INT J LANG COMMUN DISORD, XXXX 2020, VOL. 00, No. 0, 1-16

# **Research Report**

Effects of speech cues in French-speaking children with dysarthria

Erika S. Levy<sup>†</sup>, Gemma Moya-Galé<sup>†</sup><sup>‡</sup>, Younghwa Michelle Chang<sup>†</sup>, Luca Campanelli§<sup>¶</sup>, Andrea A. N. MacLeod<sup>¶</sup>, Sergio Escorial<sup>#</sup> and Christelle Maillart<sup>\*\*</sup>

<sup>†</sup>Program in Communication Sciences and Disorders, Teachers College, Columbia University, New York, NY, USA <sup>‡</sup>Department of Communication Sciences & Disorders, Long Island University, Brooklyn, NY, USA

§Haskins Laboratories, New Haven, CT, USA

¶The Graduate Center, CUNY, New York, NY, USA

||Faculty of Rehabilitation Medicine-Communication Sciences and Disorders, University of Alberta, Edmonton, AB, Canada #Departamento de Psicobiología y Metodología en Ciencias del Comportamiento, Universidad Complutense de Madrid, Madrid, Spain

\*\*Département de logopédie, Université de Liège, Liège, Belgium

(Received April 2019; accepted January 2020)

# Abstract

*Background:* Articulatory excursion and vocal intensity are reduced in many children with dysarthria due to cerebral palsy (CP), contributing to the children's intelligibility deficits and negatively affecting their social participation. However, the effects of speech-treatment strategies for improving intelligibility in this population are understudied, especially for children who speak languages other than English. In a cueing study on English-speaking children with dysarthria, acoustic variables and intelligibility improved when the children were provided with cues aimed to increase articulatory excursion and vocal intensity. While French is among the top 20 most spoken languages in the world, dysarthria and its management in French-speaking children are virtually unexplored areas of research. Information gleaned from such research is critical for providing an evidence base on which to provide treatment. *Aims:* To examine acoustic and perceptual changes in the speech of French-speaking children with dysarthria, who are provided with speech cues targeting greater articulatory excursion (French translation of 'speak with your big mouth') and vocal intensity (French translation of 'speak with your strong voice'). This study investigated whether, in response to the cues, the children would make acoustic changes and listeners would perceive the children's speech as more intelligible.

*Methods & Procedures:* Eleven children with dysarthria due to CP (six girls, five boys; ages 4;11–17;0 years; eight with spastic CP, three with dyskinetic CP) repeated pre-recorded speech stimuli across three speaking conditions (habitual, 'big mouth' and 'strong voice'). Stimuli were sentences and contrastive words in phrases. Acoustic analyses were conducted. A total of 66 Belgian-French listeners transcribed the children's utterances orthographically and rated their ease of understanding on a visual analogue scale at sentence and word levels.

Outcomes & Results: Acoustic analyses revealed significantly longer duration in response to the big mouth cue at sentence level and in response to both the big mouth and strong voice cues at word level. Significantly higher vocal sound-pressure levels were found following both cues at sentence and word levels. Both cues elicited significantly higher first-formant vowel frequencies and listeners' greater ease-of-understanding ratings at word level. Increases in the percentage of words transcribed correctly and in sentence ease-of-understanding ratings, however, did not reach statistical significance. Considerable variability between children was observed.

*Conclusions & Implications:* Speech cues targeting greater articulatory excursion and vocal intensity yield significant acoustic changes in French-speaking children with dysarthria. However, the changes may only aid listeners' ease of understanding at word level. The significant findings and great inter-speaker variability are generally consistent with studies on English-speaking children with dysarthria, although changes appear more constrained in these French-speaking children.

Keywords: cerebral palsy, dysarthria, treatment strategies, intelligibility, French, speech, children, stimulability.

Address correspondence to: Erika S. Levy, Program in Communication Sciences and Disorders, Teachers College, Columbia University, 525 West 120th St., Box 5, New York, NY 10027, USA; e-mail: elevy@tc.columbia.edu

# What this paper adds

#### What is already known on the subject

• According to the only study comparing effects of speech-cueing strategies on English-speaking children with dysarthria, intelligibility increases when the children are provided with cues aimed to increase articulatory excursion and vocal intensity. Little is known about speech characteristics in French-speaking children with dysarthria and no published research has explored effects of cueing strategies in this population.

#### What this paper adds to existing knowledge

• This paper is the first study to examine the effects of speech cues on the acoustics and intelligibility of French-speaking children with CP. It provides evidence that the children can make use of cues to modify their speech, although the changes may only aid listeners' ease of understanding at word level.

#### What are the potential or actual clinical implications of this work?

• For clinicians, the findings suggest that speech cues emphasizing increasing articulatory excursion and vocal intensity show promise for improving the ease of understanding of words produced by francophone children with dysarthria, although improvements may be modest. The variability in the responses also suggests that this population may benefit from a combination of such cues to produce words that are easier to understand.

# Introduction

Cerebral palsy (CP) is the most common motor disorder in children, with worldwide prevalence estimated at 1.5–4.0/1000 live births (Centers for Disease Control and Prevention 2013). The motor speech disorder of dysarthria is present in a substantial number of children with CP, with a wide range (21–90%) in the prevalence data reported (e.g., Mei *et al.* 2014, Nordberg *et al.* 2013). Dysarthria is often characterized by imprecise, strained, and sometimes quiet, speech, impairing the children's intelligibility and, thus, their communicative participation (Duffy 2013).

French is among the 20 most spoken languages in the world, with 68.5 million speakers in 51 countries (Lewis et al. 2013). Although dysarthria characteristics in French-speaking adults with Parkinson's disease (PD) have been described (e.g., Sauvageau et al. 2015), to the best of our knowledge, no published studies have described dysarthria characteristics in French-speaking children with CP, nor have any examined the effects of treatment strategies on the children's intelligibility. Even speech treatment for English-speaking children with dysarthria has received attention in only a small number of studies (e.g., Fox and Boliek 2012, Levy 2014, 2018, Levy et al. 2012, Miller et al. 2013, Pennington et al. 2010, 2013, 2018). As a result, speech-language pathologists in francophone environments have a weak research base from which to guide their treatment strategies for improving French-speaking children's communication. Thus, understanding the effects of speechtreatment strategies on the speech of French-speaking children with CP is essential to building a scientific foundation for treatment in this language community.

Two types of studies examine the effects of speechtreatment strategies. In cueing studies, such as the present one, talkers follow instructions to speak in a particular manner and their responses are audio recorded and analysed (e.g., Lam and Tjaden 2016). Such studies provide an important scientific foundation for the development of appropriate treatment approaches. Treatment studies, in contrast, examine longer term changes in speech production. Talkers undergo weeks of speech treatment in which particular speaking strategies are practised. The talkers are audio recorded pre- and posttreatment, without instructions to speak in a particular manner (e.g., Ramig *et al.* 2018).

Two long-standing speech-treatment strategies for dysarthria, described primarily in the literature on English-speaking adults with dysarthria, have been to increase talkers' speech clarity or vocal intensity. For English-speaking adults with PD and multiple sclerosis (MS), for example, cueing for clear or loud speech improves acoustic characteristics such as duration, vocal intensity, fundamental frequency and intelligibility (Lam and Tjaden 2016, Tjaden et al. 2014). Most treatment studies on childhood dysarthria, with research advanced primarily by Pennington and colleagues (e.g., Pennington et al. 2018), target various subsystems to improve speech production. This research has found increased duration of breath groups, for example, following a subsystems-based approach. However, the focus here is on studies involving single targets (e.g., increased articulatory excursion or vocal intensity), providing children with a cognitively simple, single instruction to follow and permitting examination of the effects of each global strategy on intelligibility.

Targeting speech clarity has shown promise for increasing intelligibility in adults with dysarthria (e.g., Park et al. 2016). In cueing studies, when native speakers of American English increase movement amplitude in clear speech, the first formant (F1) range generally increases across vowels. The second formant (F2) increases for front vowels, but not for back vowels, revealing acoustic vowel space expansion (e.g., Tjaden et al. 2013). These spectral modifications, as well as some increased vocal intensity (as measured by sound-pressure level-SPL), are thought to relate to the greater articulatory effort and increased neuromotor drive required for clear speech (Perkell et al. 2002). The durational increases that often co-occur may reflect the greater time needed to achieve vocal tract shapes for the more extreme positions in the vowel quadrilateral (Ansel and Kent 1992, Perkell et al. 2002).

Cueing for increased vocal intensity in adults with dysarthria increases intelligibility, improving audibility and decreasing spectral tilt, among other benefits (Tjaden et al. 2013, 2014). Moreover, training vocal intensity is a key element of Ramig et al.'s (2001) Lee Silverman Voice Treatment (LSVT) LOUD, which has been found to increase speech function in small studies of English-speaking children with dysarthria (Fox and Boliek 2012, Levy 2014, Levy et al. 2012). Furthermore, treatments or cues targeting increased vocal intensity result in somewhat increased duration of speech and expanded vowel-space area (Sauvageau et al. 2015). Vowel-space area expansion reflects the greater tongue and jaw displacement stemming from increased vocal effort, similar to the kinematic modifications in clear speech (Perkell et al. 2002).

In the first study comparing cueing strategies in childhood dysarthria, Levy et al. (2017) examined the effects on intelligibility of child-friendly cues targeting clear speech by means of increasing articulatory excursion ('speak with your big mouth') and cues targeting increased vocal intensity ('speak with your strong voice'). In eight English-speaking children with spastic dysarthria, both cues elicited significant changes to vocal intensity (+3.17 dB at sentence level; 5.02 dB at word level) and duration  $(+1080 \text{ ms at sentence level}; +140 \text{$ ms at word level) over a habitual condition, yielding significant improvements to intelligibility, as measured by listeners' percentage of words (orthographically) transcribed correctly (PWC), and ratings of ease of understanding (EOU) on a visual analogue scale (VAS). Acoustically, both the big mouth and strong voice conditions outperformed the habitual condition approximately equally at the sentence level. The big mouth condition revealed primarily greater duration and resulted in greater intelligibility than the strong voice condition at the word level, whereas the strong voice cue elicited primarily greater SPLs. Varying degrees and directions of F1 and F2 changes in the vowels of a subset of words in the three conditions were found, revealing no statistically significant formant changes as a function of speech cue.

#### Languages other than English

In considering the effects of cueing strategies on languages other than English, one might expect *universal* benefits to intelligibility from cues to increase vocal intensity, such as 'strong voice', because of the universality of motor impairments (Pinto *et al.* 2017) and the improvements in audibility and spectral tilt, among other acoustic benefits resulting from louder speech reported in English speakers (Tjaden *et al.* 2014). Benefits from increased vocal intensity in adult talkers have also been found in Spanish (Moya-Galé *et al.* 2018), Mandarin (Lee and McCann 2009) and French (Sauvageau *et al.* 2015), among other languages.

Alternatively, dysarthria may manifest differently across languages at segmental and prosodic levels (Hsu et al. 2017, Liss et al. 2013), as might the effects of global cueing strategies. In fact, language-specific responses to such treatment or cueing strategies are beginning to be documented (e.g., Moya-Galé et al. 2016). Moreover, although French and English lexicons contain numerous cognates, the two prosodies differ considerably. English is a Germanic language with lexical stress, such that each content word contains a stressed syllable and the position of the stressed syllable is constrained by the word. These stressed syllables differ from their unstressed counterparts with regard to syllable duration, SPL, fundamental frequency and vowel quality. In contrast, French, a Romance language, signals stress within each utterance, rather than within each word. The syllable that receives stress is constrained by its position such that the stressed syllable is the final syllable of the utterance (or of a phrase within the utterance) or penultimate if the word ends with a schwa. Stressed syllables in French are marked mainly by longer duration and greater changes in fundamental frequency compared with their unstressed counterparts (Astésano and Bertrand 2016). Because English has lexical stress, children may produce more distinct speech sounds at the word level, resulting in improved word intelligibility when cued to use intelligibility-enhancing speech strategies. In French, children cued to use such strategies may be expected to produce changes in only the final or penultimate syllable of an utterance via a stressed syllable of greater length and fundamental frequency variation, resulting in intelligibility benefit to only target words that are at the end of a phrase.

Taken together, findings from English and other languages suggest that global cues to increasing articulatory excursion and vocal intensity hold promise as strategies for increasing intelligibility. However, basic knowledge is lacking regarding dysarthria in French-speaking children, and French and English differ in their prosodic and segmental structures. Therefore, the critical question of whether the same strategies would improve the intelligibility of French-speaking children with dysarthria remain to be addressed.

#### Current study

The study examined the effects of global speech cues on the acoustic characteristics and intelligibility of speech produced by French-speaking children with dysarthria due to CP. The cues targeted greater articulatory excursion ('Parle avec ta grande bouche' ['Speak with your big mouth']) and vocal intensity ('Parle avec ta grosse voix' [loosely translated as 'Speak with your strong voice']). Specifically, we asked whether (1) the children would be able to make acoustic changes at sentence and word levels in response to these cues; and (2) blinded listeners would perceive the children's speech as more intelligible as a function of these cues. (Although EOU and PWC differ in their emphasis on effort versus accuracy, both constructs are sometimes referred to here as 'intelligibility' for convenience.)

The big mouth cue was expected to yield acoustic and perceptual gains overall. Specifically, based on Levy *et al.* (2017) and for the reasons cited in the clear speech literature (Smiljanić and Bradlow 2005), it was hypothesized that the children's sentence duration would increase. Some increase in vocal intensity was expected, as well (Tjaden *et al.* 2013). However, because of the prosodic differences between French and English, acoustic and perceptual gains might be limited in our target words, which were centrally embedded in carrier phrases, rather than positioned at the ends of phrases, where syllables would be expected to receive stress.

Hypotheses regarding spectral changes in the big mouth condition relied, in part, on the particular deficits in F2 range found in English-speaking individuals with dysarthria due to CP (Allison and Hustad 2018, Ansel and Kent 1992). These atypical formants relate to the impaired motor control for the jaw and tongue, with greater deficits in the tongue revealed in preliminary kinematic studies of children with CP. Tongue movement limitations are also present along the inferiorsuperior plane in dysarthria, reflected in restricted first formant (F1) range, but may be compensated for to some extent by jaw lowering (Nip et al. 2017). Thus, the big mouth cue might *address* the limitations in F2 range, replicating the spectral modifications found in clear speech studies, with F2 increasing for front vowels, for example, /e/ in the present study, as well as overall (Tjaden et al. 2013). Alternatively, limited F2 modifications might be hypothesized in a big mouth condition due to tongue movement restrictions across the anterior-posterior plane (Levy *et al.* 2017, Nip *et al.* 2017). Increases in F1 might be expected, as the big mouth cue itself calls for lowering of the jaw, which would be coupled with tongue lowering (Nip *et al.* 2017). Contrary to expectation of F1 increases, however, English-speaking children with dysarthria showed no statistically significant changes in this formant (Levy *et al.* 2017), rendering the hypothesis of F1 increases less evident for the present study. As found for English-speaking children, increases in intelligibility were expected following the big mouth cue for these French-speaking children, especially at sentence level (Levy *et al.* 2017).

The strong voice cue was expected to increase primarily vocal intensity (Fox and Boliek 2012, Levy 2014, Levy *et al.* 2012, 2017), but also duration (Tjaden *et al.* 2013). Hypotheses regarding spectral changes for the big mouth cue also applied to strong voice, with the acoustic changes reflecting kinematic changes accompanying greater vocal effort (Tjaden *et al.* 2013), although more limited results were expected with strong voice than with big mouth, which targets articulatory excursion more directly. Crucially, gains in intelligibility were expected (Fox and Boliek 2012, Levy *et al.* 2017).

The present study was an important first step to understanding the impact of global cues on intelligibility in children with dysarthria who speak a language other than English and, more specifically, to expand the knowledge base for speech–language pathologists working in French.

#### Methods

This study was approved by the Institutional Review Boards at Teachers College, Columbia University, New York, as well as the Université Catholique de Louvain and the Université de Liège in Belgium.

#### Participants

#### Children with CP

A total of 11 Belgian-French-speaking children (five males, six females) participated in the study. The children were taking part in a larger annual summer programme for children with CP that took place in a park in Belgium. The children were recruited from outpatient clinics specialized in CP and by means of a website of the local rehabilitation foundation (https://sites. google.com/site/intensiverehabfoundation/). Potential participants were first screened by telephone. Children who passed the phone screening had a neurologistobtained diagnosis of CP and motor skills were assessed by a physical therapist or occupational therapist. A speech–language pathologist assessed the children's

Child	Age (years;months)	Sex	Diagnosis	GMFCS	Dysarthria severity	Deviant speech characteristics
CP01	4;11	F	Spastic quadriplegia	IV	Moderate	Increased vocal intensity, moderate hypernasality, imprecise articulation, several phonological processes
CP02	5;1	М	Spastic quadriplegia	III	Mild	Breathy voice quality, mild hypernasality, imprecise articulation
CP03	7;1	М	Dyskinetic quadriplegia	III	Mild–moderate	Reduced vocal intensity, mild hypernasality, slow rate, inconsistently imprecise articulation, prosodic abnormalities (breaths within utterances)
CP04	8;9	М	Spastic quadriplegia	III–IV	Severe	Strained vocal quality, very imprecise articulation, consonant deletion
CP05	9;5	М	Dyskinetic quadriplegia	III	Mild	Monotone, slow rate, imprecise articulation
CP06	11;1	F	Dyskinetic quadriplegia	IV	Moderate–severe	Reduced intensity, breathy voice quality, slow rate, inconsistently imprecise articulation, prosodic abnormalities (breaths within utterances and syllabification of words)
CP07	11;1	М	Spastic quadriplegia	IV	Mild	Moderately fast rate
CP08	12;3	F	Spastic quadriplegia	IV	Moderate– moderately severe	Strained vocal quality, monotone pitch, imprecise articulation
CP09	14;9	F	Spastic quadriplegia	II	Mild	Reduced intensity, hypernasality, fast rate
CP10	16;2	F	Spastic quadriplegia	III	Mild	Reduced intensity, breathy voice quality
CP11	17;0	F	Spastic quadriplegia	IV	Mild	Intensity decreases during utterance, imprecise articulation, hypernasality, prosodic abnormalities (breaths within utterances)

Table 1. Participant characteristics of the children with dysarthria due to cerebral palsy

Note: GMFCS, Gross Motor Function Classification System.

speech and ability to follow tasks similar to those in the study to determine the children's speech characteristics and inclusion in the present study (Paradis *et al.* 2019).

Inclusion criteria were (1) using speech as primary means of communication, with speech considered by parents or teachers to be difficult to understand; (2) passing a bilateral pure-tone hearing screening at 20 dB HL for 500, 1000, 2000 and 4000 Hz; and (3) an ability to follow directions related to the tasks (Fox and Boliek 2012).

Table 1 lists details regarding participant characteristics. The children ranged in age from 4;11 to 17;0 years (mean = 10;8 years, SD = 4;4 years). All were native, dominant speakers of Belgian French, although six children also spoke another language. They presented with dysarthria due to dyskinetic quadriplegic CP or to spastic quadriplegic CP. Severity of dysarthria and deviant speech characteristics were determined by consensus by three certified (French-speaking) speech– language pathologists based on the children's clinical evidence of impairment, in at least one of the subsystems of speech, that was audibly and/or visually observable (Fox and Boliek 2012, Lee *et al.* 2014, 2017). The children's receptive language was judged to range from delayed to within normal limits based on an informal assessment through conversation and comprehension of simple or complex directions, and for children under 12 years, a receptive language subtest (i.e., Compréhension C2) from the Évaluation du Langage Oral (ELO; Khomsi 2001), a norm-referenced tool appropriate for the assessment of Belgian French. As in the screening, all children were judged by the speech–language pathologists to be able to perform the study tasks adequately.

# Listeners

A total of 66 Belgian-French-speaking adults (25 men, 41 women, age range = 18–29 years, mean = 22 years, SD = 2.24 years) were recruited to listen to recordings of the children with dysarthria. All participants were recruited from the Liège area in Belgium through social media and flyers and passed a bilateral pure-tone hearing screen at 20 dB HL for 500, 1000, 2000 and 4000 Hz. Listeners reported no history of speech, language or hearing problems. Additionally, they reported having no experience with individuals with motor speech disorders as verified by a language experience background questionnaire. They were paid €15 to participate.

#### Speech stimulus acquisition and selection

#### Speech stimuli

The children were recorded producing a variety of speech tasks as part of a larger study. The stimuli selected for the current experiment were three phrases or sentences from the Test of Children's Speech (TOCS+; Hodge et al. 2009) (henceforth 'sentences') translated into French (i.e., Trouve tous les crayons [Find all the crayons]; *Trois petits cochons roses* [Three little pink pigs]; *N'éclabousse pas partout* [Don't splash water everywhere]) and 15 contrastive words (Ansel and Kent 1992): gens, cent, zoo, dos, joue, sous, choux, dé, thé, chaud, chant, boule, balle, mal and fou (people, hundred, zoo, back, play, under, cabbage, dice, tea, hot, singing, ball (e.g., tennis), ball (e.g., soccer), wrong and crazy). Contrastive words were presented in the carrier phrase *Elle dit* CV(C)*peut-être* [She says CV(C) maybe] to approximate the continuous speech characteristics of children's typical communication. For examples of the children's word and sentence productions, see the additional supporting information.

# Speech recording procedure

Recordings for each child took place within a single session in a quiet room in a summer camp programme for children with CP in Brussels, Belgium. Careful control allowed the inclusion of the dimension of vocal intensity to be captured in the recordings, unlike studies in which vocal intensity is normalized (e.g., Cannito et al., 2012). A forehead Countryman EMW Lavalier microphone was placed 8 cm from the child's lips. Calibration was completed at the beginning and end of each testing session with a pure tone played via an OT 120-Korg Orchestral tuner located 8 cm from the microphone. The experimenter noted the SPL on a Galaxy Check-Mate CM140 sound-level meter adjacent to the microphone. Stimuli were recorded using a digital (ZOOM H4n handy) recorder at a sampling rate of 44.1 kHz with 16-bit resolution on a mono-channel.

Children were provided verbal and visual instructions on how to repeat recordings of utterances produced by an adult native Belgian-French speaker in the habitual, big mouth and strong voice conditions. These adult model utterances were pre-recorded to ensure consistency in the adult's production of each speaking condition. Children heard the model speaker's utterances delivered by loudspeakers (Bose SoundLink Color II) placed at a consistent distance from the child.

For the habitual condition, children were simply instructed to repeat what they heard. Photographs representing the sentences and words were provided on an iPad screen. For the big mouth condition, they were asked (in French) to 'speak with a big mouth'. For the strong voice condition, they were asked (in French) to 'speak with a strong voice'. Children were given verbal reminders if they did not repeat the utterance. They were also prompted to repeat the stimulus when extraneous noise occurred during the production or when their responses were off-task or incomplete. Breaks were provided as needed.

As is typically done in adult cueing studies (Smiljanić & Bradlow 2009; Tjaden *et al.* 2014), the habitual condition was recorded first to avoid potential carryover effects (of either cued condition). The order of presentation of the experimental conditions was counterbalanced across the children. Children were given a short break and were engaged in conversation between conditions to address potential carryover effects. The effect of presentation order was also examined quantitatively, and no significant effects emerged. For all dependent measures, *p*-values for the main effect of order were >0.1 and *p*-values for the interaction between order and condition were >0.16.

# Listening tasks

All 66 listeners completed two listening tasks in a quiet room in Liège, using custom-developed software (Chang and Chang 2015) programmed in MATLAB (Version R2015b) and presented on a laptop computer. The SPL of the calibration tone (measured before the recording of the children's productions) was reproduced at 8 cm from loudspeakers (Bose SoundLink Color II) in order to present the speech stimuli at a level representative of the children's vocal intensity. The stimuli were played through the loudspeakers, which were connected to a MacBook Air laptop computer (Model A1466). Listeners were seated 85 cm from the loudspeakers. The listeners first completed a short familiarization task, which involved the sentence and contrastive-word tasks, but each with six stimuli that were different from the experimental stimuli, recorded by a child without dysarthria. The purpose of the familiarization task was for the listeners to learn the listening task in a setting in which they could ask questions before performing the experimental listening task. Listeners took approximately 45-60 min to complete all experimental tasks.

In the sentence task, listeners rated EOU of the sentence productions. (Sentences were not transcribed, as they were predictable repetitions of previously heard sentences.) Each listener rated all 109 sentences (99 original sentences by all of the children and 10 reliability items) that were randomly presented. The final data included EOU ratings from all 66 listeners for each sentence in each condition uttered by each child. Ratings were completed on a 9-cm VAS with anchors (French translations of) 'very easy' and 'very difficult' to understand. Listeners rated EOU on this scale by sliding a cursor between the two anchors. The score corresponding to the placement of the cursor was not visible to listeners; however, for analysis purposes, the anchor 'very difficult' corresponded to 0 and 'very easy' corresponded to 100, with placements between the two anchors corresponding to scores between these endpoints.

In the contrastive-word task, listeners orthographically transcribed and rated EOU of the children's word productions (in carrier phrases). Each listener transcribed and rated 17 words in total (15 contrastive words and two reliability items produced by only one child). The 15 contrastive words included five words from each of the three speaking conditions, with no word repeated across the conditions. The final data included transcriptions and EOU ratings from two listeners per child for each word in each condition. The words in carrier phrases were randomized and played only once to each listener. Listeners were asked to transcribe each word and rate its EOU before continuing to the next word. Although this yielded rating scores that may not have been independent from the PWC, it allowed listeners to avoid learning effects by hearing each word only once. The contrastive-word task preceded the sentence task in order to avoid familiarization with the child's speech and thus perceptual learning during transcription.

# Data analysis

#### Acoustic and perceptual analyses

Four acoustic measures were examined in the habitual, big mouth, and strong voice conditions: SPL and duration were measured for each utterance (for the sentence task) and word (for the contrastive-word task). Additionally, F1, and F2 were measured for a subset of words in the contrastive-word task, as described below. These measures were selected to verify the presence of speech-production differences among the speaking conditions. Other adjustments might be associated with these speech production changes, but SPL, duration and spectral changes were the most obvious modifications expected (Levy *et al.* 2017).

The first production of each contrastive word was selected for analysis for each child. Only productions that contained noise or whose signal could not reliably be analysed were replaced by a second repetition. Every sentence and word was segmented manually (by research assistants and co-authors) at the sentence and word levels. Onsets and offsets of sentences and words were determined by the standard criteria (Lam and Tjaden 2016, Levy and Law 2010). Duration and SPLs were analysed by means of Praat (Boersma and Weenink 2006). Duration was measured in seconds, from onset to offset of target words and sentences. Input level was unchanged throughout the recording session and the average (originally produced) SPL was measured for each utterance (Lam and Tjaden 2016).

The F1 and F2 were measured by means of wideband spectrograms and a linear predictive coding (LPC) spectrum, for a 25-ms window centred at the (temporal) midpoint of the subset of vowels /u, a, e, o/ (in the words boule, balle, dé and dos) in the contrastive-word task. The average of those formant values was then obtained for each vowel. The purpose of the spectral analysis was to assess whether acoustic changes suggesting greater mouth opening and articulatory excursion would be achieved across either cued condition (e.g., Tiaden et al. 2013). Ansel and Kent (1992) found that front-back vowel contrasts were one of four parameters that account for considerable variance in intelligibility in English-speaking adults with dysarthria due to CP; thus, we investigated changes in this subset of contrastive words differing primarily along the front-back dimension (dé, dos) and in height (boule-balle).

The perceptual analysis yielded two final data sets: (1) sentence ratings of EOU; and (2) word ratings of EOU and PWC. The PWC was calculated from the transcriptions, with words considered correct if they were exact matches for the targets, homonyms or obvious misspellings of the homonym or target. For descriptive statistics on the rating task, the mean EOU rating was calculated.

# Reliability of acoustic measures

A second judge randomly selected and manually rechecked 20% of the original sentences and words to ensure the reliability of the acoustic findings. Pearson product-moment correlations and absolute measurement errors were used to index reliability. For sentences, the correlation between the first and second sets of SPL measures was 0.99 (mean absolute difference measure = 0.53 dB, SD = 1.09 dB). The correlation between the two sets of duration measures was 0.99 (mean absolute difference measure = 0.53 dB, SD = 1.09 dB). The correlation between the two sets of duration measures was 0.99 (mean absolute difference measure = 0.04 s, SD = 0.05 s). For contrastive words, the correlation between the two sets of SPL measures was 0.95 (mean absolute different measure = 1.26 dB, SD = 1.82 dB), and 0.91 for duration (mean absolute difference = 0.04 s, SD = 0.05 s).

For reliability of the F1 and F2 measurements, the vowels in the selected words *balle*, *boule*, *dé* and *dos* were manually checked by the second judge. Pearson product-moment correlation between the first and second judges was 0.99 for F1 (mean absolute difference measure = 8.38 Hz, SD = 10.30 Hz) and 0.98 for F2 (mean absolute difference measure = 58.52 Hz, SD = 113.11 Hz). The reliability of formant frequency measures was judged to be within an acceptable range and consistent with reliability reported in prior investigations (e.g., Lee *et al.* 2014).

#### Reliability of perceptual measures

For intra-listener reliability, 20% of the sentences and words were randomly selected and presented to each listener at the end of each task to be re-evaluated. The two transcriptions and EOU ratings completed by each listener were compared. For the contrastive-word task, a Pearson product-moment correlation showed strong agreement between first and second PWC for transcription (r(132) = 0.77, p < 0.001), and strong agreement for EOU rating (r(132) = 0.80, p < 0.001). For the sentence task, Pearson moment correlation for the EOU rating showed strong agreement between first and second ratings (r(645) = 0.86, p < 0.001).

Inter-listener reliability was assessed using intraclass correlation coefficient (ICC), determined from a twoway mixed model (random listener effects, fixed measure effects) for overall consistency of ratings among listeners. Aggregate listener performance was of focus in previous studies (e.g., Tjaden et al. 2014), and, therefore, the average ICC was considered the primary measure of agreement among listeners. For the contrastive-word task, agreement among listeners on PWC and EOU rating measures was calculated. The average ICC for PWC was 0.68 (95% confidence interval (CI) = [0.61,0.73]), and for word EOU rating, 0.71 (95% CI = [0.66, 0.76]), indicating moderately good inter-listener reliability. For the sentence task, agreement among listeners on EOU rating measures of each child's sentence was calculated. The average ICC for sentence EOU rating was 0.76 (95% CI = [0.71, 0.81]), indicating good inter-listener reliability. All ICCs were statistically significant (p < 0.001).

#### Statistical analyses

Data were analysed using mixed-effects regression analysis (Pinheiro and Bates 2000). Models were logistic for PWC (correct/incorrect transcription) and linear for duration, SPL, F1, F2 and EOU. For all models, condition (HA, BM, SV) was the only predictor variable. For the linear models, the dependent measures showed approximately normal distributions and therefore were kept in their original scale. No extreme values were detected, and no data were excluded.

We adopted mixed-effects regression because of its known advantages relative to traditional methods (e.g., *t*-tests or analysis of variance (ANOVA)). This approach allows flexible modelling of the variability within and between subjects, such as individual differences among children and variability in the effect of condition. It also allows proper modelling of data dependencies created by nested and crossed structures, such as observations nested within subjects and items, which avoids biases related to data aggregation (Raudenbush and Bryk 2002). Last, mixed-effects regression has been shown to be superior to traditional approaches in both large and, most importantly for the current study, small n designs (Ferron *et al.* 2009, Moeyaert *et al.* 2017).

All models included the maximal random effects structure justified by the design. For duration, SPL, F1 and F2, models included random intercepts for children and items. For EOU and PWC, models included random intercepts for children, items and listeners. Random slopes for the effect of condition were excluded only in case of convergence failures. Data were analysed with R version 3.5.1 (R Core Team 2018) using the functions *glmer* and *lmer* from the lme4 package, version 1.1-19 (Bates et al. 2015). The reported F-tests for the main effect of condition were obtained using the *joint\_tests* function from the package emmeans, version 1.3.0 (Lenth et al. 2018). For the post-hoc Tukey adjusted comparisons (Field et al. 2012), we used the emmeans function from the emmeans package (Lenth et al. 2018). Approximate r effect sizes were computed using the formula (Field et al. 2012):

$$r = \operatorname{sqrt}[t^2/(t^2 + \operatorname{df})]$$

where r > 0.1 indicates a small effect size; r > 0.3 is a medium effect size; and r > 0.5 indicates a large effect size.

#### Results

# Acoustic analysis of model speaker

Table 2 provides details of the (adult) model speaker's average duration and SPL (at 8 cm distance) for the contrastive words and TOCS+ (Hodge et al. 2009) sentences across the habitual, big mouth and strong voice conditions. For the analyses at the word level, mixedeffects regression analysis revealed main effects for both duration, F(2, 36.12) = 12.89, p < 0.001 and SPL, F(2, 36.13) = 21.74, p < 0.001. For duration, posthoc Tukey tests showed that the big mouth condition elicited significant increases in duration compared with both habitual and strong voice conditions, t(36.1) =-5.07, p < 0.001, r = 0.64, and t(36.1) = 2.79, p =0.022, r = 0.42, respectively. The difference between habitual and strong voice conditions was not statistically significant, t(36.1) = -2.28, p = 0.072, r = 0.35. For SPL, strong voice was greater than both habitual and big mouth conditions, t(36.1) = -5.25, p < 0.001, r = 0.66, and t(36.1) = -6.08, p < 0.001, r = 0.71, respectively. The difference between habitual and big mouth was not statistically significant, t(36.1) = 0.84, p = 0.684, r = 0.14.

For the analyses at the sentence level, main effects were found for both duration and SPL, F(2, 9) = 176.33, p < 0.001 and F(2, 9) = 75.99, p < 0.001,

	Habitual	Big mouth	Strong voice	Difference <sup>a</sup>
<i>Sentences</i> Duration (s) SPL (dB)	1.18 (0.24) 63.00 (0.32)	4.79 (0.69) 66.6 (1.53)	1.94 (0.3) 71.7 (1.53)	Ha <bm; bm="" ha<sv;="">SV Ha<bm; bm<sv<="" ha<sv;="" td=""></bm;></bm;>
<i>Words</i> Duration (s) SPL (dB)	0.42 (0.09) 67.9 (4.9)	0.53 (0.08) 67.2 (4.76)	0.47 (0.09) 72.7 (2.29)	Ha <bm; bm="" ha="SV;">SV Ha=BM; Ha<sv; bm<sv<="" td=""></sv;></bm;>

Table 2. Average duration and sound-pressure level (SPL) of the adult model speaker in the three speaking conditions

Notes: Values are mean (SD).

<sup>a</sup>Ha, habitual condition; BM, big mouth condition; SV, strong voice condition. The symbols '<' and '>' indicate that the first condition is significantly smaller or greater, respectively, than the second condition. The '=' sign indicates that the difference between the two conditions is not statistically significant. Effects are reported as significant for p < 0.05. See the Results section for additional information.

respectively. Post-hoc analyses showed that duration was longer in the big mouth than in the habitual and strong voice conditions, t(9) = -17.8, p < 0.001, r = 0.99, and t(9) = 14.06, p < 0.001, r = 0.98, respectively, and that duration was longer in the strong voice than in the habitual condition, t(9) = -3.76, p = 0.011, r = 0.78. For SPL, post-hoc analyses showed higher values for strong voice than habitual and big mouth conditions, t(9) = -12.3, p < 0.001, r = 0.97, and t(9) = -7.14, p < 0.001, r = 0.92, respectively. SPL was lower in the habitual than in the big mouth condition, t(9) = -5.13, p = 0.001, r = 0.86.

# Children's acoustic changes across speaking conditions

#### Sentences

Table 3 presents average group data for the children's duration and SPL (at 8 cm distance) across the three speaking conditions. A significant main effect of speaking condition was found for sentence duration, F(2, 88) = 12.26, p < 0.001. Duration was significantly longer in the big mouth condition than in the habitual and strong voice conditions, t(88) = -4.69, p < 0.001, r = 0.45, and t(88) = 3.72, p = 0.001, r = 0.37, respectively. The difference between habitual and strong voice conditions was not statistically significant, t(88) = -0.97, p = 0.6, r = 0.1. A significant main effect of speaking condition was found for SPL, F(2,11) = 9.31, p = 0.004, with SPL in the habitual condition significantly lower than both big mouth, t(12.1) =

-3.25, p = 0.018, r = 0.68, and strong voice, t(12.1) = -4.15, p = 0.004, r = 0.77. The difference between big mouth and strong voice was not statistically significant, t(12.1) = -1.67, p = 0.256, r = 0.43.

# Contrastive words

Table 4 presents the average duration and SPL of contrastive words produced by children in the three speaking conditions. A significant main effect of speaking condition was found for word duration, F(2, 472) = 5.42, p = 0.005. Duration was significantly longer in the big mouth and in the strong voice conditions than in the habitual condition, t(472) = -2.93, p = 0.01, r = 0.13, and t(472) = -2.76, p = 0.016, r = 0.13, respectively. The difference between big mouth and strong voice was not statistically significant, t(472) = 0.17, p = 0.984, r = 0.008.

A significant main effect of speaking condition was found for SPL, F(2, 472) = 47.43, p < 0.001, with SPL in the strong voice condition significantly greater than in the big mouth, t(472) = -5.05, p < 0.001, r = 0.23, and habitual conditions, t(472) = -9.74, p < 0.001, r = 0.41. SPL in the big mouth condition was also significantly greater than in the habitual condition, t(472) = -4.69, p < 0.001, r = 0.21.

Table 4 also lists the F1 and F2 values of the vowels /a/ (in *balle*), /u/ (in *boule*), /e/ (in *dé*) and /o/ (in *dos*) in the three speaking conditions. The main effect of speaking condition was significant for F1, F(2, 120) = 6.11, p = 0.003, but not for F2, F(2, 120) = 1.42, p = 0.246. Post-hoc tests for the effect of condition on F1

Table 3. Average sentence duration and sound-pressure level (SPL) of the children with dysarthria in the three speaking conditions

	Habitual	Big mouth	Strong voice	Difference <sup>a</sup>
Duration (s)	2.46 (0.85)	3 (0.95)	2.57 (0.71)	Ha <bm; bm="" ha="SV;">SV</bm;>
SPL (dB)	58.5 (6.54)	62.4 (6.85)	64.6 (5.87)	Ha <bm; bm="SV&lt;/td" ha<sv;=""></bm;>

Notes: Values are mean (SD).

<sup>a</sup>Ha, habitual condition; BM, big mouth condition; SV, strong voice condition. The symbols '<' and '>' indicate that the first condition is significantly smaller or greater, respectively, than the second condition. The '=' sign indicates that the difference between the two conditions is not statistically significant. Effects are reported as significant for p < 0.05. See the Results section for additional information.

	Habitual	Big mouth	Strong voice	Difference <sup>a</sup>
Duration (s)	0.43 (0.13)	0.47 (0.11)	0.47 (0.11)	Ha <bm; bm="SV&lt;/td" ha<sv;=""></bm;>
SPL (dB)	59.8 (6.55)	62 (4.28)	64.4 (4.7)	Ha <bm; bm<sv<="" ha<sv;="" td=""></bm;>
First formant (F1; Hz)				
balle	741 (204)	882 (194)	865 (138)	
boule	436 (133)	445 (120)	517 (105)	
dé	512 (129)	551 (117)	576 (126)	
dos	623 (199)	637 (144)	620 (160)	
Mean of four words	578 (148)	629 (115)	644 (112)	Ha <bm; bm="SV&lt;/td" ha<sv;=""></bm;>
Second formant (F2; Hz)				
balle	1786 (312)	1856 (185)	1874 (212)	
boule	1180 (253)	1119 (173)	1214 (168)	
dé	2355 (297)	2277 (425)	2483 (257)	
dos	1497 (290)	1415 (306)	1422 (236)	
Mean of four words	1705 (172)	1667 (153)	1748 (178)	Ha=BM; Ha=SV; BM=SV

Table 4. Average duration, sound-pressure level (SPL) and vowel formants (F) of contrastive words produced by children with						
dysarthria in three speaking conditions						

Notes: Values are mean (SD).

<sup>a</sup>Ha, habitual condition; BM, big mouth condition; SV, strong voice condition. The symbols '<' and '>' indicate that the first condition is significantly smaller or greater, respectively, than the second condition. The '=' sign indicates that the difference between the two conditions is not statistically significant. Effects are reported as significant for p < 0.05. See the Results section for additional information.

revealed that F1 was significantly higher in the big mouth and strong voice conditions than the habitual condition, t(120) = -2.55, p = 0.032, r = 0.23, and t(120) = -3.35, p = 0.003, r = 0.29, respectively. The difference between big mouth and strong voice was not statistically significant, t(120) = -0.79, p = 0.707, r = 0.07.

#### Perceptual changes across speaking conditions

#### Sentences

Figure 1 presents average EOU ratings for each of the children with dysarthria across the three speaking conditions at the sentence level. Descriptive statistics are reported in table 5. On the scale from 0 ('very difficult

#### Contrastive words

Figure 2 presents the average EOU ratings for each of the children with dysarthria across the three speaking conditions at word level. The mean EOU ratings for the contrastive-word task were 41.6 (SD = 20.9) for the habitual condition, 45.7 (SD = 21.5) for the big mouth

to understand') to 100 ('very easy to understand'), EOU

ratings were 53.2 (SD = 31) for the habitual condition,

increasing to 55.1 (SD = 30.8) for the big mouth condi-

tion, and to 54.3 (SD = 31.8) for the strong voice con-

dition. Mixed-effects regression revealed that the main

effect of speaking condition was not statistically signifi-

cant, F(2, 11.1) = 0.48, p = 0.634.

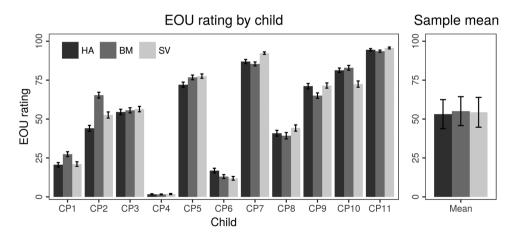


Figure 1. Listeners' average ratings of ease of understanding (EOU) of sentences for the 11 children with dysarthria across habitual (HA), big mouth (BM) and strong voice (SV) conditions. Sentences were rated from 'very difficult' to understand (corresponding to 0) to 'very easy' to understand (corresponding to 100). Error bars:  $\pm$  SE.

	1	8		
	Habitual	Big mouth	Strong voice	Difference <sup>b</sup>
Sentences EOU	53.2 (31)	55.1 (30.8)	54.3 (31.8)	Ha=BM; Ha=SV; BM=SV
Words EOU	41.6 (20.9)	45.7 (21.5)	45.5 (22.4)	Ha <bm; bm="SV&lt;/td" ha<sv;=""></bm;>
Words PWC <sup>a</sup>	38.2 (22.5)	43 (29.1)	43 (29.6)	Ha=BM; Ha=SV; BM=SV

 Table 5. Ease-of-understanding (EOU) ratings for words and sentences and percentage of words transcribed correctly in the three speaking conditions. Mean (Standard Deviation)

Notes: Values are mean (SD).

<sup>a</sup>PWC, average percentage of words orthographically transcribed correctly.

<sup>1</sup> We, average precentage of words or inorganized value concerns. <sup>b</sup> Ha, habitual condition; BM, big mouth condition; SV, strong voice condition. The symbols '<' and '>' indicate that the first condition is significantly smaller or greater, respectively, than the second condition. The '=' sign indicates that the difference between the two conditions is not statistically significant. Effects are reported as significant for p < 0.05. See the Results section for additional information.

condition, and 45.5 (SD = 22.4) for the strong voice condition, indicating an increase in perceived intelligibility in both the big mouth and strong voice conditions. Descriptive statistics for the sample are also reported in table 5. Mixed-effects regression revealed a significant main effect for the speaking conditions, F(2, 924) = 3.94, p = 0.02. Post-hoc analyses showed significant increases from habitual to big mouth, t(926) = -2.47, p = 0.037, r = 0.08, and from habitual to strong voice, t(926) = -2.39, p = 0.045, r = 0.08. The difference between big mouth and strong voice was not statistically significant, t(926) = 0.08, p = 0.997, r = 0.003.

The average PWC is presented in figure 3 and table 5. PWC for contrastive words was 38.2% (SD = 22.5) in the habitual condition, and 43% in both big mouth (SD = 29.1) and strong voice (SD = 29.6) conditions. The effect of condition did not reach statistical significance, F(2, Inf) = 1.55, p = 0.211.

# Individual data

The children's performance on acoustic and perceptual measures was variable. Thus, in order to provide more data on the children's variability and to inform future research, individual data are provided in tables S1–S18 and figures S1–S2 in the additional supporting information. These include descriptive statistics by child, tables of descriptive statistics for the effect of the cues and scatter plots with non-parametric correlations regarding the relationships between age and severity and effects of cues for PWC.

#### Discussion

This study investigated the consequences of cueing French-speaking children with dysarthria to speak with their 'big mouth' or with their 'strong voice'. Findings indicate that: (1) the children were able to vary their speech styles in response to models and cues, including increasing SPLs and durations of their utterances, although the acoustic effects varied as a function of the cue and the linguistic (i.e., sentence versus word) level; (2) at sentence level, big mouth and strong voice cues did not increase EOU ratings significantly and considerable variability between children was observed; and (3) at word level, both big mouth and strong voice cues yielded significantly greater EOU, but increases in PWC did not reach statistical significance.

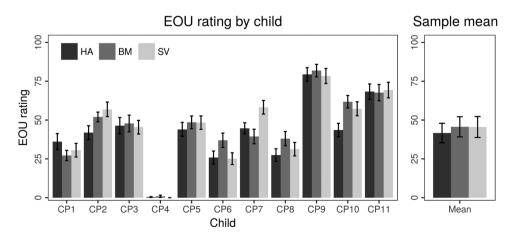


Figure 2. Listeners' average ratings of ease of understanding (EOU) of words for the 11 children with dysarthria across habitual (HA), big mouth (BM) and strong voice (SV) conditions. Words were rated from 'very difficult' to understand (corresponding to 0) to 'very easy' to understand (corresponding to 100). Error bars:  $\pm$  SE.

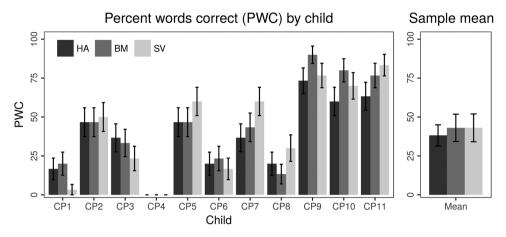


Figure 3. Average percentage of words orthographically transcribed correctly (PWC) for the 11 children with dysarthria across habitual (HA), big mouth (BM) and strong voice (SV) conditions. Error bars:  $\pm$  SE.

#### Acoustic changes in response to speech cues

Based on the indications of change in response to similar cues in English-speaking children with dysarthria (Levy *et al.* 2017), the French-speaking children were expected to modify their speech styles, increasing primarily duration for big mouth condition (at sentence level) and vocal intensity for strong voice condition, with overlap in acoustic modifications anticipated across the cues. The extent of potential changes in this study was difficult to anticipate as this was the first such study on French-speaking children, and French rhythmic characteristics were thought potentially to constrain durational adjustments. Limited changes were thus anticipated for target words, which were not in a prosodic position to receive stress.

Acoustic analyses of the children's speech revealed significantly greater durations at the sentence level following the big mouth cue and at the word level following cues for both big mouth and strong voice conditions compared with the habitual speaking condition. This was a somewhat different outcome from the adult model's significant word duration increase in big mouth, but not in strong voice, condition.

The children's large increases in duration following the big mouth cue at sentence level, but not at word level, suggest that durational increases in response to this cue may be executed less within words themselves, and more within the larger sentence context. Because stress placement in French is not used for distinguishing words (as it is in English), these French-speaking children may not manipulate word duration as is done in English. Sentence-level duration, in contrast, is manipulated in French stress, as final syllable prominence in an utterance contributes to marking phrase boundaries (Duez 2014). As a result, the children may have maximized utterance-final syllable lengthening to preserve prosodic boundaries. Identifying the locus of the sentence-level changes in response to the big mouth cue, potentially in utterance-final syllable lengthening or in lengthened pause duration, is an important direction for future research.

While the children's increase in word duration in both cued conditions was statistically significant, the effect size was small. In contrast, Levy et al.'s (2017) English study revealed larger changes in duration (approximately 140 ms) than the approximately 40 ms increase in French words in a big mouth condition over habitual condition. In English, words produced in a big mouth condition revealed greater duration than those in strong voice, pointing to benefits from the big mouth cue above and beyond those provided by strong voice. In French, the big mouth cue elicited word productions with the same durational increases as following the strong voice cue, suggesting that word-level prosodic constraints may have limited durational increases in the big mouth condition. Comparisons across studies should be made with caution, however, because of the studies' intrinsic differences, including the children's ages and dysarthria characteristics.

Increased vocal intensity was observed primarily in response to the strong voice cue, with greater increases than in the big mouth condition at word level, although big mouth also yielded significant increases in SPL relative to the habitual condition. These findings are in line with our hypotheses, as well as with previous treatment and cueing studies involving English-speaking children with dysarthria (Fox and Boliek 2012, Levy *et al.* 2017). SPL changes as a function of such cues may represent acoustic changes that are less specific to the target language, although whether cultural or other constraints impact vocal intensity changes remains to be explored further.

In response to both cues, significantly higher F1 values were observed in the subset of vowels measured,

reflecting greater jaw and tongue displacement likely due to increased articulatory displacement and vocal effort. In English-speaking children with dysarthria, in contrast, although F1 means appeared to be higher for the cued conditions, no significant group differences were found (Levy *et al.* 2017). It appears that the vowels produced by the French-speaking children in response to both cues more closely approximate target vocal tract shapes for the vowels. Individuals with dysarthria may exaggerate jaw movement to compensate for reduced tongue function (Nip *et al.* 2017); thus, the changes elicited by these cues may similarly help compensate for tongue movement limitations along the inferior– superior plane in some children with dysarthria.

The absence of statistically significant F2 changes may reflect previously reported limitations in tongue movement in the anterior–posterior plane for individuals with CP (Ansel and Kent 1992, Nip *et al.* 2017). Clearly, a better understanding is needed regarding articulatory–acoustic relationships, as well as regarding the degree to which modifying articulatory movements might improve intelligibility in dysarthria (Mefferd and Green 2010).

# Intelligibility changes in response to the cues

Counter to our hypotheses, at sentence level, gains in EOU in the cued conditions were not statistically significant, despite significant gains in duration and SPL. Explanations such as greater predictability of sentence level (compared with word level) stimuli, especially given that listeners heard all children producing the same sentences across conditions, will be evaluated in future work.

The finding of greater EOU in response to both big mouth and strong voice cues at the word level, hypothesized at the outset, was in line with Levy et al. (2017) and with adult studies on cued clear or loud speech (Tjaden et al. 2014). Thus, these cueing strategies show promise for increasing the EOU of words produced by Frenchspeaking children with dysarthria. Most children benefited from one cue or the other, although three children did not seem to benefit from either cue. Certainly, questions remain for further study regarding why one cue may benefit a particular child more than another. PWC followed a similar pattern, increasing from 38% in habitual condition to 43% following both big mouth and strong voice cues, but with increases not reaching significance, likely reflecting the considerable inter-speaker differences in this small sample.

The modest word-level gains in EOU, with no significant increases in PWC and large individual differences, highlight the challenge certain children with motor-based disorders face in improving their word intelligibility. Further research is needed to understand the source of the enhanced EOU in French at word level, in the absence of significant PWC increases. While more subjective than PWC, ratings are more sensitive to aspects of speech impairment than PWC in adults with dysarthria (Sussman and Tjaden 2012). Relevant to the present study, for two children who received similar PWC scores, for example, the child with more severe dysarthria might have required greater effort for the listeners to understand, resulting in differences in the children's EOU ratings, but not in their PWC. Examination of contrastive words (in various prosodic contexts), rhythms and intonation within and across languages may aid in evaluating effects of such cues on various vowel and consonant contrasts in children with dysarthria.

For clinicians, our findings suggest that treatment strategies emphasizing increasing articulatory excursion and vocal intensity show promise for improving EOU of words uttered by francophone children with dysarthria, although gains may be modest. In the present study, some children's intelligibility increased more in response to the big mouth cue and others' increased following the strong voice cue. Clinicians might consider including, or at least testing, clients' responses to, both cues.

# Limitations and future directions

The findings must be viewed with awareness of the study's limitations. First, the number of participants was small, and the children were heterogeneous with regard to age and severity of dysarthria, among other characteristics, limiting the conclusions that can be drawn. Future studies with larger numbers of participants should examine quantitively the effects of key factors, such as age and dysarthria severity, that may moderate the cueing effects. It is possible that the techniques were more effective for some children than for others. Qualitative inspection and the significant moderate-strong PWC correlations in figures S1 and S2 in the additional supporting information, for example, indicate that older children and those with milder dysarthria seem to benefit from the cues, whereas this is less the case for younger children and those with more severe dysarthria. Such information may have clinical relevance for determining appropriate therapeutic approaches for children with particular characteristics.

Second, for the sentence task, listeners heard various speakers producing the same sentences, potentially affecting their ratings.

Third, the French translation of TOCS+ sentences (Hodge *et al.* 2009) and the contrastive words of interest in French necessarily differed from their American English counterparts, as did participant characteristics. While these differences would not impact the withinsubject design of this study, differences in stimuli, participants, and design across studies, limit comparisons of results across studies and languages. Moreover, it is known that various instructions yield different acoustic and intelligibility changes (Lam and Tjaden 2016); thus, different terminology across languages (e.g., that *fort* means strong and loud in French) may affect speech differentially across languages. Therefore, examining responses to variations of the instructions could provide insight on optimizing the cueing terminology.

A final limitation is that, while speech modification studies are important for comparing the effects of cues on speech production, they do not necessarily predict long-term changes as a function of treatment. For example, although the articulation-focused LSVT ARTIC has shown increases in SPL and intelligibility in adults with PD, gains from LSVT LOUD (Ramig *et al.* 2018) have been longer lasting. While the present study examined productions immediately following modelling and cueing, treatment studies do not provide cues at preor post-test; therefore, any changes revealed represent longer term changes in speech. Thus, it is imperative to take the treatment-related studies to the next step and to test change following weeks of training, investigating children's retention of new speech behaviours.

# Conclusions

In this first study of the effects of global speech cues (French translations of 'speak with your big mouth' or 'speak with your strong voice') on acoustics and intelligibility in children with dysarthria who speak a language other than English, the overall findings for these French-speaking children suggest advantages and therefore, potential clinical utility, of both cues at word level. That is, the children can make use of cues to modify their speech, although the changes may only aid listeners' EOU at word level. Changes were limited and considerable speaker variability was also observed in intelligibility measures and ranking of speaking conditions, suggesting that this population may benefit from a combination of such cues to produce words that are easier to understand. Transcription accuracy and sentence-level EOU did not improve despite the significant acoustic changes; thus, the need remains for strategies that change speech acoustics sufficiently to enhance various measures of intelligibility in this population at both word and sentence levels. Understanding the impact of such cues is expected not only to expand the knowledge base for speech-language pathologists working in French but also to contribute to a database on which to base assessments of language-specific and more universal consequences of speech cues for intelligibility.

The consequences of the big mouth and strong voice cues for intelligibility and EOU of French-speaking children with dysarthria may provide preliminary evidence of language-specific characteristics potentially modulating the effects of cueing strategies. Examining how language-specific and more universal responses to speech cues relate to dysarthria treatment efficacy across languages and in bilinguals may advance research toward the goal of helping children with dysarthria of various linguistic backgrounds learn to speak with greater intelligibility.

#### Acknowledgments

Funding was provided by the Global Investment Fund and a Dean's Competitive Grant for Faculty Research at Teachers College, Columbia University, for the first author. Thanks to the participants, their families, the clinicians, the research assistants, and Joséphine Ancelle, Yannick Bleyenheuft, Joelle Coen, Lola Romero Gomez, Andrew Gordon, Bethany Hetrick, and Kyung Hae Hwang. Declaration of interest: The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

#### References

- ALLISON, K. M. and HUSTAD, K. C., 2018, Acoustic predictors of pediatric dysarthria in cerebral palsy. *Journal of Speech*, *Language, and Hearing Research*, **61**, 462–478.
- ANSEL, B. M. and KENT, R. D., 1992, Acoustic-phonetic contrasts and intelligibility in dysarthria associated with mixed cerebral palsy. *Journal of Speech, Language, and Hearing Research*, 35, 296–308.
- ASTÉSANO, C. and BERTRAND, R., 2016, Accentuation et niveaux de constituance en français: enjeux phonologiques et psycholinguistiques. *Langue Française*, 3, 11–30.
- BATES, D., MACHLER, M., BOLKER, B. and WALKER, S., 2015, Fitting linear mixed-effects models using lme4. *Journal of Statistical Software*, 67, 1–48.
- BOERSMA, P. and WEENINK, D., 2006, *Praat: Doing Phonetics by Computer (Version 4.4.11)* [computer program] (available at: http://www.praat.org)
- CANNITO, M. P., SUITER, D. M., BEVERLY, D., CHORNA, L., WOLF, T. and PFEIFFER, R. M., 2012, Sentence intelligibility before and after voice treatment in speakers with idiopathic Parkinson's disease. *Journal of Voice*, **26**, 214–219.
- CENTERS FOR DISEASE CONTROL AND PREVENTION (CDC), 2013, Cerebral Palsy (CP). [online] (available at: http://www.cdc.gov/ncbddd/cp/data.html)
- CHANG, E. W. and CHANG, Y. M., 2015, Word Transcription VAS & Sentence VAS (Version 1.0) [computer software].
- DUEZ, D., 2014, Some segmental and prosodic aspects of motor speech disorders in French. In N. Miller and A. Lowit (eds.), *Motor Speech Disorders: A Cross-Language Perspective* (Tonawanda, NY: Multilingual Matters), pp. 168–194.
- DUFFY, J. R., 2013, Motor Speech Disorders: Substrates, Differential Diagnosis, and Management (St. Louis, MO: Elsevier Mosby).
- FERRON, J. M., BELL, B. A., HESS, M. R., RENDINA-GOBIOFF, G. and HIBBARD, S. T., 2009, Making treatment effect inferences from multiple-baseline data: The utility of multilevel modeling approaches. *Behavior Research Methods*, 41, 372–384.
- FIELD, A. P., MILES, J. and FIELD, Z., 2012, *Discovering statistics using R* (Thousand Oaks, CA: Sage).
- FOX, C. M. and BOLIEK, C. A., 2012, Intensive voice treatment (LSVT LOUD) for children with spastic cerebral palsy and dysarthria. *Journal of Speech, Language, and Hearing Research*, 55, 930–945.

- HODGE, M., DANIELS, J. and GOTZKE, C. L., 2009, TOCS Intelligibility Measures (Version 5.3) [computer software] (Edmonton, Canada: University of Alberta).
- HSU, S. C., JIAO, Y., MCAULIFFE, M. J., BERISHA, V., WU, R. and LEVY, E. S., 2017, Acoustic and perceptual speech characteristics of native Mandarin speakers with Parkinson's disease. *Journal of the Acoustical Society of America*, **141**, 293–299.
- KHOMSI, A., 2001, *ELO: Evaluation du Langage Oral* (Paris: Les Editions du Centre de Psychologie Appliquée).
- LAM, J. and TJADEN, K., 2016, Clear speech variants: an acoustic study in Parkinson's disease. *Journal of Speech, Language, and Hearing Research*, **59**, 631–646.
- LEE, J., HUSTAD, K. C. and WEISMER, G., 2014, Predicting speech intelligibility with a multiple speech subsystems approach in children with cerebral palsy. *Journal of Speech, Language, and Hearing Research*, **57**, 1666–1678.
- LEE, T. and MCCANN, C., 2009, A phonation therapy approach for Mandarin-English bilingual clients with dysarthria. *Clinical Linguistics & Phonetics*, 23, 762–779.
- LENTH, R., LOVE, J. and HERVE, M., 2018, *emmeans: estimated* marginal means, aka least-squares means [R Package] (available at: https://CRAN.R-project.org/package=emmean)
- LEVY, E. S., 2014, Implementing two treatment approaches to childhood dysarthria. *International Journal of Speech-Language Pathology*, 16, 344–354.
- LEVY, E. S., 2018, 'Implementing speech intelligibility treatment for children with dysarthria'. American Speech Language and Hearing Annual Convention, Boston, MA.
- LEVY, E. S., CHANG, Y. M., ANCELLE, J. and MCAULIFFE, M., 2017, Acoustic and perceptual consequences of speech cues for children with dysarthria. *Journal of Speech, Language, and Hearing Research*, **60**, 1766–1779.
- LEVY, E. S. and LAW II, F. F., 2010, Production of French vowels by American-English learners of French: language experience, consonantal context, and the perception-production relationship. *Journal of the Acoustical Society of America*, **128**, 1290–1305.
- LEVY, E. S., RAMIG, L. O. and CAMARATA, S. M., 2012, The effects of two speech interventions on speech function in pediatric dysarthria. *Journal of Medical Speech-Language Pathology*, **20**, 82–87.
- LEWIS, M. P., SIMONS, G. F. and FENNIG, C. D., 2013, *Ethnologue: Languages of the World* (17 ed.) (Dallas, Texas: SIL International) [online] (available at: http://www.ethnologue.com)
- LISS, J. M., UTIANSKI, R. and LANSFORD, K., 2013, Crosslinguistic application of English-centric rhythm descriptors in motor speech disorders. *Folia Phoniatrica et Logopaedica*, 65, 3–19.
- MATLAB, 2015, Version 8.6 (R2015b) (Natick, Massachusetts: The MathWorks Inc.).
- MEFFERD, A. S. and GREEN, J. R., 2010, The articulatory-to-acoustic relationship in response to speaking rate and loudness manipulations. *Journal of Speech, Language, and Hearing Research*, 53, 1206–1219.
- MEI, C., REILLY, S., REDDIHOUGH, D., MENSAH, F. and MORGAN, A., 2014, Motor speech impairment, activity, and participation in children with cerebral palsy. *International Journal of Speech-Language Pathology*, 16, 427–435.
- MILLER, N., PENNINGTON, L., ROBSON, S., ROELANT, E., STEEN, N. and LOMBARDO, E., 2013, Changes in voice quality after speech–language therapy intervention in older children with cerebral palsy. *Folia Phoniatrica et Logopaedica*, 65, 200–207.
- MOEYAERT, M., RINDSKOPF, D., ONGHENA, P. and VAN DEN NOORT-GATE, W., 2017, Multilevel modeling of single-case data: A

comparison of maximum likelihood and Bayesian estimation. *Psychological Methods*, **22**, 760–778.

- MOYA-GALÉ, G., GOUDARZI, A., BAYÉS, A., MCAULIFFE, M., BULTÉ, B. and LEVY, E. S., 2018, The effects of intensive speech treatment on conversational intelligibility in Spanish speakers with Parkinson's disease. *American Journal of Speech-Language Pathology*, 27, 154–165.
- MOYA-GALÉ, G., MCAULIFFE, M. and LEVY, E. S., 2016, 'The effects of loudness and speaking rate manipulation on vowel production in Spanish speakers with Parkinson's disease'. Motor Speech Conference, Newport Beach, CA.
- NIP, I. S. B., ARIAS, C. R., MORITA, K. and RICHARDSON, H., 2017, Initial observations of lingual movement characteristics of children with cerebral palsy. *Journal of Speech, Language, and Hearing Research*, **60**, 1780–1790.
- NORDBERG, A., MINISCALCO, C., LOHMANDER, A. and HIMMEL-MANN, K., 2013, Speech problems affect more than one in two children with cerebral palsy: Swedish population-based study. Acta Paediatrica, International Journal of Paediatrics, 102, 161–166.
- PARADIS, J., DISPA, D., DE MONTPELLIER, A., EBNER-KARESTINOS, D., ARANEDA, R., SAUSSEZ, G. and BLEYENHEUFT, Y., 2019, Interrater reliability of activity questionnaires after an intensive motor-skill learning intervention for children with cerebral palsy. Archives of physical medicine and rehabilitation, 100, 1655–1662.
- PARK, S., THEODOROS, D., FINCH, E. and CARDELL, E., 2016, Be clear: a new intensive speech treatment for adults with nonprogressive dysarthria. *American Journal of Speech-Language Pathology*, 25, 97–110.
- 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**, 337–44.
- 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, 464–471.
- PENNINGTON, L., LOMBARDO, E., STEEN, N. and MILLER, N., 2018, Acoustic changes in the speech of children with cerebral palsy following an intensive program of dysarthria therapy. *International Journal of Language & Communication Disorders*, 53, 182–195.
- PERKELL, J. S., ZANDIPOUR, M., MATTHIES, M. L. and LANE, H., 2002, Economy of effort in different speaking conditions: I. A preliminary study of intersubject differences and modeling issues. *Journal of the Acoustical Society of America*, **112**, 1627– 1641.
- PINHEIRO, J.C. and BATES, D.M., 2000, *Mixed-effects models in S and S-PLUS* (New York: Springer).
- PINTO, S., CHAN, A., GUIMARÃES, I., ROTHE-NEVES, R. and SA-DAT, J., 2017, A cross-linguistic perspective to the study of dysarthria in Parkinson's disease. *Journal of Phonetics*, 64, 156– 167.
- R CORE TEAM, 2018, *R: A language and environment for statistical computing* (Vienna, Aaustria: R foundation for statistical computing).
- RAMIG, L.O., HALPERN, A., SPIELMAN, J., FOX, C. and FREEMAN, K., 2018, Speech treatment in Parkinson's disease: randomized controlled trial (RCT). *Movement Disorders*, 33, 1777–1791.
- RAMIG, L. O., SAPIR, S., COUNTRYMAN, S., PAWLAS, A. A., O'BRIEN, C., HOEHN, M. and THOMPSON, L. L., 2001, Intensive voice treatment (LSVT) for patients with Parkinson's disease: a 2 year follow up. *Journal of Neurology, Neurosurgery & Psychiatry*, 71, 493–498.

- RAUDENBUSH, S.W. and BRYK, A.S., 2002, *Hierarchical linear models: Applications and data analysis methods.* 2nd ed. (Thousand Oaks, CA: Sage Publications).
- SAUVAGEAU, V. M., ROY, J. P., LANGLOIS, M. and MACOIR, J., 2015, Impact of the LSVT on vowel articulation and coarticulation in Parkinson's disease. *Journal of Clinical Linguistics & Phonetics*, **29**, 424–440.
- SMILJANIĆ, R. and BRADLOW, A. R., 2005, Production and perception of clear speech in Croatian and English. *Journal of the Acoustical Society of America*, **118**, 1677–1688.
- SMILJANIC, R. and BRADLOW, A. R., 2009, Speaking and hearing clearly: Talker and listener factors in speaking style changes. *Language and Linguistics Compass*, 3, 236–264.
- SUSSMAN, J. E. and TJADEN, K., 2012, Perceptual measures of speech from individuals with Parkinson's disease and multiple sclerosis: Intelligibility and beyond. *Journal of Speech, Language, and Hearing Research*, 55, 1208–1219.
- TJADEN, K., LAM, J. and WILDING, G., 2013, Vowel acoustics in Parkinson's disease and multiple sclerosis: comparison of clear, loud, and slow speaking conditions. *Journal of Speech, Language, and Hearing Research*, **56**, 1485– 1502.
- TJADEN, K., SUSSMAN, J. E. and WILDING, G. E., 2014, Impact of clear, loud, and slow speech on scaled intelligibility and speech severity in Parkinson's disease and multiple sclerosis. *Journal of Speech, Language, and Hearing Research*, **57**, 779–792.

# **Supporting Information**

Additional supporting information may be found online in the Supporting Information section at the end of the article.

**Figure S1.** Percentage of words orthographically transcribed correctly (PWC): scatterplot showing the relationship between differences between conditions (effect of cues) and age.

**Figure S2.** Percentage of words orthographically transcribed correctly (PWC): scatterplot showing the relationship between differences between conditions (effect of cues) and dysarthria severity.

**Table S1.** Listeners' ratings of ease of understanding(EOU) of words by child and condition.

**Table S2.** Listeners' ratings of ease of understanding (EOU) of words: descriptive statistics for the differences between conditions.

**Table S3.** Listeners' ratings of ease of understanding (EOU) of sentences by child and condition.

**Table S4.** Listeners' ratings of ease of understanding (EOU) of sentences: descriptive statistics for the differences between conditions.

**Table S5.** Percentage of words orthographically transcribed correctly (PWC) by child and condition.

**Table S6.** Percentage of words orthographically transcribed correctly (PWC): descriptive statistics for the differences between conditions.

**Table S7.** Descriptive statistics for vowel first formant (F1) of contrastive words by child and condition.

**Table S8.** Vowel first formant (F1) of contrastive words: descriptive statistics for the differences between conditions.

**Table S9.** Descriptive statistics for vowel second formant (F2) of contrastive words by child and condition.

**Table S10.** Vowel second formant (F2) of contrastive words: descriptive statistics for the differences between conditions.

**Table S11.** Descriptive statistics for word duration in seconds by child and condition.

**Table S12.** Word duration in seconds: descriptive statistics for the differences between conditions.

 
 Table S13. Descriptive statistics for word soundpressure level (SPL) in dB by child and condition.

**Table S14.** Word sounds-pressure level (SPL) in dB: descriptive statistics for the differences between conditions.

**Table S15.** Descriptive statistics for sentence duration in seconds by child and condition.

 Table S16.
 Sentence duration in seconds: descriptive statistics for the differences between conditions.

 
 Table S17. Descriptive statistics for sentence soundpressure level (SPL) in dB by child and condition.

**Table S18.** Sentence sound-pressure level (SPL) in dB: descriptive statistics for the differences between conditions.