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Investigation of Motor Programming in Childhood Apraxia of Speech

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INVESTIGATION OF MOTOR PROGRAMMING IN CHILDHOOD APRAXIA OF SPEECH

BY

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ABSTRACT

INVESTIGATION OF MOTOR PROGRAMMING IN CHILDHOOD APRAXIA OF SPEECH

By

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The underlying nature of the motor programming disorder in stroke-related apraxia of speech (AOS) has received extensive investigation. This is not the case for childhood apraxia of speech (CAS). CAS and AOS present with many of the same perceptual characteristics (increased intersegment and segment duration ((i.e., segmentation)), equal lexical stress, and speech sound distortions), however, little work has been done to investigate whether the disorders have the same mechanism of action (McNeil, Robin, & Schmidt, 2009). Previous studies investigating motor programming in AOS have utilized a two-stage model developed by Klapp (2003). This model breaks down the motor programming process into two stages. The first is INT which involves the organization of the internal timing and spatial information for a movement. The INT process also involves the movement being loaded into a motor buffer. The second is SEQ which involves the sequencing of the motor units into accurate serial order. The present study investigates motor programming in CAS by utilizing a self-select paradigm. The self-select paradigm enables both INT and SEQ to be measured in each trial (Wright et al., 2009). The INT process is termed study time (ST) and reflects time that a participant takes to prepare their response. The SEQ process is termed reaction time (RT) and reflects the amount of time that it takes for a participant to execute their response after the presentation of a "go" signal. Maas et al. (2008) utilized this self-select paradigm in adults with stoke-induced AOS compared to typical controls and individuals with a language impairment (aphasia). They found that for

participants with AOS the impairment was specific to the INT (ST) stage of motor programming. This translated to participants with AOS having overall longer ST in comparison to typical and aphasic controls. In contrast, participants with AOS had comparable SEQ (RT) with typical controls. These findings were demonstrated across nonspeech (finger-tapping) and speech movements. This indicates that AOS is due to a central, rather than modality-specific impairment (Maas et al., 2008).

In this study, an experimental task was designed to investigate whether the impairment seen in CAS is that same as that observed in AOS, and whether the impairment is central or modality-specific. The experiment tested speech movements (vocalization of the syllable /bɔ/). Again, like in Maas et al. (2008) three groups of participants were included in the study: children with a diagnosis of CAS, typically developing children (TD), and children with a language-based phonological impairment (PI). There were 26 participants in total ranging from ages 5-12: CAS (n=6), PI (n=9), TD (n=11).

Results from the experiment (vocalizations of the syllable /bɔ/) demonstrated a negative correlation of ST and RT with age for TD and PI participants. This was not the case for participants with CAS who did not demonstrate a decrease in ST and RT with age. This suggests that for participants with CAS, the motor programming process does not become more efficient as they get older. These findings provide insight into the underlying motor programming impairment in CAS and inform the need for theory-driven treatment approaches which specifically target the disorder. Another finding demonstrates that as the experiment progresses, children with CAS demonstrate a dramatic increase in ST while TD and PI participants do not. Conversely, TD and CAS participants demonstrate similar patterns of RT across the experiment. These findings support the hypothesis that the impairment in CAS is specific to the INT stage of the motor programming process, while the SEQ phase is intact. Given that these findings are similar to what was found in Maas et al. (2008) with adults with AOS, the results also support the hypothesis that CAS and AOS arise from the same mechanism of action.

INTRODUCTION

Stroke-related apraxia of speech (AOS) is a disorder of motor programming differentially identified by increased duration of segments (i.e., segmentation) between syllables and words, equal lexical stress, and distortion of speech sounds (McNeil, Robin, & Schmidt, 2009; Ballard, Granier, & Robin, 2000; Ballard & Robin, 2007; Deger & Ziegler, 2002; Kent & Rosenbek, 1983). The mechanism underlying AOS in adults results in an inability to translate the phonological representations of utterances into the spatiotemporal parameters required to execute the message (McNeil, Robin, & Schmidt, 2009; Maas et al., 2008). This deficit manifests itself in perceptual, acoustic, and kinematic levels of analysis (e.g., Robin, Bean, & Folkins, 1989; McNeil, Robin, & Schmidt, 2009; Seddoh et al., 1996). Far less is known about the mechanisms underlying childhood apraxia of speech (CAS) and though it is considered by most to be a motor programming impairment, some continue to view it as a phonological disorder. As a motor programming disturbance, CAS has been hypothesized to have the same underlying deficits as found in AOS (McNeil, Robin, & Schmidt, 2009; Miller et al., 2021). Moreover, clients with both AOS or CAS show positive responses to treatments based in motor learning, suggesting a close overlap between the two. However, little research had been conducted to investigate this claim. This gap in our knowledge of CAS is particularly striking given the fact that the differential characteristics of the two disorders are the same. The underlying mechanisms of CAS must be determined to improve diagnostic and treatment approaches for the disorder. In other words, data are needed to support theory driven clinical practice in CAS.

In order to take the first step in filling this important knowledge gap, the present study was designed to identify the motor programming deficits in CAS and if they are the same as those found in AOS. The primary research questions addressed were (1) Are the impairments in

CAS localized to a particular stage of the motor programming process? (2) Is this impairment the same as what has been observed in individuals with AOS?

Previous research has indicated that for individuals with AOS, the deficit in motor programming can be localized to a stage of motor programming representing a working memory buffer (Maas et al., 2008). This stage is impaired or inefficient for both speech and nonspeech movements in AOS. Maas et al. (2008) utilized a self-select paradigm (described below) to identify the specific motor programming deficit in AOS. This paradigm was derived from a two-stage model established by Klapp (2003) which breaks down the motor programming process. This model delineates two processes that are involved in motor programming (Klapp, 2003). The first process is INT. During the INT phase of motor programming, the movement's spatiotemporal structure is organized, then that unit is loaded into to a motor buffer (working memory store). At this point, the SEQ process begins. During the SEQ process, the units of movement in the motor buffer are sequenced into the appropriate serial order. This process occurs after the initiation of movement. Results from Maas et al. (2008) identified that the aspect of motor programming impaired in AOS is INT. Individuals with AOS had a deficit in their organization of the spatiotemporal structure of units of movement and loading the units into the motor buffer. In contrast, the sequencing and initiation of the motor units (SEQ) were similar to those of controls. The impairment in INT but intact SEQ were present in speech and nonspeech movements (finger tapping and vocalizing the syllable /bɔ/). These findings support the hypothesis that AOS is caused by a motor programming disorder that is central, rather than modality specific. This corresponds with a growing body of literature that suggests that AOS is caused by a central motor programming disorder (Ballard, Robin, & Folkins, 2003).

Specifically, INT organizes the internal spatiotemporal units for a movement prior to execution and loads those units into a motor buffer. SEQ is responsible for the on-line sequencing of the motor units once the intended movements are initiated. Therefore, sequencing the motor programs that comprise the intended speech output occurs as speech is initiated as a separate process of programming. Again, INT is preprogrammed before the beginning of the movement. Wright et al. (2009) found that the length of INT is impacted by motor unit complexity. When the movement was more complex, the INT duration increased. Conversely, SEQ was unaffected by motor unit complexity but increased in length with more units in the working memory buffer. Klapp (2003) found that syllables of words are concatenated (joined) and input into the working memory buffer as one unit. How complex the unit depends on how many syllables are in the word.

The self-select paradigm utilized in Maas et al. (2008) allows for the measurement of INT and SEQ in each trial (Wright et al., 2009). The self-select paradigm requires the participant to produce a skilled movement (e.g., speech) after an auditory or visual model is shown. Participants are provided as much time as they need to prepare their response, and when ready, they are instructed to press the spacebar. The time that it takes for the participant to prepare their response is called study time (ST). This is reflective of the INT process of motor programming. After the participant presses the spacebar, there is a period of variable delay, then they are presented with a “go” signal. The time between when the “go” signal is presented and the participant executes their response is known as reaction time (RT) and is reflective of the SEQ process. Previous studies have shown that for non-speech movements when the duration of the response is longer, the ST period is longer, but the RT period is not (Klapp, 1995; Immink &

Wright, 2001, Wright et al., 2004). When the length of the sequence for the response is longer, RT was impacted but ST was not.

Previous research in AOS across speech and non-speech movements has found that the deficit in INT is central vs. modality-specific (Maas et al., 2008). Therefore, there was initially an interest in applying the self-select paradigm in the present study to both speech and nonspeech movements in children. The paradigm was initially run on several typically developing children. Participants demonstrated no difficulties learning the experimental tasks for the speech movements, but when presented with the same patterns and asked to execute them via tapping their finger on the f-key, accuracy was low. One reason for the discrepancy in performance could be that while children use speech motor movements daily for communication, this is not the case for sequences of finger tapping. Tapping of a finger in a particular sequence is a skill specific to certain activities like playing the piano, and for most children was completely novel. Due to low accuracy across typically developing participants, the decision was made to discontinue testing for the nonspeech task. Therefore, the hypotheses that follow are specific to the speech task.

According to the hypothesis that AOS and CAS have the same mechanism of action, when using the self-select paradigm with similar stimuli to Maas et al. (2008), a strong prediction is that the INT process (working memory buffer where programs are assembled) is disrupted. This would correspond to participants with CAS having a longer ST in comparison to individuals with a language impairment (PI) and typically developing controls (TD). We would also expect that more complex motor units would translate to a longer ST across groups due to the nature of the INT process. A second hypothesis is that SEQ (RT) will be comparable between TD and CAS participants. A third hypothesis is that children with phonological impairments will have an intact INT but will have a disruption of the sequencer (SEQ). This would translate to a longer RT for PI participants when compared to TD and CAS participants.

II. METHODS

Participants

The study consisted of 26 participants between 5 and 12 years of age. The participants were assigned to one of three groups: CAS (n=6), PI (n=9), and TD (n=11). The average age was 8.9 years (SD=1.92) for the TD participant group, 7.2 years (SD =2.39) for PI, and 9.8 years (SD=2.32) for CAS. Although 8 children were recruited in the CAS group, two were unable to accurately execute the experimental tasks due to apraxia severity. One child demonstrated difficulty following the instructions of the task, and the other child could not accurately program the CV combination to produce the speech syllables for the experiment. These children were excluded from analysis and are not reflected in the group numbers above. The parent or guardian of all participants was provided informed parental consent and the child provided informed assent. All procedures were approved by the University of New Hampshire Institutional Review Board (Title: *An Investigation of Motor Programming in Childhood Apraxia of Speech*. Protocol number: IRB-FY2022-184)

Inclusion criteria were (1) age-typical language scores on the *Clinical Evaluation of Language Fundamentals- Fifth Edition Screening Test* (CELF-5 Screening Test) (Wiig, Secord, & Semel, 2013), (2) normal hearing as assessed by a pure-tone audiometric screening at 500-4,000 Hz at 20 dB (*Childhood Hearing Screening*, n.d.), (3) normal vision based on parent report, (4) native English speaker, (5) no other developmental, speech, or genetic disorders by parent report. Participants were excluded if they presented with (1) oro-facial muscle weakness, (2) altered reflexes or the presence of reflexes that developmentally would not be present such as a positive Babinski sign, (3) hyper- or hypotonic muscle tone, (4) extrapyramidal signs (e.g., hyperkinetic movements), or (5) orofacial structural abnormalities.

It should be noted that one of the participants with CAS who was included in analysis scored one point below the age criterion language score on the CELF screener. However, due to how close the child was to meeting the language criterion for his age, and the desire to keep the number of participants as high as possible for the CAS group, the decision was made to keep him in the study. Another participant with CAS had a diagnosis of Tourette's syndrome, which presented as hyperkinetic movements of mild facial grimacing/eye-blinking. Again, to keep the numbers of participants in the CAS group as high as possible, the decision was made to include this participant in the study results.

For the children with speech impairments, the diagnosis of CAS or PI was made by two experts in motor speech disorders based on the key differential features of CAS being segmentation, equal lexical stress, inconsistent type of error, and distortion of speech sounds (McNeil, Robin, & Schmidt, 2009). Segmentation is defined as increased intersegment and segment duration. This is especially true in polysyllabic words where there are inter-syllabic pauses. Equal lexical stress is defined as equal or diminished stress between syllables of polysyllabic words or across phrases. Finally, distortions of speech sounds are errors in the production of phonemes that cannot be classified as a clear substitution. Examples of speech sound distortions include vowel, voicing, and consonant substitution distortions (Miller et al., 2021).

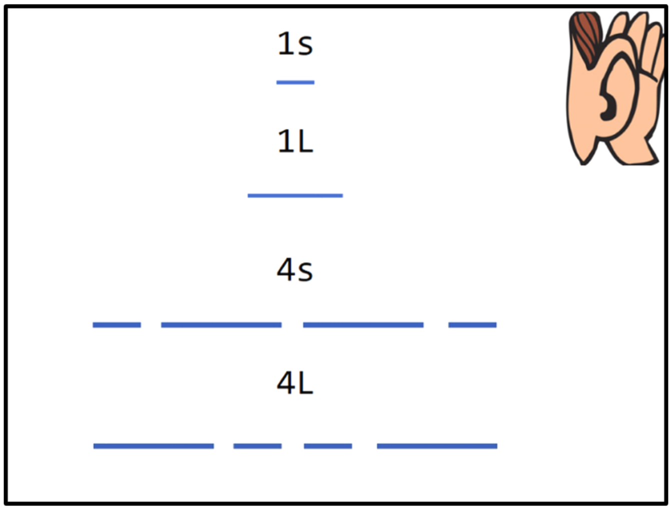
Phonological impairments stem from a difficulty in learning the rule behind a particular group of sounds. Error patterns are seen across sounds and are predictable in nature. Phonological impairments are linguistically based (*Speech Sound Disorders: Articulation and*

Phonology., n.d.). Children were diagnosed with phonological impairments based upon presentation of these predictable error patterns during the language screening evaluation. It is important to note that children who had a history of phonological impairment were also included in the group.

Procedures

The children were asked to complete an experiment that followed the procedures outlined in Maas et al. (2008). This experiment was designed to investigate the INT and SEQ processes of motor programming (Klapp, 2003). INT is the organization of the internal spatiotemporal motor unit structure. That motor unit is then loaded into a motor buffer. SEQ is the sequencing into serial order the motor units in the buffer. The children were trained on four different pattern responses (vocalizations of the syllable "/bɔ/") which had both an auditory cue and a visual cue presented on a computer screen (see Figure 1). After they were familiar with these patterns, they were provided a visual cue only and asked to accurately produce their own model to match the auditory cues that they had heard (Immink & Wright, 2001; Klapp, 1995; Wright et al., 2009). The patterns either had 1 or 4 tones and each tone was either short (150 ms) or long (450 ms). To indicate to the child which pattern to execute, a visual model was displayed on the screen (see Figure 1). A 1s pattern indicated one vocalization for a short duration (150 ms). A 1L pattern indicated one vocalization for a long duration (450 ms). A 4s pattern involved pressing the key in a short-long-long-short sequence (150-450-450-150). Finally, a 4L pattern involved pressing the key in a long-short-short-long sequence (450-150-150-450). The model pattern had 100 ms interstimulus intervals.

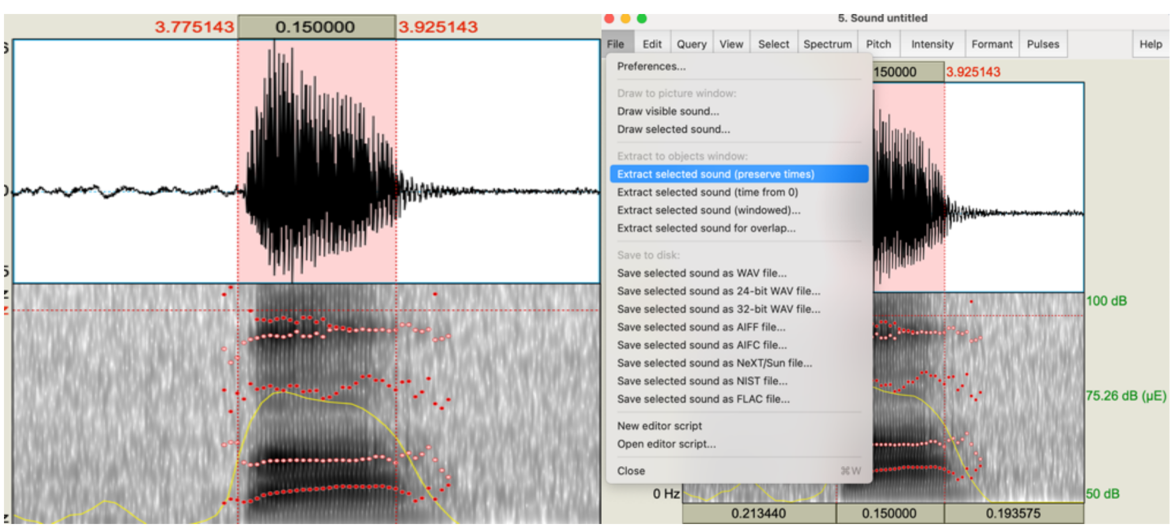
Figure 1. Trained Key Pattern and Vocal Responses



Note. S=150 ms, L=450 ms, 1= one press response, 4= four press response.

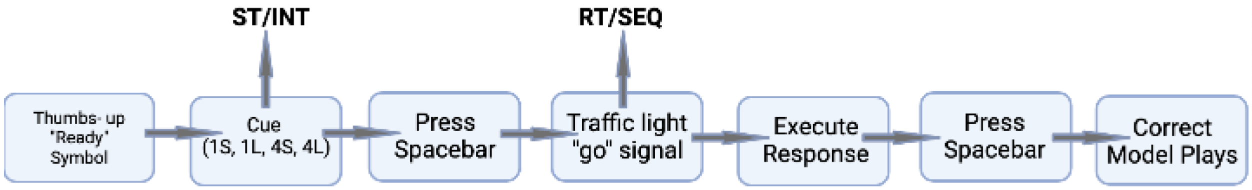
Auditory models of the target were created using recordings of a male native English speaker. The models were altered so that they were exactly 150 or 450 ms long as measured from plosive burst to the offset of vocalization. This truncation was done using Praat software. The speaker produced a syllable that was approximately 150 ms or 450 ms then a selection of the appropriate length was highlighted and extracted (see Figure 2). Four pattern sequences contained 100 ms pauses between each syllable.

Figure 2. Praat syllable selection and truncation



The experiment was carried out using E-prime software (Psychology Software Tools Inc.) on a Dell laptop and was coded at the University of New Hampshire. Speech input was collected using the Chronos hardware box (Psychology Software Tools Inc.) as well as a Koss SB/45 headset with microphone. The child wore the headset, and the microphone was positioned approximately 5 cm away from their mouth. Data was stored in a computer file. Each trial of the experiment followed the same sequence of events (see Figure 3). The trial began with a presentation of a thumbs-up icon on the screen for 500 ms. Prior to the trial, the children were instructed that this symbol means that they should "get ready" to respond. After the thumbs-up icon was presented, the visual cue appeared on the screen (1s, 1L, 4s, 4L). The child took the time that they needed to prepare their response based upon the visual cue on the screen. When they were ready to respond, they were instructed to press the space bar. The period of time that it took for the child to prepare their response was Study Time (ST). This period reflected the INT process or the internal working memory buffer for motor programs. Once the child pressed the space bar there was a variable delay time (800-1200 ms), then a symbol of a green traffic light was presented on the screen for 300 ms. This indicated to the child that they should execute their response by vocalizing the pattern of the syllable "/bɔ/" to accurately match the cue presented. The time that it took the child to vocalize after the presentation of the green traffic light symbol was reaction time (RT). This reflected the SEQ process.

Figure 3. Self-Select Paradigm



Note. Self-Select Paradigm (e.g., Immink and Wright, 2001; Wright et al., 2004; Maas et al., 2008). ST, study time; RT, reaction time.

Prior to testing the child was provided with auditory instructions read by the investigator (Appendix A). Each child was tested individually in a sound isolated room. Following the instructions, auditory models of each of the patterned responses were played to help the child understand the target responses that they would produce. Each pattern was played 4 times in a blocked manner (1s, 1L, 4s, 4L). After the child has listened to each of the target responses, the production portion of the experiment began. There were 6 blocked trials that contained patterns presented in random order. If a child produced a trial incorrectly it was re-presented in a "rerun block". Incorrect trials were automatically scored in the E-Prime software based upon the timing of the child's responses. Each of the blocks contained 16 trials. The number of rerun blocks presented depended on the number of trials that the child initially executed correctly. Incorrect trials were rerun to ensure that ST and RT measures were only calculated on correct pattern executions. In the first block, the experimenter re-explained the instructions to the child and provided additional examples of correct responses.

During both the training and test portions of the paradigm, the child received feedback after each response about whether they were correct or incorrect. If the child produced a correct response, they received a message on the screen reading "correct!" If the child produced an incorrect response, an error message was presented. The error message was specific to the child's response. The message may have read: (1) "Too slow!" if the child initiated their response >1000ms after the go-signal was presented; (2) "Too Early! Wait for the go-signal." If the child initiated the response during the variable delay period prior to go-signal presentation; (3) "Too long" or "too short" if the child executed a response that was overall outside the specified range (+/- 100ms outside target length for individual press patterns or +/- 500ms outside target length for 4 sequence patterns); (4) "Pause too long", if the child's pause between presses was longer than 200ms; and (5) "Incorrect response", if the child executed a 4s response instead of a 4L or vice versa. The timing of the child's response was automatically measured by the E-Prime software and was judged for accuracy based upon the parameters above. Error messages were read aloud to the child by the experimenter. After the child finished vocalizing their response, they pressed the spacebar again and were played back the correct auditory model. This happened whether the child's response was correct or incorrect and was for the purpose of solidifying the child's acquisition of accurate responses. There were 2 seconds between trials. The experiment took approximately 30 minutes.

During the production phase, the child was provided with rests after each block. The experimenter determined how long the rest period should be, based on the child's level of engagement or signs of fatigue. Breaks were approximately 2 minutes in length. During the break, the child was encouraged to drink water to keep their voice healthy. At the end of each rest period, the examiner reminded the child of the instructions to "respond quickly and accurately".

Analytic Approach

Several measures of the child's performance were recorded for analysis. The percent accuracy of trials was recorded. This measure was calculated based upon the number of trials that were re-presented after an incorrect response. The dependent variables were the child's time spent preparing their response (ST) and the time that it took them to initiate their response by tripping the voice key after the imperative go-signal was presented (RT). The ST and RT were only recorded on responses that the child accurately executed. The independent variable was group (i.e., CAS, PI, TD). The duration of both single syllables and 4 sequence patterns was recorded.

For the visualizations, a best fit modeling approach was utilized. Linear, exponential, 1^{st}-, 2^{nd}-, and 3^{rd}- order quadratic fits were tested. Linear fit yielded the highest r^2 values and was subsequently the fit chosen.

III. RESULTS

Accuracy of Responses

The accuracy of participant responses was identified based on the number of rerun trials presented at the end of the session. Excluded from analysis were blocks 10 and 11 of rerun trials. The number of rerun trials that the children were presented with was based on their accuracy during the first six blocks. The rerun blocks were similarly each composed of 16 trials. No participants with CAS were presented with more than nine blocks total, with three rerun blocks. Three TD participants and one PI participant were presented with 10 total blocks, two PI participants were presented with 11 total blocks. Therefore, blocks 10 and 11 were not representative of each group (CAS, TD, PI) as a whole. Data from these blocks were excluded from the analysis and overall percent accuracy scoring.

The response accuracy for participants with CAS was compared to the accuracy of responses for TD and PI participants. For participants with CAS, the percent accuracy at the group level was 48.99%. For the PI group, 49.20% Finally, for TD 55.10% at the group level. Accuracy of responses was further broken down based upon cue type (1s, 1L, 4s, 4L). For 1s response type, accuracy was highest across groups (CAS; 69.94%, PI; 67.40%, TD; 70.93%). For 1L accuracy slightly decreased across groups (CAS; 59.46%, PI; 50.67%, TD; 59.13%). There was an additional decrease in accuracy for 4L responses as complexity of syllable cue increased (CAS; 38.79%, PI; 39.68%, TD; 44.5%). Finally, accuracy was lowest across groups for the 4s syllable cue (CAS; 33.49%, PI; 41.37%, TD; 48.62%).

ST/RT across press cue types

ST across press cue types.

Outliers in ST were excluded. Outliers were calculated as data points that were greater than 1.5 group standard deviations away from the individual participant mean. A total of 54 data points or 2.98% of data set were considered outliers and removed. Average ST for the TD and PI participants were calculated at the group level. The PI group had the longest study time (M= 3545.38 ms, SD= 2553.75). Next was TD (M=3193.61 ms, SD= 2395.53). Contrary to the hypothesis that ST would be the longest for individuals with CAS, children in this group had the shortest ST at the group level (M=2810.71, SD=1926.85). ST was further delineated based on press type (1s, 1L, 4s, 4L). ST was shortest for 1s cues across groups, with an increase in ST across groups as the complexity of response increased. ST was the longest in each group with the 4s cue type which was the most complex.

RT across press cue types

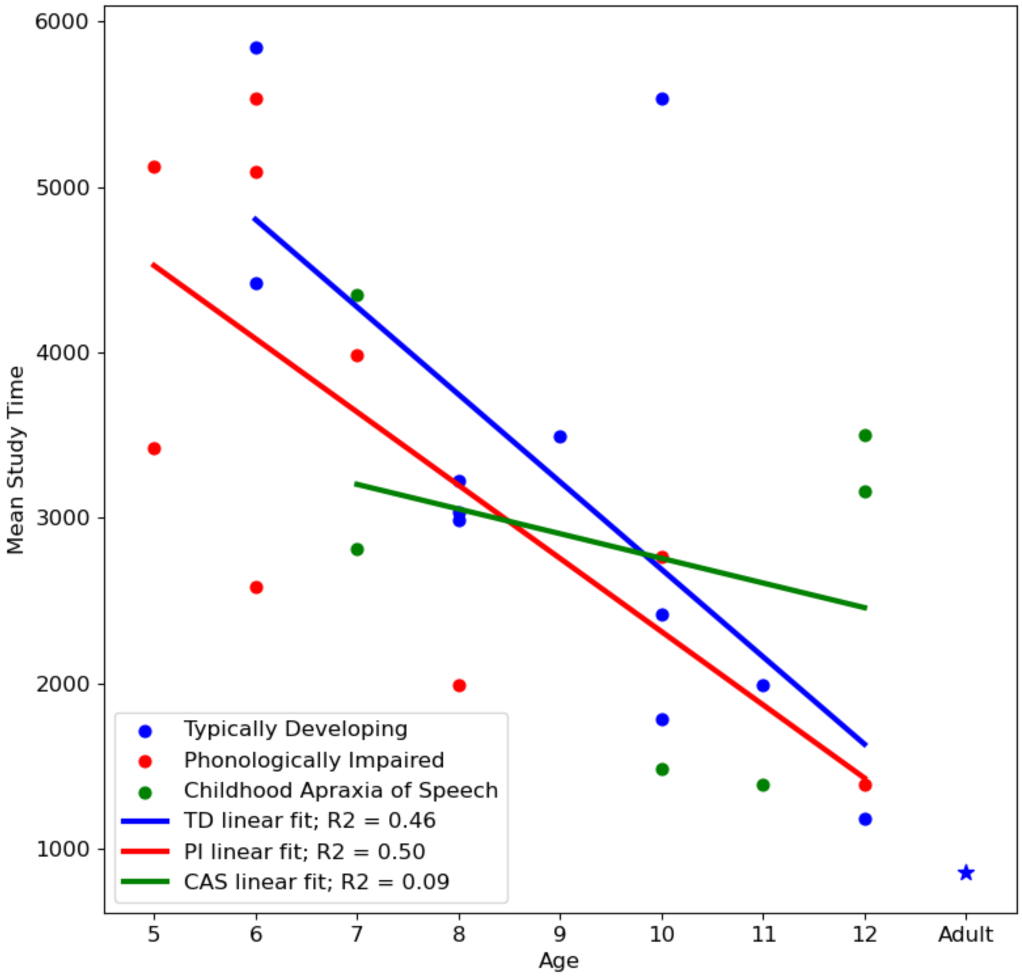
Outliers in the study were excluded from the analysis using the same method as ST. Including the data points from blocks 10 and 11 which were excluded from analysis, 126 data points or 6.96% of the total data set was removed. Average RT for each group was calculated and further delineated based on cue type. At the group level, average RT across groups was similar. For TD, (M=434.77 ms, SD=127.28); for PI, (M=424.83 ms, SD=133.39); for CAS, (M=428.07 ms, SD=135.14). When broken down by press type, RTs were similar across groups.

Age and ST/RT

Correlation between age and ST

Next, the relationship between ST and age was investigated using a linear best fit model (Figure 4). For TD (r^2=0.46) and PI (r^2=0.50) participants, the correlation between age and ST was similar, with a negative correlation as age increased. This correlation was not as clear in participants with CAS (r^2=0.09), indicating a low correlation between age and ST. Data from typically developing adults who completed similar experimental tasks using the self- select paradigm were included in this figure (Wright et al., 2009). Data shows that for TD and PI participants at age 12, their ST is similar to that of an adult. This is not the case for the CAS participants who demonstrate a substantially higher ST at age 12.

Figure 4. Study time by age.

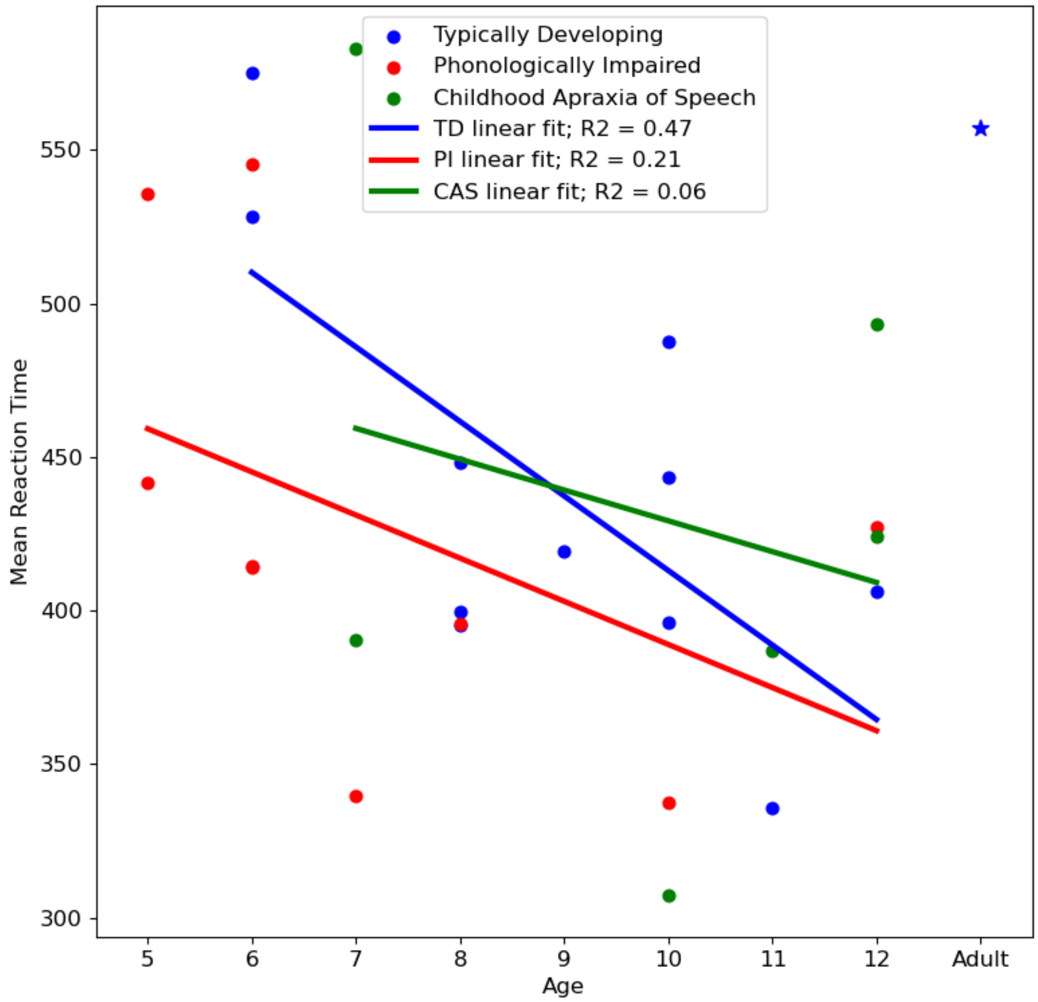


Note. Adult data from Wright et al. (2009), Taken using self-select paradigm on 12 typical young adults.

Correlation between age and RT

Next, the relationship between RT and age was investigated. This was done using a linear best fit modeling approach (Figure 5). The correlation between age and RT differed by group. For TD participants there was a negative correlation as age increased (r^2=0.47). For PI participants this correlation was weaker (r^2=0.21). For participants with CAS, the correlation between RT and age is weak (r^2=0.06). These results are similar to those noted with the correlation between ST and age. RT data from typical adults was included in the figure (Wright et al., 2009). Results demonstrate that RT is substantially higher for adults than it is for children across any of the age groups at age 12.

Figure 5. Reaction time by age.



Note. Adult data from Wright et al. (2009), Taken using self-select paradigm on 12 typical young adults.

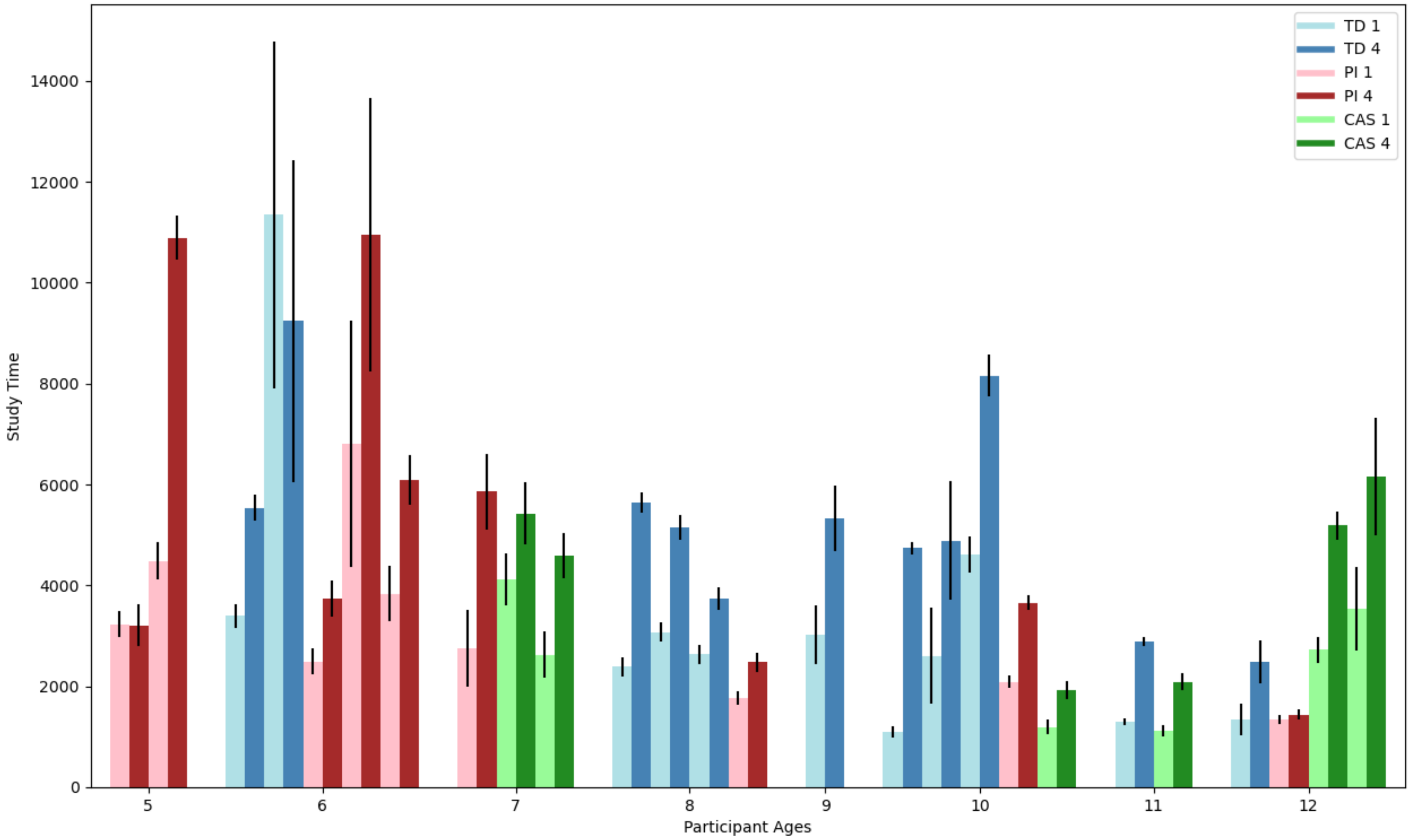
1 vs. 4 syllable cues ST/RT

1 vs. 4 syllable cues ST

The ST for both 1 and 4 press cue responses were recorded for each participant (Figure 6). These STs were then organized by participant age. While there was not an equal distribution of participants across ages, there are two notable observations that can be drawn regarding age and

ST. The first is that except for one TD 6-year-old participant, ST is longer for four press responses than one press. Another notable aspect of the figure is the clarity of the age effect in TD and PI participants. There is a visible reduction in ST as the age of the participant increases. For participants with CAS, however, the trend is relatively flat. ST for 12-year-old participants with CAS is comparable to that of 7-year-olds in the same group.

Figure 6. 1 vs. 4 syllable cues ST

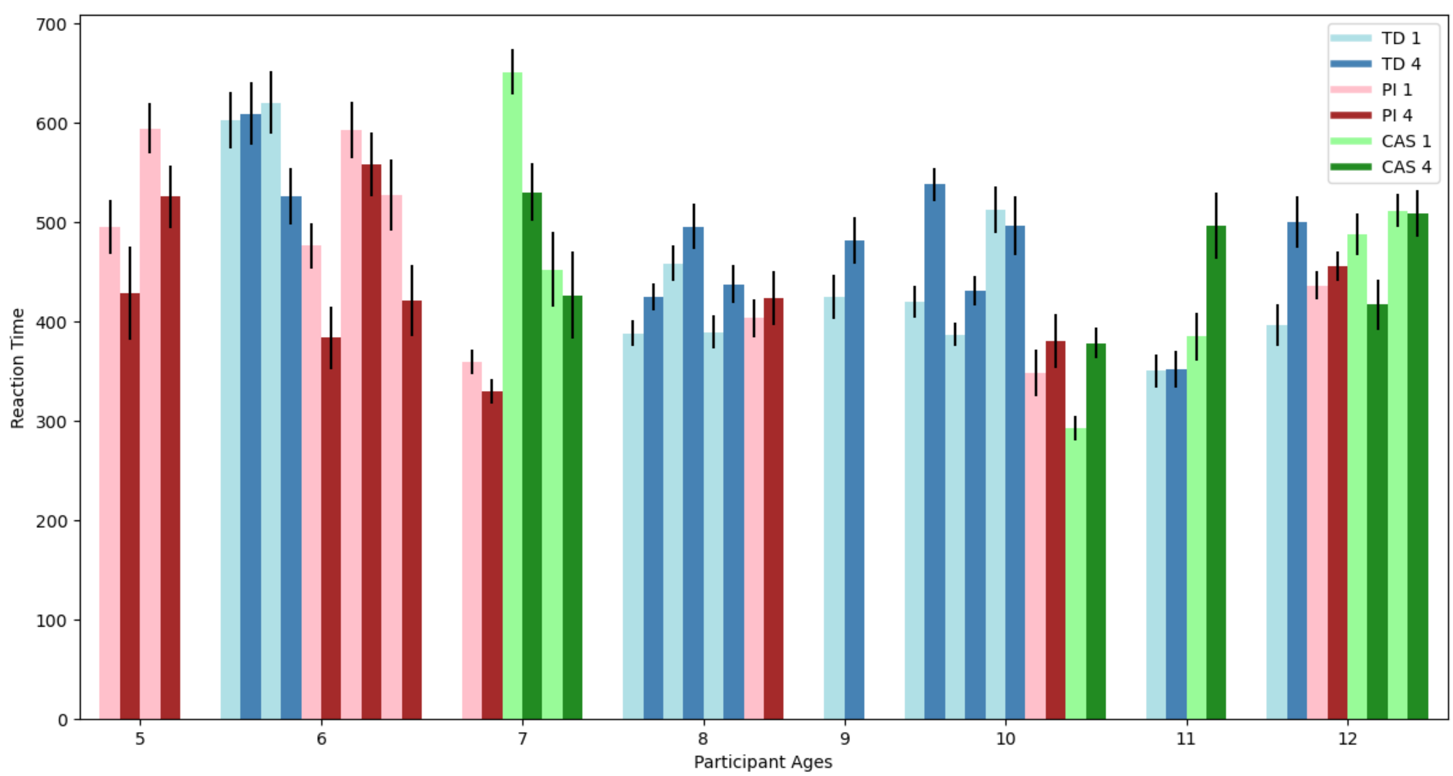


Note. TD=typically developing group, PI= phonological impairment group, CAS= childhood apraxia of speech group. 1=one syllable cue response, 4=four syllable cue response.

1 vs. 4 syllable cues RT

The RTs for each participant were similarly recorded for both one and four syllable cue types across each age (Figure 7). Interestingly, for RT it is not always the case that one-syllable cue responses are shorter than the corresponding four-syllable cues. With the exception of one TD participant, the rest of the participants across groups between the ages of 5 and 7 demonstrate longer RT for 1 syllable cue types than 4. This trend begins to shift at age 8, when for most participants across groups, RT becomes either approximately equal across response types, or longer for four-syllable response types. Another takeaway is that across groups, RT is relatively comparable. Unlike with ST, there is not a presence of an age effect, nor any substantial differences in RT between groups.

Figure 7. 1 vs. 4 syllable cues RT



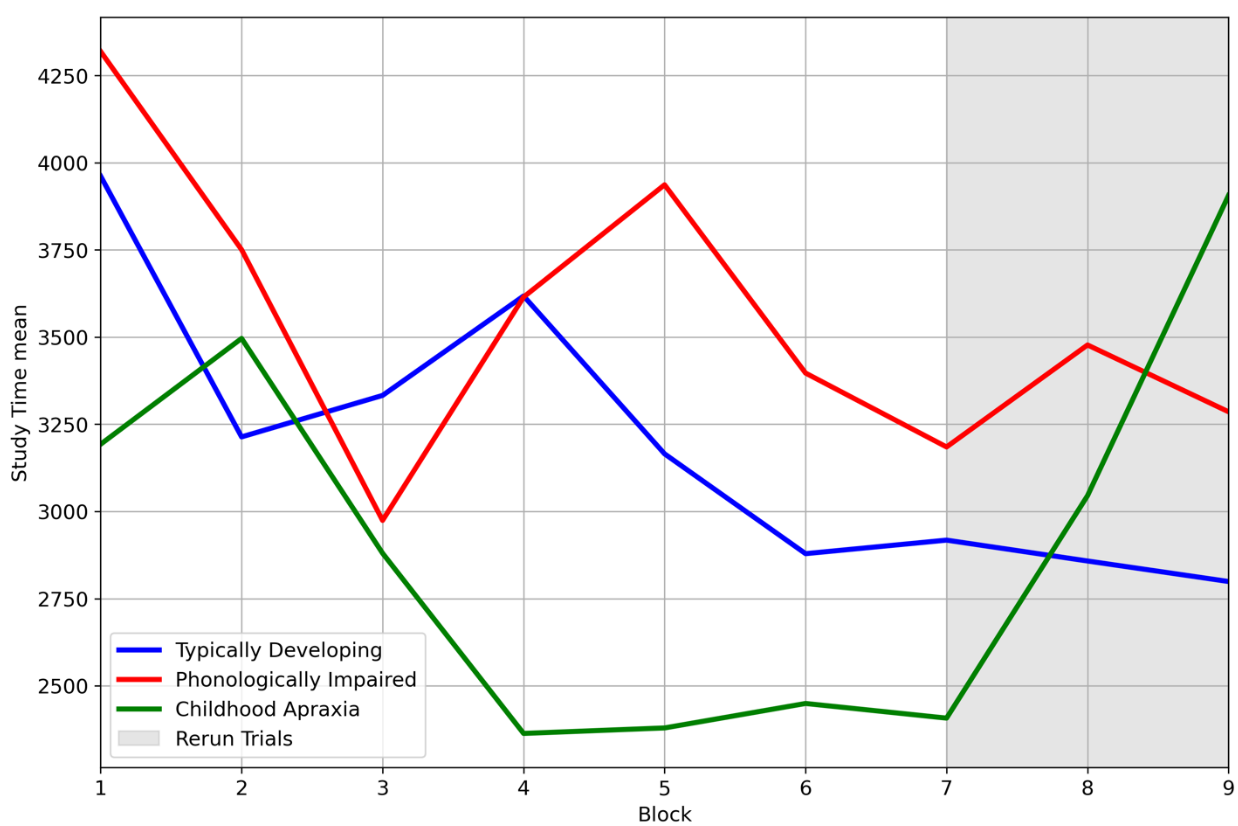
Note. TD=typically developing group, PI= phonological impairment group, CAS= childhood apraxia of speech group. 1=one syllable cue response, 4=four syllable cue response.

ST/RT across blocks

ST across blocks

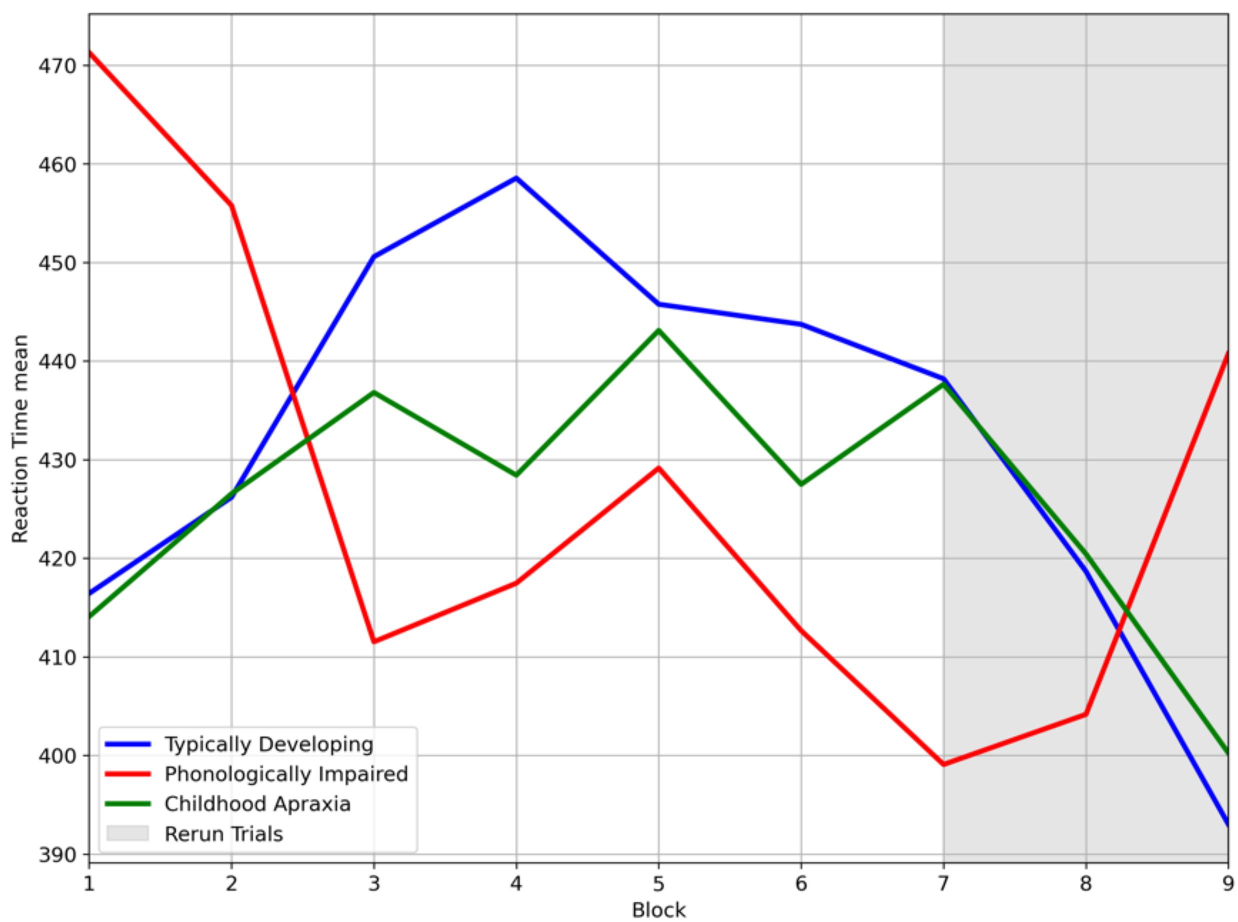
Average study time at each block was evaluated per group (Figure 8). Blocks were composed of 16 trials, with random presentation of each of the four cues (1s, 1L, 4s, 4L) four times. This was to determine the effects of learning or fatigue on ST over the course of the experiment. While all three groups demonstrate a significant decrease in study time as the blocks progress, it is interesting to note that this decrease begins immediately for TD and PI participants, while participants with CAS first demonstrate an increase in ST across the first blocks. Also notable, is that while TD and PI participants show a second peak in study time duration around blocks four and five, this is not the case with CAS. Participants with CAS maintain low study time across blocks four through seven. Finally, during the rerun trials TD and PI participants demonstrate relatively stable STs. Conversely, Participants with CAS show a dramatic increase in ST between blocks 7 and 9.

Figure 8. Mean group study time across blocks.



RT across blocks. Next, reaction time across blocks was calculated to determine whether there was an effect of learning or fatigue on RT across groups as the experiment progressed (Figure 9). Of note is that while TD participants and participants with CAS follow a similar trajectory across blocks, PI participants follow a different path. This is especially apparent during the rerun trials, where PI participants demonstrate a dramatic increase in RT, while TD and CAS participants demonstrate a decrease in RT.

Figure 9. Mean group reaction time across blocks



IV. Discussion

This study compared motor programming in children with apraxia of speech (CAS), those with phonological impairments (PI), and typically developing children. The main purpose was to identify whether the impairment in CAS is the same as that in adult, stroke-related AOS. To accomplish this, the study used the self-select paradigm to determine whether the ST (reflective of INT, a working memory buffer) and RT (reflective of SEQ, an on-line sequencing component), predicted impairments in ST in CAS and RT in PI.

Age and ST/RT

A primary finding of this study was that motor programming as represented by ST and RT showed a strong age effect in children with PI and those who are typically developing, but not those with CAS. In fact, in PI and TD groups the correlation line was a trajectory into adulthood based on data from Wright et al. (2009). These data show a developmental course for motor programming for which the processing time decreases from 5-young adulthood. However, the relatively flat developmental curve for the children with CAS, demonstrates difficulty developing motor programs at the same rate as children with PI or those that are typically developing. Given that ST reflects the processing demand placed on INT, these data support the findings in adults with AOS (Maas et al., 2008). These data demonstrate that over time, individuals with PI and those who are typically developing automatize the motor programming process for both ST and RT, likely because they become more efficient with speech programming. Thus, those two groups of participants can rapidly designate the internal spatiotemporal parameters of a particular movement in INT and then sequence the output rapidly. This also suggests that processing load and effort associated with this load remains relatively high in CAS. Moreover, given the slope of the developmental trajectory into adulthood, it is likely that individuals with CAS continue to feel high effort during speech production and struggle with developing a strong working memory buffer for motor units. These data demonstrate that without intervention (or the appropriate intervention) inefficient processing results in the need to use a higher amount of resources for computation and storage in working memory in CAS. These results demonstrate the need for treatments that target a rapid increase in speech sound development by reducing the load on INT.

The negative correlation that is seen for ST in TD participants is also seen with RT. This is notable because it may indicate that, like ST, as TD participants get older, the RT process becomes more efficient. For participants with CAS, because the output from the INT has not changed (i.e., has not become more efficient through the concatenation of individual syllables into words) there is a need to sequence the same number of individual units as the child grows older. This demonstrates the implications of the higher processing load associated with the impairment in INT, and again highlights the importance of treatment which is specific to the underlying impairment.

While observing the RT for each individual participant, we see that visually across all three groups RT is similar. This supports findings from Maas et al. (2008) where RT between AOS and typical controls is similar. It is also interesting to interpret the RT results with age in the light of the Wright et al. (2009) results of motor programming in adults. They found that the average RT for young adults was 557 ms. This is higher than the RT for any of the 12-year-old children who participated. This finding is interesting given the negative correlation between RT and age observed with TD participants. One reason for this discrepancy could be that in Wright et al. (2009) participants were provided with different stimuli, some of which were more complex. For example, participants in this study executed different patterns of the same syllable "ba." In Wright et al. (2009), patterns included repetitions of patterns containing different syllables (i.e., "ta-stra-ru-ta"). This additional complexity of target may translate to the observed longer RTs in adults than the children in this study.

ST/RT Reveals Increased Challenges in Last Two Blocks for Speech Sound Disorders

The last two blocks of the experimental testing served to rerun those trials that were inaccurate. Specifically, consistent with predictions, it was the CAS group that demonstrated much higher ST than the other two groups, indicating that when getting fatigued, the processing load on INT becomes quite high resulting in a breakdown in motor programming. Also consistent with our predictions, the children with CAS demonstrated a similar pattern for RT in the last two blocks as the typically developing children. Conversely, PI participants demonstrated an increase in RT during the last two blocks. The definition of apraxia of speech (adult and child) uses sequential errors as a differential symptom that excludes the diagnosis of apraxia but that is consistent with phonological errors. That the PI children were the only ones to have much greater processing load on the SEQ but not INT, provides empirical evidence in support of this differential criterion.

These findings support what was seen in the Maas et al. (2008) paper investigating motor programming in adults with AOS compared to typical controls and adults with aphasia, thereby supporting the hypothesis that the mechanism of action in CAS is the same as in stroke-related AOS. Differences between groups on various speech and/or language measures is likely due to when the apraxia emerged (from birth versus acquired later in life). The developmental trends for both INT and SEQ found here are consistent with the strengthening of the motor programming process over time, which is substantially diminished in CAS. This likely results in a reduction in learning associated with language and speech milestones. It should be noted that each of the participants with CAS had been enrolled in therapy for years, and yet did not demonstrate a decrease in the processing load on the INT process, which in turn, impacted the SEQ process. Given this information, it is critical that appropriate treatment approaches which target the mechanism of action in CAS be implemented.

Limitations and Future Directions

This study is limited by a relatively small number of participants in each group. Additionally, results from the CAS group are representative of children with a moderate to mild presentation of apraxia, who have been in speech therapy for years. The two children with severe CAS who were recruited were unable to complete the task. Therefore, a future research direction would be to modify the task, perhaps through incorporation of a longer training period. This would allow for the involvement of younger children with CAS or children with a more severe presentation of the disorder. Children would additionally benefit from a longer training period for the non-speech motor tasks, spread across several sessions. This would allow for the inclusion of non-speech motor programming data and could provide evidence regarding the hypothesis of CAS being a central, not modality-specific, disorder. Another limitation is that the age effect across participants was so pronounced that it was difficult to draw conclusions about ST/RT at the group level. Future directions would involve recruiting a large group of participants across a much small age range (1-2 years). This would allow for comparisons to be made at the group level. Despite the limitations, this data can be interpreted through the lens of previous work, namely Maas et al. (2008), to provide insight as to the nature of motor programming impairments in children with CAS.

Conclusions

Motor programming in children with CAS was investigated using a self-select paradigm (Wright et al., 2009) derived from the Klapp (2003) model. While TD and PI children demonstrated a decrease in ST with age, children with CAS did not show this age effect. Additionally, as the experiment progressed, participants with CAS demonstrated a dramatic increase in ST while TD and PI participants did not. Conversely, RT followed a similar pattern for TD and CAS participants. The increase in ST, indicating a higher processing load on the INT stage of motor programming, with comparable RT between CAS and TD participants is similar to what was found by Maas et al. (2008) in adults with AOS. These findings support the hypotheses that 1) the impairment in CAS is localized to the INT stage of motor programming, 2) that the SEQ phase of motor programming is intact, and 3) that CAS and AOS arise from the same mechanism of action.

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APPENDIX

1. Appendix A: Paradigm Instructions Script.

Appendix A: Paradigm Instructions Script

Welcome to the experiment! Press the spacebar when you are ready to hear the instructions. (Word “Welcome”)

This picture is of a listening ear. When this symbol is on the screen it is a signal to listen carefully to the directions that I am going to read to you! If you are ready to hear the directions press the spacebar! (Listening ear)

During this experiment, you are going to see one of four cues appear on the computer screen. These cues will tell you how you should say “ba” into the computer. The cues are 1s, 1L, 4s, and 4L. If 1s appears on the screen, you will say “ba” once for a short amount of time. If 1L appears, you will say “ba” once for a longer amount of time If 4s appears, you will say “ba” 4 times in a short-long-long-short pattern. If 4L appears you will say “ba” in a long-short-short-long pattern. (diagram with different length lines, listening ear in corner)

Do you have any questions? When you are ready to hear more, you can press the spacebar.

When the experiment begins, a picture of a “thumbs-up” hand is going to appear on the screen. This is the “ready” signal, and it means that you should get ready for the cue that is going to appear on the screen soon! After the thumbs-up signal the cue of 1s, 1L, 4s, or 4L will show up on the screen. Take your time looking at the cue and when you are ready to respond you will press the spacebar. (thumbs-up picture with listening ear in corner)

Questions? When you are ready you can press the spacebar.

When you see a picture of a green light appear on the screen, you can go ahead and respond by saying “ba” the number of times, and for how long the cue said to. Try to answer as quickly and correctly as you can! (green light picture with listening ear in corner)

Questions? When you are ready you can press the spacebar.

Once you have responded, you are going to get a message in green if you said “ba” correctly, and a message in red if you responded incorrectly. I will read to you and explain what the message in red means, and then you will hear a model of the correct response so that you can learn the targets. After the model, the “thumbs-up” signal will come up on the screen again and the next trial will begin. (Example of “correct!” in green and “too slow” in red with listening ear in corner)

Questions? When you are ready you can press the spacebar.

Next, we will hear models of the correct press responses for each of the 4 cues. The sounds you will hear represent how long, and in what order you will say “ba” Press the spacebar when you are ready to hear the models and remember to listen carefully! (Listening ear)

Now you are going to practice responding to the cues! This is like practicing a musical instrument or practicing a sport! The more that you practice these cues, the more you will learn about the correct way to respond! During this section, your responses will not count, but still try to answer as accurately as you can. At the end of this section, the experiment will begin.

Do you have any questions? Press the spacebar when you are ready to begin the practice session.

Now you are ready to begin the experiment! Pay close attention to the cues that appear on the screen and try to answer as quickly and accurately as possible. (Experiment pic)

Questions? Press the spacebar when you are ready to begin the experiment.