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Swallowing and Motor Speech Skills in Unilateral Cerebral Palsy: Novel Findings From a Preliminary Cross-Sectional Study

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ABSTRACT

Purpose: Our purpose was to start examining clinical swallowing and motor speech skills of school-age children with unilateral cerebral palsy (UCP) compared to typically developing children (TDC), how these skills relate to each other, and whether they are predicted by clinical/demographic data (age, birth history, lesion type, etc.).
Method: Seventeen children with UCP and 17 TDC (7–12 years old) participated in this cross-sectional study. Feeding/swallowing skills were evaluated using the Dysphagia Disorder Survey (DDS) and a normalized measure of mealtime efficiency (normalized mealtime duration, i.e., nMD). Motor speech was assessed via speech intelligibility and speech rate measures using the Test of Children's Speech Plus. Analyses included nonparametric bootstrapping, correlation analysis, and multiple regression.
Results: Children with UCP exhibited more severe (higher) DDS scores (p = .0096, Part 1; p = .0132, Part 2) and reduced speech rate than TDC (p = .0120). Furthermore, in children with UCP, total DDS scores were moderately negatively correlated with speech intelligibility (words: r = −.6162, p = .0086; sentences: r = −.60792, p = .0096). Expressive language scores were the only significant predictor of feeding and swallowing performance, and receptive language scores were the only significant predictor of motor speech skills.
Conclusions: Swallowing and motor speech skills can be affected in school-age children with UCP, with wide variability of performance also noted. Preliminary cross-system interactions between swallowing, speech, and language are observed and might support the complex relationships between these domains. Further understanding these relationships in this population could have prognostic and/or therapeutic value and warrants further study.

Cerebral palsy (CP) represents a group of disorders of the development of movement and posture caused by diverse nonprogressive abnormalities in the developing brain, which are frequently accompanied by disorders in sensation, cognition, perception, communication, and behavior (Bax et al., 2005). It affects 1.5 to 4/1000 live births (Arneson et al., 2009; Durkin et al., 2016; Paneth et al., 2006), and it is the most common pediatric motor disability. Based on the diversity of its clinical features, CP can be classified using several classification schemes. A popular classification is based on the anatomical distribution of the disorder (i.e., hemiplegia, quadriplegia, diplegia, and monoplegia; Cans, 2007; Gorter et al.,

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2007). Although these anatomically based terms are widely used, they focus mainly on describing motor symptoms affecting the limbs, and thus, they fail to capture critical head, neck, and trunk involvement. Therefore, a more inclusive binary anatomical distribution classification of unilateral versus bilateral CP, which can include detailed descriptions of symptoms affecting the limbs and the head, neck, and trunk areas, has been more recently adopted (Barber et al., 2016; Pagnozzi et al., 2020).

Unilateral CP is a CP subtype where primarily one side of the body has been affected and is typically caused by lesions/abnormalities that predominantly affect one hemisphere (Fehlings et al., 2021). It is the most common form of CP, and as such, it has been studied much more than other CP subtypes (Jonsson et al., 2019; Reid et al., 2015). Specifically, several studies on the neuroplastic capacity of gross motor, manual, and somatosensory functions in UCP have revealed differential patterns of neural reorganization (Kuo et al., 2017; Wilke et al., 2009) that have helped develop interventions in these domains. Much less attention has been devoted to the so-called bulbar functions of children with UCP, such as swallowing and motor speech. This is likely because these functions have bilateral neural control and rely in part—though not entirely—on brainstem circuitry (Behroozmand et al., 2015; Malandraki et al., 2009); therefore, it is assumed that, in UCP, they are spared or compensated for by the less impacted hemisphere. However, when these functions are disrupted, swallowing and motor speech disorders (dysphagia and dysarthria, respectively) can occur (Benfer et al., 2013; Hustad et al., 2014) with negative consequences to health and quality of life (Huang et al., 2013; Michelsen et al., 2014).

A limited number of early observational or parent-report studies have found that dysphagia and dysarthria can co-occur even in children with UCP (Parkes et al., 2010). In a series of studies on a convenience sample of self-feeding school-age children with unilateral and bilateral spastic CP, children with UCP often exhibited clinical signs of dysphagia—albeit milder than the bilateral group (Kantarcigil et al., 2016; Mishra et al., 2018). The most common signs observed in children with UCP included reductions in pre-oral stage components, such as feeding independence and postural control; difficulties in reception of food in the mouth and reduced oral containment; and infrequent signs of penetration/aspiration (e.g., throat clearing/coughing during meals; Kantarcigil et al., 2016; Mishra et al., 2018). Speech motor impairments have not been extensively described in this subpopulation of children with UCP. In a recent longitudinal study including many types of children with CP, speech motor impairments were identified in up to 40% of 5-year-old children with hemiplegia (UCP; Braza et al., 2019). These manifested as reductions in speech rate and in intelligibility of words (Braza et al., 2019).

Indeed, focused experimental research on swallowing and motor speech and on the potential relationship between these functions in UCP is limited. Focused research in this area has both clinical significance and the potential to provide neurodevelopmental and cross-system insights. From a clinical perspective, school-age children with UCP rarely receive services for swallowing or motor speech deficits likely because it is assumed that they have developed functional swallowing and motor speech skills by school age. However, based on the earlier described observational work (Braza et al., 2019; Kantarcigil et al., 2016; Mishra et al., 2018) and our clinical experience, swallowing and motor speech deficits can be observed in this population as well, and therefore, this assumption may need to be, at least in part, challenged. Therefore, to ensure that this population does not remain underrecognized or underserved, more focused research would be pertinent.

From a theoretical and neurophysiological perspective, this population offers a potentially very useful in vivo model to understand neurophysiological reorganization for optimal swallowing and motor speech development. Specifically, in UCP, where primarily one hemisphere is affected and lesions are variable, it is expected that some children will show deficits in swallowing and/or motor speech performance while others may not. Examining this range of performance and the underlying physiological pathways that lead to these variable outcomes may help us identify adaptive versus maladaptive swallowing and motor speech pathways that these children develop. Determining these pathways could be critical in our efforts to develop more effective behavioral or neurostimulation interventions for each domain, targeting, for example, the enhancement of the adaptive pathways. This could be an important step toward the establishment of more physiologically based interventions that are glaringly lacking for this population (Arvedson, 2013; Khamis et al., 2020; Snider et al., 2011). Finally, UCP consists of a relatively cohesive CP subgroup and offers a less complex system structurally and functionally compared to bilateral CP, thereby serving as a good basis on which to begin understanding these underlying pathways before more work in other CP subgroups can be initiated.

Furthermore, swallowing and speech share many anatomical and physiological substrates, muscles, cranial nerves, and central pattern generator circuitry (Lund et al., 1998; Martin et al., 2004; McFarland & Lund, 1995). In healthy individuals, it is known that the central control of swallowing and speech consists of a largely shared bilateral brain network including the primary lateral sensorimotor cortex, premotor area, insula, inferior frontal gyrus/frontal operculum, and anterior cingulate cortex (Eickhoff et al., 2009; Malandraki et al., 2009; Martin et al., 2004; Riecker et al., 2005; Saarinen et al., 2006). Despite these shared mechanisms, empirical evidence of the relationship among the systems involved in both typical development and in CP is sparse. Specifically, early evidence stemming from electromyographic and kinematic studies focused on coordinative patterns of orofacial muscle activity during speech and nonspeech tasks (e.g., chewing and swallowing) in typically developing infants, toddlers (Green et al., 1997, 2000; Ruark & Moore, 1997; Steeve et al., 2008; Steeve & Moore, 2009), and adults (Green et al., 2000; Moore et al., 1988; Wohlert & Goffman, 1994). This work has generally supported distinct peripheral neural control patterns for speech versus nonspeech tasks. However, many of these earlier studies included tasks that were noncontrolled and difficult to compare between domains (e.g., variable speech utterances and variable foods) and focused on healthy populations only. Developing carefully designed experimental paradigms including swallowing and speech outcome variables that are parallel (i.e., have similar functional elements) and examining these variables in children with UCP could provide critical insights about separate and common/cross-system pathways of swallowing and speech in this population that could be targeted in treatment. In the adult literature, we have some evidence for treatments in one domain that have resulted in cross-system gains in the other system or its subsystems (e.g., the Lee Silverman Voice Treatment, the Recline and Head Lift exercises, or Expiratory Muscle Strength Training; Fox et al., 2006; Fujiki et al., 2019; Pitts et al., 2009); similar work in pediatrics and in CP is scarce and would be worth exploring once we have a better understanding of the separate and/or common/cross-system pathways that these children may develop.

To start examining these clinical and theoretical postulations, we conducted a preliminary prospective cross-sectional study including a battery of behavioral swallowing and motor speech assessments, an electromyography evaluation, and a brain magnetic resonance imaging (MRI) protocol. This article focuses on the behavioral data. The primary aim of the behavioral arm of this study was to examine the clinical swallowing and motor speech skills of school-age children with UCP as compared to those of typically developing children (TDC). Based on prior work (Braza et al., 2019; Kantarcigil et al., 2016; Mishra et al., 2018; Parkes et al., 2010), we hypothesized that children with UCP would exhibit reduced swallowing and motor speech skills compared to their typically developing peers. We further hypothesized that this involvement would correlate within and across domains (swallowing and motor speech) in the UCP group. Furthermore, prior work has shown that some clinical/demographic data such as lesion type and side, birth history, gross motor function levels, language scores, and age may be associated with swallowing and/or motor speech skills in CP and other neurologic populations (Hidecker et al., 2018; Hustad et al., 2019; Mourão et al., 2017; Pirila et al., 2007; Selley et al., 2001). Therefore, a secondary exploratory aim was to examine the relationship between clinical/demographic data (e.g., side of body affected, birth history, type of lesion, gross motor involvement, language scores, sex, and age) and behavioral indices of swallowing and speech difficulty.

Method

Participants

From October 2015 to December 2019, two groups of children were recruited, that is, children with UCP and TDC, from two academic clinical centers through flyers, ads, word of mouth, social media posts, and through the CP Research Network Clinical Registry. In addition to age (7–12 years old), inclusion criteria for the UCP group included (a) diagnosis of spastic or ataxic UCP confirmed by an MD, (b) mean length of utterance greater than/or equal to 3.5 morphemes (to ensure that children had adequate speech productions to participate in the motor speech assessment), (c) being orally fed, (d) having a score of greater than 70 on the Test of Nonverbal Intelligence–Fourth Edition (TONI-4; to ensure that findings would not be confounded by the presence of intellectual disability; Brown et al., 2010), and (e) being able to lie still for at least 30 min (criterion for the brain MRI of the larger study). Inclusion criteria for the TDC included developmental test scores within normal limits and being able to lie still for at least 30 min (criterion for the brain MRI of the larger study). Exclusion criteria for both groups included history of neurological diagnoses (other than CP for the UCP group), head/neck surgery or radiation, hearing impairment, or MRI contraindications. Children with UCP were further excluded if they were diagnosed with dyskinetic UCP, as excessive movements could interfere with MRI data collection (conducted as part of the larger study).

Additionally, adult listeners were recruited to complete orthographic transcriptions of children's speech for the speech intelligibility measures detailed below. Inclusion criteria for the adult listeners were age (18–45 years old); being native speakers of American English; passing a pure-tone hearing screening at 25 dB HL for 500, 1000, 2000, and 4000 kHz bilaterally; no history of speech/language/cognitive problems; and no experience listening to children with speech disorders. A total of 215 adult listeners were screened, and 100 adult listeners participated (26 men and 74 women; M_{age}=25 years, SD = 6.9, range: 18–44).

Data were collected at two sites: (a) Purdue University, West Lafayette, IN, and (b) Nationwide Children's

Hospital, Columbus, OH. Ethics approval was obtained from the Purdue University Institutional Review Board (Protocol 1410015417) for both institutions. Adult listeners and children's parents provided informed consent, and all children provided assent.

Data Collection and Analysis

Data collection was completed in a university or hospital research-dedicated space.

Initial Assessments/Screenings

Children were screened for hearing and nonverbal intelligence using the TONI-4 (Brown et al., 2010). For the UCP group, a pediatric neurologist confirmed the UCP diagnosis and type, after reviewing each child's case history, structural brain MRI scan, and video recordings of their gait and conversational speech. Evaluations of gross motor function, manual ability, and functional communication and eating levels were completed by certified speech-language pathologists (SLPs) trained in these measures (see Table 1). Finally, language comprehension and production were screened using the Recalling Sentences and Following Directions subtests of the Clinical Evaluation of Language Fundamentals–Fifth Edition (CELF-5), respectively, by a certified SLP as well (Wiig et al., 2013). The Recalling Sentences subtest was selected because it is a simple index that incorporates many aspects of language processing (Klem et al., 2015). Language assessment was added to the protocol as part of the screening procedure only. Language outcomes were not included in the primary outcome variables for this study, as the focus is on swallowing and motor speech skills.

Table 1. Demographics and clinical data.

Characteristic	UCP (*n* = 17)	TDC (*n* = 17)
Sex	10 males/7 females	10 males/7 females
Age range (years;months)	7;2–12;2	7;6–12;3
Race	White = 11 Black = 3 Asian or Pacific Islander = 2 NR = 1	White = 17
TONI scores: range (*M*)	75–113 (93.19)*	88–136 (107.59)*
CELF-5 FD: range (*M*)	1–14 (7.06)[a,*]	10–19 (12.88)[a,*]
CELF-5 RS: range (*M*)	1–14 (7.75)[a,*]	8–19 (13.18)[a,*]
Left/right side of the body most affected	8/9	N/A
Brain lesion type	Cortical/subcortical = 10 Malformation = 2 PVL/WM = 3 No lesion identified = 1 No MRI = 1	N/A
CP type	Spastic = 16 Mixed = 0 Unknown/NR = 1	N/A
GMFCS level	Level I = 11 Level II = 6	N/A
MACS level	Level I = 2 Level II = 14 Level III = 1	N/A
EDACS level	Level I = 9 Level II = 5 Level III = 3	N/A
CFCS level	Level I = 12 Level II = 2 Level III = 2 Level IV =1	N/A

Note. UCP = unilateral cerebral palsy; TDC = typically developing children; NR = not reported; TONI = Test of Nonverbal Intelligence; CELF-5 FD = Clinical Evaluation of Language Fundamentals–Fifth Edition, Following Directions; CELF-5 RS = Clinical Evaluation of Language Fundamentals–Fifth Edition, Recalling Sentences; N/A = not applicable; PVL/WM = periventricular/white matter lesions; MRI = magnetic resonance imaging; CP = cerebral palsy; GMFCS = Gross Motor Function Classification System; MACS = Manual Ability Classification System; EDACS = Eating and Drinking Ability Classification System; CFCS = Communication Function Classification System.

[a]CELF-5 scores displayed are subtest scaled scores (normal range [within 1 *SD* of *M*] for CELF-5 RS and CELF-5 FD is 7–13).

*Difference between groups is significant ($p < .05$).

Feeding and Swallowing Assessment and Outcome Variables

Feeding and swallowing were assessed using a validated clinical dysphagia assessment, the Dysphagia Disorder Survey (DDS; Sheppard et al., 2014), and a refined measure of eating efficiency (normalized mealtime duration [nMD]) adapted from the study of Mishra et al. (2018). For both assessments, children participated in a "snack time" and were asked to self-feed standardized volumes/quantities of a liquid (water), a nonchewable solid (pudding), and a chewable solid (pretzel). Children were seated on a Rifton chair in front of a height-adjustable table and fed themselves. Snack times were video-recorded using two high-definition cameras (Panasonic HC-V770) that were placed on tripod stands: one positioned directly in front of the children (approximately 3 ft from the child) and the other positioned in a lateral view (approximately 3 ft from the side of the child).

A certified SLP and trained DDS user scored each child's feeding and swallowing competencies using the DDS. The DDS has two parts including 19 items scored using a binary system (0 = *normal*, 1 = *abnormal*). DDS Part 1 evaluates pre-oral swallowing components and feeding, that is, diet level, independence during feeding, and so forth (see Figure 2; Sheppard et al., 2014). DDS Part 2 evaluates oropharyngeal swallowing competencies, that is, orienting to food, oral reception, chewing, postswallow signs of difficulty, and so forth (see Figure 2; Sheppard et al., 2014). In addition, 20% of the recordings were re-analyzed by another trained DDS user to enable interrater reliability measures.

Video recordings also allowed for calculations of total mealtime duration (TMD) and nMD. TMD is defined as the time from the video frame at which the first bolus or bolus-containing utensil touches the child's lips to the video frame at which the child signals meal conclusion (Mishra et al., 2018). The timer on the video-viewing software (QuickTime) was used to record exact start and end times. We normalized TMD by dividing it by the total number of sips and bites the children took to allow comparison of average time per bite/sip. We refer to this new measure as normalized MD (nMD). These measurements were completed by two undergraduate research assistants (RAs) who were trained and achieved > 95% agreement with a senior laboratory member in these measures before initiating data analysis. At the end of data analysis, 20% of the recordings were re-analyzed by the same and another rater to enable intra- and interrater reliability measures. Furthermore, the raters of nMD were blinded to group.

Motor Speech Skills Assessment and Outcome Variables

Motor speech skills were assessed using speech intelligibility indices (Hodge & Gotzke, 2014a; Hustad et al., 2015) and speech rate (Hodge & Gotzke, 2014a). Speech stimuli were taken from the Test of Children's Speech Plus (TOCS+; Hodge et al., 2009). Using a delayed imitation paradigm, children repeated a prerecorded model of randomly generated sets of words, as well as sentences that systematically varied in length from two to seven words. Phrases/sentences were developmentally appropriate for young children in lexical, phonetic, syntactic, and morphological features (Hodge & Gotzke, 2014a). Samples were recorded using the TOCS+ system in a quiet laboratory room (university setting) or a quiet hospital therapy room (hospital setting) using a Dell computer and a built-in microphone array (Realtek High Definition Audio) approximately 14 in. from the child's mouth. The audio signal was recorded at 48000-Hz sampling rate with 16-bit quantization. The signal was monitored during initial trial recordings through the TOCS+ software to ensure that it was audible and was not clipped. Also, at the end of each recording session, an RA listened to all recordings to confirm audio integrity of the data.

For speech intelligibility, each child's recording was orthographically transcribed by three different adult listeners, blinded to group, per TOCS+ procedures and guidance (Hodge et al., 2009; Hodge & Gotzke, 2014a). One hundred adult listeners who were native American English speakers, had normal hearing, and had no prior experience with speech or language disorders were recruited to each transcribe one child's productions. Specifically, for all, but two children, three different adult listeners listened and transcribed their productions. For two children, two adult listeners, instead of three, participated in the transcription of their productions because the data collection sessions of the last two adult listeners had to be canceled when our research operations were halted abruptly due to COVID-19 in March 2020. Orthographic transcriptions were completed through the TOCS+ software installed on a desktop computer in a quiet laboratory room at the university setting. The listeners were wearing headphones, and audio output was approximately at 75–80 dB. Speech stimuli (words and sentences) were delivered in random order to each listener using the TOCS+ software. For word stimuli, adult listeners were provided with a list of potential words prior to starting the task and instructed to read silently through the list of words, per TOCS+ guidance (Hodge & Gotzke, 2014a, 2014b). For sentence stimuli, adult listeners had the option to listen to the sentence a second time, again according to TOCS+ guidance (Hodge & Gotzke, 2014a, 2014b). Intelligibility scores were obtained by calculating percent of words correctly transcribed for words and sentences (Hodge & Gotzke, 2014a; Hustad et al., 2015). Average word and sentence intelligibility scores across the three listeners (or the two listeners for two children) were calculated.

Also, 20% of the stimuli were transcribed twice by each listener for intrareliability calculations (Hustad et al., 2012, 2015).

Speech rate provides an index of speech subsystem coordination and speech motor timing (Hustad et al., 2010), and it offers a parallel measure to the nMD swallowing measure. Speech rate (including pauses) was calculated by syllables produced per minute for the five longest utterances produced by each child via the TOCS+ sentences (Hodge & Gotzke, 2014b). Sentence duration was determined in Praat (Boersma, 2001). The number of syllables was divided by the duration of each sentence to determine the number of syllables produced per minute. To determine speech rate, syllables produced per minute were averaged across the five sentences. Moreover, 20% of the recordings were re-analyzed by the same and another rater to enable intra- and interrater reliability measures.

Statistical Analyses

Statistical analyses were performed using R (R Core Team, 2017). A priori power analysis using G*Power 3.0 identified that a sample size of 15 in each group was needed to detect differences between group means in our main outcome variable of our larger study (i.e., normalized mean amplitude during swallow and speech tasks obtained via surface electromyography) with 80% power, with $a = .05$. Descriptive statistics were calculated for demographic, clinical, and initial assessment data. We analyzed intra- and interrater reliability for 20% of all primary outcome variables (DDS scores, nMD, intelligibility indices, and speech rate) using intraclass correlation coefficients (ICCs).

Differences between groups in our primary swallowing and speech variables (DDS scores, nMD, intelligibility, and speech rate) were assessed using the nonparametric bootstrap test in order to relax the normality and constant variance conditions given the preliminary nature of the study. A Bonferroni correction was applied to account for multiple comparisons. Similar adjustment was made to the 95% confidence intervals (CIs) describing the difference in mean scores. To examine correlations within and between domains (swallowing and speech) within the UCP group, Spearman correlations were used. Multiple regression was used to examine whether clinical/demographic data (side of body affected, type of lesion, birth history, language scores, body mass index [BMI], Gross Motor Function Classification System [GMFCS] and Manual Ability Classification System [MACS] levels, sex, age, etc.) predict our primary swallowing and speech variables. Model selection based on Akaike information criterion was used to determine best fitting model.

Results

Demographics and Sample Characteristics

Demographics, screening, and clinical data are presented in Table 1. A total of 80 children with CP and 25 TDC were phone screened at both sites. The majority of children with CP who were phone screened did not qualify based on comorbidities present (e.g., seizures), followed—in order of frequency—by being diagnosed with other types of CP (e.g., bilateral), travel distance to study sites, family schedules, or concerns about the MRI. Similarly, family schedules and concerns about the MRI were the two primary reasons for the TDC who were phone screened and did not qualify. Seventeen children with UCP and 17 TDC qualified and participated in the behavioral arm of this study. As shown in Table 1, the two groups did not differ significantly in age or sex. Although the two groups differed in nonverbal intelligence scores, they were all within the normal range. In the UCP group, eight children had primarily left-side body involvement and nine children had primarily right-side body involvement; 10 lesions were categorized as cortical/subcortical, two lesions were malformations, and three were white matter lesions. One child did not complete the MRI protocol, and for one child, the lesion was not identifiable in the structural MRI scan.

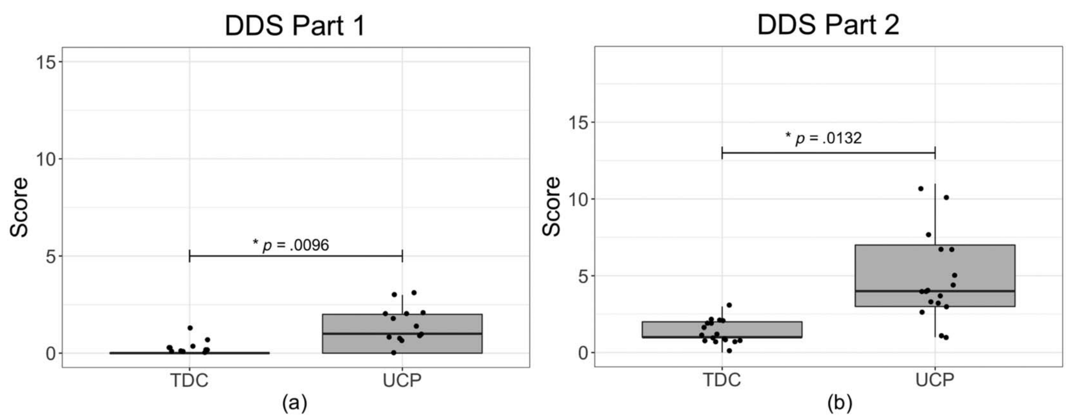
Reliability

ICCs revealed excellent interrater reliability for DDS Part 1 (1.000) and Part 2 ratings (0.933). Excellent intra- and interrater reliability for nMD (0.990 and 0.998, respectively) and speech rate (0.962 and 0.998, respectively) were also observed. Intra- and interrater reliability were good for percent intelligibility–words (0.887 and 0.781, respectively) and percent intelligibility–sentences (0.929 and 0.712, respectively).

Feeding/Swallowing Skills

Children with UCP had more severe (higher) scores than TDC on DDS Part 1 (CI [0.35, 1.65]; $p = .0096$) and Part 2 (CI [1.40, 5.18]; $p = .0132$), indicating more signs of feeding and swallowing difficulties in children with UCP (see Figure 1). For DDS Part 1, reduced postural control and the use of adaptive or maladaptive feeding techniques were the most common symptoms in the children with UCP (see Figure 2). For DDS Part 2, abnormalities were primarily seen in reception, oral containment, and chewing for chewable solids and in reception, containment, and oropharyngeal clearance for liquids for the group with UCP (see Figure 2).

Figure 1. (a) DDS Part 1 scores for both groups (possible range 0–15; *p = .0096). (b) DDS Part 2 scores for both groups (possible range 0–19; *p = .0132). DDS = Dysphagia Disorder Survey; TDC = typically developing children; UCP = unilateral cerebral palsy.



Children with UCP took on average 13.02 s per bite/sip (SD = 3.87), whereas TDC took 10.44 s per bite/sip (SD = 1.88) during snack time. This difference was not statistically significant (CI [−0.02, 5.279]; p = .1416); however, nMD values in UCP varied widely (from 5.65 up to 19.2 s per bite/sip; see Figure 3a).

Motor Speech Skills

The UCP group also had numerically lower word and sentence intelligibility indices than the TDC, but this difference did not reach statistical significance (words: CI [−39.42, 2.53]; p = .3732; sentences: CI [−22.15, 5.00]; p = .9756) likely due to the variability in the UCP group's scores (see Figure 4). Specifically, the UCP group's intelligibility scores ranged from 2.14% to 97.86% for single words and from 6.67% to 99.58% for sentences. Regarding speech rate, children with UCP produced on average 183.77 syllables/min (SD = 34.29), whereas TDC produced 218.65 syllables/min (SD = 24.32), and this difference was statistically significant (CI [−56.32, −18.21]; p = .0120; see Figure 3b).

Relationship Within and Across Domains Within the UCP Group

For the UCP group, we further explored correlations within and across domains for the four main outcome measures (DDS, nMD, intelligibility, and speech rate). Within domain (i.e., within measures of swallowing [i.e., DDS and nMD] and within measures of speech [i.e., intelligibility and speech rate]), no correlations were identified. Across domains, we explored correlations between the two parallel main outcome measures in each domain (i.e., DDS and intelligibility, and nMD and speech rate). We found that total DDS scores were moderately negatively correlated with both speech intelligibility indices (words: r = −.6162, p = .0086; sentences: r = −.60792, p = .0096; see Figure 5).

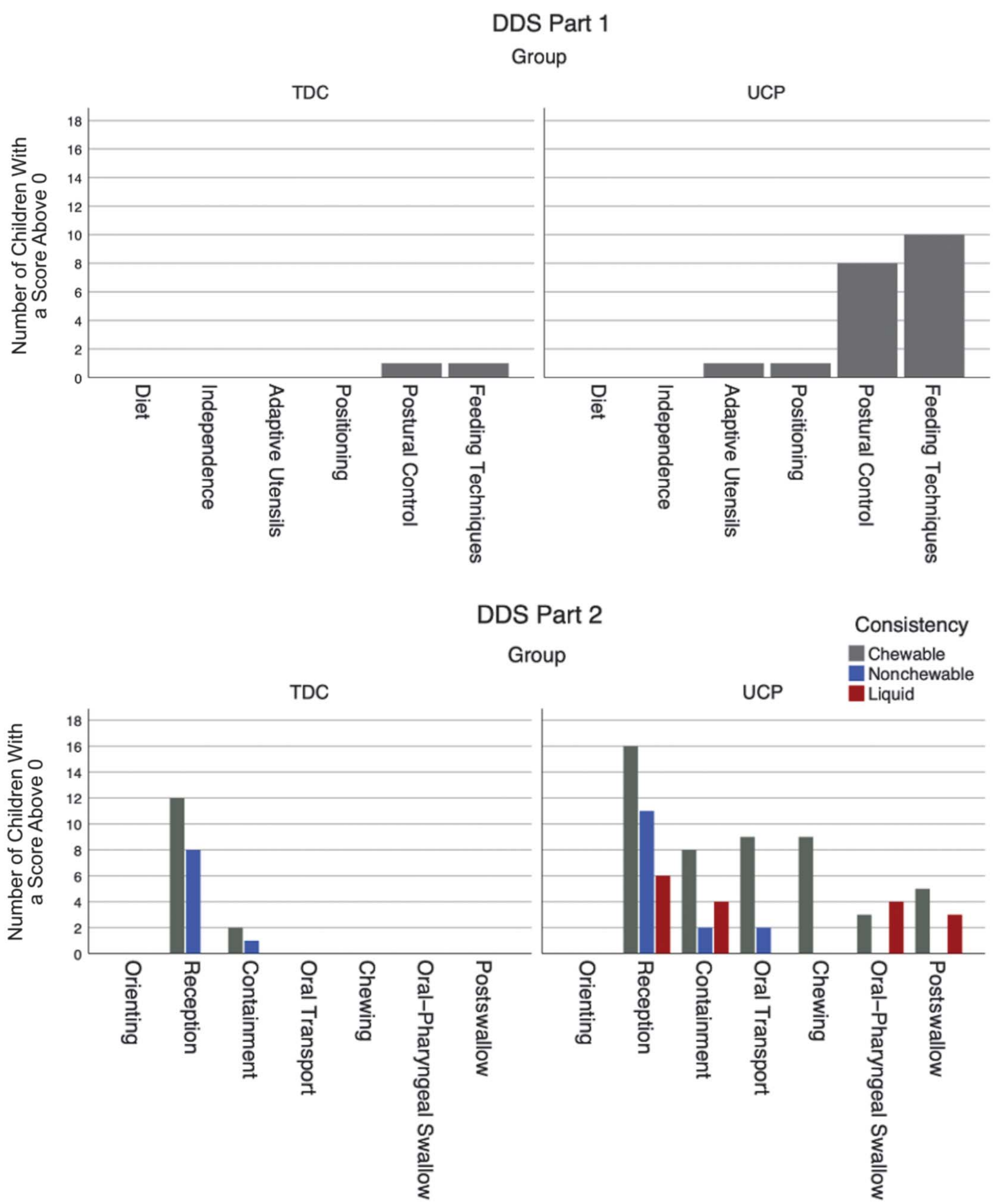
Relationship Between Clinical/Demographic Data and Behavioral Scores in UCP

Multiple regression analyses investigated whether clinical/demographic data (side of body affected, type of lesion, birth history, language scores, BMI, gross motor and manual abilities [GMFCS and MACS levels], sex, age, etc.) predicted our primary swallowing (DDS and nMD) and speech (intelligibility and speech rate) outcome variables. All multiple regressions resulted in no more than one significant predictor per domain. Within the swallowing domain, the expressive language score significantly predicted total DDS scores (b = −0.09731, p = .00263) after controlling for all other variables, indicating a relationship between feeding/swallowing difficulties and reduced expressive language abilities (see Figure 6a). In the motor speech domain, the receptive language score significantly predicted intelligibility scores (words: b = 3.791, p = .02092; sentences: b = 3.167, p = .0419) and speech rate (b = 5.073, p = .0312), after controlling for all other variables, specifically finding that better receptive language skills were associated with more intelligible and faster speech rate (see Figures 6b–d).

Discussion

In this study, we aimed to start examining the clinical swallowing and motor speech skills and potential cross-system interactions of school-age children with UCP compared to typically developing peers. Our primary hypothesis was that children with UCP would exhibit reduced swallowing and motor speech skills compared to

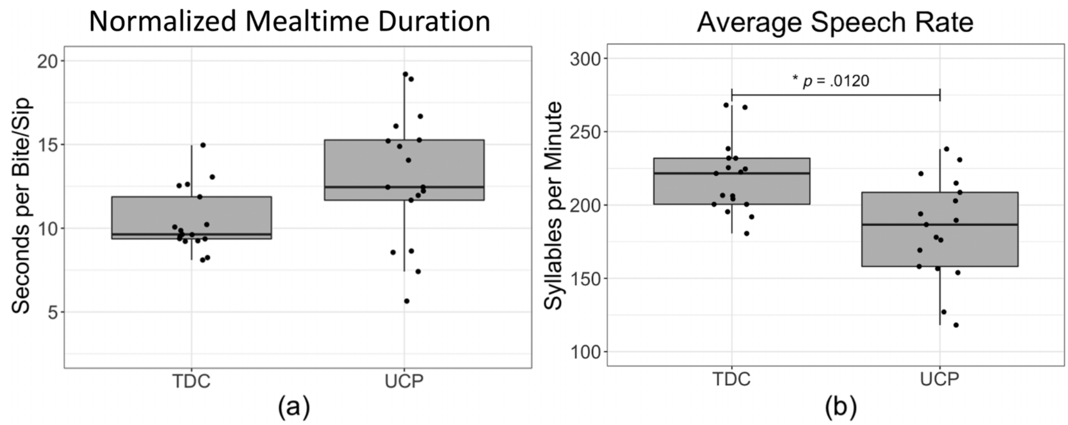
Figure 2. (a) Number of children with DDS Part 1 scores over 0 in each subsection. (b) Number of children with DDS Part 2 scores over 0 in each subsection. DDS = Dysphagia Disorder Survey; TDC = typically developing children; UCP = unilateral cerebral palsy.



TDC. This hypothesis was partially supported by our findings.

First, regarding the swallowing domain, we found that feeding and swallowing abilities were overall reduced in our UCP sample, with some variability also observed. These findings partially validate previous work focused on younger age groups and/or other CP subtypes (Benfer et al., 2013, 2014; Kantarcigil et al., 2016; Mishra et al., 2018). Overall, in children with CP, feeding and swallowing development is known to occur within an abnormally developing system, and swallowing deficits may affect all or some of the swallowing phases and are often present from birth (e.g., Benfer et al., 2013; Calis et al., 2008; Love et al., 1980; Mishra et al., 2019; Rogers et al., 1994). However, it has also been documented that children with GMFCS Levels I and II (i.e., milder gross motor involvement) are likely to show improvements or fully functional feeding and swallowing skills by age

Figure 3. Swallowing and motor speech efficiency measures: (a) normalized mealtime duration (i.e., number of seconds per bite/sip) for both groups (not significant) and (b) average speech rate for both groups (*p = .0120). TDC = typically developing children; UCP = unilateral cerebral palsy.



5 years (Benfer et al., 2017). In our sample (also GMFCS Levels I and II), we observed persistent feeding and swallowing difficulties in some children, highlighting the variability of swallowing performance that can be observed in this subgroup even at older ages.

Although eating efficiency, measured using the new nMD measure we developed, was descriptively reduced in UCP (see Figure 3a), this difference was not significant. Prior studies have provided mixed findings on this topic (Benfer et al., 2014; Reilly & Skuse, 1992; Wilson et al., 2009). In addition to using parent report measures (Benfer et al., 2014; Wilson et al., 2009), most previous works have focused on children of preschool age. In a recent study using direct measures in a sample of school-age children with spastic CP, including UCP, DDS scores have been significantly correlated with TMD (Mishra et al., 2018). In this study, we did not find a correlation between DDS scores and nMD. This could be because of the distribution of nMD values in children with UCP (see Figure 2) in our sample, showing that some children required more seconds per bite/sip and some took fewer seconds per bite/sip than TDC for their snack. This finding may suggest that both slow and fast eating paces could be an adaptive/maladaptive behavior for these children. Also, TMD measures used in prior studies can be affected by multiple variables (e.g., foods being consumed, feeder, and environment); therefore, we normalized this value by dividing it by the number of sips/bites children took. We believe that this process improved data integrity and should be considered in future research.

Regarding motor speech skills, children with UCP descriptively demonstrated reduced speech intelligibility

Figure 4. (a) Percent intelligibility of single words on Test of Children's Speech Plus for both groups (not significant). (b) Percent intelligibility of sentences on Test of Children's Speech Plus for both groups (not significant). TDC = typically developing children; UCP = unilateral cerebral palsy.

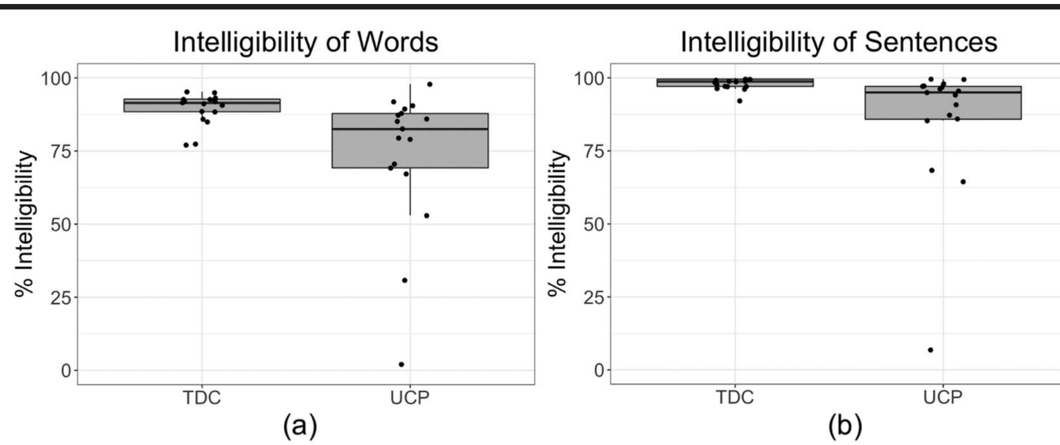
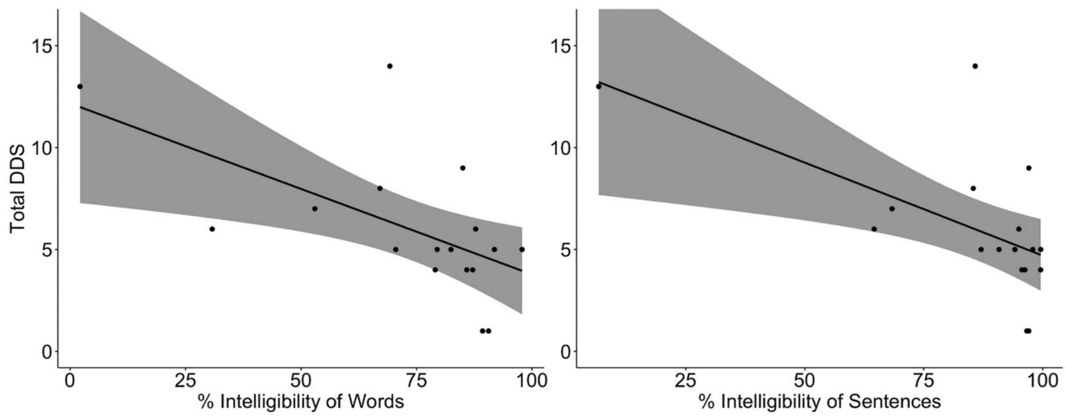
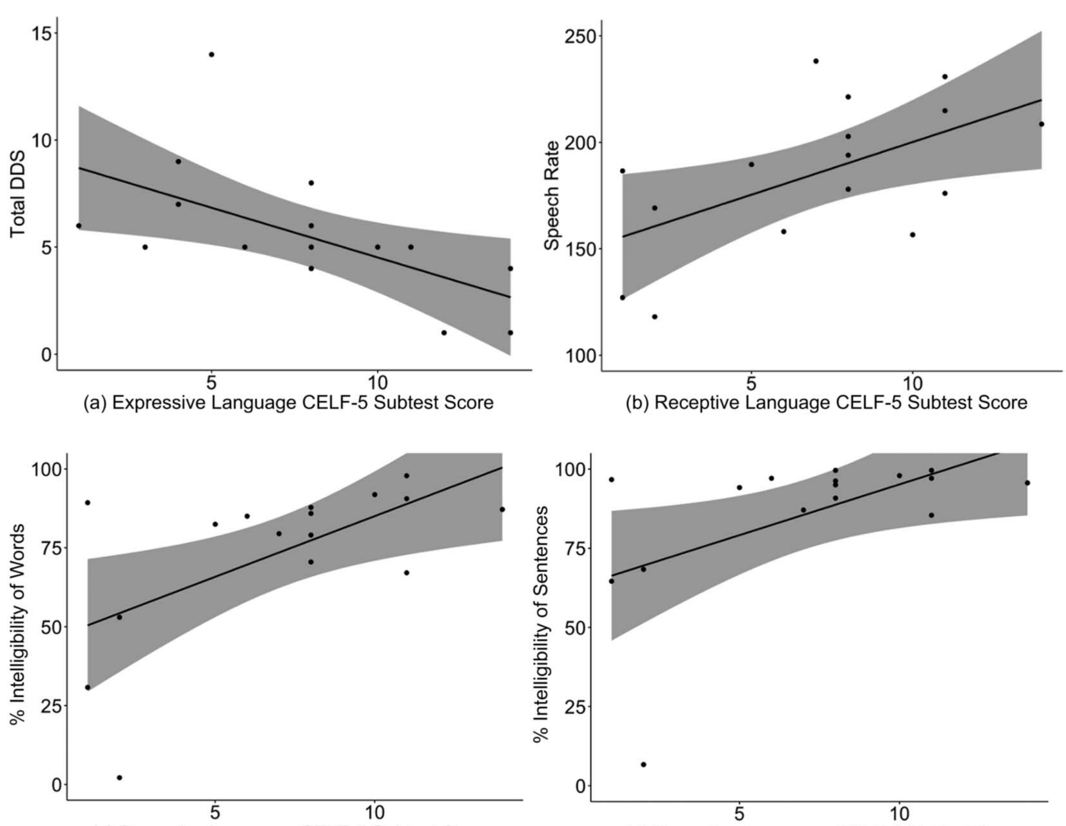


Figure 5. Relationship between total Dysphagia Disorder Survey (DDS) score and (a) percent intelligibility of words ($r = -.6162$; $p = .0086$) and (b) intelligibility of sentences ($r = -.60792$; $p = .0096$) for the unilateral cerebral palsy group.



compared to TDC. However, this difference was also not significant. Speech intelligibility is essential for successful speech communication, relies on all involved systems (articulation, phonation, resonance, and respiration; Yorkston et al., 2010), and provides an index of severity of the motor speech disorder (Hodge & Gotzke, 2014b; Weismer & Martin, 1992). Previous studies show that young children with CP frequently exhibit reduced speech

Figure 6. (a) Relationship between total Dysphagia Disorder Survey (DDS) score and expressive language subtest of the Clinical Evaluation of Language Fundamentals–Fifth Edition (CELF-5) scores ($b = -0.09731$, $p = .00263$). (b) Relationship between speech rate and receptive language subtest of the CELF-5 scores ($b = 5.073$, $p = .0312$). (c) Relationship between percent intelligibility of words and receptive language subtest of the CELF-5 scores ($b = 3.791$, $p = .02092$). (d) Relationship between percent intelligibility of sentences and receptive language subtest of the CELF-5 scores ($b = 3.167$, $p = .0419$), for the unilateral cerebral palsy group.



intelligibility, particularly when they have a motor speech impairment (Hustad et al., 2012, 2015). More recent work has revealed that speech intelligibility may continue to improve in both TDC and those with CP even at 7 or 8 years of age (Braza et al., 2019). In our school-age sample, 11/17 children with UCP had intelligibility scores similar to TDC, whereas the remaining six children had markedly lower scores for word intelligibility (< 75% and as low as 2.14%), and three of those six remained below 75% intelligibility for sentences (lowest score: 6.67%; see Figure 4), suggesting that, for a subgroup of children with UCP, these deficits can be more impactful and persist into later childhood. Furthermore, we found that even 7- to 12-year-old TDC did not have perfect intelligibility on words (range: 76.92%–95.30%) but did have near-perfect intelligibility on sentences (range: 92.08–100%), supporting the argument that TDC continue to improve their intelligibility beyond the age of 4 years (Braza et al., 2019).

Speech rate indicates speech coordination and efficiency and was used as a parallel measure to nMD. We found that speech rate of children with UCP was significantly reduced compared to TDC, and again, the UCP group exhibited noticeable variability (see Figure 3b). Younger children with CP are known to exhibit reduced speech rate when compared to TDC (Braza et al., 2019; Darling-White et al., 2018). With this preliminary study, we further show that speech rate can be reduced even in school-age children with CP who have mild gross motor involvement (GMFCS Levels I and II). Furthermore, there was no correlation between speech rate and intelligibility scores. This finding suggests that even largely intelligible children with UCP may have reduced speech rate, indicating some persisting, albeit mild, deficits in speech motor control in this sample.

Our findings in each of these two domains (swallowing and motor speech) partially validate our clinical theorem that older children with UCP may exhibit swallowing and motor speech deficits, with wide variability also observed in both domains. Although these reductions may not always reach functional/clinical significance, even mild difficulties or reduced efficiency in swallowing and/or motor speech skills may have an impact in these children's current or future social integration and life participation and need to be further understood. Specifically, children with UCP have been shown to exhibit reduced socioemotional functioning and increased peer problems compared to typically developing peers (Whittingham et al., 2014). Furthermore, it has been observed that only about 25% of young adults with CP and GMFCS Level I (i.e., most commonly associated with UCP) have competitive employment and less than 23% have a partner (Pettersson & Rodby-Bousquet, 2021). Given the critical importance of swallowing and speech/communication in social life and participation (Farri et al., 2007; Lefton-Greif et al., 2014; Schoon et al., 2010), one wonders if mild or even subtle deficits in these domains could be associated with some of these negative social outcomes in childhood or adulthood. This further highlights the importance of conducting longitudinal research in this area in the future. Overall, our study provides preliminary support that clinicians need to recognize that these children may need swallowing and motor speech services and deserve appropriate clinical attention.

Furthermore, swallowing and motor speech partially share several anatomical and physiological substrates and neural control elements (Green et al., 2000; Malandraki et al., 2009; Martin et al., 2004; Saarinen et al., 2006; Steeve et al., 2008), and difficulties in both domains frequently co-occur in CP (Parkes et al., 2010). Despite these shared mechanisms and relatively frequent co-occurrence of deficits in both systems in CP, swallowing and motor speech have been traditionally studied and treated individually and potential cross-system interactions have not been explored in this population. Evidence from the adult literature suggests that treatments targeting one domain may have a positive therapeutic effect on the other domain as well (Fox et al., 2006; Fujiki et al., 2019; Pitts et al., 2009), posing the question if similar cross-system gains can be seen in pediatrics and in CP specifically. To start exploring these potential cross-system interactions at the behavioral level, we explored relationships (correlations) across domains between the two parallel main outcome measures in each domain (DDS and intelligibility, and nMD and speech rate, respectively). We identified that total DDS scores were moderately negatively correlated with both speech intelligibility indices (words and sentences), possibly supporting the notion that when feeding and swallowing difficulties are present, speech intelligibility may also be affected. However, given the wide distribution of data (see Figure 5) and the small sample size in this preliminary work, this relationship needs to be validated with a larger data set. Furthermore, what could be more elucidating regarding cross-system interactions would be to investigate not only the correlations between these parallel behavioral outcome variables but also commonalities and differences in the underlying neuromuscular and neurophysiological mechanisms needed to produce these outcomes, which is the aim of our larger study.

Finally, we found that expressive language abilities, measured using only the Recalling Sentences subtest of the CELF-5, predicted DDS scores, and receptive language abilities, measured using only the Following Directions subtest of the CELF-5, predicted intelligibility and speech rate in school-age children with UCP. Given the fact that language assessment using only these two subtests of the CELF-5 was part of our screening procedures and not the focus of this study and given the relatively small sample size and variability in our UCP sample, these results have to be interpreted with caution. The relationship between motor speech skills and receptive language is not novel and has been shown in prior research both in CP and other populations (Darling-White et al., 2018; Hustad et al., 2018). Specifically, preschool children with CP and speech motor impairment have been found to demonstrate a slight but constant receptive language delay (about 6-month delay) that persists through 4.5 years of age, compared to peers without speech motor impairment or typically developing controls (Hustad et al., 2018). In our sample, this relationship appeared to likely be driven by few children/data points (see Figure 6), and therefore, further larger scale studies are needed before definitive conclusions can be made.

What was probably more surprising was the relationship identified between feeding and swallowing skills and expressive language scores. The co-occurrence of feeding/swallowing deficits and language impairment has been priorly documented in children with CP (Goh et al., 2018). Furthermore, through a retrospective design, it has been recently shown that a history of early feeding and swallowing difficulties may predict language impairment later in life (Malas et al., 2015, 2017). Our study provides prospective preliminary data that support a potential relationship between deficits in swallowing and language. This finding could have several explanations. First, feeding disorders may negatively impact caregiver–child relationships, which could impact language input during daily interactions and, therefore, language development. For example, a study investigating the influence of mother–child interactions in the development of behavioral and communicative outputs in infants found that infants with both feeding and language disorders had more negative and conflict-oriented mother–child interactions than infants who had either feeding or language disorders alone (Fabrizi et al., 2010). Additionally, swallowing and language production also share some neurophysiological substrates (e.g., frontal operculum), and it is likely that deficits in these shared areas could impact both domains (Malas et al., 2017). Albeit preliminary, these findings could have cross-system implications and should be further explored as well.

We acknowledge that this is a preliminary study that has a relatively small sample size. However, our recruitment efforts were extensive and involved two large academic centers and a national patient registry and spanned a 4-year period. Furthermore and despite the relatively small sample, using strict statistical corrections, we were able to find robust differences between groups for several key variables, and we believe that these findings will be helpful in guiding future research in this area. In addition, for the regression analyses, we had some missing values for certain clinical/demographic variables (birth history, lesion type, and BMI for three, one, and two participants, respectively) for which we used imputation and which could have impacted our prediction models. Furthermore, we did not use instrumental methods (i.e., videofluoroscopy) to comprehensively evaluate swallowing in the UCP group; given the milder nature of these children's swallowing difficulties, it was deemed unethical to expose them to radiation. Also, we have used caution in interpreting our findings relating to language skills and how they might be associated with our swallowing or motor speech outcome variables because we only completed two subtests of a full language assessment, and language was not a primary variable of interest in this study. As aforementioned, for the intelligibility testing, for all, but two children, three adult listeners transcribed their speech productions; for two children, only two adult listeners transcribed their speech data. This was because the last two adult listeners' data collection session had to be canceled due to COVID-19, and when in-person data collection resumed, the grant period had been completed. This is a preliminary cross-sectional behavioral study, and as such, we cannot make inferences on the underlying mechanisms of the preliminary relationships identified. Our ongoing larger study investigates the underlying neuromuscular and neurophysiological correlates of these behaviors and will be able to provide further insights. Future research following a longitudinal design would be critical to further understand the developmental patterns and additional clinical implications of these findings.

To summarize, swallowing and motor speech skills can be affected in school-age children with UCP, with wide variability also observed in both domains. The variability and the correlational patterns across domains observed in this study provide a framework for future research on the relationship of these functions and their underlying shared and separate mechanisms. Finally, the preliminary cross-system interactions between bulbar functions (swallowing and speech) and language may suggest complex shared correlates that could have prognostic or therapeutic value, and they also warrants further study.

Data Availability Statement

The data that support the findings of this study are available on request from the corresponding author. De-identified data will become openly available to a public repository upon completion of the larger study.

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