the study ($z(1)=0.86$, $p=0.353$), as demonstrated in Figures 1 and 2.

Group Differences in Language Outcomes

At follow-up four of the children did not complete PLS-5 testing resulting in measures from 62 children being used in this analysis. In comparing a group of children with multiple disabilities to a group of children with only hearing loss, there were no significant differences in PLS-5 Total Language Score at baseline ($t(54)=1.68$, $p=0.099$) or follow-up ($t(60)=1.84$, $p=0.071$). This was also true for the PLS-5 AC Score at baseline ($t(54)=2.00$, $p=0.050$) and follow-up ($t(54)=1.56$, $p=0.125$). Additionally, there were no significant differences on the PLS-5 EC Score at baseline ($t(59)=1.82$, $p=0.075$) and follow-up ($t(60)=1.66$, $p=0.102$). These results are demonstrated in Figure 3.

Furthermore, in examining the change in PLS-5 Total Language Scores from baseline to follow-up divided by the change in age from baseline to follow-up (the slope), there were no significant differences between the groups ($t(52)=1.44$, $p=0.155$). No significant differences in the change in PLS-5 AC Score from baseline to follow-up ($t(51)=1.06$, $p=0.293$) and the change in PLS-5 EC Score from baseline to follow-up ($t(55)=1.54$, $p=0.130$) were also noted. Specifically, the average change in the PLS-5 Total Language Score for children with only hearing loss was 9.88 months, while the average change Score for children with multiple disabilities was 8.32 months. The average change in the PLS-5 AC Scores for children in the control group was 8.60 months, while the average change for children with multiple disabilities was 7.97 months. In examining the PLS-5 EC Score the average change for the control group was 9.48 months, while the average change for children with multiple disabilities was 9.05 months (refer to Figure 4).

Correlations between P1 Latency and PLS-5

Correlations were first performed by collapsing across the groups. P1 latencies at baseline were not correlated with the PLS-5 AC scores ($r(38)=0.04$, $p=0.214$), EC scores ($r(38)=0.05$, $p=0.149$), or Total Language scores ($r(38)=0.05$, $p=0.185$) at baseline. P1 latencies at baseline were also not significantly correlated with follow-up scores obtained on the PLS-5 AC test ($t(36)=1.10$, $p=0.278$), EC test ($t(36)=1.72$, $p=0.095$), or the PLS-5 Total Language scores ($t(36)=1.72$, $p=0.094$). However, in examining P1 latencies at follow-up weak positive correlations between PLS-5 AC scores ($r(42)=0.13$, $p=0.014$), EC scores ($r(42)=0.17$, $p=0.006$), and Total Language scores ($r(42)=0.18$, $p=0.004$) obtained at follow-up.

When examining the correlations by group, it was found that P1 latencies at baseline in the disabilities group were not correlated with PLS-5 AC scores ($r(17)=0.09$, $p=0.201$), EC scores ($r(17)=0.11$, $p=0.176$), or Total Language scores ($r(17)=0.10$, $p=0.185$) at baseline in the disabilities group nor with PLS-5 AC scores ($r(16)=0.13$, $p=0.147$), EC scores ($r(16)=0.09$, $p=0.241$), or Total Language scores ($r(16)=0.11$, $p=0.171$) at the follow-up in the disabilities group. P1 latencies at follow-up were not correlated with PLS-5 AC scores ($r(20)=0.17$, $p=0.053$), EC scores ($r(20)=0.13$, $p=0.101$), or Total Language scores ($r(20)=0.16$, $p=0.070$) obtained at follow-up.

In the control group, P1 latencies at baseline were not correlated with PLS-5 AC scores ($r(19)=0.01$, $p=0.692$), EC scores ($r(19)=0.02$, $p=0.547$), or Total Language scores ($r(19)=0.01$, $p=0.620$) at baseline nor with PLS-5 AC scores ($r(18)=0.02$, $p=0.598$), EC scores ($r(18)=0.02$,

$p=0.593$), or Total Language scores ($r(18)=0.01$, $p=0.631$) at the follow-up. At follow-up, P1 latency were not correlated with follow-up PLS-5 AC scores ($r(20)=0.04$, $p=0.357$), EC scores ($r(20)=0.04$, $p=0.358$), or Total Language scores ($r(20)=0.04$, $p=0.358$).

Discussion

Our main finding is that there are no significant differences in auditory cortical development (as measured by P1 latencies) or language development (as measured by PLS-5 scores) between the control group of children with hearing loss and the group of children with hearing loss and additional disabilities. We also found both groups to be equivalent in terms of the portions of children who showed a change from abnormal to normal P1 latency classifications from baseline to follow-up. Furthermore, there were no strong correlations between P1 latencies and PLS-5 scores.

Our study found no statistically significant difference in P1 latencies or PLS-5 scores between the control group and disability group. In contrast, a previous study by Mesallam, Yousef, and Almasaad (2019) exploring auditory and language skills development after cochlear implantation in children with multiple disabilities found that outcomes of only some of their multiple disabilities' subgroups were comparable to the control group. The authors suggested that due to these findings' children with hearing loss and additional disabilities cannot be directly compared to the children with hearing loss and no additional disabilities. Furthermore, it was suggested that due to variable degrees of outcomes for children with additional disabilities the degree of benefit from CI may depend on the type of additional disability (Mesallam, Yousef, & Almasaad, 2019). It has also been reported that children with multiple disabilities show improvement on PLS scores in receptive, but not expressive language (Beer et al., 2012). In addition, even with these improvements, children with multiple disabilities have been reported to obtain lower scores on language outcomes than children with only hearing loss (Beer et al., 2012). Cupples, et al. (2018) demonstrated differential results depending on the specific additional disability. Specifically, children with more cognitive impairments showed declines in language ability tests over a 2-year period, while children with other disabilities demonstrated improvements. Therefore, differences in our results from those that have previously been reported could be attributable to the fact that we did not analyze the data according to the type of disability or that additional types of disabilities were included in our sample. However, this study adds novel findings regarding P1 latencies, as to our knowledge, this is the first study to look at cortical auditory maturation in a group of children with multiple disabilities.

There was no significant difference between the disability group and control group for change from abnormal to normal P1 latencies. 31% went from abnormal to normal in the control group and 28% for the disability group. This is significant because it shows when children receive an additional dose of therapy (either in-person or via telemedicine) in the short amount of time of six months, a child can go from abnormal to normal even with additional disabilities. It is important to note that in this study the average age for implantation in the control group was relatively equal, i.e., 19.43 months and for the disability group was 23.23 months. Many previous studies have stressed the importance of early implantation in this population (Cupples et al., 2018; Sharma, Dorman, & Spahr, 2002). Outcomes across the two groups of children may differ if the two groups of children showed a difference in age of implantation.

There were almost no correlations between P1 latencies and PLS-5 scores. We hypothesize that P1 latencies are a better reference for oral listening skills such as speech perception, rather than language development. Language development in this study was not restricted to oral language development. A previous study by Cardon and Sharma, published in 2013, found that P1 latencies correlated with listening skills (IT-MAIS). Children with earlier P1 latencies tended to have higher IT-MAIS scores (Cardon and Sharma, 2013). Contrary to the present findings, Birman, Elliott, & Gibson (2012) reported on an auditory skills task (Categories of Auditory Performance), in which they found poorer performance for children with additional disabilities compared to those with only hearing loss. They also found lower scores on auditory skills tasks for children with developmental delays in comparison to those without developmental delays. In our study, 68% of children from our study are sign language users, for either some or most of the time. Cupples et al., (2018) noted the use of oral communication only increased improvements in receptive vocabulary. Therefore, it makes sense that P1 latencies and PLS-5 did not correlate well in our sample because PLS-5 scores are not restricted to oral language. Future research may examine listening skill differences in these populations alongside CAEP measures.

A limitation of this study was that we were unable to separate the disability group into subcategories of disabilities. Due to the diverse range of disabilities and small sample size, we had to put all disabilities into one group, our disability group, and thus could not give a clearer picture of how specific disabilities may impact our results. Another limitation was that the disabilities were self-reported from parents instead of being formally evaluated. It would be interesting to see how this could have affected the accuracy of the participants' listed disabilities. Despite these limitations, we have found that overall, the disability group is not different from the control group during a time period when both groups were provided with an additional dose of language therapy.

In conclusion, our key takeaway from this study is there were no significant differences in developmental trajectories for auditory cortical development and language development as measured by P1 latencies and PLS-5 scores between children with hearing loss and children with hearing loss and additional disabilities. Our main finding of the lack of difference in cortical and language development between these two groups is useful information for clinicians and parents on what kind of developmental trajectories to expect when assessing the effect of intervention with hearing aids and cochlear implants in children with hearing loss who have additional disabilities.

Disability	N
Pendred's Syndrome	2
CMV	7
Waardenburg Syndrome	5
Fine Motor Problems	10
Gross Motor Problems	10
Heart Defect	5
Epilepsy	2
Asthma	2

Table 1: Disability distribution table. The left column displays the type of disability, and the right column displays the number of participants with that disability.

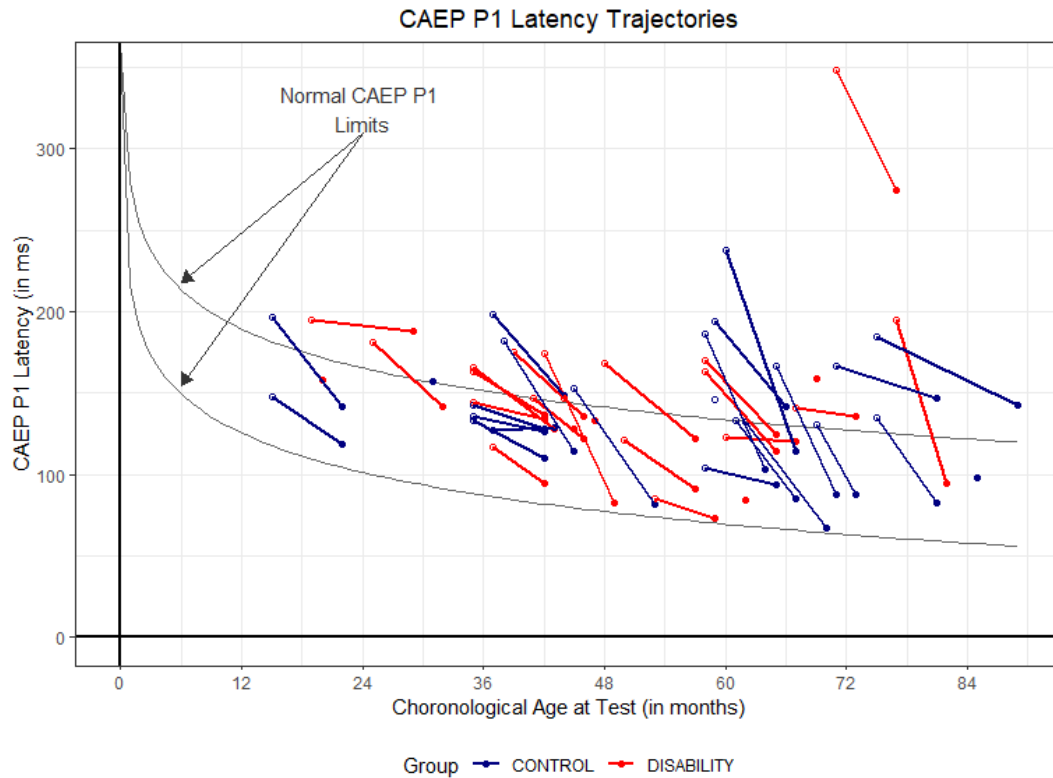


Figure 1: CAEP P1 Latency Trajectories. The Y-axis displays participants' CAEP P1 latency in ms and the X-axis displays participants' chronological age at test in months. In this graph, the control group (blue) and disability group (red) are being compared on their P1 latency changes from baseline to follow-up. Every participant's P1 latency decreased at follow-up and the majority of the children demonstrated P1 latencies within normal limits at follow-up. As can be seen in this graph, significant differences between groups were not evident.

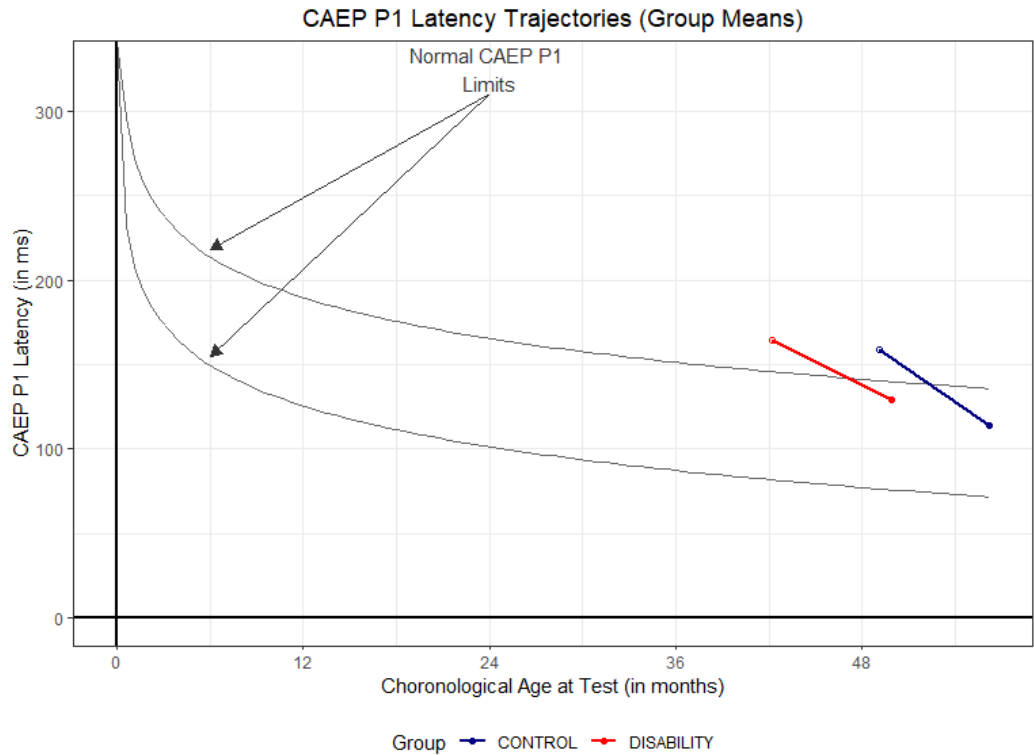


Figure 2: CAEP P1 Latency Trajectories (Group Means). The Y-axis displays the CAEP P1 latency in ms and the X-axis displays the chronological age at test in months with the control group (blue) and disability group (red) mean P1 latencies visualized against 95% confidence intervals for age matched normal P1 latencies (Sharma, Dorman, & Spahr, 2002). From this graph we can see the average P1 latency trajectory for the two groups is very similar.

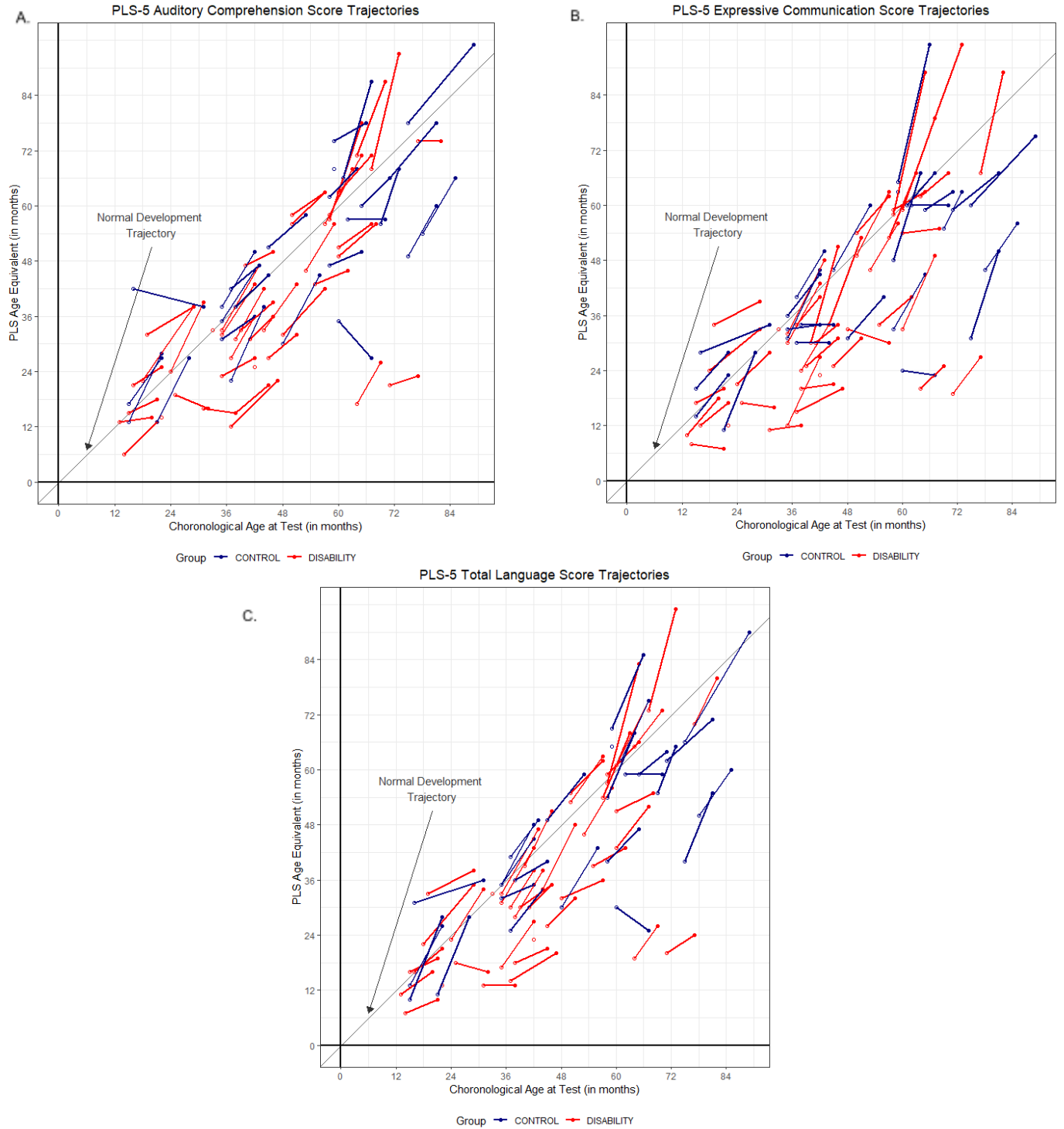


Figure 3: PLS-5 Score Trajectories. The Y-axis displays the participants' PLS age equivalent scores in months and the X-axis displays the participants' chronological age at test in months for the control group (blue) and disability group (red). The linear gray line shows the normal development trajectory. From this graph it can be seen that the majority of participants increased in PLS age equivalent scores from baseline to follow-up. This change was not different amongst our two groups for (A.) Auditory Comprehension Scores, (B.) Expressive Communication Scores, and (C.) Total Language Scores.

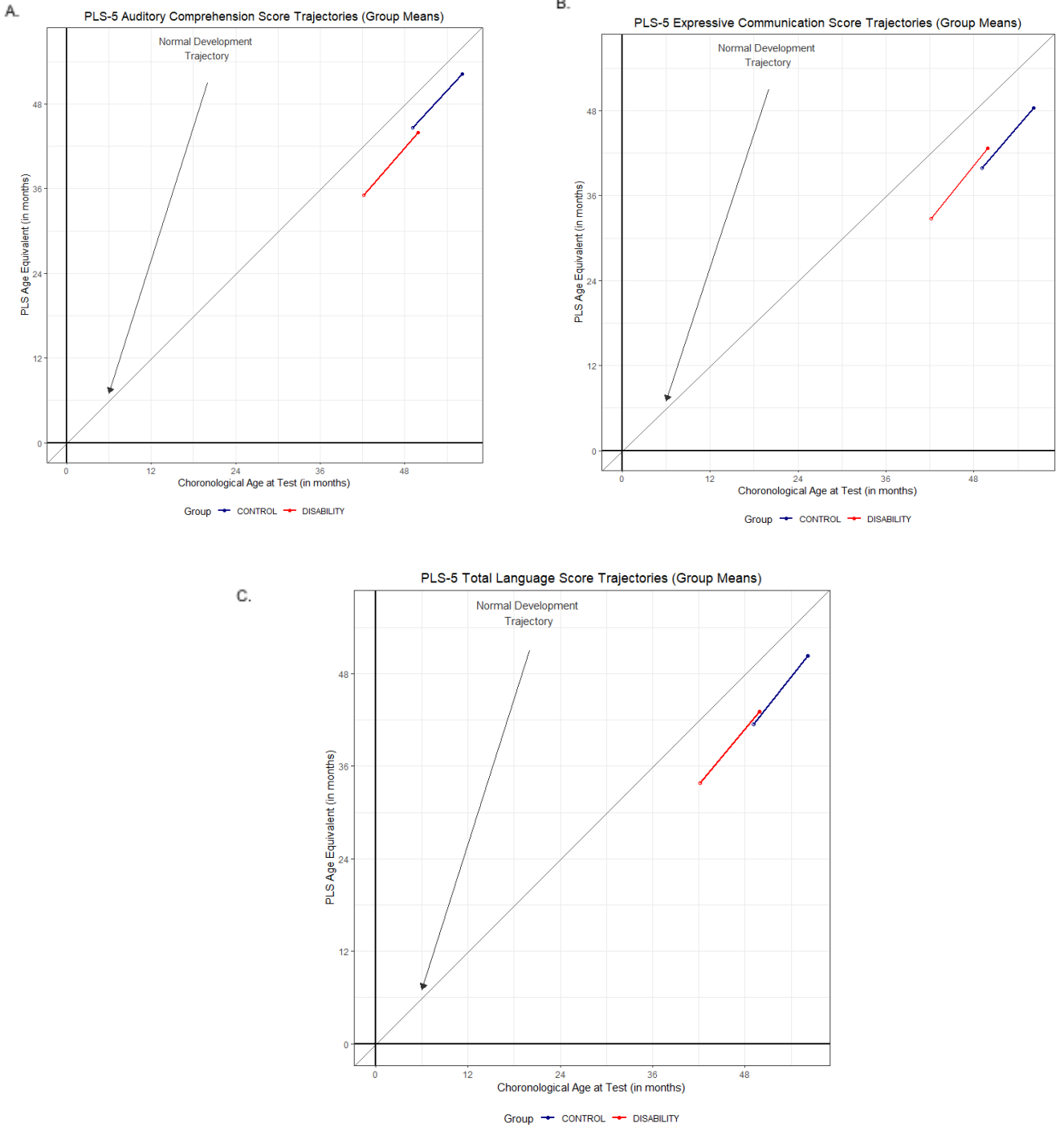


Figure 4: PLS-5 Score Trajectories (Group Means). The Y-axis displays the participants' PLS age equivalent scores in months and the X-axis displays the participants' chronological age at test in months for the disability group (red) and control group (blue). The linear gray line shows the normal development trajectory. From this graph it can be seen that both the control group and disability group increased in PLS age equivalent scores from baseline to follow-up. There were no significant differences between our two groups for (A.) Auditory Comprehension Scores, (B.) Expressive Communication Scores, and (C.) Total Language Scores.

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