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*. Advanced online publication.
https://doi.org/10.1044/2020_JSLHR-20-00258
(Accepted for publication Dec. 8, 2020)

Speech Treatment Effects on Narrative Intelligibility in French-Speaking Children with Dysarthria

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The authors declare no conflict of interest at the time of publication.
This project was supported by Teachers College, Columbia University, through a Global Investment Fund and a Dean’s Competitive Grant for Faculty Research (awarded to Erika S. Levy).

Abstract

Purpose: This study examined the effects of Speech Intelligibility Treatment (SIT) on intelligibility and naturalness of narrative speech produced by francophone children with dysarthria due to cerebral palsy (CP).

Method: Ten francophone children with dysarthria were randomized to one of two treatments, Speech Intelligibility Treatment or Hand-arm Bimanual Intensive Therapy Including Lower Extremities, a physical therapy (PT) treatment. Both treatments were conducted in a camp setting and were comparable in dosage. The children were recorded pre- and post-treatment producing a story narrative. Intelligibility was measured by means of 60 blinded listeners' orthographic transcription accuracy (percentage of words transcribed correctly). The listeners also rated the children's naturalness on a visual analogue scale.

Results: A significant pre- to post-treatment increase in intelligibility was found for the SIT group, but not for the PT group, with great individual variability observed among the children. No significant changes were found for naturalness ratings or sound pressure level in the SIT group or the PT group post-treatment. Articulation rate increased in both treatment groups, although not differentially across treatments.

Conclusion: Findings from this first treatment study on intelligibility in francophone children with dysarthria suggest that SIT shows promise for increasing narrative intelligibility in this population. Acoustic contributors to the increased intelligibility remain to be explored further.

Introduction

Cerebral palsy (CP) is the most common motor disability in children and is estimated to occur in 1.5-4 per 1,000 live births worldwide (Centers for Disease Control and Prevention, 2018). The motor speech disorder of dysarthria is frequently associated with CP (Schölderle et al., 2016), with prevalence data reported ranging from 21% to 90% (Nordberg et al., 2013; Mei et al., 2014).

Dysarthria in CP is characterized by decreased consonantal and vocalic contrasts (Ansel & Kent, 1992; Coleman & Meyers, 1991; Kim et al., 2011; Platt, Andrews, & Howie, 1980), reduced prosodic control (Patel, 2002, 2003, 2004), reduced breath support (Redstone, 2004), decreased speaking rate (Allison & Hustad, 2018; Darling-White, Sakash, & Hustad, 2018), and atypical resonance (Allison & Hustad, 2018). Concomitant with these physiological and acoustic variables, decreased speech intelligibility is a prominent characteristic of dysarthria (Darley et al., 1969). Intelligibility refers to the extent to which the acoustic signal produced by a speaker can be decoded by a listener (Kent et al., 1989; Hustad et al., 2012; Yorkston & Beukelman, 1980) and is therefore crucial for communication, social participation, and quality of life (Walshe & Miller, 2011).

Speech intelligibility in children with dysarthria due to CP is reduced compared to that of typically-developing children (Braza et al., 2019; Hustad et al., 2010; Hustad et al., 2012) and of children with CP with no speech motor impairment (Hustad et al., 2020). Moreover, intelligibility in children with CP with no speech motor deficits is also reduced compared to that of typically developing children (Hustad et al., 2019). Decreased intelligibility not only hinders social participation, but it has also been reported to impact school success (Dickinson et al., 2007). The importance of intelligibility in children with dysarthria, therefore, cannot be overestimated.

Intelligibility in this population has been studied at the single word level, as well as in repeated phrases (e.g., Hustad et al., 2012; Pennington et al., 2013) and in connected speech in response to simple questions (Pennington et al., 2013). However, these methods do not provide a full representation of how children communicate on a daily basis. In narrative language tasks, children are typically asked to compose or retell a story (Danahy Ebert & Scott, 2014), allowing the examiner to gain insight into how children use language in real-world situations (Costanza-Smith, 2010; Danahy Ebert & Scott, 2014). Despite the stronger ecological validity of narrative intelligibility (compared to words in isolation or repeated phrases, for example), very little is known about how speech treatment may lead to improvements in this construct.

In an increasingly globalized world, evidence-based speech treatment research in languages other than English is surprisingly lacking, especially in the area of motor speech disorders (Miller et al., 2014). French is spoken by 68.5 million speakers across 51 countries and is one of the 20 most spoken languages in the world (Lewis et al.2013). To our knowledge, no published studies have examined the effects of any treatment program on the intelligibility or acoustic speech characteristics of francophone children with dysarthria. Consequently, speech-language pathologists (SLPs) in this language community do not have evidence-based guidelines by which to tailor their treatments for French-speaking children's communication. Thus, conducting treatment research with this population is essential to establishing a scientific foundation for their treatment.

Cueing and treatment studies on dysarthria in CP

Recently, a speech cueing study on eight American English (AE) speaking children with dysarthria secondary to CP found increases in the children's vocal intensity, speech duration and intelligibility when the children were cued to "speak with a big mouth" or "speak with a strong voice" (Levy et al., 2017). These cues are aimed to increase articulatory excursion and vocal intensity, respectively.

Little is known about the speech characteristics of French speakers with dysarthria, and even less about the possible acoustic and perceptual consequences of speech treatment for this population. From a physiological perspective, it could be assumed that a cue targeting increased vocal intensity would elicit the same changes in any language, i.e., increased sound pressure level (SPL). Consistent with the notion of cross-linguistic changes due to dysarthria, vowel space has been reported to be reduced in speakers with dysarthria from various language backgrounds (e.g., Baumann et al., 2018), including French (Martel Sauvageau et al., 2015). In a study on Quebec French speakers with Parkinson's disease, vowel space area expanded after intensive speech treatment targeting increased vocal intensity (Martel Sauvageau et al., 2015), paralleling findings from English studies (e.g., Sapir et al., 2007). It is possible then that speech treatment might also result in vowel space expansion in other populations with motor speech impairments, such as children with CP. Findings such as these could point towards a possible universal benefit of treatments that promote louder and clearer speech.

How such benefits would translate into intelligibility gains across languages is subject to further exploration and may be mediated by language-specific constraints. For example, Levy, Moya-Galé, Chang, Campanelli et al. (2020) hypothesized that words at the end of sentences might be most likely to show intelligibility gains in dysarthria in French speakers, given the stress patterns in this language. In the first cueing study examining the effects of global cues in francophone children with dysarthria, Levy, Moya-Galé, Chang, Campanelli et al. (2020) found that cues aimed to increase articulatory excursion and vocal intensity show promise for improving ease of understanding of the children's words, although improvements might be modest. Specifically, increased listener visual analogue scale (VAS) ratings of ease of understanding at the word level were found when the children were cued to speak with their *grande bouche* ['big mouth'] or *grosse voix* ['strong voice'], consistent with the AE cueing study. Both cues also elicited increased SPLs and word duration, as well as higher first-formant frequencies for select vowels measured, suggesting a lower tongue position. In response to the "big mouth" cue, sentence duration increased. However, no significant increases in transcription accuracy at the word level or ease of understanding at the sentence level were observed, suggesting that the children with dysarthria may require more intensive practice to achieve significant motor changes. The children's diverse responses to the cues suggest that a combination of such cues may benefit francophone children with dysarthria.

In speech cueing studies, investigators provide global cues to participants during data collection and examine changes in the participants' immediate communication. Treatment research, in contrast, examines changes as a result of a longer-term intervention. In speech treatment studies, speakers are not cued to modify their speech during data collection.

Treatment research is sparse in children with dysarthria. However, promising results have been found for American English-speaking children with dysarthria in response to global speech treatments, such as the Lee Silverman voice treatment (LSVT LOUD®; Fox & Boliek, 2012) and Speech Intelligibility Treatment (SIT; Levy, 2014, 2018; Levy et al., in press), which focus on one or two cues or instructions (e.g., "speak loud" or "speak with your big mouth and strong voice”). Positive results have also been reported for more traditional multi-system treatments, such as the speech systems approach (e.g., Pennington et al., 2010). A brief overview of findings from these treatment approaches is provided below.

Treatment Studies on English-Speakers With Dysarthria Due to CP

Speech Systems Approach

The speech systems approach (Pennington et al., 2010; Pennington et al., 2006; Yorkston et al., 1999) targets increased breath support and phonation and decreased speech rate. This approach has revealed positive results for children with dysarthria. The premise of this approach is that dysarthria affects not only articulation (Strand, 1995) but also the respiratory and phonatory subsystems, which should therefore also be addressed in treatment. Recent studies focusing on a speech systems approach have explored improving intelligibility and communicative participation in younger children with dysarthria in traditional settings (Pennington et al., 2013) as well as via teletherapy (Pennington et al., 2019). Overall, significant gains in intelligibility were observed, but results also revealed the variability in performance in these children. Acoustic consequences of speech systems treatment in older children with dysarthria have also been examined; however, only changes in vocal intensity at the single word level and decreased fundamental frequency during connected speech were associated with previously reported intelligibility gains (Pennington et al., 2018).

Lee Silverman Voice Treatment (LSVT LOUD)

LSVT LOUD is an intensive speech treatment that utilizes a single focus on healthy vocal loudness to improve vocal function and was originally designed for individuals with hypokinetic dysarthria secondary to Parkinson's disease (PD; Ramig et al., 1994; Ramig et al., 2001). Throughout the decades, LSVT LOUD has been implemented with various other populations, including adults with multiple sclerosis (Sapir et al., 2001), cerebellar dysfunction (Sapir et al., 2003), stroke (Mahler & Ramig, 2012) or down syndrome (Mahler & Jones, 2012), and children with CP (Boliek & Fox, 2017; Fox & Boliek, 2012). In a multiple baseline single subject study, Fox and Boliek (2012) investigated the effects of this treatment on perceptual and acoustic voice characteristics of four children with spastic CP. Results from seven raters indicated increased listener preference for post-treatment speech samples for vocal intensity, voice quality and pitch variability, with minimal changes observed in the acoustic measures analyzed, such as SPL and fundamental frequency (F0) range. Furthermore, in a Phase I small group pre-to-post treatment design with three children with CP, Levy et al. (2013) compared a systems-based approach to LSVT LOUD. The systems-based approach focused on respiration-phonation coordination, posture, speech clarity, and regulation of speech and vocal intensity. Results indicated similar post-treatment improvements in listeners’ ease of understanding of the children’s speech in both groups, although only the children who received LSVT LOUD increased vocal intensity post-treatment. Similar results were found in a Phase I treatment validation study with seven children with CP who received LSVT LOUD and seven neurotypical controls (Boliek & Fox, 2017). This study included acoustic measures of vocal fold vibration (i.e., jitter and shimmer), which decreased significantly post-treatment, suggesting positive physiological changes in the children with CP after treatment. No objective measure of intelligibility (i.e., transcription accuracy), however, was provided in these studies.

Speech Intelligibility Treatment (SIT)

Based on the literature on the benefits of global speech cues in dysarthria (Fox & Boliek, 2012; Levy et al., 2017; Tjaden & Wilding, 2004), Speech Intelligibility Treatment (SIT; Levy, 2014, 2018; Levy et al., in press) was developed specifically for children with dysarthria due to CP. SIT is a dual-focus treatment approach that aims to increase intelligibility by targeting increased articulatory working space and vocal intensity. This treatment follows a camp format, in which intensive speech treatment is delivered for 6.5 hours daily, five days a week for three weeks, plus 15 minutes of homework daily. During treatment, children are instructed to produce speech in a variety of linguistic tasks using their "big mouth" and "strong voice." In a recent treatment study on the effects of SIT on intelligibility and communicative participation in 17 children with dysarthria, Levy et al. (in press) found that intelligibility of narrative speech, as measured by ease of understanding ratings, improved significantly following treatment, with gains maintained at six-week follow-up. Additionally, a statistically significant improvement was found in all but one of the categories of communicative participation assessed immediately post-treatment and at a six-week follow-up. Despite the variability observed among the children with dysarthria, these results provide preliminary evidence of beneficial effects of SIT for improving intelligibility and overall communication in AE-speaking children with dysarthria. However, to our knowledge, no research has yet examined treatment-related changes in the speech of francophone children with dysarthria.

The present study investigated the effects of SIT on the narrative intelligibility of francophone children with dysarthria due to CP. We also investigated the effects of the treatment on two acoustic variables associated with the cues implemented in SIT (i.e., "big mouth" and "strong voice"), SPL and articulation rate. It was hypothesized that children receiving this intensive speech treatment would increase their intelligibility during narrative speech and that this increase would be accompanied by increased vocal intensity and reduced articulation rate.

The hypothesis of increased intelligibility was based on the intelligibility gains yielded when (French- and English-speaking) children with CP were cued to speak with a "big mouth" or "strong voice" (Levy et al., 2017; Levy, Moya-Galé, Chang, Campanelli et al., 2020), as well as when English-speaking children were trained in the speech systems treatment approach (Pennington et al., 2013) or LSVT LOUD (Boliek & Fox, 2017). Vocal intensity also increased when children with dysarthria were trained in LSVT LOUD (Boliek & Fox, 2014) or cued to speak with a "big mouth" or "strong voice" (Levy et al., 2017; Levy, Moya-Galé, Chang, Campanelli et al., 2020). Decreased articulatory rate was expected based on increased utterance duration found by Levy and her colleagues when (French- and English-speaking) children with dysarthria were cued to speak with their "big mouth" (Levy et al., 2017; Levy-Moya-Galé et al., 2020). Rate reduction may stem from the longer time required to reach targets in an expanded articulatory space, as has been found in studies of clear speech (Bradlow et al., 2003; Lam et al., 2012; Lam & Tjaden, 2016).

Naturalness was a secondary variable of interest in the study. Naturalness can be operationalized as the degree to which speech "conforms to the listener's standards of rate, rhythm, intonation, stress patterning, and …to the syntactic structure of the utterance being produced" (Yorkston et al., 1999, p. 464). Perception of naturalness has been negatively correlated with the psychoacoustic variable of monopitch (Anand & Stepp, 2015). Prosody-based treatment approaches have been traditionally perceived as promising strategies for improving naturalness (Liss, 2007). It was expected that SIT, with its "strong voice" cue, might have a positive effect on perceived naturalness. Typical vocal intensity is a component of naturalsounding speech (Patel et al., 2011), and increasing vocal intensity has been observed to yield benefits beyond SPL, such as increased pitch variation. Boliek and Fox (2014) found a positive effect on pitch variation at the sentence level following LSVT LOUD in a single case study of a child with dysarthria due to CP. While the improved pitch variation could be attributed, at least in part, to the pitch gliding exercises in the LSVT LOUD protocol, greater pitch variation as a result of simply following cues to speak loud has also been documented in healthy adults (e.g., Dromey & Ramig, 1998; Patel & Schell, 2008; Watson & Hughes, 2006) and in adults with dysarthria secondary to PD and multiple sclerosis (Tjaden & Wilding, 2011). Thus, the greater intensity elicited by the "strong voice" cue might be accompanied by greater pitch variation and therefore, naturalness. Because naturalness has barely been explored in treatment studies with children with dysarthria and CP, however, our hypothesis regarding this secondary outcome variable was guarded.

To the authors' knowledge, this is the first speech treatment study on French-speaking children with dysarthria. Additionally, it is the first dysarthria treatment study to include an active treatment comparator matched on all variables except the treatment target of speech.

Methods

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

Children with dysarthria

Ten Belgian-French speaking children with dysarthria secondary to CP (six males and four females) participated in this study. Recruitment occurred in outpatient clinics that specialized in CP and through a local rehabilitation foundation website (https://sites.google.com/site/intensiverehabfoundation/). Seven children had a medical diagnosis of spastic quadriplegia and three, of dyskinetic quadriplegia. Motor abilities were assessed by a physical or occupational therapist. The children's speech was evaluated by three licensed SLPs, who determined presence, characteristics and severity of dysarthria via consensus based on clinical signs of motor speech deficits perceived auditorily and/or visually (Fox & Boliek, 2012; Levy et al., 2017; McAuliffe et al., 2014). In order to be included in the study, children had to have speech as their primary mode of communication, be identified as having speech that was "difficult to understand" by their parents and/or teachers, pass a bilateral pure tone hearing screening at 20dB HL for 500, 1000, 2000 and 4000Hz, and be able to follow clinicians' directions (Fox & Boliek, 2012). Additionally, the children's parents had to be willing to enroll their children in the randomized controlled trial. Nine of the ten children in this study also participated in Levy, Moya-Galé, Chang, Campanelli et al.'s (2020) cueing study; however, the tasks, stimuli, and timeline differed between the two studies. In the present investigation, for example, treatment was conducted, post-tests were performed, and speech cues were not provided during testing.

The children's characteristics are presented in Table 1 below.

Child	Age	Sex	Diagnosis	GMFCS	Dysarthria Severity	Perceptual Impression	Treatment Group
CPF1	4;11	F	Spastic quadriplegia	IV	Moderate	Increased vocal intensity, moderate hypernasality, imprecise articulation, several phonological processes	PT
CPF2	5;1	M	Spastic quadriplegia	III	Mild	Breathy voice quality, mild hypernasality, imprecise articulation	SIT
CPF3	7;1	M	Dyskinetic quadriplegia	III	Mild-moderate	Reduced vocal intensity, mild hypernasality, slow rate, inconsistently imprecise articulation, prosodic abnormalities (breaths within utterances)	SIT
CPF4	8;9	M	Spastic quadriplegia	III-IV	Severe	Strained vocal quality, very imprecise articulation, consonant deletion	SIT
CPF5	9	M	Spastic quadriplegia	IV	Moderate-moderately severe	Reduced vocal intensity, imprecise articulation, strained vocal quality	PT
CPF6	9;5	M	Dyskinetic quadriplegia	III	Mild	Monotone, slow rate, imprecise articulation	PT
CPF7	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)	SIT
CPF8	11;1	M	Spastic quadriplegia	IV	Mild	Moderately fast rate	PT

CPF9	12;3	F	Spastic quadriplegia	IV	Moderate-moderately severe	Strained vocal quality, monotone pitch, imprecise articulation	SIT
CPF10	16;2	F	Spastic quadriplegia	III	Mild	Reduced intensity, breathy voice quality	PT
Note. Perceptual impressions were determined by three experienced speech-language pathologists. GMFCS = Gross Motor Function Classification System; M = male; F = female; SIT = Speech Intelligibility Treatment; PT = physical therapy							

Procedure

This study took place within the environment of a summer camp program conducted in Parc Parmentier, a park in Brussels, Belgium, dedicated to activities fostering the development of youth from disadvantaged backgrounds. Children with dysarthria were recorded engaging in a variety of speech tasks pre-treatment and immediately post-treatment. The current study reports on their production of a story narrative, which was elicited with sequence picture cards. (Other speech measures were also collected, but not analyzed for this study.) Testing took place in a quiet room, which was a dormitory room transformed into a recording space by the experimenters. This room was in a separate building from the treatment space. The testing was conducted by a local SLP graduate student, blinded to the children's treatment conditions, so as to maximize experimental control and reduce bias.

Treatment Protocols: Speech Intelligibility Treatment and Physical Therapy

The 10 children with dysarthria were randomized into either SIT or the Hand-arm Bimanual Intensive Therapy Including Lower Extremities (Bleyenheuft & Gordon, 2014; henceforth referred to as physical therapy or PT), which served as an active treatment comparator. The treatment groups were matched in terms of service delivery model (group and individual therapy provided in camp format) and intensive treatment dosage, as treatment was provided for 6.5 hours a day for 15 days over three weeks. SIT targets increased articulatory working space and vocal intensity by means of child-friendly cues aimed at increasing intelligibility. During speech treatment, children were instructed to speak with their *grande bouche* ['big mouth'] and *grosse voix* ['strong voice'] in a variety of tasks that increased in linguistic complexity and cognitive processing demands over time. Individual and group activities were designed by the graduate clinicians and tailored to the children's interests, eliciting as much speech output as possible. Breaks during treatment were provided as needed. A summary of the protocol is provided in Levy et al. (in press).

The PT treatment consisted of intensive training that included postural control and upper and lower extremity (hand-arm and leg) function. The full protocol can be found in Bleyenheuft and Gordon (2014). All children who were enrolled in the current study completed their treatment program, with none lost to follow-up.

Speech Stimuli

The stimuli described here are a subset of a larger battery of stimuli used to examine speech production of francophone children with dysarthria secondary to CP. Recording procedures are described in Levy, Moya-Galé Chang, Campanelli et al. (2020). In short, children wore an EMW Omnidirectional Lavalier microphone that was taped to their forehead and was secured with a headband. Stimuli were obtained using a ZOOM H4n digital recorder and microphone-to-mouth distance was consistently maintained at 8cm across children. Calibration was conducted at the beginning and end of each recording session to ensure the SPL level during presentation of the speech stimuli to listeners was representative of the child's vocal intensity (Fox & Boliek, 2012; Levy et al., 2017; Ramig et al. 2001; Švec & Granqvist, 2018). Children were instructed to narrate a set of seven pictures depicting an age-appropriate story. Verbal prompts were provided if the child appeared to be off task or fatigued. If extraneous noise occurred during the recording, the child was prompted to repeat the utterance. Breaks were provided as needed.

In order to prepare the stimuli for the perceptual tasks, two francophone graduate research assistants, who were SLP students, selected three pre-treatment and three post-treatment utterances from the story narrative. Utterances selected were the first three semantically and grammatically correct sentences produced by each child in each session. The number of words pre-to-post treatment was not significantly different within or across treatment groups (mean = 15.4, SD = 5.86 at pre-test and mean = 16.6, SD = 4.83 at post-test for SIT group; mean = 19.6, SD = 7.09 at pre-test and mean = 23.4 , SD = 9.71 at post-test for PT group). These utterances were subsequently orthographically transcribed by the experimenters and compared for inter-scorer reliability (Gordon-Brannan & Hodson, 2000; Kwiatkowski & Shriberg, 1992). Discrepancies (approximately 4% of the stimuli) were discussed with the first and last authors and reanalyzed to reach a consensus.

Listeners

Sixty Belgian French-speaking neurotypical adults (32 men, 28 women; average age = 25 years; range = 18-52 years) participated in the intelligibility and naturalness assessment task. These listeners were different from those described in Levy, Moya-Galé, Chang, Campanelli et al. (2020). They were recruited from the Liège area in Belgium and passed a bilateral pure-tone hearing screening at 25dB HL 500, 1000, 2000 and 4000 Hz (American National Standards Institute, 2004). Listeners reported no history of speech or language problems and no prior experience with motor speech disorders. Listeners were paid 15 euros for their participation.

Intelligibility and naturalness assessment:

Listeners completed two perceptual tasks free-field on an HP Pavilion laptop computer using custom-developed software (Chang & Chang, 2015) programmed in MATLAB (Version R2018b, The MathWorks, Inc., 2015). Listeners were seated by a desk with the laptop on it. They were instructed to sit comfortably and not to lean towards the stereo loudspeakers (Logitech Z150), which were directly connected to the laptop. A distance of 85 cm was maintained between listeners and the loudspeakers to represent an average distance between conversational partners (Hall, 1966). The experiment took approximately 30 minutes to complete.

A familiarization task was completed preceding both tasks. Listeners were presented with three utterances produced by a child with dysarthria who was not part of the study. For each utterance, listeners were instructed to write down word for word what they heard (sentence transcription task). Next, they were instructed to rate the naturalness of the same utterance on a VAS (naturalness rating task) from 0-100 with anchors 0 = *pas du tout naturelle* ['not at all natural'] and 100 = *très naturelle* ['very natural'] before they were presented with the next utterance. In order to avoid familiarity effects, listeners always completed the sentence transcription task first (Borrie, McAuliffe, & Liss, 2012). Listeners were able to ask any clarification questions before the beginning of the test. None of the listeners requested a break during completion of the perceptual tasks.

Sentence transcription and naturalness ratings tasks

In the sentence transcription task, listeners heard a total of 60 utterances from the story narratives produced by the children with dysarthria (3 utterances per data collection point x 10 speakers). Presentation of utterances was randomized and blocked by child in order to minimize order effects. Pre- and post-treatment utterances within-child were randomized, as well. For each utterance, listeners were instructed to write down what they heard to the best of their ability and then to rate the utterance for naturalness on the VAS. Utterances were only played once in order to avoid learning effects (Wilson, Bell, & Koslowski, 2003).

Data Analysis

Perceptual and Acoustic Analysis

Transcription accuracy, considered an objective measure of intelligibility (Hustad, 2006), was calculated as percentage of words correctly transcribed. Orthographic transcriptions were considered correct if words matched the target exactly or if differences were due to homonyms or obvious misspellings (Cannito et al., 2012; Hustad, 2007; Levy, Moya-Galé, Chang, Freeman, et al., 2020). Mean naturalness ratings were calculated from the VAS responses.

Two acoustic variables were examined across all narrative utterances: SPL and articulation rate. These measures were chosen as they correspond to acoustic changes expected in SIT, i.e., increased vocal intensity and decreased speech rate (Levy et al., 2017; Levy, Moya-Galé, Chang, Campanelli et al., 2020), and have been widely reported in the literature on loud and clear speech (Smiljanić, & Bradlow, 2009; Tjaden et al., 2014; Tjaden & Wilding, 2004). Utterances were manually analyzed by two graduate research assistants using Praat software (Boersma & Weenink, 2006). Standard criteria for acoustic measurements were used to establish onset and offset (Klatt, 1975; Levy & Law, 2010). Because input levels were not changed during the recording sessions, vocal intensity was extracted by measuring average SPL in each utterance using both the wideband spectrogram and waveform from the software. Pauses of greater than 0.150s were removed for SPL and articulation rate analysis (Robb et al., 2004). Pause and sentence durations were measured in seconds (Darling-White et al., 2018).

Reliability

Inter-rater reliability for transcription accuracy (TA) and naturalness (NA) outcomes was calculated using intraclass correlations for correspondence via a two-way random effects approach treating listeners as random. Because the average rating was of interest, we present estimates for both a single listener and the average of 60 listeners; that is, ICC (2,1) and ICC (2,60) (cf., Shrout & Fleiss, 1979). For transcription accuracy, the ICC for a single listener was .87, while for the average of 60 listeners, it was >.99. For naturalness ratings, the ICC for a single listener was .25, while for the average of 60 listeners, it was .95. That is, single listeners were very reliable when transcribing but not reliable when rating naturalness. Averaging over all 60 raters, however, both transcription accuracy and naturalness ratings were quite reliable.

A second judge randomly selected and reanalyzed 20% of the data in order to evaluate the reliability of the acoustic findings (Levy et al., 2017). Cronbach's α for the acoustic measures was estimated to be 0.95.

Statistical Analysis

Perceptual Analysis

Mixed-effects regression models were fit with crossed random effects for Listener and Child. The data were organized in long format to allow the PRE/POST time factor, the PT/SIT treatment factor, and their two-way interaction to be included as fixed effects in the regression models. Separate models were run for both primary perceptual outcomes: transcription accuracy and naturalness ratings. As an example, transcription accuracy (TA) is modeled as

$$TA_{ijk} = \beta_0 + \beta_1 TIME_{ijk} + \beta_2 SIT_{ijk} + \beta_3 (TIME * SIT)_{ijk} + u_{0j} + u_{0k} + \epsilon_{ijk}$$

where TA_{ijk} is the TA for sample i from participant j as rated by listener k at time $TIME_{ijk}$, $u_{0j} \sim N(0, \tau_0^2)$ and $u_{0k} \sim N(0, \tau_1^2)$ are normally distributed random intercepts for Child and Listener, respectively, $\epsilon_{ijk} \sim N(0, \sigma^2)$ is residual error, and the random effects are modeled as mutually independent.

The mixed effects models formulated as above are essentially generalizations of the random effects ANOVA model with the advantage that the mixed-effects regression approach can accommodate missing data and potentially unbalanced data sets without requiring listwise deletion of cases.

Acoustic Analysis

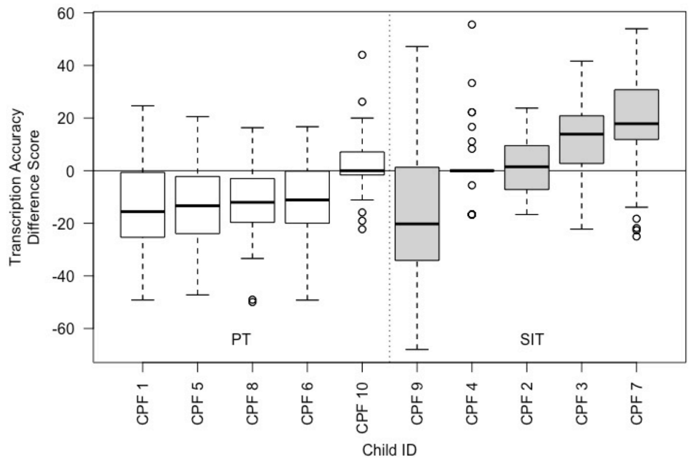
The assessment of sound pressure level (SPL) and articulation did not involve the set of 60 listeners; however, three utterances per child were recorded (i.e., children's data were repeatedly measured). Thus, for the acoustic analyses, a random intercept for Child (but not for Listener) was included in the mixed-effects regression model, which was otherwise formulated as described above.

Results

Transcription accuracy and naturalness ratings

Table 2 shows descriptive statistics of mean and SD for TA and naturalness ratings. Table 3 shows results from the mixed effects model fits. For TA, estimates of variance components reveal that child-to-child variability explains about 81% of total variance in transcription accuracy, listener-to-listener variability contributes only about 2%, and the remaining 17% is residual variation that cannot be explained by either Child or Listener. Fixed effects results for transcription accuracy show a significant treatment x time interaction ($\hat{\beta}_3 = 12.4; p < .001$). That is, the average pre-to-post gain in transcription accuracy in the SIT arm is about 12 points higher than in the PT arm. The significant coefficient for the TIME main effect ($\hat{\beta}_1 = -9.4; p < .001$), suggests that for those assigned to the PT arm (i.e., SIT = 0), the average TA decreased by approximately 9 points from pretest to post-test. Thus, the differential (pre-to-post) average change in TA of approximately 12 points was driven both by losses in the PT arm and by gains in the SIT arm; see Figure 1 for boxplots of the pre-to-post gain scores, stratified by participant and treatment group.

Figure 1. Individual speakers' gain score difference for transcription accuracy in Speech Intelligibility Treatment (SIT) and physical therapy (PT) groups.



For naturalness we find no evidence of a treatment effect in either direction ($\hat{\beta}_3 = 1.64; p = .41$). This finding may be verified by visual inspection of Figure 2, which shows boxplots of difference scores for the naturalness outcome; note that the profiles are nearly identical across treatment conditions. The significant coefficient for the main effect of the treatment ($\hat{\beta}_2 = -11.99; p = .04$), suggests that the baseline naturalness score for the SIT group was, on average, about 12 points lower than for the PT group. This finding is consistent with the analogous coefficient for the transcription accuracy outcome ($\hat{\beta}_2 = -22.7; p = .24$), which shows that the SIT group scored about 23 points lower than the PT group, on average, at pretest. Despite randomized assignment to treatment arms, chance baseline imbalances such as these are not uncommon when the sample sizes are very small. By focusing on the test of the coefficient on the time by treatment interaction, our analyses focus on gain scores, rather than absolute scores, which inherently control for baseline imbalance on the outcome.

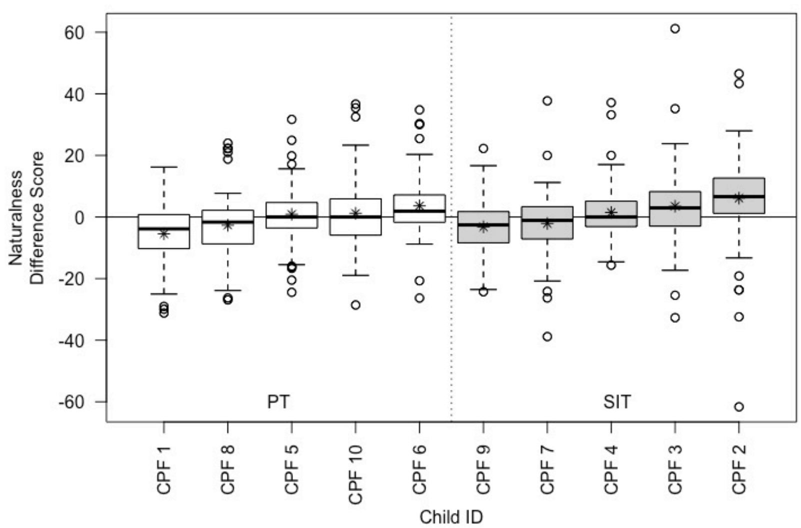
Table 2. Transcription accuracy (TA) and naturalness ratings for the Speech Intelligibility Treatment (SIT) and Physical Therapy (PT) groups at two timepoints

Condition	Variable	PRE Mean (SD)	POST Mean (SD)
SIT	Transcription Accuracy	48.82 (33.38)	51.83 (35.74)
	Naturalness Ratings	44.91 (24.74)	46.06 (26.33)
PT	Transcription Accuracy	71.54 (28.04)	62.32 (31.62)
	Naturalness Ratings	56.89 (23.47)	56.41 (23.22)

Table 3. Results from the mixed effects model for transcription accuracy and naturalness

Outcome	**Transcription Accuracy**		**Naturalness**	
Fixed Effects	**Coeff (SE)**	**p-val**	**Coeff (SE)**	**p-val**
Intercept	71.54 (13.02)	<.001	56.89 (4.22)	<.001
SIT	-22.72 (18.40)	.24	-11.99 (5.26)	.04
Time	-9.39 (1.09)	<.001	-0.48 (1.41)	.73
SIT x Time	12.41 (1.54)	<.001	1.64 (1.99)	.41
Variance Components				
σ^2 (residual)	177.91		240.82	
τ_0^2 (child ID)	843.37		64.17	
τ_1^2 (listener ID)	16.69		296.64	

Figure 2. Individual speakers' gain score difference for naturalness ratings in Speech Intelligibility Treatment (SIT) and physical therapy (PT) groups.



Figures illustrating raw scores in TA and naturalness can be found as Supplemental Materials.

SPL and articulation rate

Table 4 shows the descriptive statistics (mean and SD) for SPL (dB) and articulation rate (syllables/second). Table 5 shows results from the mixed effects model fits. For SPL, after controlling for the fixed effects, Child accounted for about 46% of residual variation in sound pressure level. The treatment by time interaction was not significant for the SPL outcome (p = .79), indicating lack of evidence for differential change in SPL across treatment groups from baseline to post-test. The main effects for time and treatment were not significant.

Table 4. Average sound pressure level (SPL) in dB and articulation rate in syllables/second for the Speech Intelligibility Treatment (SIT) and Physical Therapy (PT) groups at two timepoints

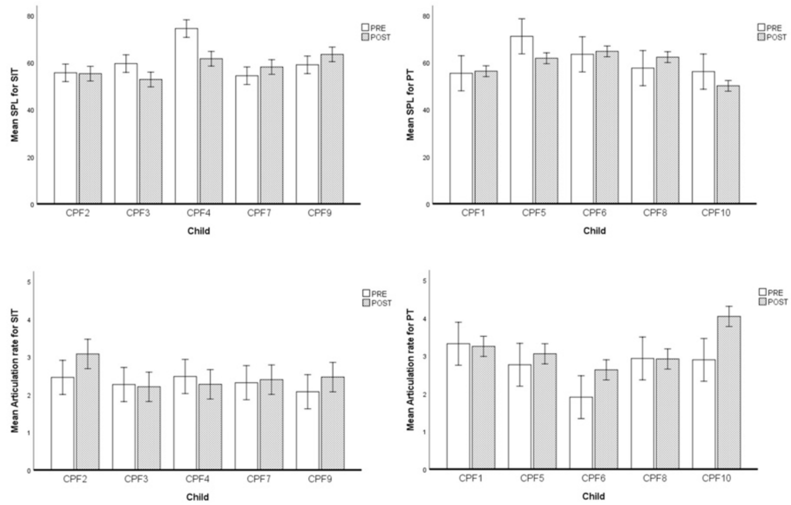
Condition	Variable	PRE Mean (SD)	POST Mean (SD)
SIT	SPL	60.61 (7.91)	58.25 (4.66)
	Articulation rate	2.32 (.36)	2.48 (.43)
PT	SPL	60.74 (8.20)	59.03 (5.69)
	Articulation rate	2.76 (.64)	3.17 (.53)

Note: SPL was measured 8cm from the children's mouth

Table 5. Results from the mixed model effects for sound pressure level (SPL) and articulation rate

Outcome	**Sound Pressure Level**		**Articulation Rate 1**		**Articulation Rate 2**	
Fixed Effects	**Coeff (SE)**	**p-val**	**Coeff (SE)**	**p-val**	**Coeff (SE)**	**p-val**
Intercept	60.74 (2.35)	<.001	2.81 (0.18)	<.001	2.85 (0.17)	<.001
SIT	-0.13 (3.32)	.97	-0.64 (0.25)	.02	-0.73 (0.22)	.008
Time	-1.71 (1.75)	.34	0.37 (0.16)	.03	0.29 (0.12)	.02
SIT x Time	-0.65 (2.48)	.79	-0.17 (0.23)	.48		
Variance Components						
σ^2 (residual)	23.10		0.20		0.20	
τ_0^2 (child ID)	19.81		0.09		0.09	

For articulation rate, about 31% of residual variability was due to Child. The treatment by time interaction was not significant for the articulation rate outcome either (p = .48); however, main effects for treatment and time were significant in this model. In a follow-up model, run without the treatment by time interaction, the main effect for time and treatment were both significant, indicating (a) that the SIT group had a lower articulation rate at baseline ($\hat{\beta}_2 = -0.73; p = .008$), and (b) that both groups increased their articulation rate over time on average ($\hat{\beta}_1 = 0.29; p = .02$). There was no evidence of a differential effect over time based on treatment group assignment.



Discussion

Speech treatment research on children with dysarthria due to CP is sparse, especially on those who speak languages other than English (Miller & Lowit, 2014). This is the first study to explore perceptual and acoustic effects of intensive speech treatment in francophone children with dysarthria. To the authors' knowledge, it is also the first study to examine treatment-related changes in narrative intelligibility in this linguistic population. Our findings indicate a positive impact of SIT on intelligibility in French-speaking children with dysarthria. These results are compared to previously-reported findings on related populations and clinical considerations across languages are discussed.

Intelligibility and naturalness changes

Intelligibility changes as a result of intensive speech treatment have been reported in the literature on English-speaking children with dysarthria due to CP (Levy et al., in press; Pennington et al., 2006; Pennington et al., 2010; Pennington et al., 2013). Our findings from blinded listeners' (n = 60) transcription accuracy scores parallel results from Levy et al.'s (in press) SIT study of American English-speaking children with dysarthria, who also showed significant gains in intelligibility, as measured by VAS ratings of ease of understanding, for narrative speech post-treatment. Of note, narrative language samples present with strong ecological validity, as they provide insight into how children use language in real-world situations (Costanza-Smith, 2010; Danahy Ebert & Scott, 2014). Although our results are preliminary and involve a small number of children, findings from the French and English SIT studies provide encouraging evidence for the ability of children with dysarthria to improve their speech intelligibility in a relatively demanding and naturalistic task.

The results from the present study extend Levy, Moya-Galé, Chang, Campanelli et al.'s (2020) findings on the use of the speech cues "big mouth" and "strong voice" with French speaking children with CP. In that study, improvements in VAS ratings were found at the word level but not at the sentence level. Word-level TA did not improve significantly in response to either cue. In our current treatment study, TA increased significantly at the sentence level after three weeks of SIT. Unlike in cueing studies, the children in the present treatment study were not provided cues during data collection: thus, the recordings may better represent the children's actual speech performance. It is possible that practicing the techniques during the three weeks of treatment increased the children's ability and stamina to make speech adjustments during narrative speech, thereby yielding greater changes. That is, the children may have increased their speech motor control through the intensive speech treatment that implements select motor learning principles and fosters task-dependent neuroplasticity (Kleim & Jones, 2008). The reasons for the decrease in TA scores for the children in the PT group remain open to speculation, but it is possible that the PT camp led to greater physical fatigue, resulting in a reduction in intelligibility (Hodge, 2013). The mean difference in TA across groups from pre to post was approximately 12%. This indicates that, on average, although the accuracy for those in the SIT group increased by only approximately 3%, the accuracy for those in the PT group, which served as the randomized counterfactual in the present study, decreased by about 9%. Thus, it is plausible that in the absence of SIT, those in the SIT group would have regressed to the level of the PT group. This might have been due to fatiguing effects of the three-week camp, as suggested above. However, these results suggest that speech treatment also had positive effects in improving intelligibility. Treatment effects on the intelligibility of speakers with dysarthria have been documented reporting a wide range of values for the gains (Stipancic et al., 2018), with some studies reporting statistically significant increases in intelligibility scores of 4% post-treatment (Cannito et al., 2012). Three of the five children in the SIT group in our study evidenced moderate dysarthria, while a fourth presented with a severe motor speech disorder. An increase in their narrative intelligibility, therefore, suggests a potential benefit from intensive speech treatment for this population. Replication of this and similar studies will be helpful in specifying the extent of treatment gains and clinical relevance.

Improvements in naturalness were not observed in either group in the present study, suggesting that speech naturalness may not be enhanced by this intelligibility-focused treatment. Moreover, this result may not be unexpected if we take into account that treatment approaches targeting prosody may best promote changes in naturalness (Liss, 2007; Scott & Caird, 1983). SIT, with its dual focus, aims at increasing not only vocal intensity, a component of prosody, but also articulatory working space. The vocal parameters that contribute to changes in naturalness remain to be further explored.

Considerable speaker variability was observed in intelligibility and naturalness in both the SIT and PT groups. Individual differences in performance of children with dysarthria have been widely reported (Fox & Boliek, 2012; Levy et al, 2017), which may speak to the diverse representation of motor deficits in this population. The more severe speech motor deficits observed in the SIT group likely account for the more limited, albeit significant, increase in intelligibility post-treatment. Our results also suggest an independent relationship between intelligibility and naturalness when we consider individual data. For example, CPF4, the child with the most severe dysarthria in the SIT group, obtained a score of 4% in TA post-treatment whilst his naturalness rating averaged 39%. A trade-off between intelligibility and naturalness has been reported in the literature on dysarthric speech (Patel et al., 2013). In particular, it is thought that perceived naturalness may be dependent on suprasegmental information (e.g., intonational contour; Patel & Schell, 2008; Vojtech et al., 2019). It is then possible that the listeners in our study perceived prosodic improvements in the child with severe dysarthria even if those gains did not achieve an increase of intelligibility.

Acoustic changes

SPL did not change significantly post-treatment for either SIT or PT, contrary to our hypothesis of increased SPL resulting from intensive speech treatment. This finding, however, is in line with other treatment research studies that have focused on increasing vocal intensity. For example, in Fox and Boliek's (2012) LSVT LOUD study, vocal SPL in repeated utterances did not increase significantly for most of the children with dysarthria in their sample. The authors speculate that inclusion of the entire speech envelope in the SPL measure could have captured "whispered" elements (Fox & Boliek, 2012), as might also have been the case here. Our results are also consistent with Levy et al.'s (in press) finding of no significant increase in SPL post SIT. It should be noted that the neuromuscular limitations experienced by children with CP may impact the extent to which potential treatment gains in vocal intensity may be generalized to everyday communication (Fox & Boliek, 2012). Our findings, however, are not consistent with some previously reported studies in which SPL increased either as a result of intensive speech treatment (Boliek & Fox, 2017) or of stimulability cues (Levy et al., 2017; Levy, Moya-Galé, Chang, Campanelli et al., 2020).

One of the cues in SIT, "big mouth" is an adaptation of techniques for eliciting clear speech (Bradlow et al., 2003) and most closely resembles cues to enunciate and overenunciate, typically reported in the literature on Parkinson's disease (Lam & Tjaden, 2013; Ramig et al., 2015). It was expected that articulation rate would decrease after treatment, as is found with clear speech (Bradlow et al., 2003), given the increased time required to reach targets in an expanded articulatory space (Bradlow et al., 2003; Lam et al., 2012; Lam & Tjaden, 2016). Instead, and contrary to our initial hypothesis, findings in the current study indicated an increase in articulation rate for both groups post-treatment. However, this change was not found to be due to a particular treatment. Although rate control modifications involving a reduced articulation rate have been prototypically reported in the literature as techniques to improve intelligibility in speakers with dysarthria (e.g., Turner et al., 1995), some studies have reported opposite results (Van Nuffelen et al., 2009) or gains limited to only those children with a more moderate-severe motor impairment (Sakash et al., 2019). Our findings indicate that both groups of children spoke faster after three weeks of treatment. These results parallel those in Levy et al.'s (in press) SIT study of 17 English-speaking children with dysarthria. Replication of these studies is needed to elucidate the role of articulation rate as it relates to intelligibility in children with dysarthria.

The acoustic results of the current study, as well as those reported in Levy et al. (in press), suggest that the significant increase in intelligibility in the SIT group may be accounted for by variables beyond SPL and articulation rate.

Strengths and limitations

This first speech treatment study conducted with francophone children with dysarthria secondary to CP was rigorously designed. For example, the study included an active comparator treatment, which followed the same treatment delivery model (in a camp format) and intensive dosage as the SIT group. The significant gains in narrative intelligibility observed in the SIT group, but not in the PT group, can therefore be directly associated with the intensive speech treatment rather than with the social aspect of the camp. An additional strength in our study design was the control for bias. Testing took place in a separate building from the treatment space. Furthermore, testers were blinded to treatment conditions and listeners for the perceptual tasks had no prior experience with or exposure to motor speech disorders and were also blinded to the study conditions. Additionally, speech recording methodology permitted the children's original SPL to be captured in their utterances (Švec & Granqvist, 2018) and be subsequently presented to blinded listeners (n= 60). Lastly, the use of a story narrative strengthened the ecological validity of the study, as this type of task may represent how children use speech in their daily lives (Costanza-Smith, 2010; Danahy Ebert & Scott, 2014). A limitation to the use of the narrative task, however, was the loss of some experimental control, such as which particular words would be produced by the children. Moreover, although articulation rate was utilized as an indirect measure of the “big mouth” cue during the narrative task, a more direct observation of the effects of this cue on articulatory working space would have been changes in the first (F1) and second (F2) formants of selected vowels (Levy et al., 2017; Levy, Moya-Galé, Chang, Campanelli et al., 2020). Variable results for formant changes resulting from this cue have been reported in both cueing (Levy et al., 2017) and treatment studies (Levy et al., in press). Hence, the effects of the “big mouth” cue on spectral characteristics of vowels remain to be further examined. Future studies should also investigate potential language specific treatment-related changes, such as duration and pitch prominence of the stressed syllable of an utterance (Duez, 2014), and their relationship with intelligibility.

A further limitation in this study was the small sample size in each treatment group. Our results, although promising, can therefore be considered only preliminary. Replication of this study with a larger sample size is needed to confirm these results and strengthen the study’s external validity.

Conclusion and Future Directions

Findings from this study, together with findings from examinations of English-speakers with dysarthria, inform our speech-treatment research on English- and French-speaking monolinguals and bilinguals and provide preliminary support for the use of SIT to improve intelligibility in the francophone population. Predictors of improved intelligibility and naturalness changes beyond SPL and articulation rate remain a subject for further analysis.

Acknowledgements

Our heartfelt thanks to the children and their families, as well as to Joséphine Ancelle, Marie-Claude Bergeron, Yanick Bleyenheuft, Ernest Chang, Younghwa Michelle Chang, Andrew Gordon, Sandrine Harrison, Bethany Hetrick, Eunice Hong, Sabrina Kalin Martínez, and Maud le Gall. Special thanks to Andrea MacLeod, who provided administrative support and contributed to the initial brainstorming of ideas for the study, and Christelle Maillart, who assisted with administrative support and initial brainstorming, clinician recruitment and supervision, and data collection.

This project was supported by Teachers College, Columbia University, through a Global Investment Fund and a Dean’s Competitive Grant for Faculty Research (awarded to Erika S. Levy)

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Captions

Figure 1. Individual speakers' gain score difference for transcription accuracy (TA) in Speech Intelligibility Treatment (SIT) and Physical Therapy (PT) groups

Figure 2. Individual speakers' gain score difference for naturalness ratings in Speech Intelligibility Treatment (SIT) and Physical Therapy (PT) groups

Figure 3. Individual speaker data within the SIT and PT groups for both sound pressure level (SPL) and articulation rate. Error intervals represent standard errors.

Supplemental Material:

Figure 4. Individual speakers' raw score for transcription accuracy (TA) in Speech Intelligibility Treatment (SIT) and Physical Therapy (PT) groups

Figure 5. Individual speakers' raw score for naturalness in Speech Intelligibility Treatment (SIT) and Physical Therapy (PT) groups