

Research Article

Sentence Length Effects on Intelligibility in Two Groups of Older Children With Neurodevelopmental Disorders

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https://doi.org/10.1044/2023_AJSLP-23-00093**ABSTRACT****Purpose:** The purpose of this study was to examine the impact of sentence length on intelligibility in two groups of older children with neurodevelopmental disabilities.**Method:** Nine children diagnosed with cerebral palsy (CP) and eight children diagnosed with Down syndrome (DS), between the ages of 8 and 17 years, repeated sentences varying in length from two to seven words. Three hundred forty adult listeners (20 listeners per child) provided orthographic transcriptions of children's speech, which were used to calculate intelligibility scores.**Results:** There was a significant main effect of sentence length on intelligibility for children with CP. Intelligibility significantly increased from two- and three-word sentences to four-, five-, and six-word sentences, then significantly decreased from four-, five-, and six-word sentences to seven-word sentences. There was a main effect of sentence length on intelligibility for children with DS. Intelligibility significantly increased from two-word sentences to four-, five-, and six-word sentences.**Conclusions:** The primary findings of this study include the following: (a) Unlike in typically developing children, sentence length continues to influence intelligibility well into adolescence for children with neurodevelopmental disorders, and (b) sentence length may influence intelligibility differently in children with CP than in children with DS; however, other factors besides the type of neurodevelopmental disorder (e.g., severity of speech motor involvement and/or cognitive-linguistic impairment) could play a role in the relationship between sentence length and intelligibility and must be investigated in future studies.

At least half of children with neurodevelopmental disorders, such as cerebral palsy (CP) and Down syndrome (DS), demonstrate decreased speech intelligibility (Hustad et al., 2020; Wild et al., 2018; Wilson et al., 2019b). Decreased speech intelligibility can negatively impact engagement in daily life activities that require communication (Connaghan et al., 2022; Pennington & McConachie, 2001), which may lead to loneliness and a host of negative mental and physical health consequences (Lyyra et al., 2018; Stickley et al., 2016; Tillinger, 2013).

Thus, it is no surprise that the primary goal of speech production-focused interventions for children with CP and DS is increased speech intelligibility (Moya-Galé et al., 2021; Pennington et al., 2010, 2013; Sakash et al., 2020; Swift & Rosin, 1990).

Intelligible speech does not mean that speech is perfect or “typical.” Rather, speech is considered intelligible when listeners are able “to map the acoustic signal onto the intended lexical units in spite of segmental- or suprasegmental-level problems” (Hustad et al., 2012, p. 1177). Intelligibility is often quantified by the percentage of words a listener is able to correctly identify from the speaker's message (e.g., Hustad et al., 2012). Intelligibility is a complex construct that is influenced by numerous

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factors including the speaker's impairment(s), the listener's ability to understand degraded speech, the environment in which the communication occurs, and the linguistic features of the message itself (e.g., Hustad & Borrie, 2021). Speech production-focused interventions designed for children with CP and DS focus almost exclusively on the use of speaker-centered approaches in which the child is taught to modify their speech production (e.g., increase vocal loudness) to enhance intelligibility (Pennington et al., 2016). An emerging area of interest in speech production-focused interventions is a focus on the length of the message the child produces. Changing the length of the message is an appealing compensatory strategy because it does not require the child to physically change the way their body produces speech, which may be difficult and unreliable.

Sentence length impacts intelligibility in young typically developing children. Starting around 4 years of age, multiword utterances are typically more intelligible than single-word utterances (Hustad et al., 2012, 2021). This is likely because multiword utterances provide more linguistic context for the listener, resulting in better intelligibility. Within multiword utterances, shorter sentences tend to be more intelligible than longer sentences (e.g., four words > seven words; Hustad et al., 2012). Thus, the increased speech motor demands of longer sentences may negate the benefit of increased linguistic context (Hustad et al., 2012). The impact of sentence length on intelligibility in young children with CP is influenced by the presence or absence of speech motor involvement (Allison & Hustad, 2014; Hustad et al., 2012). At 4 years of age, children with CP and no speech motor involvement have reduced intelligibility as compared with typically developing peers but follow similar patterns of intelligibility as it relates to sentence length (Hustad et al., 2012). Children with CP and speech motor involvement demonstrate descriptively different patterns than both typically developing children and children with CP and no speech motor involvement. At 4 years of age, children with CP and speech motor involvement do not appear to benefit from the added linguistic context of multiword utterances. Intelligibility was highest in single-word utterances with a tendency to decrease or plateau with each increase in sentence length (Hustad et al., 2012). It is possible that multiword utterances, regardless of the length of those utterances, were too taxing on the speech motor systems of such young children with CP.

Starting around 5 years of age, sentence length no longer impacts intelligibility in typically developing children (Hustad et al., 2021). This does not appear to hold true for 5-year-olds with CP with or without speech motor involvement as their fluctuations in intelligibility across multiword utterances are similar to those of 4-year-olds with CP (Allison & Hustad, 2014). There are no studies

examining the impact of sentence length on intelligibility in children with CP older than 5 years of age. It is unknown when or if children with CP reach an age at which sentence length no longer affects intelligibility. Intelligibility deficits in young children with CP often persist into adolescence and adulthood, indicating a need for speech production-focused intervention throughout the life span (Hustad, 2007; Hustad et al., 2019, 2020; Mahr et al., 2020; Schölderle et al., 2016). If individuals with CP reach a particular age at which sentence length no longer affects intelligibility, that information would help refine inclusionary criteria for interventions that utilize sentence length modifications to improve intelligibility.

There are no studies, to our knowledge, that examine the impact of sentence length on intelligibility in children with DS of any age. Similar to children with CP, intelligibility deficits that begin in early childhood often persist into adolescence and adulthood in individuals with DS (Barnes et al., 2009; Chapman et al., 1998; Rosin et al., 1988; Wild et al., 2018). However, it would be unwise to assume that children with DS will respond to interventions targeting sentence length as a way to improve intelligibility in the same manner as children with CP. Decreased intelligibility in children with DS is the result of a constellation of causes, including anatomic differences that impact the size and shape of the vocal tract (Rodrigues et al., 2019; Sforza et al., 2012), motor speech disorders (Kumin, 2006; Wilson et al., 2019a, 2019b), and cognitive-linguistic disorders (Chapman & Hesketh, 2001; Lukowski et al., 2019). Although there is considerable individual variability, these deficits are considered part of the clinical phenotype of DS, meaning that almost all children with DS will exhibit these deficits to some degree (Daunhauer & Fidler, 2011). On the other hand, the clinical phenotype for children with CP varies widely depending on the etiology of the disorder (Metz et al., 2022). For children with CP, speech motor involvement affects approximately 50% (Nordberg et al., 2013), cognitive deficits affect approximately 40% (Odding et al., 2006; Sigurdardottir et al., 2008), and language deficits affect 30%–74% (Mei et al., 2016; Otapowicz et al., 2005; Parkes et al., 2010; Pirila et al., 2007; Voorman et al., 2010). These deficits can appear individually, in combination, or not at all (Hustad et al., 2010).

Differences in the clinical phenotype of children with DS and children with CP may lead to differences in speech production behavior and response to intervention (Darling-White & Jaeger, 2023; Yoder & Warren, 2002). Of particular importance to this study, Darling-White and Jaeger (2023) found that sentence length impacted acoustic measures related to speech rate differently in older children with CP than in older children with DS. Articulation rate increased with increased sentence length in children

with CP but not in children with DS. Furthermore, children with DS paused for a greater amount of time with increases in sentence length, but children with CP did not. It is imperative to understand how sentence length impacts intelligibility in each group of children with neurodevelopmental disorders (CP and DS) in order to tailor speech production-focused interventions appropriately.

The purpose of this study was to examine the impact of sentence length on intelligibility in two groups of older children with neurodevelopmental disorders. Children were between the ages of 8 and 17 years and diagnosed with either CP or DS. Typically developing children were not included in this study, because we do not expect sentence length to impact intelligibility after 5 years of age (Hustad et al., 2021). Data from children with CP and children with DS were examined separately, and any differences between the groups are described descriptively. This approach will inform the design and implementation of etiology-specific interventions targeting sentence length as a way to improve speech intelligibility.

Based on the data from younger children with CP detailed above, it was hypothesized that older children with CP would demonstrate decreasing intelligibility with increasing sentence length. Given the lack of information about sentence length and intelligibility in children with DS regardless of age, it was not possible to form directional hypotheses regarding the performance of older children with DS. Based on the findings of Darling-White and Jaeger (2023), it was hypothesized that the relationship between sentence length and intelligibility would present differently in children with CP than in children with DS.

Method

Participants

Children With Neurodevelopmental Disorders

Nine children diagnosed with CP ($M_{\text{age}} = 12;9$ [years;months]) and eight children diagnosed with DS ($M_{\text{age}} = 13;2$) participated in this study. Children from both groups appear in previous publications (Darling-White, 2022; Darling-White & Jaeger, 2023; Kovacs & Darling-White, 2022), but the examination of intelligibility by sentence length in these children is unique to this study. Legal guardians provided written consent and participants provided verbal assent prior to data collection. Study procedures were approved by the University of Arizona Human Subjects Review Board (Protocol 16055837A005).

Participants were recruited via community postings, specialty medical clinics, and online forums. Inclusionary criteria for this study were as follows: (a) speak fluent

English and (b) be able to repeat sentences up to seven words in length. Two children with CP (M06CP and M08CP) and two children with DS (F03DS and M09DS) were bilingual (i.e., regularly used two languages; Grosjean, 1992). Three of these children were early bilinguals (i.e., learned both languages when they were younger than 5 years of age; Paradis et al., 2021) and spoke primarily Spanish in the home and English at school. One child, M08CP, was a Chinese-English bilingual and began learning English approximately 3 years prior to the study. English was M08CP's primary language at home and at school. There are no differences between bilingual and monolingual children with neurodevelopmental disorders on measures of language, cognition, or adaptive functioning when tested in the majority language (i.e., English) regardless of age of acquisition (Edgin et al., 2011; Kay-Raining Bird et al., 2005, 2016).

Demographic characteristics including age, race, ethnicity, language status, speech motor status, intelligibility, gross motor impairment (children with CP), and adaptive behavior skills (children with DS) are presented in Table 1. These demographic characteristics were not used as inclusionary or exclusionary criteria but, rather, are a way to characterize our sample to allow for cross-study comparison. For the majority of children, the presence or absence of language impairment was determined by the Core Language score of the Clinical Evaluation of Language Fundamentals-Fifth Edition (Wiig et al., 2013). In three instances, language impairment status was based on parent report. The first author, a certified speech-language pathologist, made a dichotomous classification of each child as having or not having speech motor involvement based on perceptual assessment during a variety of connected speech tasks (e.g., conversation, reading, and single-sentence production). The dichotomous classification system was based on the Speech Language Profile Groups paradigm developed by Hustad and colleagues for children with CP (e.g., Hustad et al., 2010). Severity of speech motor involvement was based on the mean intelligibility scores across sentence lengths. The procedure followed to obtain intelligibility scores is described below. Three levels of intelligibility were defined: high (no or mild speech motor involvement) = 81%–100%, moderate (moderate speech motor involvement) = 61%–80%, and low (severe speech motor involvement) = 0%–60% (Natzke et al., 2020).

All children with CP, except F09CP, passed a bilateral pure-tone hearing screening at 20 dB HL for 500, 1000, 2000, and 4000 Hz. Per parent report, there were no concerns regarding F09CP's hearing status and no history of failed hearing screenings. Only two of the children with DS participated in the hearing screening due to time constraints. M02DS passed the screening at all frequencies,

Table 1. Participant demographic information.

Participant	Age	Race/ethnicity	Language impairment	Speech motor impairment	Intelligibility	Type of CP	GMFCS	Adaptive Behavior Composite of Vineland-3
F01CP	13;5	White/non-Hispanic	Severe impairment	Yes	91% – High	Spastic diplegia	II	
F02CP	14;6	White/non-Hispanic	No impairment	No	96% – High	Spastic hemiplegia	I	
F03CP	16;11	Did not report/ Hispanic–Latino	Severe impairment	Yes	33% – Low	Spastic quadriplegia	II	
M04CP	11;9	More than one/ non-Hispanic	No impairment	No	93% – High	Spastic	I	
F05CP	14;10	White/non-Hispanic	No impairment	Yes	89% – High	Spastic	II	
M06CP	12;3	White/Hispanic– Latino	Severe impairment	Yes	91% – High	Spastic hemiplegia	I	
F07CP	9;2	White/non-Hispanic	Moderate impairment	Yes	65% – Moderate	Spastic quadriplegia	II	
M08CP	13;8	Asian/non-Hispanic	No impairment ^a	Yes	70% – Moderate	Spastic quadriplegia	II	
F09CP	8;3	White/non-Hispanic	Borderline impairment	Yes	74% – Moderate	Mixed – ataxic and hypotonic quadriplegia	III	
F01DS	16;1	White/Hispanic– Latino	Severe impairment	Yes	78% – Moderate			75
M02DS	11;10	Did not report/ Hispanic–Latino	Severe impairment	Yes	74% – Moderate			74
F03DS	17;11	White/Hispanic– Latino	Severe impairment	Yes	42% – Low			68
F04DS	10;7	White/non-Hispanic	Severe impairment	Yes	47% – Low			78
F05DS	13;6	White/non-Hispanic	Severe impairment	Yes	74% – Moderate			67
F06DS	12;2	White/non-Hispanic	Impairment ^a	Yes	35% – Low			65
F07DS	11;9	White/Hispanic– Latino	Severe impairment	Yes	76% – Moderate			63
M09DS	10;7	White/Hispanic– Latino	Impairment ^a	Yes	24% – Low			62

Note. Age is provided in years;months. Language impairment classifications are based on the Core Language score of the Clinical Evaluation of Language Fundamentals–Fifth Edition (Wiig et al., 2013) or parent report. M04CP and F05CP did not report the topographical distribution of their spasticity. Intelligibility is reported as the mean intelligibility across sentence lengths with no multitalker babble noise. Intelligibility ratings are as follows: High = 81% and up, Moderate = 61%–80%, and Low = 0%–60% (Natzke et al., 2020). Level I indicates little to no gross motor impairment. The Adaptive Behavior Composite of the Vineland-3 (Sparrow et al., 2016) is expressed as a standard score with a mean of 100 and an SD of 15. CP = cerebral palsy; F = female; M = male; DS = Down syndrome; GMFCS = Gross Motor Function Classification System (Palisano et al., 1997).

^aParent-reported language impairment, which was not rated by severity level.

and F05DS had an elevated threshold of 25 dB at 500 Hz in the right ear but passed at all other frequencies. F01DS and M09DS wore bilateral hearing aids, and their parents had no concerns about their hearing that were not addressed by the hearing aids. No parent concerns regarding hearing status were reported for the remaining five children with DS.

Adult Listeners

Three hundred forty adult listeners (20 listeners per child) participated in this study. Listeners provided orthographic transcriptions of the sentence-level Test of Children's Speech (TOCS+; Hodge & Daniels, 2009) described below as the basis for intelligibility measures. Listeners were recruited from Amazon Mechanical Turk (MTurk), an online crowdsourcing platform. The use of crowdsourcing platforms for auditory-perceptual studies in the speech sciences has been validated (Lansford et al., 2016; McAllister Byun et al., 2015; Ziegler et al., 2021) and is becoming more frequent in the literature (e.g., Borrie et al., 2017; Jiao et al., 2019; McAllister Byun, 2017; McAllister Byun et al., 2016; Nightingale et al., 2020). Participation was limited to MTurk workers designated by Amazon as Masters (i.e., have high approval ratings) with a U.S.-based IP address. Participation requirements were as follows: (a) use of Firefox, Chrome, or Safari browsers; (b) between the ages of 18 and 45 years; (c) native speaker of American English; (d) no history of speech, language, learning, or hearing disorders; (e) no more than incidental experience listening to children with speech sound disorders; and (f) a pair of headphones to wear while completing the task. The confirmation of these requirements was based on self-report.

Acquisition of Speech Samples From Children

Children wore an omnidirectional headset microphone (Shure WBH53) during the completion of the sentence-level TOCS+ (Hodge & Daniels, 2009). The acoustic signal was recorded via a digital audio recorder (Marantz PMD671) with a compact flash card, then transferred to a computer and resampled at 18 kHz with a low-pass filter at 9 kHz for anti-aliasing using GoldWave. Data were collected in a quiet space at the University of Arizona or at the child's home depending on parent preference. Children completed several speech tasks during the session, the order of which was counterbalanced. Speech tasks typically took between 30 and 45 min to complete. Frequent breaks were provided to prevent fatigue and increase compliance.

During the sentence-level TOCS+, the TOCS+ software generates a list of 34 sentences varying in length

from two to seven words from a pool of 2,000 phrases (Hodge & Daniels, 2009). The order of sentence length presentation is randomized. Prior to data collection, the TOCS+ software was used to create 30 unique lists. Each TOCS+ list had different numbers of sentences at each length, but a representative TOCS+ list contained four two-word, five three-word, six four-word, seven five-word, six six-word, and six seven-word sentences. No TOCS+ list was repeated within a group of children (CP and DS). In two instances, a child with CP and a child with DS produced the same TOCS+ list. TOCS+ sentences are appropriate for use with individuals with a developmental language age as young as 3 years.

Children repeated each stimulus sentence using their comfortable pitch and loudness following a prerecorded adult model. Stimulus sentences were prerecorded by the first author in a sound-attenuating booth. The text and accompanying audio of each stimulus sentence were presented via a laptop computer. Sentence repetition is a commonly used elicitation technique for intelligibility testing in children with neurodevelopmental disorders regardless of age (e.g., Allison & Hustad, 2014; Darling-White & Jaeger, 2023; Hustad et al., 2010, 2019; Wild et al., 2018). It is particularly useful for children with neurodevelopmental disorders who do not have the literacy skills necessary to read the stimulus sentence aloud without assistance, as was the case for many of the children with DS in this study. The first author monitored children's productions online to ensure collection of sentences that were as error free as possible. If errors (e.g., word omissions or substitutions, laughing or coughing during the sentence, forgetting the sentence halfway through and asking for clarification, overlap with the prerecorded adult model, background noise) were detected during data collection, the child was asked to repeat the sentence. If a repetition was necessary, children frequently produced the sentence correctly on the second attempt although a third attempt was occasionally required. All children except one, F04DS, produced sentences at each sentence length. F04DS did not produce any seven-word sentences. When seven-word sentences were presented, F04DS consistently omitted one or two words. The number of sentences included in the intelligibility analysis by sentence length and group is presented in Table 2.

Acquisition and Scoring of Intelligibility Data

Each stimulus sentence was separated into its own WAV file and amplitude-normalized via a customized MATLAB script (B. Story) prior to presentation. Stimulus sentences for each child were orthographically transcribed by 20 adult listeners. Stimulus sentences were presented via a Qualtrics survey, and the order of stimulus sentences

Table 2. Number of sentences included in intelligibility analysis by sentence length and group.

Sentence length	CP	DS
2 words	36	34
3 words	45	43
4 words	54	59
5 words	67	54
6 words	54	45
7 words	48	36

Note. CP = cerebral palsy; DS = Down syndrome.

was randomized for each listener. The task took approximately 30 min. Listeners were instructed to listen to each sentence and type the words that they heard (without the use of abbreviations) in the textbox provided. Stimulus sentences were only presented to each listener one time. Listeners were told they would only hear real English words and were encouraged to guess if they were unsure.

Prior to scoring the listener responses, the authors created a scoring key for each child by listening to each stimulus sentence, comparing it with the target sentence, and writing down the words the child produced. If the child was unintelligible, the target word or sentence was used. Listener responses were scored as correct if they were an exact phonemic match with the target. Homonyms and misspellings were counted as correct as long as they were an exact phonemic match with the target. A team of two to four undergraduate research assistants scored the listener responses. Each team member scored a particular response set individually, and then the group met to discuss their scores. If discrepancies arose, the team would discuss the discrepancy and make a group decision about the final score based on the scoring rules. Discrepancies were primarily due to human error (e.g., typos and miscalculations). A percent intelligibility score was calculated by dividing the total number of words correctly identified at each sentence length by the total number of words produced at each sentence length and multiplying by 100.

For five children with CP, their average percent intelligible score across all sentence lengths was 89% or higher (see Table 1). Given their high level of intelligibility and the strong possibility that differences in sentence length would not be detected due to ceiling effects, a decision was made to mix their stimulus sentences with multitalker babble noise using MATLAB and rerun the listening experiment. Based on pilot testing, 0 dB SNR (noise is the same level of intensity as the child's speech) was applied to each sentence. This ratio minimized floor and ceiling effects. The 0 dB SNR ratio reduced children's average intelligibility across all sentence lengths by an average of 33% (range: 23%–45%). For example, F01CP went from 91% intelligible to 46% intelligible after her

sentences were mixed with multitalker babble noise at 0 dB SNR. For these children, only the percent intelligibility scores from stimulus sentences mixed with noise were included in the statistical analyses outlined below.

We calculated the interrater reliability of intelligibility measurements via the intraclass correlation coefficient (ICC) using SPSS. The ICC examines the agreement between percent intelligibility scores across all sentence lengths for each listener of each child. Using an average-score, one-way random effects model, we found strong agreement among listeners for both groups, CP: ICC = .98, 95% confidence interval (CI) [.94, .99]; DS: ICC = .99, 95% CI [.99, 1].

Statistical Analysis

A general linear mixed-model analysis of variance was used to examine the relationship between intelligibility and sentence length within each group. For each child, the percent intelligibility scores obtained from each listener by sentence length (20 listeners per child) were used in the statistical model (as opposed to an average percent intelligibility score across listeners for each sentence length). Subject was modeled as a random effect. Sentence length was modeled as the fixed effect. Bonferroni post hoc comparisons were used to examine statistically significant pairwise comparisons. The main effect of utterance length was considered significant at an α level of .05. Based on Bonferroni correction procedures (.05/15), pairwise comparisons were considered significant at $\alpha \leq .003$.

Results

Figure 1 depicts mean intelligibility and standard error by sentence length for both groups of children.

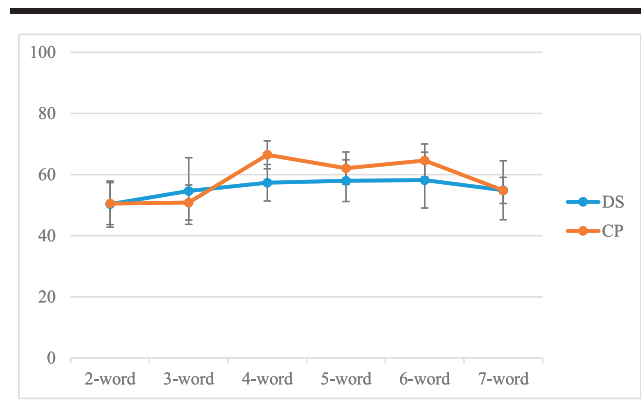
Children With CP

There was a significant main effect of sentence length on intelligibility for children with CP, $F(5, 1072) = 33.69$, $p < .001$. Post hoc testing revealed a significant difference between two- and three-word sentences and four-, five-, and six-word sentences as well as a significant difference between four-, five-, and six-word sentences and seven-word sentences (see Table 3). Intelligibility significantly increased from two- and three-word sentences to four-, five-, and six-word sentences, then significantly decreased from four-, five-, and six-word sentences to seven-word sentences.

Children With DS

There was a significant main effect of sentence length on intelligibility for children with DS, $F(5, 927.04) = 6.62$,

Figure 1. Mean intelligibility and standard error by sentence length and group. DS = Down syndrome; CP = cerebral palsy.



$p < .001$. Post hoc testing revealed a significant difference between two-word sentences and four-, five-, and six-word sentences (see Table 4). Intelligibility significantly increased from two-word sentences to four-, five-, and six-word sentences.

Discussion

The purpose of this study was to examine the impact of sentence length on intelligibility in two groups of older children with neurodevelopmental disorders. The primary findings of this study, which will be discussed in detail below, include the following: (a) Unlike in typically developing children, sentence length continues to influence intelligibility well into adolescence for children with

Table 3. Results of post hoc testing for the significant main effect of sentence length on intelligibility in children with cerebral palsy.

Contrast	Mean difference	SE	<i>p</i>
2 words vs. 3 words	−0.36	1.72	1
2 words vs. 4 words	−15.93	1.72	< .001*
2 words vs. 5 words	−11.56	1.72	< .001*
2 words vs. 6 words	−14.01	1.72	< .001*
2 words vs. 7 words	−4.22	1.72	.21
3 words vs. 4 words	−15.57	1.72	< .001*
3 words vs. 5 words	−11.20	1.72	< .001*
3 words vs. 6 words	−13.66	1.72	< .001*
3 words vs. 7 words	−3.86	1.72	.37
4 words vs. 5 words	4.37	1.72	.17
4 words vs. 6 words	1.92	1.72	1
4 words vs. 7 words	11.71	1.72	< .001*
5 words vs. 6 words	−2.45	1.72	1
5 words vs. 7 words	7.34	1.72	< .001*
6 words vs. 7 words	9.79	1.72	< .001*

* $p \leq .003$.

Table 4. Results of post hoc testing for the significant main effect of sentence length on intelligibility in children with Down syndrome.

Contrast	Mean difference	SE	<i>p</i>
2 words vs. 3 words	−4.29	1.68	.16
2 words vs. 4 words	−7	1.68	< .001*
2 words vs. 5 words	−7.68	1.68	< .001*
2 words vs. 6 words	−7.86	1.68	< .001*
2 words vs. 7 words	−3.63	1.75	.57
3 words vs. 4 words	−2.71	1.68	1
3 words vs. 5 words	−3.39	1.68	.65
3 words vs. 6 words	−3.57	1.68	.50
3 words vs. 7 words	0.67	1.75	1
4 words vs. 5 words	−0.68	1.68	1
4 words vs. 6 words	−0.86	1.68	1
4 words vs. 7 words	3.38	1.75	.80
5 words vs. 6 words	−0.18	1.68	1
5 words vs. 7 words	4.05	1.75	.31
6 words vs. 7 words	4.24	1.75	.23

* $p \leq .003$.

neurodevelopmental disorders, and (b) sentence length may influence intelligibility differently in children with CP than in children with DS.

Children With CP

The hypothesis that older children with CP would demonstrate decreasing intelligibility with increasing sentence length was partially supported by the results. Although there were some similarities with previous studies, sentence length seems to impact intelligibility in older children with CP differently than in younger children with CP, particularly for those with speech motor involvement. A key difference is the sentence length at which peak intelligibility was achieved. Within multiword utterances, young children with CP and speech motor involvement with or without language impairment demonstrated their highest levels of intelligibility in two- or three-word sentences, and then intelligibility decreased with each subsequent sentence length (Allison & Hustad, 2014; Hustad et al., 2012). Older children with CP demonstrated their highest levels of intelligibility during sentences in the middle range (four-, five-, and six-word sentences).

Unlike previous studies of the impact of sentence length on intelligibility, we were unable to separate our sample of children with CP based on the presence or absence of speech motor impairment and/or language impairment because of the small size of each group. Our sample included children with CP with and without speech motor involvement and children with CP and speech motor involvement with and without co-occurring

language impairment. Therefore, examining the patterns of individual means is of utmost importance (see Table 5). We examined the group and individual patterns through the lens of clinical significance as an additional check on the statistical results. Clinically meaningful results (i.e., results that likely reflect a true change in function) were defined as a difference in intelligibility greater than or equal to 10% (Allison, 2020).

Group results for children with CP indicated that two-, three-, and seven-word sentences were produced with significantly lower levels of intelligibility than four-, five-, and six-word sentences. There were no significant differences between two- and three-word sentences and seven-word sentences. As a group, all statistically significant comparisons, except five- and six-word sentences versus seven-word sentences, were also clinically significant. For all statistically significant comparisons, except one, at least five of the nine children with CP demonstrated a clinically significant difference in the same direction as the statistical result. The only exception was the difference between five-word sentences and seven-word sentences. Only three of the nine children with CP demonstrated a clinically significant difference in the same direction as the statistical result. Four-word sentences were produced with the highest levels of intelligibility for six of the nine children with CP. The other three children with CP produced their highest levels of intelligibility in five- or six-word sentences. Despite the variability, the individual patterns match the group-level statistics and indicate that most of the statistically significant changes were also clinically significant and likely reflect a true change in function.

One explanation for the difference in peak intelligibility between younger and older children with CP is development. Previous studies hypothesized that young children with CP and speech motor impairment were unable to capitalize on the added linguistic context of sentences longer than two or three words in length, because these sentences were overly burdensome for their developing

speech motor systems, particularly in the context of simultaneous language impairment (Allison & Hustad, 2014; Hustad et al., 2012). For example, 4-year-old children with CP and co-occurring speech motor involvement and language impairment were unable to repeat sentences greater than four words in length (Hustad et al., 2012). This could have been due to the fact that several sentence lengths were beyond the expected mean length of utterance in words for typically developing 4- and 5-year-olds (~4; Rice et al., 2010) and thus well beyond what would have been expected from a 4- or 5-year-old with language impairment. This study used the same speech task (i.e., sentences from the TOCS+) as the previous studies with young children with CP, but our participants were at least 3–4 years older. None of the participants had difficulty completing the speech task, and all sentence lengths were within the mean length of utterance expected for children over the age of 8 years (Lenhart et al., 2022). It is known that the majority of children with CP regardless of speech motor involvement or language impairment make gains in intelligibility with age, which have generally been attributed to the maturation of the speech motor, phonological, and cognitive–linguistic systems (Hustad et al., 2019, 2020; Mahr et al., 2020). Thus, older children with CP likely benefited from the added linguistic context of the sentences in the middle range regardless of speech motor involvement or language impairment because the speech task was less burdensome.

The benefit of added linguistic context with increasing sentence length was overridden once older children with CP reached the longest sentence length, however. One potential explanation for this phenomenon is the documented change in speech rate in response to sentence length in older children with CP. When acoustic measures related to speech rate were examined for the same sample of children performing the same task as presented in this study, Darling-White and Jaeger (2023) reported significantly increased articulation rate with increased sentence length for children with CP. Since the seven-word

Table 5. Mean intelligibility for each child with cerebral palsy by sentence length.

Participant	2 words	3 words	4 words	5 words	6 words	7 words
F01CP ^a	23%	35%	59%	51%	46%	47%
F02CP ^a	66%	49%	76%	59%	74%	48%
F03CP	13%	29%	38%	26%	41%	31%
M04CP ^a	42%	66%	79%	69%	76%	68%
F05CP ^a	59%	74%	59%	66%	75%	44%
M06CP ^a	56%	36%	61%	68%	46%	61%
F07CP	68%	39%	71%	67%	66%	68%
M08CP	56%	56%	72%	69%	85%	64%
F09CP	72%	72%	83%	83%	72%	63%

^aParticipants whose sentences were mixed with noise to prevent ceiling effects during the listening study.

sentences were produced with the fastest articulation rates, it is possible that seven-word sentences were perceived as less intelligible due to consequences of increased articulation rates, such as vowel centralization.

Children With DS

This is the first study, to our knowledge, to investigate the impact of sentence length on intelligibility in children with DS of any age. Although there was a statistically significant increase in intelligibility from two-word sentences to four-, five-, and six-word sentences, an examination involving individual data (see Table 6) and clinical significance levels reveals a much more complicated picture. None of the statistically significant group-level comparisons were clinically significant. Upon examination of individual data, it is apparent that there is no clear pattern or trend from child to child. In fact, no two children demonstrated the same pattern of intelligibility across sentence lengths. The highest intelligibility values were observed anywhere between three- and six-word sentences, whereas the lowest intelligibility values were observed across all sentence lengths except four-word sentences. For each statistically significant comparison, only two to four of the eight children with DS demonstrated a clinically significant difference in the same direction as the statistical result. In fact, for the comparison between two- and six-word sentences, the same number of children with DS demonstrated a clinically significant difference in the opposite direction as the statistical result as those who demonstrated a clinically significant difference in the same direction as the statistical result. Given the amount of individual variability and general lack of clinical significance, we conclude that sentence length did not systematically alter intelligibility in the sample of children with DS included in this study.

The hypothesis that the relationship between sentence length and intelligibility would present differently in children with CP than in children with DS was supported

by the results. Although the clinical phenotype of DS is much different than the clinical phenotype of CP, heterogeneity of speech impairment is an issue common in both types of disorders. According to auditory-perceptual and objective measures of speech production, speech impairments demonstrated by children with CP and children with DS do not conform to a syndrome-specific pattern and represent deficits across all speech subsystems (e.g., Allison & Hustad, 2018; Hustad et al., 2010, 2014; Jones et al., 2019; Kent et al., 2021). Despite this heterogeneity, the impact of sentence length on intelligibility was systematic in older children with CP (intelligibility increased from short to midlength sentences then decreased with the longest sentence), with the majority of children in our sample following similar patterns. Although intelligibility did fluctuate across sentence lengths, there was no discernable systematic pattern for the group of children with DS. There is clearly something unique about the impairment profile of DS that is interfering with a listener's ability to interpret running speech using traditional strategies like linguistic context.

It could be argued that a lack of change across sentence lengths in the children with DS is developmentally appropriate given that around 5 years of age, sentence length no longer impacts intelligibility in typically developing children (Hustad et al., 2021). However, sentence length likely stops being a significant factor in intelligibility around 5 years of age, because the average 5-year-old child is greater than or equal to 90% intelligible. The children with DS in this study all had significant intelligibility deficits, with half demonstrating peak intelligibility levels lower than 60%.

The patterns of acoustic change observed in children with DS in response to sentence length may be influencing intelligibility patterns. As discussed previously, Darling-White and Jaeger (2023) examined acoustic measures related to speech rate for the same sample of children performing the same task as presented in this study. Unlike in children with CP, increases in sentence length did not

Table 6. Mean intelligibility for each child with Down syndrome by sentence length.

Participant	2 words	3 words	4 words	5 words	6 words	7 words
F01DS	73%	96%	74%	60%	83%	80%
M02DS	43%	52%	70%	77%	86%	79%
F03DS	39%	45%	47%	54%	36%	34%
F04DS	55%	38%	44%	57%	49%	—
F05DS	73%	87%	69%	84%	64%	73%
F06DS	43%	10%	50%	35%	37%	34%
F07DS	68%	83%	74%	70%	88%	66%
M09DS	10%	26%	30%	28%	22%	18%

Note. The em dash indicates data not obtained, as F04DS did not produce any seven-word sentences.

impact articulation rate but did impact pausing in children with DS. Children with DS paused for a greater amount of time with increases in sentence length. If increases in pause time were a primary driver to intelligibility in this task, we would have seen more systematic changes in intelligibility (regardless of the direction of change) in the children with DS. As observed with the children with CP, patterns of change to articulation rate (or lack thereof) seem to be similar to the patterns of change (or lack thereof) in intelligibility.

The lack of a systematic response to sentence length could also be due to the way in which the speech motor and cognitive–linguistic systems interact in children with DS. All children with DS demonstrated moderate to low levels of intelligibility, indicating that speech motor involvement was likely a significant factor in performance. Most of the children with CP had high levels of intelligibility, indicating no or mild speech motor involvement. Although cognitive status was not specifically tested in this study, cognitive impairment is part of the clinical phenotype of DS, with verbal short-term memory being an area of particular challenge (Chapman & Hesketh, 2001; Daunhauer & Fidler, 2011; Jarrold et al., 2004). Cognitive impairment is much less frequent in children with CP (Odding et al., 2006; Sigurdardottir et al., 2008). During the sentence repetition task, the sentence was presented both visually and verbally. However, many of the children with DS were forced to rely solely on their verbal short-term memory skills to complete the task, because they did not have the literacy skills to utilize the written sentence as a memory aide. This was not the case with the children with CP in our sample. Furthermore, the longer sentences may have been beyond the expected mean length of utterance in words for some of the children with DS (Chapman et al., 1998; Miles et al., 2006). All of the children with DS in our sample had language impairment, with the majority having severe language impairment. Only five of nine children with CP in our sample demonstrated language impairment, and only three of those five had severe language impairment. It is likely that the speech task was much more difficult for the children with DS from both a speech motor and cognitive–linguistic perspective. Thus, it is possible that the demands of the task interfered with the ability of the speech motor and cognitive–linguistic systems to coordinate with one another in a typical fashion.

Limitations and Future Directions

Both groups, CP and DS, are heterogeneous, and it is difficult to form definitive conclusions about the impact of sentence length on intelligibility on older children with neurodevelopmental disorders from the small sample presented in this study. This is particularly true since several

of the children with CP were intelligible enough to require the use of multitalker babble noise mixed with their sentences to prevent ceiling effects in the listening study. Due to its size, our sample does not include the full spectrum of speech motor and cognitive–linguistic impairments that are present in these groups of children. A data set representative of a wide range of speech motor and cognitive–linguistic impairments within each population may reveal more nuanced information about the interaction of sentence length and intelligibility. This type of representative data set may also help refine inclusionary and exclusionary criteria for interventions that utilize sentence length modifications to improve intelligibility. This study included mostly children with CP who demonstrated high levels of intelligibility (without multitalker babble), indicating no or mild speech motor involvement. Although the children with CP who demonstrated moderate or low levels of intelligibility followed similar patterns of clinical and statistical significance, it is unclear if this will be the case for the majority of children with CP and moderate to severe speech motor involvement.

The intelligibility data in this study mimic the articulation rate data of Darling-White and Jaeger (2023) for both children with CP and children with DS, strengthening the hypothesis that intelligibility changes with sentence length are related to articulation rate. Pausing behavior, on the other hand, does not appear to be a significant factor in intelligibility in this sentence repetition task, since there was no change in intelligibility with increases in pausing behavior for children with DS and children with CP did not alter pausing behavior. Future work should seek to elucidate the relationship between changes in acoustic measures of speech production across sentence lengths and changes in intelligibility across sentence lengths. This type of investigation is missing from the literature and could shed light on the apparent lack of intelligibility changes across sentence lengths in children with DS.

Currently, it is unclear if sentence length is the primary driver of the intelligibility changes (or lack thereof) in this study. Other sentence-level factors, such as the phonetic characteristics of each sentence, may impact intelligibility (Allison & Hustad, 2014). Allison and Hustad (2014) found that both sentence length and phonetic complexity were significant predictors of intelligibility in 5-year-old children with CP and speech motor involvement. Interestingly, the effect each sentence-level factor had on intelligibility varied widely based on the individual child. Sentence length contributed independently of phonetic complexity in some younger children with CP and not others. Future large-scale studies should examine multiple sentence-level factors, in addition to sentence length, to provide a clearer understanding of the role sentence-level factors play in intelligibility.

Clinical Implications

Training older children with CP to communicate using messages between four and six words in length may be an effective compensatory strategy to improve intelligibility in this population. Speaking in these midrange sentence lengths improved intelligibility to a clinically significant degree (greater than or equal to 10%) for most of the children with CP in this study. Caution may be needed when using this approach with children with CP and moderate to severe speech motor involvement as most children with CP in this study had no or mild speech motor involvement. More work needs to be done to investigate the likelihood of intervention success for children with CP across the speech motor severity spectrum.

The strategy of altering sentence length to improve intelligibility is unlikely to work with older children with DS. Fluctuations in intelligibility across sentence lengths were observed in our sample of children with DS, but they were not systematic enough to create a specific intervention approach. This is now the second study to report etiology-specific differences in the relationship between sentence length and speech production using a speech task with high ecological validity. These data support the idea that etiology-specific adaptations may be necessary when designing and implementing speech production-focused interventions for children with neurodevelopmental disorders (Darling-White & Jaeger, 2023).

Data Availability Statement

The datasets generated during and/or analyzed during the current study are available from the corresponding author on reasonable request.

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References

- Allison, K. M. (2020). Measuring speech intelligibility in children with motor speech disorders. *Perspectives of the ASHA Special Interest Groups*, 5(4), 809–820. https://doi.org/10.1044/2020_PERSP-19-00110
- Allison, K. M., & Hustad, K. C. (2014). Impact of sentence length and phonetic complexity on intelligibility of 5-year-old children with cerebral palsy. *International Journal of Speech-Language Pathology*, 16(4), 396–407. <https://doi.org/10.3109/17549507.2013.876667>
- Allison, K. M., & Hustad, K. C. (2018). Data-driven classification of dysarthria profiles in children with cerebral palsy. *Journal of Speech, Language, and Hearing Research*, 61(12), 2837–2853. https://doi.org/10.1044/2018_JSLHR-S-17-0356
- Barnes, E., Roberts, J., Long, S. H., Martin, G. E., Berni, M. C., Mandulak, K. C., & Sideris, J. (2009). Phonological accuracy and intelligibility in connected speech of boys with fragile X syndrome or Down syndrome. *Journal of Speech, Language, and Hearing Research*, 52(4), 1048–1061. [https://doi.org/10.1044/1092-4388\(2009\)08-0001](https://doi.org/10.1044/1092-4388(2009)08-0001)
- Borrie, S. A., Lansford, K. L., & Barrett, T. S. (2017). Generalized adaptation to dysarthric speech. *Journal of Speech, Language, and Hearing Research*, 60(11), 3110–3117. https://doi.org/10.1044/2017_JSLHR-S-17-0127
- Chapman, R., & Hesketh, L. J. (2001). Language, cognition, and short-term memory in individuals with Down syndrome. *Down Syndrome Research and Practice*, 7(1), 1–7. <https://doi.org/10.3104/reviews.108>
- Chapman, R. S., Seung, H.-K., Schwartz, S. E., & Kay-Raining Bird, E. (1998). Language skills of children and adolescents with Down syndrome II. Production deficits. *Journal of Speech, Language, and Hearing Research*, 41(4), 861–873. <https://doi.org/10.1044/jslhr.4104.861>
- Connaghan, K. P., Baylor, C., Romanczyk, M., Rickwood, J., & Bedell, G. (2022). Communication and social interaction experiences of youths with congenital motor speech disorders. *American Journal of Speech-Language Pathology*, 31(6), 2609–2627. https://doi.org/10.1044/2022_AJSLP-22-00034
- Darling-White, M. (2022). Comparison of respiratory calibration methods for the estimation of lung volume in children with and without neuromotor disorders. *Journal of Speech, Language, and Hearing Research*, 65(2), 525–537. https://doi.org/10.1044/2021_JSLHR-21-00333
- Darling-White, M., & Jaeger, A. (2023). Differential impacts of sentence length on speech rate in two groups of children with neurodevelopmental disorders. *American Journal of Speech-Language Pathology*, 32(3), 1083–1098. https://doi.org/10.1044/2022_AJSLP-22-00209
- Daunhauer, L. A., & Fidler, D. J. (2011). The Down syndrome behavioral phenotype: Implications for practice and research in occupational therapy. *Occupational Therapy in Health Care*, 25(1), 7–25. <https://doi.org/10.3109/07380577.2010.535601>
- Edgin, J. O., Kumar, A., Spanò, G., & Nadel, L. (2011). Neuropsychological effects of second language exposure in Down

- syndrome. *Journal of Intellectual Disability Research*, 55(3), 351–356. <https://doi.org/10.1111/j.1365-2788.2010.01362.x>
- Grosjean, F. (1992). Another view of bilingualism. *Advances in Psychology*, 83, 51–62. [https://doi.org/10.1016/S0166-4115\(08\)61487-9](https://doi.org/10.1016/S0166-4115(08)61487-9)
- Hodge, M. M., & Daniels, J. (2009). *TOCS+ intelligibility measures* (Version 5.3) [Computer software]. University of Alberta.
- Hustad, K. C. (2007). Effects of speech stimuli and dysarthria severity on intelligibility scores and listener confidence ratings for speakers with cerebral palsy. *Folia Phoniatrica et Logopaedica*, 59(6), 306–317. <https://doi.org/10.1159/000108337>
- Hustad, K. C., Allison, K., McFadd, E., & Riehle, K. (2014). Speech and language development in 2-year-old children with cerebral palsy. *Developmental Neuropsychology*, 17(3), 167–175. <https://doi.org/10.3109/17518423.2012.747009>
- Hustad, K. C., & Borrie, S. A. (2021). Intelligibility impairment. In J. S. Damico, N. Müller, & M. J. Ball (Eds.), *The handbook of language and speech disorders* (1st ed., pp. 81–94). Wiley. <https://doi.org/10.1002/9781119606987.ch4>
- Hustad, K. C., Gorton, K., & Lee, J. (2010). Classification of speech and language profiles in 4-year-old children with cerebral palsy: A prospective preliminary study. *Journal of Speech, Language, and Hearing Research*, 53(6), 1496–1513. [https://doi.org/10.1044/1092-4388\(2010/09-0176\)](https://doi.org/10.1044/1092-4388(2010/09-0176))
- Hustad, K. C., Mahr, T. J., Broman, A. T., & Rathouz, P. J. (2020). Longitudinal growth in single-word intelligibility among children with cerebral palsy from 24 to 96 months of age: Effects of speech-language profile group membership on outcomes. *Journal of Speech, Language, and Hearing Research*, 63(1), 32–48. https://doi.org/10.1044/2019_JSLHR-19-00033
- Hustad, K. C., Mahr, T. J., Natzke, P., & Rathouz, P. J. (2021). Speech development between 30 and 119 months in typical children I: Intelligibility growth curves for single-word and multiword productions. *Journal of Speech, Language, and Hearing Research*, 64(10), 3707–3719. https://doi.org/10.1044/2021_JSLHR-21-00142
- Hustad, K. C., Sakash, A., Natzke, P. E. M., Broman, A. T., & Rathouz, P. J. (2019). Longitudinal growth in single word intelligibility among children with cerebral palsy from 24 to 96 months of age: Predicting later outcomes from early speech production. *Journal of Speech, Language, and Hearing Research*, 62(6), 1599–1613. https://doi.org/10.1044/2018_JSLHR-S-18-0319
- Hustad, K. C., Schueler, B., Schultz, L., & DuHadway, C. (2012). Intelligibility of 4-year-old children with and without cerebral palsy. *Journal of Speech, Language, and Hearing Research*, 55(4), 1177–1189. [https://doi.org/10.1044/1092-4388\(2011/11-0083\)](https://doi.org/10.1044/1092-4388(2011/11-0083))
- Jarrold, C., Cowan, N., Hewes, A. K., & Riby, D. M. (2004). Speech timing and verbal short-term memory: Evidence for contrasting deficits in Down syndrome and Williams syndrome. *Journal of Memory and Language*, 51(3), 365–380. <https://doi.org/10.1016/j.jml.2004.06.007>
- Jiao, Y., LaCross, A., Berisha, V., & Liss, J. (2019). Objective intelligibility assessment by automated segmental and supra-segmental listening error analysis. *Journal of Speech, Language, and Hearing Research*, 62(9), 3359–3366. https://doi.org/10.1044/2019_JSLHR-S-19-0119
- Jones, H. N., Crisp, K. D., Kuchibhatla, M., Mahler, L., Risoli, T., Jones, C. W., & Kishnani, P. (2019). Auditory-perceptual speech features in children with Down syndrome. *American Journal on Intellectual and Developmental Disabilities*, 124(4), 324–338. <https://doi.org/10.1352/1944-7558-124.4.324>
- Kay-Raining Bird, E., Cleave, P., Trudeau, N., Thordardottir, E., Sutton, A., & Thorpe, A. (2005). The language abilities of bilingual children with Down syndrome. *American Journal of Speech-Language Pathology*, 14(3), 187–199. [https://doi.org/10.1044/1058-0360\(2005/019\)](https://doi.org/10.1044/1058-0360(2005/019))
- Kay-Raining Bird, E., Genesee, F., & Verhoeven, L. (2016). Bilingualism in children with developmental disorders: A narrative review. *Journal of Communication Disorders*, 63, 1–14. <https://doi.org/10.1016/j.jcomdis.2016.07.003>
- Kent, R. D., Eichhorn, J., Wilson, E. M., Suk, Y., Bolt, D. M., & Vorperian, H. K. (2021). Auditory-perceptual features of speech in children and adults with Down syndrome: A speech profile analysis. *Journal of Speech, Language, and Hearing Research*, 64(4), 1157–1175. https://doi.org/10.1044/2021_JSLHR-20-00617
- Kovacs, S., & Darling-White, M. (2022). A descriptive study of speech breathing in children with cerebral palsy during two types of connected speech tasks. *Journal of Speech, Language, and Hearing Research*, 65(12), 4557–4576. https://doi.org/10.1044/2022_JSLHR-22-00295
- Kumin, L. (2006). Speech intelligibility and childhood verbal apraxia in children with Down syndrome. *Down Syndrome Research and Practice*, 10(1), 10–22. <https://doi.org/10.3104/reports.301>
- Lansford, K. L., Borrie, S. A., & Bystricky, L. (2016). Use of crowdsourcing to assess the ecological validity of perceptual-training paradigms in dysarthria. *American Journal of Speech-Language Pathology*, 25(2), 233–239. https://doi.org/10.1044/2015_AJSLP-15-0059
- Lenhart, M. H., Timler, G. R., Pavelko, S. L., Bronaugh, D. A., & Dudding, C. C. (2022). Syntactic complexity across language sampling contexts in school-age children, ages 8–11 years. *Language, Speech, and Hearing Services in Schools*, 53(4), 1168–1176. https://doi.org/10.1044/2022_LSHSS-21-00187
- Lukowski, A. F., Milojevich, H. M., & Eales, L. (2019). Cognitive functioning in children with Down syndrome: Current knowledge and future directions. *Advances in Child Development and Behavior*, 56, 257–289. <https://doi.org/10.1016/bs.acdb.2019.01.002>
- Lyyra, N., Välimaa, R., & Tynjälä, J. (2018). Loneliness and subjective health complaints among school-aged children. *Scandinavian Journal of Public Health*, 46(Suppl. 20), 87–93. <https://doi.org/10.1177/1403494817743901>
- Mahr, T. J., Rathouz, P. J., & Hustad, K. C. (2020). Longitudinal growth in intelligibility of connected speech from 2 to 8 years in children with cerebral palsy: A novel Bayesian approach. *Journal of Speech, Language, and Hearing Research*, 63(9), 2880–2893. https://doi.org/10.1044/2020_JSLHR-20-00181
- McAllister Byun, T. (2017). Efficacy of visual-acoustic biofeedback intervention for residual rhotic errors: A single-subject randomization study. *Journal of Speech, Language, and Hearing Research*, 60(5), 1175–1193. https://doi.org/10.1044/2016_JSLHR-S-16-0038
- McAllister Byun, T., Halpin, P. F., & Szeredi, D. (2015). Online crowdsourcing for efficient rating of speech: A validation study. *Journal of Communication Disorders*, 53, 70–83. <https://doi.org/10.1016/j.jcomdis.2014.11.003>
- McAllister Byun, T., Harel, D., Halpin, P. F., & Szeredi, D. (2016). Deriving gradient measures of child speech from crowdsourced ratings. *Journal of Communication Disorders*, 64, 91–102. <https://doi.org/10.1016/j.jcomdis.2016.07.001>
- Mei, C., Reilly, S., Reddihough, D., Mensah, F., Pennington, L., & Morgan, A. (2016). Language outcomes of children with cerebral palsy aged 5 years and 6 years: A population-based

- study. *Developmental Medicine & Child Neurology*, 58(6), 605–611. <https://doi.org/10.1111/dmcn.12957>
- Metz, C., Jaster, M., Walch, E., Sarpong-Bengelsdorf, A., Kaindl, A. M., & Schneider, J. (2022). Clinical phenotype of cerebral palsy depends on the cause: Is it really cerebral palsy? A retrospective study. *Journal of Child Neurology*, 37(2), 112–118. <https://doi.org/10.1177/08830738211059686>
- Miles, S., Chapman, R., & Sindberg, H. (2006). Sampling context affects MLU in the language of adolescents with Down syndrome. *Journal of Speech, Language, and Hearing Research*, 49(2), 325–337. [https://doi.org/10.1044/1092-4388\(2006/026\)](https://doi.org/10.1044/1092-4388(2006/026))
- Moya-Galé, G., Keller, B., Escorial, S., & Levy, E. S. (2021). Speech treatment effects on narrative intelligibility in French-speaking children with dysarthria. *Journal of Speech, Language, and Hearing Research*, 64(6S), 2154–2168. https://doi.org/10.1044/2020_JSLHR-20-00258
- Natzke, P., Sakash, A., Mahr, T., & Hustad, K. C. (2020). Measuring speech production development in children with cerebral palsy between 6 and 8 years of age: Relationships among measures. *Language, Speech, and Hearing Services in Schools*, 51(3), 882–896. https://doi.org/10.1044/2020_LSHSS-19-00102
- Nightingale, C., Swartz, M., Ramig, L. O., & McAllister, T. (2020). Using crowdsourced listeners' ratings to measure speech changes in hypokinetic dysarthria: A proof-of-concept study. *American Journal of Speech-Language Pathology*, 29(2), 873–882. https://doi.org/10.1044/2019_AJSLP-19-00162
- Nordberg, A., Miniscalco, C., Lohmander, A., & Himmelmann, K. (2013). Speech problems affect more than one in two children with cerebral palsy: Swedish population-based study. *Acta Paediatrica*, 102(2), 161–166. <https://doi.org/10.1111/apa.12076>
- Odding, E., Roebroek, M. E., & Stam, H. J. (2006). The epidemiology of cerebral palsy: Incidence, impairments and risk factors. *Disability and Rehabilitation*, 28(4), 183–191. <https://doi.org/10.1080/09638280500158422>
- Otapowicz, D., Sobaniec, W., & Okurowska-Zawada, B. (2005). Time of cooing appearance and further development of speech in children with cerebral palsy. *Roczniki Akademii Medycznej w Białymstoku*, 50(Suppl. 1), 78–81.
- Palisano, R., Rosenbaum, P., Walter, S., Russell, D., Wood, E., & Galuppi, B. (1997). Development and reliability of a system to classify gross motor function in children with cerebral palsy. *Developmental Medicine & Child Neurology*, 39(4), 214–223. <https://doi.org/10.1111/j.1469-8749.1997.tb07414.x>
- Paradis, J., Genesee, F., & Crago, M. (2021). *Dual language development & disorders: A handbook on bilingualism and second language learning* (3rd ed.). Brookes.
- Parkes, J., Hill, N., Platt, M. J., & Donnelly, C. (2010). Oromotor dysfunction and communication impairments in children with cerebral palsy: A register study. *Developmental Medicine & Child Neurology*, 52(12), 1113–1119. <https://doi.org/10.1111/j.1469-8749.2010.03765.x>
- Pennington, L., & McConachie, H. (2001). Interaction between children with cerebral palsy and their mothers: The effects of speech intelligibility. *International Journal of Language & Communication Disorders*, 36(3), 371–393. <https://doi.org/10.1080/13682820110045847>
- Pennington, L., Miller, N., Robson, S., & Steen, N. (2010). Intensive speech and language therapy for older children with cerebral palsy: A systems approach. *Developmental Medicine & Child Neurology*, 52(4), 337–344. <https://doi.org/10.1111/j.1469-8749.2009.03366.x>
- Pennington, L., Parker, N. K., Kelly, H., & Miller, N. (2016). Speech therapy for children with dysarthria acquired before three years of age. *Cochrane Database of Systematic Reviews*, 7(7), Article CD006937. <https://doi.org/10.1002/14651858.CD006937.pub3>
- Pennington, L., Roelant, E., Thompson, V., Robson, S., Steen, N., & Miller, N. (2013). Intensive dysarthria therapy for younger children with cerebral palsy. *Developmental Medicine & Child Neurology*, 55(5), 464–471. <https://doi.org/10.1111/dmcn.12098>
- Pirila, S., van der Meere, J., Pentikainen, T., Ruusu-Niemi, P., Korpela, R., Kilpinen, J., & Nieminen, P. (2007). Language and motor speech skills in children with cerebral palsy. *Journal of Communication Disorders*, 40(2), 116–128. <https://doi.org/10.1016/j.jcomdis.2006.06.002>
- Rice, M. L., Smolik, F., Perpich, D., Thompson, T., Rytting, N., & Blossom, M. (2010). Mean length of utterance levels in 6-month intervals for children 3 to 9 years with and without language impairments. *Journal of Speech, Language, and Hearing Research*, 53(2), 333–349. [https://doi.org/10.1044/1092-4388\(2009/08-0183\)](https://doi.org/10.1044/1092-4388(2009/08-0183))
- Rodrigues, M., Nunes, J., Figueiredo, S., Martins de Campos, A., & Geraldo, A. F. (2019). Neuroimaging assessment in Down syndrome: A pictorial review. *Insights into Imaging*, 10(1), Article 52. <https://doi.org/10.1186/s13244-019-0729-3>
- Rosin, M. M., Swift, E., Bless, D., & Kluppel Vetter, D. (1988). Communication profiles of adolescents with Down syndrome. *Communication Disorders Quarterly*, 12(1), 49–64. <https://doi.org/10.1177/152574018801200105>
- Sakash, A., Mahr, T. J., Natzke, P. E. M., & Hustad, K. C. (2020). Effects of rate manipulation on intelligibility in children with cerebral palsy. *American Journal of Speech-Language Pathology*, 29(1), 127–141. https://doi.org/10.1044/2019_AJSLP-19-0047
- Schölderle, T., Staiger, A., Lampe, R., Strecker, K., & Ziegler, W. (2016). Dysarthria in adults with cerebral palsy: Clinical presentation and impacts on communication. *Journal of Speech, Language, and Hearing Research*, 59(2), 216–229. https://doi.org/10.1044/2015_JSLHR-S-15-0086
- Sforza, C., Dellavia, C., Allievi, C., Tommasi, D. G., & Ferrario, V. F. (2012). Anthropometric indices of facial features in Down's syndrome subjects. In V. R. Preedy (Ed.), *Handbook of anthropometry: Physical measures of human form in health and disease* (pp. 1603–1618). Springer. https://doi.org/10.1007/978-1-4419-1788-1_98
- Sigurdardottir, S., Eiríksdóttir, A., Gunnarsdóttir, E., Meintema, M., Arnadóttir, U., & Vik, T. (2008). Cognitive profile in young Icelandic children with cerebral palsy. *Developmental Medicine & Child Neurology*, 50(5), 357–362. <https://doi.org/10.1111/j.1469-8749.2008.02046.x>
- Sparrow, S. S., Cicchetti, D. V., & Saulneir, C. A. (2016). Vineland Adaptive Behavior Scales, Third Edition (Vineland-3). Pearson.
- Stickley, A., Koyanagi, A., Kaposov, R., Blatný, M., Hrdlička, M., Schwab-Stone, M., & Ruchkin, V. (2016). Loneliness and its association with psychological and somatic health problems among Czech, Russian and U.S. adolescents. *BMC Psychiatry*, 16(1), Article 128. <https://doi.org/10.1186/s12888-016-0829-2>
- Swift, E., & Rosin, P. (1990). A remediation sequence to improve speech intelligibility for students with Down syndrome. *Language, Speech, and Hearing Services in Schools*, 21(3), 140–146. <https://doi.org/10.1044/0161-1461.2103.140>
- Tillinger, M. (2013). *The influence of friends and family on well-being for children and adolescents with developmental disabilities* [Doctoral dissertation]. Boston College.
- Voorman, J. M., Dallmeijer, A. J., Van Eck, M., Schuengel, C., & Becher, J. G. (2010). Social functioning and communication in children with cerebral palsy: Association with disease

- characteristics and personal and environmental factors. *Developmental Medicine & Child Neurology*, 52(5), 441–447. <https://doi.org/10.1111/j.1469-8749.2009.03399.x>
- Wiig, E. H., Semel, E., & Secord, W. A.** (2013). *Clinical Evaluation of Language Fundamentals—Fifth Edition (CELF-5)*. NCS Pearson.
- Wild, A., Vorperian, H. K., Kent, R. D., Bolt, D. M., & Austin, D.** (2018). Single-word speech intelligibility in children and adults with Down syndrome. *American Journal of Speech-Language Pathology*, 27(1), 222–236. https://doi.org/10.1044/2017_AJSLP-17-0002
- Wilson, E. M., Abbeduto, L., Camarata, S. M., & Shriberg, L. D.** (2019a). Estimates of the prevalence of speech and motor speech disorders in adolescents with Down syndrome. *Clinical Linguistics & Phonetics*, 33(8), 772–789. <https://doi.org/10.1080/02699206.2019.1595735>
- Wilson, E. M., Abbeduto, L., Camarata, S. M., & Shriberg, L. D.** (2019b). Speech and motor speech disorders and intelligibility in adolescents with Down syndrome. *Clinical Linguistics & Phonetics*, 33(8), 790–814. <https://doi.org/10.1080/02699206.2019.1595736>
- Yoder, P. J., & Warren, S. F.** (2002). Effects of prelinguistic milieu teaching and parent responsivity education on dyads involving children with intellectual disabilities. *Journal of Speech, Language, and Hearing Research*, 45(6), 1158–1174. [https://doi.org/10.1044/1092-4388\(2002/094\)](https://doi.org/10.1044/1092-4388(2002/094))
- Ziegler, W., Lehner, K., & KommPaS Study Group.** (2021). Crowdsourcing as a tool in the clinical assessment of intelligibility in dysarthria: How to deal with excessive variation. *Journal of Communication Disorders*, 93, Article 106135. <https://doi.org/10.1016/j.jcomdis.2021.106135>