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Voluntary Cough and Clinical Swallow Function in Children with Spastic Cerebral Palsy and Healthy Controls

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Abstract

Dysphagia and resulting pulmonary sequelae are frequently observed in children with spastic cerebral palsy (SCP). However, physiological evidence regarding airway protective behaviors (specifically swallowing and cough function) in these children is sparse. The aim of this investigation was to quantify specific feeding, swallowing and cough impairments in children with SCP compared to controls. Eleven children with SCP (mean age: 7 $\pm$ 2 years; GMFCS: I-V; MACS: I-V) and 10 age-matched controls participated. Clinical feeding and swallowing performance was evaluated with the Dysphagia Disorder Survey (DDS) using standardized liquid, puree, and chewable solid consistencies. Suprahyoid muscle activity and respiratory-swallow patterns were assessed with simultaneous surface electromyography and respiratory inductance plethysmography as children swallowed the various consistencies. Voluntary cough airflow measures were also obtained. Nonparametric tests were used for group comparisons and correlational analyses. Compared to controls, children with SCP demonstrated more signs of clinical feeding and swallowing impairment ($p < .0001$, $\eta^2 = .771$), heightened suprahyoid muscle activity for puree ($p = .014$, $\eta^2 = .305$) and chewable solids ($p = .001$, $\eta^2 = .528$), more frequent post-swallow inhalation across liquid ($p = .005$, $\eta^2 = .401$), puree ($p = .014$, $\eta^2 = .304$) and chewable solids ($p = .035$, $\eta^2 = .223$), and lower cough volume acceleration ($p = .019$, $\eta^2 = .289$). Post-swallow inhalation for chewable solids was correlated with the DDS Part 1 ($r_s = .734$, $p = .010$), DDS Part 2 ($r_s = .610$, $p = .046$) and the DDS Total scores ($r_s = .673$, $p = .023$). This study is the first to provide evidence of specific physiological deficits of both swallowing and voluntary cough in children with SCP. Potential hypotheses explaining these deficits and implications for physiologically driven management are explored.

Keywords: cerebral palsy; dysphagia; dystussia; pediatrics; deglutition; deglutition disorders

Introduction

Cerebral palsy (CP) describes a group of disorders that adversely affect motor development and are attributable to non-progressive brain damage [1]. It is the most common physical disability of childhood, with a prevalence of 2-3/1000 births [2, 3]. Spastic cerebral palsy (SCP) is the most common variant of the disorder, comprising approximately 80% of cases and characterized by velocity-dependent resistance to passive stretch resulting in hypertonia and impaired voluntary movement patterns [3, 4].

Children with CP may present with deficiencies across the pre-oral, oral preparatory, oral transport, pharyngeal and esophageal phases of swallowing [5-7]. These disorders impact the safety and efficiency of oral intake required for adequate nutrition and hydration. Dysphagia has been reported in 19-99% of this population and frequently results in aspiration and subsequent respiratory tract infections [8, 9]. The variability in reported prevalence of dysphagia in CP likely reflects differences in level of motoric impairment, intellectual disability, presence of concomitant impairments in the samples studied, as well as differences in methodologies used across studies [8, 9]. Mirrett and colleagues [6] found that greater than 70% of children with SCP in their sample demonstrated aspiration events. However, the physiological components that underlie oropharyngeal dysphagia in this population remain largely unexplored. This has likely contributed to the paucity of physiologically motivated dysphagia management approaches currently available to clinicians for use in this population.

Effective function of swallowing and cough is important for maintaining pulmonary health [10]. In contrast to typically developing children, children with CP develop their feeding and swallowing skills on an abnormal neuromotor system that often includes pathological reflexes [11]. While the majority of literature has documented deficits in children with bilateral motoric involvement, recent evidence demonstrates that children with unilateral deficits also exhibit feeding and swallowing difficulties [12, 13]. In fact, a positive stepwise relationship between oropharyngeal dysphagia and gross motor function has been documented [12]. Therefore, children across levels of motor function should be included in investigations pertaining to dysphagia.

The reported frequency of dysphagia in CP coupled with clinical reports of abnormal cough function (dystussia) places this population at high-risk for medical/pulmonary complications. While Wang and colleagues [14] showed that children with CP demonstrate reduced maximum inspiratory and expiratory pressures compared to controls, no objective measures of cough airflow have been reported. Associations between physiological and clinical measures of both swallowing and cough dysfunction are warranted to provide a more comprehensive understanding of airway protection in this population. To begin addressing these gaps, we aimed to a) assess clinical feeding and swallowing performance in children with SCP compared to a typically developing control (TDC) group, b) quantify airway protective behaviors (swallow muscle activity, respiratory-swallow coordination, cough) in children with SCP as compared to a TDC group, and c) identify correlations between clinical feeding and swallowing performance in children with SCP and physiological measures. Compared to controls, we hypothesized that children with SCP would demonstrate impaired clinical feeding and swallowing performance, heightened suprahyoid muscle activity during swallowing secondary to spasticity, impaired respiratory-swallow coordination, and reduced cough effectiveness. We further hypothesized that the physiological measures would be correlated with clinical feeding and swallowing performance in children with SCP.

Methods

Participants

This study was approved by the Institutional Review Board of Teachers College, Columbia University. Participants with SCP were recruited from a CP upper extremity rehabilitation camp and the United Cerebral Palsy Foundation of New York City. Typically developing controls were recruited through flyers posted throughout the University and the community. For children with SCP, inclusion criteria were: a) a confirmed, primary diagnosis of SCP by neurological report and b) 4 - 11 years of age. Exclusion criteria included: a) neurological disorders unrelated to SCP, b) current respiratory infections or asthma, c) unaided auditory-visual impairments, d) reduced language comprehension as determined via the Test for Auditory Comprehension of Language (TACL) [15], and e) history of tracheostomy and/or head-neck surgery. The majority of children with SCP were independent, self-feeders on unrestricted oral diets. Gross motor function and manual ability were assessed by trained research assistants using the Gross Motor Function Classification Scale (GMFCS) and Manual Ability Classification Scale (MACS) [16, 17]. These levels provided a description of each child's current level of motor function.

For the TDC group, inclusion criteria were: a) typical development and language comprehension as documented by pediatric intake form and the TACL [15] and b) 4 - 11 years of age. Exclusion criteria included: a) neurological disorder, b) respiratory disorder, c) unaided auditory-visual impairments, d) history of tracheostomy and/or head-neck surgery, and e) history of feeding and swallowing disorders.

All data were collected in the Laboratory for the Study of Upper Airway Dysfunction at Teachers College, Columbia University. Data were obtained and analyzed by the primary investigator (PI) with assistance from a master's level speech-language pathology research assistant. The PI and research assistant had successfully completed training in the administration of the Dysphagia Disorder Survey (DDS) [18]. The PI further trained the research assistant in data collection and analysis procedures for all assessments through demonstration, verbal instruction and written manuals.

Clinical Feeding and Swallowing Assessment

Clinical feeding and swallowing performance was evaluated using the DDS, a standardized mealtime assessment used frequently in the CP population [8, 12, 13, 18, 19]. The DDS has two parts that yield the Total score. The DDS Part 1 contains six components that are related to feeding and swallowing. These include diet level, independence level, adaptive utensils used, positioning, postural control, and feeding techniques. The DDS Part 2 contains seven components that comprise a task analysis of feeding and swallowing. These include orientation, reception, containment, oral transport, chewing, oral-pharyngeal swallow, and post-swallow signs of penetration and aspiration.

Children were seated comfortably to allow for a natural eating position and were instructed to eat and drink in their natural manner. Each child consumed the following standardized consistencies: one cup of water (three ounces), one pudding cup (3.25 ounces), and two cookies (one ounce/cookie). Each child consumed a minimum of five sips/bites of each consistency for accurate scoring [18]. A Canon HD video camera was placed three feet in front of the child, with a view that allowed for visualization of the child and food items [19]. This allowed for reliability testing. The DDS scoring was completed in real time during the mealtime assessment using the DDS scoring form and instruction manual. Outcomes included the DDS Part 1 (0-15), DDS Part 2 (0-19), and the DDS Total Score (0-34). The DDS Total Score is the

sum of DDS Part 1 and DDS Part 2. Higher values on the DDS indicate more signs of clinical feeding and swallowing difficulties.

Physiological Assessment

To evaluate underlying physiological components of swallowing (electrophysiology) and respiratory-swallow coordination, surface electromyography (sEMG) and Respiratory Inductance Plethysmography (Respitrace) were employed**.** These recordings were obtained simultaneously using the PowerLab bio-amp system (ADInstruments). These measures were collected in a manner consistent with commonly employed techniques described in the literature [20-22]. In short, children were seated comfortably and provided with back and foot support to promote postural stability. Children were familiarized with the equipment. Bipolar, pediatric disposable EMG electrodes were placed bilaterally over the suprahyoid muscle group (mylohyoid, geniohyoid, anterior belly of the digastric), and a ground electrode was placed on the clavicle. The inter-electrode distance was fixed at 1.5cm. The bio-amp was calibrated to record at a 2KHz-sampling rate with a 500μV range. A mains filter was used to attenuate extraneous noise in the environment in order to maximize the signal to noise ratio. For plethysmography, a cotton elastic thoracic band was then placed beneath the axilla and an abdominal band was placed at the umbilicus level. Following a period of tidal breathing, children were instructed to inhale deeply and exhale, and then maximally displace the abdomen inwardly and outwardly to ensure accurate detection of movement by the elastic bands. Children were then instructed to perform three trials of maximum mandibular depression, which yielded a maximum amplitude signal later used for EMG signal normalization [20]. Following baseline measures, children were presented with the following consistencies in a randomized order: two trials of 5ml water (syringe), 2 trials of 5cc pudding (spoon), and 2 trials of ¼ cookie (hand). All boluses were presented to the children by the examiner in order to reduce any impact of being fed versus being self-fed between groups or within increased disease severity.

LabChart 7 Pro software (ADInstruments) was used for analysis of electromyograhic and respiratory-swallow coordination data. By means of the arithmetic feature of LabChart, raw sEMG swallow signals were rectified and smoothed through the root mean square equation. For children who swallowed more than once per consistency, the first swallow was analyzed in order to make comparisons across all participants and to maintain consistency with prior pediatric literature [23, 24]. Start and end values corresponding to each swallow signal were determined using the Respitrace signal as this was synchronized with the sEMG signal. Once the area of the signal was selected and corresponding values were obtained through the data pad feature in LabChart, normalized values (swallow amplitude divided by maximum [mandibular depression] amplitude and multiplied by 100) for each swallow were recorded. Visual detection of the Respitrace signal was then used to determine the respiratory-swallow pattern corresponding to each swallow. A positive slope represented inhalation, a zero slope represented the period of respiratory cessation, and a negative slope represented exhalation. Pre- and post-swallow breathing patterns were documented across liquid, puree, and chewable solid swallows.

Voluntary Cough Assessment

Voluntary sequential cough was elicited and measured in a manner similar to Hegland and colleagues [25]. Children were comfortably seated with arms at their side. A facemask was fitted over the child's mouth and nose and was coupled to a pneumotachograph system that input differential pressure change to a digital spirometer (ADInstruments) (Fig. 1). The airflow signal was digitized at 2KHz with a range of 2V, and samples were low pass filtered at 150Hz. Following 20 seconds of tidal breathing, the investigator modeled four coughs in a row and

instructed the children to take a deep breath and cough sequentially as if something was stuck in the throat. Three trials of sequential cough were performed with a 20 second rest period between each cough epoch.

"Insert Fig. 1 here"

All de-identified measures of voluntary cough airflow were analyzed using LabChart 7 Pro Software (ADInstruments). Those who analyzed the data were blinded to participant group as they analyzed digitized airflow data. Table 1 provides definitions of the airflow measures analyzed in this study, which included cough inspired volume (CIV), inspiratory phase duration (IPD), compression phase duration (CPD), peak expiratory flow rate rise time (PEFRT), peak expiratory flow rate (PEFR), cough volume acceleration (CVA), and cough expired volume (CEV).

"Insert Table 1 here"

Statistical Analyses

Intra-and inter-rater reliability of all variables was assessed between the PI and trained research assistant on 20% of the data using intra-class correlation coefficients for continuous variables and Cohen's kappa coefficient for binary variables. Non-parametric statistical analyses were conducted with SPSS statistical software (IBM Corp. Released 2016. IBM SPSS Statistics for Windows, Version 24.0. Armonk, NY: IBM Corp) given limited power and non-normal distribution of the data [26]. Descriptive statistics including median and range of GMFCS and MACS levels were calculated for all children with SCP. A Bonferroni corrected Mann Whitney test was used to determine between-group differences in demographic data including age, sex, body mass index, and age-equivalency on the TACL. The Kruskal-Wallis test was used to assess between-group differences in measures of clinical feeding and swallowing performance, and the physiological measures of suprahyoid muscle activity, respiratory-swallow patterns, and voluntary cough airflow. Regarding the surface EMG measures, for children with SCP, a comparison was first made between the less impaired versus more impaired side of function as determined by caregiver report and clinical observation, and no differences in normalized values were detected ($\chi^2 = .838$, $p = .360$, $\eta^2 = .084$). Therefore, the data were collapsed and used for comparison with the TDC group. Spearman's Rank Order Correlations were used to assess relationships between the DDS and physiological measures: sEMG activity, respiratory-swallow coordination, and voluntary cough airflow. Effect sizes (η^2) were calculated for all applicable measures. The alpha level of significance was set at $p < .05$ for all statistical tests.

Results

Reliability

Intraclass correlation coefficients for intra-rater and inter-rater reliability exceeded .80 for all variables as follows: DDS (.992, .961), sEMG Left (.978, .863), sEMG Right (.989, .987), CIV (.999, .999), IPD (.996, .918), CPD (1.00, .841), PEFRT (1.00, .868), PEFR (1.00, 1.00), CVA (.995, .982), and CEV (.999, .996). Additionally, Cohen's kappa for intra-rater and inter-rater reliability for pre-swallow respiratory pattern (1.00, .895) and post-swallow respiratory pattern (1.00, 882) demonstrated high-level of agreement.

Participant Characteristics

A total of 21 children completed the protocol, including 11 children with SCP and 10 controls. The three children with SCP and bilateral motoric involvement (GMFCS IV-V) had received feeding and swallowing therapy in infancy, but no child was engaged in active therapy at the time of the present investigation. Table 2 displays participant characteristics. The two groups were well matched with no significant differences in age, sex, body mass index, or age-

level of language comprehension. The majority of children in both groups demonstrated a comprehension level within the average range for their age ($\geq 50^{th}$ percentile).

"Insert Table 2 here"

Clinical Feeding and Swallowing Performance

The SCP group presented with significantly more impaired clinical feeding and swallowing dysfunction compared to the TDC group as evidenced by higher scores on the DDS Part 1 ($\chi^2 = 17.098$, $p < .0001$, $\eta^2 = .855$), DDS Part 2 ($\chi^2 = 14.291$, $p < .0005$, $\eta^2 = .715$) and the DDS Total ($\chi^2 = 15.421$, $p < .0001$, $\eta^2 = .771$). A qualitative, between-group comparison of the feeding and swallowing task analysis portion of the DDS (Part 2) is presented in Fig. 2. Compared to the TDC group, children with SCP had more difficulties across all feeding and swallowing domains. No child in the TDC group displayed overt signs of aspiration, whereas three children with SCP each exhibited a single instance of cough during the DDS assessment.

"Insert Fig. 2 here"

Swallowing Physiology Measures

Children with SCP required a significantly greater percent of maximum amplitude during puree ($\chi^2 = 6.091$, $p = .014$, $\eta^2 = .305$) and chewable solid swallows ($\chi^2 = 10.555$, $p = .001$, $\eta^2 = .528$), but not during liquid swallows ($\chi^2 = .077$, $p = .782$, $\eta^2 = .004$). Specifically, the percent of maximum amplitude required by the SCP group was 54.5% (liquid), 64.4% (puree), and 74.9% (chewable solid) whereas the TDC group required 46.4% (liquid), 51.9% (puree), and 55.8% (chewable solid).

Regarding the respiration-swallowing coordination, a significantly greater number of children in the SCP group demonstrated a post-swallow inhalation across liquid ($\chi^2 = 8.015$, $p = .005$, $\eta^2 = .401$), puree ($\chi^2 = 6.086$, $p = .014$, $\eta^2 = .304$), and chewable solid consistencies ($\chi^2 = 4.455$, $p = .035$, $\eta^2 = .223$) compared to children in the TDC group (Fig. 3). No significant pre-swallow differences were observed.

"Insert Fig. 3 here"

Voluntary Cough Effectiveness

The median number of coughs produced per cough epoch for both groups was three. No significant between-group differences were found on measures of CIV, IPD, CPD, PEFRT, PEFR or CEV. However, the SCP group demonstrated significantly reduced CVA ($\chi^2 = 5.491$, $p = .019$, $\eta^2 = .289$) compared to the TDC group for the first cough response (Cr1) of the cough epoch. Fig. 4 displays a representative cough from a TDC (A) and a child with SCP (B). Table 3 presents group means and standard deviations of voluntary cough airflow measures (CIV, IPD, CPD, PEFRT, PEFR, CVA, and CEV) for Cr1. No significant differences in the second cough response (Cr2) were detected.

"Insert Table 3 here"

"Insert Fig. 4 here"

Correlations Between Clinical and Physiological Measures

sEMG activity was not correlated with the DDS, though significant correlations were observed between post-swallow inhalation for chewable solids and the DDS Part 1 ($r_s = .734$, $p = .010$), DDS Part 2 ($r_s = .610$, $p = .046$) and DDS Total scores ($r_s = .673$, $p = .023$). No significant correlations between CVA and the DDS were identified.

Discussion

Clinical feeding and swallowing difficulties of children with CP have been well documented. However, physiological information regarding swallowing and cough function and their associations with clinical feeding and swallowing performance is sparse. With this study we aimed to begin addressing these areas. To our knowledge, this is the first study to objectively measure multiple aspects of airway protection (voluntary cough, respiratory-swallow coordination, and clinical swallow function) in children with SCP and healthy controls. As hypothesized, we found significant between-group differences on measures of clinical feeding and swallow performance, suprahyoid muscle activity, respiratory-swallow coordination, and voluntary cough effectiveness. In addition, within the SCP group, correlations were found between post-swallow inhalation for chewable solids and all components of the DDS.

Results of the electromyographical assessment suggest abnormalities in force generation providing indirect evidence for discoordination of swallowing and voluntary cough. Children with SCP display neuromechanistic and developmental deficits [27-29]. At the neurodevelopmental level, there is disruption in the maturation of the corticobulbar and corticospinal pathways, which are essential to developing muscle tone and posture, mediating flexion/extension patterns and reflexes, and promoting muscle development [30]. Significantly greater suprahyoid muscle activity was exhibited by the SCP group compared to the TDC group only for the puree and chewable solid consistencies, and not for liquids. This is in agreement with prior research demonstrating greater activity during mastication of solids [31, 32]. Additional muscular effort is naturally required for these thicker consistencies [33]. However, individuals with CP have difficulty inhibiting muscle activity in the presence of spasticity and co-activation of muscles with increased muscle tension [29]. As has been shown with muscles of mastication, the submental muscles appear to follow a similar pattern of excessive activation.

Furthermore, our cohort of children with SCP demonstrated abnormal respiratory-swallow coordination across consistencies, though these patterns were significantly correlated with clinical feeding and swallowing performance for chewable solids. Abnormal respiratory-swallow coordination in CP has been documented in prior literature [34, 35]. McPherson et al. [34] found that children with CP (5-12 years of age) frequently exhibited a post-swallow inhalation following liquid swallows more often than chewable solid swallows. Rempel and Moussavi [35] found that individuals with CP (13-30 years of age) exhibited post-swallow inhalation following liquid consumption (180 ml) 50% of the time. However, these studies utilized larger liquid volumes whereas the present investigation selected volumes that were deemed safer and easier to manage by the participants. Children with SCP are prone to shallow and altered breathing patterns, given spasticity of the respiratory musculature along with difficulty sequencing volitional movement, and postural limitations [36]. Post-swallow exhalation is thought to facilitate clearance of residue from the airway, thereby decreasing the likelihood and/or severity of penetration and aspiration events [37]. More than 50% of the children with SCP in this study displayed post-swallow inhalation across consistencies, which may place them at risk for post-swallow penetration/aspiration events across consistency types. Upon further validation with a larger sample size, this finding is of high clinical interest, as it identifies a physiological component (i.e., abnormal breathing-swallowing coordination) that needs to be more thoroughly evaluated and, if needed, treated in these children.

Another potentially clinically-relevant finding of our study was the impaired voluntary cough function seen in the sample of children with SCP. Specifically, the SCP group demonstrated decreased cough volume acceleration (PEFR/PEFRT) compared to children in the TDC group. CVA is a measure of cough effectiveness and relates to the shearing force potential

of the cough to clear the airway [38]. While CVA has been associated with aspiration and used to detect penetration and aspiration in neurogenic populations, additional research is necessary to demonstrate such associations in children with SCP [39, 40]. The deficient voluntary cough response observed in this sample of children with SCP may stem from inadequate control and coordination of laryngeal, thoracic, and abdominal musculature. Children with SCP present with spasticity of the respiratory musculature, chest wall rigidity, impaired posture and abnormal timing of muscle activity [36, 41]. These factors may also contribute to insufficient intrathoracic and subglottic pressure. Voluntary cough function was not correlated with clinical feeding and swallowing performance. However, future research is needed to assess the relationships between voluntary cough, cough in response to aspiration, and swallow safety in children with SCP as it may provide a rationale for inclusion of cough testing during the clinical examination.

Furthermore, the SCP group presented with dysfunction across feeding and swallowing components of the DDS as compared to the TDC group. Prior literature has documented dysphagia in 19-99% of individuals with CP. Our findings are in agreement with research representing the higher end of that range [8, 9, 42]. The DDS was not significantly correlated with EMG or cough data, though this could be due to the small sample size, or due to the nature of this clinical assessment. The DDS is a clinical evaluation tool that does not provide extensive information regarding the pharyngeal phase of swallowing. The physiological abnormalities detected in this cohort serve to provide a more comprehensive view of the detrimental effects of spasticity on swallow, voluntary cough, and respiratory-swallow coordination. We postulate that these physiological deficits may not be captured clinically, but are nonetheless important factors to consider, particularly when assessing swallow safety and efficiency and when guiding management.

The observed dysfunctions in the present sample are likely related to spasticity and oral motor difficulties, which are linked to pyramidal tract damage and are associated with reduced nutritional intake [43-48]. Therefore, it is not surprising that the children in the SCP group were observed to display compensatory strategies while eating. For example, they often placed the bolus-containing utensil more posteriorly into the oral cavity as well as produced a posterior head-tilt in a likely attempt to facilitate the oral transport phase of the swallow and to prevent anterior spillage. Some children were also observed to place chewable solid consistencies on the molar surfaces in order to counter difficulties of lingual lateralization and mastication. Finally, these children frequently swallowed multiple times per bolus to promote effective clearance from the oral cavity. While the DDS does not allow for direct detection of pharyngeal phase impairments, the three children who coughed during this clinical assessment also displayed a post-swallow inhalation pattern across consistencies. The relationship between clinical performance, physiological measures, and swallow safety in this population requires further investigation.

Results of this study serve to further increase our understanding of the clinical presentation of oropharyngeal dysphagia in children with SCP. While clinical deficits have been described previously and were found to be in agreement with the present findings, the physiological factors contributing to such observations are beginning to be demonstrated in both swallow and voluntary cough function [8, 9, 42]. Given that all tasks were grounded in volitional movement, the primary difficulty appears to stem from aberrant force generation. The extent to which children were able to perform skilled movements of the oropharyngeal and respiratory musculature was limited, likely resulting in the observed differences with controls. Research

assessing the effects of voluntary cough and swallow deficits in relation to pulmonary outcomes is warranted.

Limitations

This study was not without limitations. The small sample size, power and non-normal distribution of the data required the use of non-parametric statistical analyses. This limitation may have decreased our ability to detect further between-group differences across variables. In the present sample, all participants were fed by the investigator during the sEMG / respiratory inductance plethysmography task in order to increase standardization of the protocol across participants. However, this may have influenced muscle activity and respiratory-swallow coordination by reducing proprioceptive cues and control over bolus volume and rate of eating [49].

Additionally, this investigation did not incorporate an instrumental evaluation of swallowing. The children with SCP did not have a history of respiratory infections, nor did they demonstrate signs of pharyngeal phase deficits that would warrant such an evaluation. Therefore, the use of videofluoroscopy or endoscopy would have been unethical [50]. However, all children received a clinical evaluation report with swallowing-related recommendations from the PI, a licensed speech-language pathologist with several years of pediatric experience. Assessment of feeding and swallowing performance was based on the DDS, a standardized tool that has been frequently utilized in prior research to evaluate clinical feeding and swallow performance in children with CP [8, 12, 13, 18, 19].

Conclusion

This investigation is the first to objectively evaluate and quantify voluntary cough and clinical swallow function in children with SCP, and to make comparisons with a typically developing control group. The observed deficits provide insight into physiological impairments. Clinical dysphagia is compounded by dystussia in our cohort of children with SCP. Failure to clear the airways from aspirate material may contribute to respiratory complications. Dysphagia management can benefit from the inclusion of objective assessment of clinical and physiological measures that incorporate both swallow and cough function. Future research exploring the coordination of muscle activity and effectiveness of swallowing and cough will further advance our understanding of airway protection in this population.

Compliance with Ethical Standards

Conflict of Interest The authors declare no conflicts of interest.

Human Participation This research involved human participation at the Laboratory for the Study of Upper Airway Dysfunction at Teachers College, Columbia University and was approved by the Institutional Review Board of Teachers College, Columbia University. All participants volunteered for this research investigation.

Informed Consent In addition to verbal consent, all caregivers and children (as dictated by IRB guidelines) signed informed consent forms following a thorough explanation of the clinical research protocol.

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Table 1 Objective measures of cough airflow

Cough Airflow Measure	**Abbreviation**	**Unit of Measurement**	**Definition**
Cough Inspired Volume	CIV	Liters (L)	Amount of air inspired during the inspiratory phase of cough
Inspiratory Phase Duration	IPD	seconds (s)	Time between post-tidal volume breathing inspiratory onset and compression phase onset
Compression Phase Duration	CPD	seconds (s)	Time between inspiratory phase conclusion and expiratory phase onset
Peak Expiratory Flow Rate Rise Time	PEFRT	seconds (s)	Time between expiratory phase onset and peak expiratory flow
Peak Expiratory Flow Rate	PEFR	Liters/second (L/s)	Peak flow of the expiratory phase
Cough Volume Acceleration	CVA	Liters/second^2 (L/s^2)	Peak Expiratory Flow Rate / Peak Expiratory Flow Rate Rise Time
Cough Expired Volume	CEV	Liters (L)	Amount of air expired during the expiratory phase of cough

Table 2 Participant characteristics

Variable	**TDC group (n=10)**	**SCP group (n=11)**	***p*-value**
Primary Etiology (%) (number)	NA	IVH 36.4 (4) PVH 63.6 (7)	NA
Motor Involvement (%) (number)	NA	Hemiplegia 72.7 (8) Diplegia 18.2 (2) Quadriplegia 9.1 (1)	NA
Median, Range (GMFCS)	NA	II, I-V	NA
Median, Range (MACS)	NA	II, I-V	NA
Mean age (SD) (years)	7.6 (2)	7.7 (2.4)	.973
Males (%) (number)	50 (5)	72.7 (8)	.378
Mean BMI (SD)	15.5 (3)	18.2 (3.8)	.114
Median TACL (IQR)	11 (2.3)	7 (2.5)	.061

TDC typically developing control, *SCP* spastic cerebral palsy, *IVH* intraventricular hemorrhage, *PVH* periventricular hemorrhage, *SD* standard deviation, *TACL* test for auditory comprehension of language, *IQR* interquartile range, *BMI* body mass index, *GMFCS* gross motor functional classification scale, *MACS* manual ability classification scale, *NA* not applicable

Table 3 Between-group differences on measures of voluntary cough airflow

Cough Response 1 Variable	**TDC** Mean (SD)	**SCP** Mean (SD)
CIV	.389 (.300)	.543 (.417)
IPD	.798 (.525)	.998 (.528)
CPD	.282 (.203)	.283 (.171)
PEFRT	.044 (.015)	.107 (.182)
PEFR	2.14 (.658)	1.79 (.182)
CVA*	52.7 (18.5)	31.5 (19.3)
CEV	.360 (.240)	.265 (.264)

*$p < .05$

Figure Legends

Fig. 1 Participant fitted with facemask coupled to a pneumotachograph completing the voluntary cough assessment

Fig. 2 Between-group differences on percent of participants with dysfunction across domains of the Dysphagia Disorder Survey
TDC Typically Developing Control, *SCP* Spastic Cerebral Palsy. [$p < .0005$]

Fig. 3 Between-group differences on percent of participants exhibiting pre- and post-swallow inhalation across consistencies
TDC Typically Developing Control, *SCP* Spastic Cerebral Palsy. [$p < .05$]

Fig. 4 Representative cough from a typically developing control (A) and a child with spastic cerebral palsy (B) demonstrating discoordinated inspiratory, compression, and expiratory phases of cough observed in the child with spastic cerebral palsy compared to the typically developing control
A Cough Inspired Volume, *B* Inspiratory Phase Duration, *C* Compression Phase Duration, *D* Peak Expiratory Flow Rate Rise Time, *E* Peak Expiratory Flow Rate, *F* Cough Expired Volume

Fig. 1

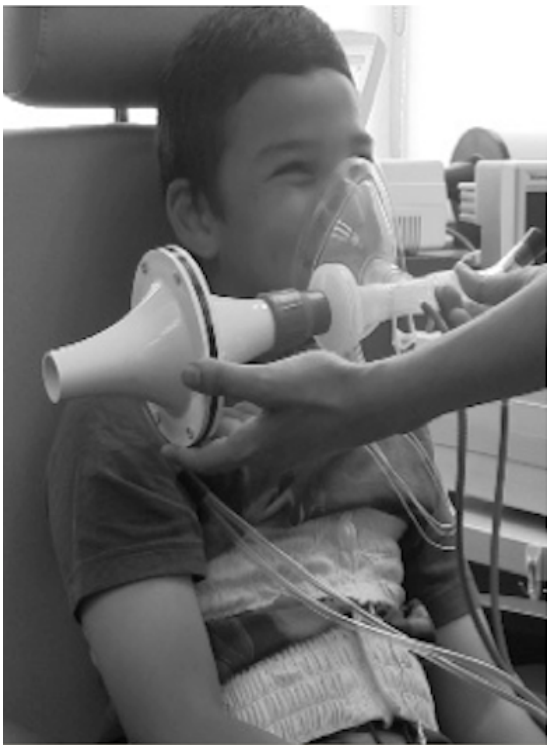

Fig. 2

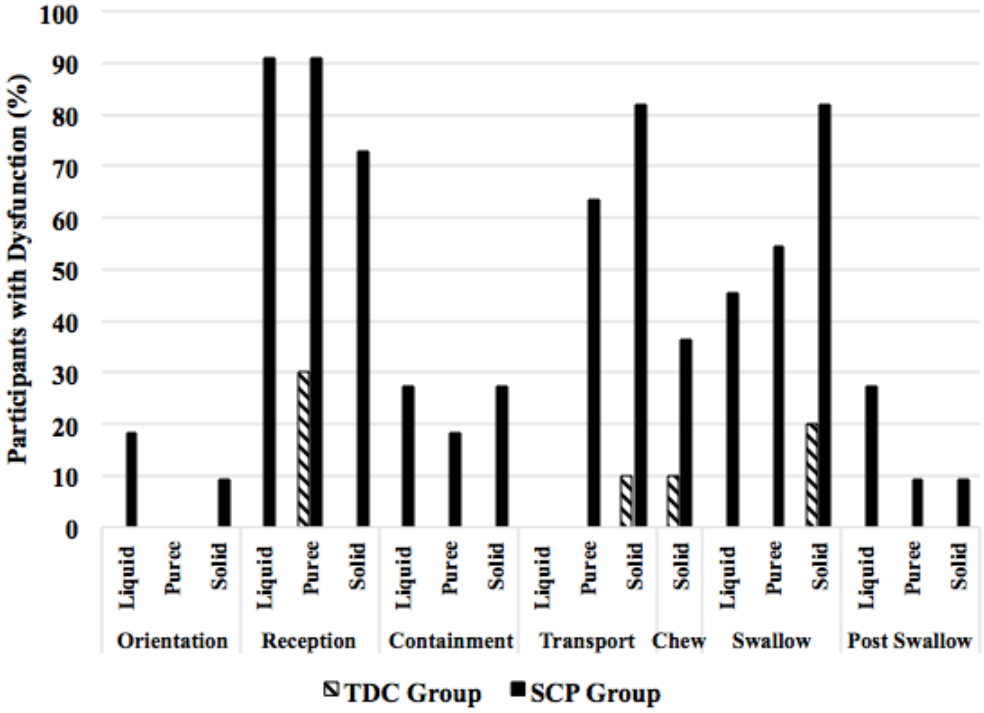
Participants with Dysfunction (%)
100
90
80
70
60
50
40
30
20
10
0
Liquid
Puree
Solid
Orientation
Reception
Containment
Transport
Chew
Swallow
Post Swallow
TDC Group
SCP Group

Fig. 3

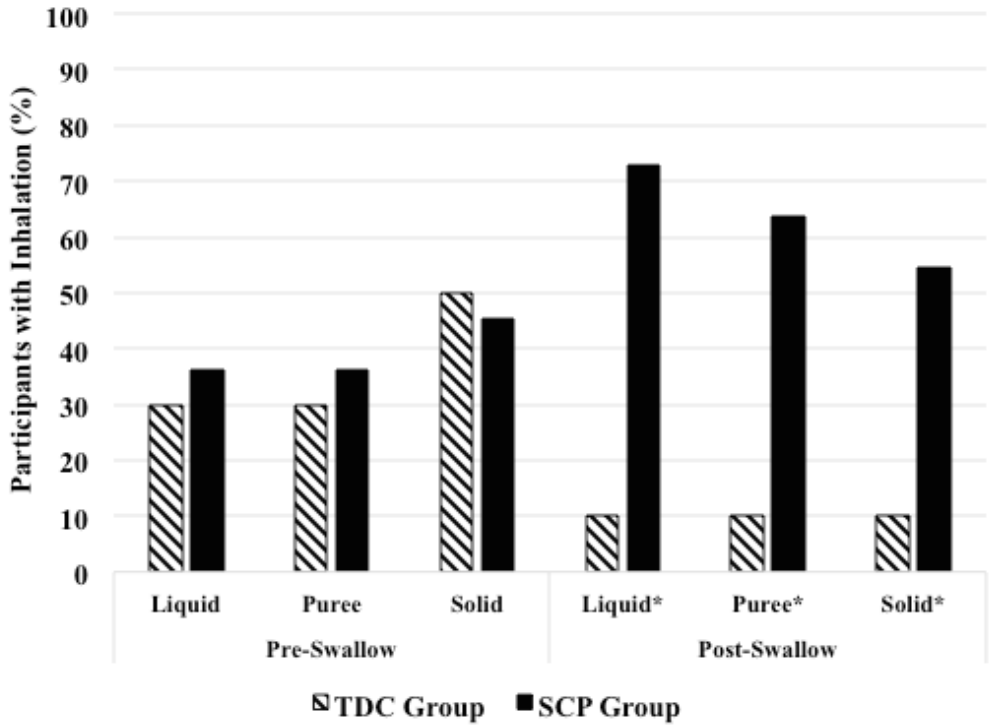
Participants with Inhalation (%)
100
90
80
70
60
50
40
30
20
10
0
Liquid
Puree
Solid
Liquid*
Puree*
Solid*
Pre-Swallow
Post-Swallow
TDC Group
SCP Group

Fig. 4