# AIRWAY PROTECTIVE BEHAVIORS AND MEALTIME PERFORMANCE IN CHILDREN WITH SPASTIC CEREBRAL PALSY AND TYPICALLY DEVELOPING CONTROLS

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**Submitted in partial fulfillment of the
requirements for the degree of Doctor of
Philosophy
under the Executive Committee
of the Graduate School of Arts and Sciences**

**COLUMBIA UNIVERSITY**

**2017**

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## ABSTRACT

AIRWAY PROTECTIVE BEHAVIORS AND MEALTIME PERFORMANCE IN CHILDREN WITH SPASTIC CEREBRAL PALSY AND TYPICALLY DEVELOPING CONTROLS

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This investigation aimed to objectively measure physiological deficiencies across the continuum of airway protection and clinical feeding and swallowing severity in children with spastic cerebral palsy (SCP), and to make comparisons with a healthy control group. Eleven children with SCP (GMFCS I-V) and 10 controls (mean age: 7$\pm$ 2 years) were tested. Results revealed significantly impaired oropharyngeal function and greater clinical feeding and swallowing severity for children with SCP. These children also demonstrated impaired respiratory-swallow coordination evidenced by frequent post-swallow inhalation across all bolus types. Reduced cough volume acceleration was also observed in children with SCP compared to controls. In the SCP group, significant correlations with clinical feeding and swallowing severity were observed with oral praxis and post-swallow inhalation. Additionally, clinical feeding and swallowing severity and peak expiratory flow rate were significantly correlated with gross motor function. This is the first study to objectively identify the co-occurrence of dysphagia and dystussia in children with SCP, and to quantify factors underlying these deficits. Future research should address functional deficits along the continuum of airway protection in both assessment and treatment paradigms in order to maximize the quality of life for this population.

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## LIST OF ABBREVIATIONS

| | |
|---|---|
| ADL | Activities of Daily Living |
| CP | Cerebral Palsy |
| SCP | Spastic Cerebral Palsy |
| USCP | Unilateral Spastic Cerebral Palsy |
| BSCP | Bilateral Spastic Cerebral Palsy |
| TDC | Typically Developing Control |
| SSB | Suck-Swallow-Breathe |
| SLP | Speech-Language Pathologist |
| KSPT | Kaufman Speech Praxis Test |
| IOPI | Iowa Oral Performance Instrument |
| sEMG | surface electromyography |
| MUAP | Motor Unit Action Potential |
| SAD | Swallow Apnea Duration |
| RSP | Respiratory Swallow Pattern |
| CIV | Cough Inspired Volume |
| IPD | Inspiratory Phase Duration |
| CPD | Compression Phase Duration |
| PEFRT | Peak Expiratory Flow Rate Rise Time |
| PEFR | Peak Expiratory Flow Rate |
| CVA | Cough Volume Acceleration |
| CEV | Cough Expired Volume |
| DDS | Dysphagia Disorder Survey |
| TACL | Test for Auditory Comprehension of Language |
| GMFCS | Gross Motor Function Classification System |
| MACS | Manual Ability Classification System |
| EDACS | Eating and Drinking Ability Classification System |
| SPSS | Statistical Package for the Social Sciences |

## ACKNOWLEDGEMENTS

My doctoral program was certainly unique. It was filled with surprises, challenges, and accomplishments along the way. Throughout this journey, I was always able to rely on my family and friends for support.

I feel extremely blessed to have been mentored by the best professionals in our field. I thank Shawn Goodman for giving me my first exposure to research as an undergraduate student. I thank Michael Karnell for developing my clinical research acumen as I completed my senior thesis. This experience permanently fueled my passion for clinical research. During my graduate program, I was mentored in both research design and clinical practice by Jeri Logemann. She not only helped me with academic inquiries, but was a strong support during all difficulties. Her enthusiasm and passion for our field will forever be an inspiration to me.

I am very thankful to all members of my dissertation committee. Michelle Troche is a pioneer in our field. She invested her time and energy into challenging me and shaping me into a competent researcher. She challenged me to think critically and creatively. Georgia Malandraki is a consummate professional. She trained me in the fundamentals of research and teaching while always being a source of support and guidance. She amazes me with her work ethic and has encouraged me throughout my doctoral program. Joan Sheppard is one of the best pediatric dysphagia educators in our field. She introduced me to world of pediatrics and is the reason I have chosen my professional path. I thank Andy Gordon and Erika Levy, both of whom are prolific researchers. Their assistance and guidance has been invaluable.

## DEDICATION

I dedicate this work to all of the children who participated in this novel research. Their spirit and contribution has made this project most fulfilling.

# I. INTRODUCTION AND LITERATURE REVIEW

## i. Cerebral Palsy (General)

Cerebral palsy (CP) describes "a group of permanent disorders of the development of movement and posture causing activity limitation that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain. The motor disorders of cerebral palsy are often accompanied by disturbances of sensation, perception, cognition, communication, and behavior, by epilepsy, and by secondary musculoskeletal problems" (Rosenbaum et al., 2007, p. 9). Cerebral palsy is the most common neuromotor disability of childhood (Accardo, 2008). In CP, one observes a non-progressive encephalopathy involving damage to the neuromotor pathways occurring during the early stages of development. Children with CP must learn to develop and acquire feeding and swallowing skills on an abnormal motor system that may involve spasticity, dyskinesia, and/or ataxia (Bax et al., 2005). Among CP types, spastic cerebral palsy (SCP) is the most common and comprises nearly 80% of all cases (Himmelmann & Uvebrant, 2014; Kirby et al., 2011). Additionally, within the umbrella of spastic cerebral palsy (SCP), roughly half of the children demonstrate unilateral involvement and half demonstrate bilateral involvement (Himmelmann & Uvebrant, 2014; Kirby et al., 2011).

Children across CP types demonstrate difficulties with feeding and swallowing as well as underlying competencies that can result in malnutrition and detrimental psychosocial impacts (Berlni et al., 2009; Morrow, Quine, Loughlin, & Craig, 2008; Sheppard, 2011). Feeding skills and mobility are the most powerful prognostic indicators for survival in this population (Strauss et al., 1999). Deficiencies across the oral preparatory, oral transport, pharyngeal and esophageal phases of swallowing have been well documented in this population (Rogers et al., 1994; Mirrett et al., 1994; Arvedson et al., 1994; Del Giudice et al., 1999). In particular, children with CP

demonstrate swallowing problems that result in aspiration of food and liquids. Aspiration, which is food or liquid traveling below the true vocal folds and into the airway, can result from deficits in the continuum of behaviors that are involved in airway protection (Troche et al., 2014). This continuum of airway protective behaviors includes swallowing and cough at either end; with swallowing being essential for preventing the bolus from entering the airway and cough being necessary for ejecting food or liquid from the airway. Clinically, it has been reported that children with CP exhibit abnormal cough (dystussia) or no cough response to aspiration (atussia). The cause of this is likely multifactorial and may be influenced by poor posture, reduced motor control, respiratory muscle weakness, and possible reduced airway sensitivity (Seddon & Khan, 2003; Sullivan, 2013). The decreased ability to clear secretions through an effective cough response would increase the likelihood of lung disease (Arvedson, Rogers, Buck, Smart, & Msall, 1994). The effect of impaired airway protective behaviors increases the risk of long-term health problems in individuals with CP.

Despite the prevalence of reduced airway protection and mealtime deficiencies in children with cerebral palsy, little work has objectively investigated these deficiencies in this population. In fact, there have been no studies objectively testing cough effectiveness in these children. Additionally, there is a paucity of literature assessing the underlying factors contributing to mealtime deficiencies in this vulnerable population. This scientific gap in our knowledge has, in part, resulted in speech-language pathologists (SLPs) implementing assessment and treatment protocols for children with CP that are largely based on anecdotal evidence. In fact, Snider, Majnemer, and Darsaklis (2011) found that many feeding and swallowing interventions for children with CP are not evidence-based and are not guided by objective data. Therefore, the present study aims to objectively assess oropharyngeal function,

respiratory-swallow coordination, cough effectiveness, and clinical feeding and swallowing severity in children with SCP and typically developing controls (TDCs). Specific characteristics of cerebral palsy and a review of the literature are presented below, followed by the specific aims and predictions. The implemented methodology, statistical tests, and results for all aims are then provided followed by an in-depth discussion that highlights the insights gained from the present investigation.

### ii. Cerebral Palsy: Definition, Etiology, and Classification

Cerebral palsy (CP) describes a static encephalopathy to the developing brain that affects neuromotor function. The prevalence of cerebral palsy is approximately 2-3/1000 births (Kirby et al., 2011; Reid et al., 2011; Nordmark et al., 2001; Wichers et al., 2001; Colver et al., 2000). Goldstein (2004) described injuries of the developing brain observed in CP to include poor cell migration, reduced myelination of nerve fibers, reduced synapses and gray matter cell death. A variety of pre-, peri-, and post-natal risk factors have been associated with cerebral palsy, and these may include prematurity/low birth weight, asphyxia, neurological malformations, hemorrhages, strokes, and infections (Wu & Colford, 2000; Wu et al., 2003; Schendel et al., 2002; O'Shea et al., 2000; Suvanand et al., 1997). For children with low birth weight (i.e., <2500 g), the most common causes include intraventricular hemorrhage, periventricular hemorrhage, periventricular leukomalacia, and cerebral infarction; for children having normal birth weight (>2500 g), the most common cause appears to be hypoxic-ischemic encephalopathy (Meberg & Broch, 2004). Certainly, multiple etiologies may coexist in relation to cerebral palsy. Once CP has been determined, it is often further divided into subtypes.

Cerebral palsy types have traditionally been classified by extremity involvement (e.g., quadriplegia, diplegia, hemiplegia) or by neurologic movement disorder (e.g., spastic, dyskinetic, ataxic, mixed) (Cans, 2000; Sanger et al., 2003). In terms of limb involvement, quadriplegia refers to the involvement of all four limbs, diplegia refers to greater involvement of the lower extremities, and hemiplegia refers to primarily unilateral weakness. However, it has been recommended that the terms quadriplegia and diplegia not be used for separate classifications given their ambiguous interpretations, but that categorization of CP should be based on unilateral versus bilateral involvement as this has shown high reliability of classification (Cans, 2000;

Colver & Sethumadhavan, 2003; Rosenbaum et al., 2007). Bilateral spastic cerebral palsy (BSCP) is defined as encompassing both sides of the body and often results from a multifocal cystic encephalomalacia leading to extensive damage to the cerebral cortex (Kerrigan et al., 1991). Greater lower limb involvement is often associated with periventricular leukomalacia (Leviton & Gilles, 1984). Unilateral spastic cerebral palsy (USCP) has single sided involvement and is characterized by primarily unilateral deficits involving grey and white matter damage (Korzeniewski et al., 2008; Stallings et al., 1995).

CP is also categorized based on neurologic movement disorder. Specifically, the underlying movement disorders associated with cerebral palsy have been documented in relation to spasticity, dyskinesia, and ataxia. Spasticity is the result of abnormal muscle tone and function. It has been defined as when "resistance to externally imposed movement increases with increasing speed of stretch and varies with the direction of joint movement, and/or resistance to externally imposed movement rises rapidly above a threshold speed or joint angle" (Sanger et al., 2003, p. e91). It is associated with hypertonia (i.e., resistance to passive stretch), hyperreflexia (exaggerated myotatic reflex), and may lead to impaired voluntary (praxis) movement (Albright, 1995; Katz & Rymer, 1989; Young & Delwaide, 1981). Spasticity is thought to arise from excessive excitability of alpha motor neurons caused by impairments in descending corticospinal, corticobulbar, reticulospinal, and vestibulospinal tracts (Sanger et al., 2003; Harrison, 1989). Though spastic CP is most common, other clinical subtypes include dyskinetic CP and ataxic CP. Dyskinetic cerebral palsy is characterized by active primitive reflexes (e.g., asymmetric tonic neck, tonic labyrinthine), fluctuating muscle tone, and involuntary movements (Krägeloh-Mann et al., 2005). It is further divided into dystonic cerebral palsy and choreo-athetoid cerebral palsy. This type of CP results from damage to the basal ganglia and/or the

thalamus, often during the perinatal period, which can include intracranial hemorrhage and/or stroke, hypoxic-ischemic encephalopathy, or cerebral infections (Himmelmann et al., 2007). Ataxic CP is evidenced by abnormal posture with disrupted muscular coordination resulting in abnormal accuracy, sequencing, and force of movements (Rosenbaum, 2003). In ataxic CP, there is predominately white matter damage (Korzeniewski et al., 2008) with cerebellar hypoplasia and cerebellar malformations (Aisen et al., 2011).

The aforementioned classifications are often used clinically and have been useful for research purposes; however, many children do not fall into a specific category and may show signs and symptoms characteristic of multiple CP types. Regardless of the classification, one can clearly recognize the influence of CP on multiple sensorimotor functions including the development of feeding and swallowing. The effects of cerebral palsy on feeding and swallowing will be described following a review of typical development.

### iii. Cerebral Palsy: Neuromechanistic and Developmental Components

Children with CP must develop motor control on an abnormal physiological system. These children demonstrate abnormal regulation of muscle activity resulting in impaired volitional motor control and coordination (praxis) (Damiano et al., 2000). This is observed across gross and fine motor skills and certainly includes feeding and swallowing. Functional deficits are dependent on degree of motor impairment. While emphasis in the current manuscript will be given to motoric function, it is known that there are sensory deficits which can contribute to co-contraction of agonist/antagonist musculature that further hinder voluntary movement (Gibbs, Harrison, Stephens, & Evans, 1999).

The Neuronal Group Selection Theory (NGST) accounts for the combination of neuromechanistic and developmental deficits in CP (Edelman, 1993; Hadders-Algra, 2000a). According to NGST, networks within the cortex and subcortex are based on physiological and developmental components. Each network is comprised of collections of neurons (functional units) that selectively respond to movement patterns. The responses are based on experiences early in life. Therefore, the developmental experiences in CP contribute to aberrant neuronal responses to movement. Specifically, the cortiobulbar and corticospinal pathways are of interest given that they are not completely myelinated until 2 years of age. There is clear disruption in pathway development, and the timing of neurological insult would dictate further developmental experiences. These pathways contribute to developing tone and posture, mediating flexion/extension patterns and reflexes, and promoting muscle development (Sarnat, 1989). Children with CP develop abnormal physiological bases of motor control. This disruption can be applied to early (suck-swallow-breathe) and later (mastication) skills. There is increased motor variation with more complex skills and varying amounts of adaptation that

must occur in order to effectively preform the skill. According to the NGST, the motor control processes do not achieve complete maturation until adolescence. It is at this time that healthy populations are able to adapt in an efficient and precise manner to task-specific situations (Hadders-Algra, 2000b). However, in CP, the age at which maturation of skills are achieved are known to be delayed (Jan, 2006). An intact motor system requires precise communication among the cerebral cortex, thalamus, brain stem, cerebellum and spinal cord. Ascending and descending neural pathways must be functional. Children with CP demonstrate limited range of adaptation (reduced flexibility) when performing goal-directed movement as a result of programming deficits and neurological damage. This limited ability to adapt within a task has been associated with white matter damage (Vaal et al. 2000). Pathways that travel through the pyramids of the medulla, with the majority of fibers decussating and traveling down the contralateral spinal cord and synapsing, are compromised. Pyramidal tract damage often results in oral motor discoordination (Russman & Ashwal, 2004). The integrity of the presynaptic fibers (upper motor neurons) is compromised in CP and results in spasticity and impaired volitional movement as difficulties in modulating motor responses are observed given motor programming dysfunction. Additionally, at the level of the neurotransmitters, one can identify a decrease in inhibitory function (GABA, glycine) resulting in prolonged muscle contractions with uncontrolled movement. A primary and life threatening consequence of discoordination is dysphagia. However, the precise mechanisms as related to oropharyngeal and airway protective components have yet to be explored in relation to motor programming deficits in children with cerebral palsy.

### iv. Normal Development of Feeding and Swallowing

Consistent with definitions from Delaney & Arvedson (2008) and Logemann (2007), feeding is defined as "anticipatory events, such as motivation and readiness for eating, food acceptance, and also the important interactions developed between the infant and the caregiver/feeder during the feeding process" (Sheppard & Malandraki, 2015, p. 167-168). Swallowing, on the other hand, is defined as "the complex sensorimotor sequence of events that are initiated by recognizing the presence (touch), taste, temperature, and viscosity of food or fluid in the oral cavity, followed by the preparation, in the case of food, to a consistency that can be swallowed, and finalized by its safe transportation through the oral, pharyngeal, and esophageal anatomic structures to the stomach" (Sheppard & Malandraki, 2015, p. 167-168).

Following birth, the healthy infant receives nutrition through the nipple (breast or bottle). During nipple feeds, an efficient and coordinated suck-swallow-breathe (SSB) pattern is necessary for airway protection while providing nutrition necessary for growth and development (Koenig et al., 1990). In typically developing children, developmental milestones with subsequent feeding and swallowing skills are acquired from birth through three years of age and are then refined with experience (Arvedson & Brodsky, 2002). Sheppard (2008) highlighted the importance of motor learning in skills acquisition with emphasis on the interactions between the individual, environment and task. Table 1 presents a chronology of typical feeding and swallowing development over the first two years of life (Arvedson & Brodsky, 2002; Ayoob & Barresi, 2007; Carruth et al., 2004; Delaney & Arvedson, 2008; Morris & Klein, 2000; Institute of Medicine-Food and Nutrition Board, 2001).

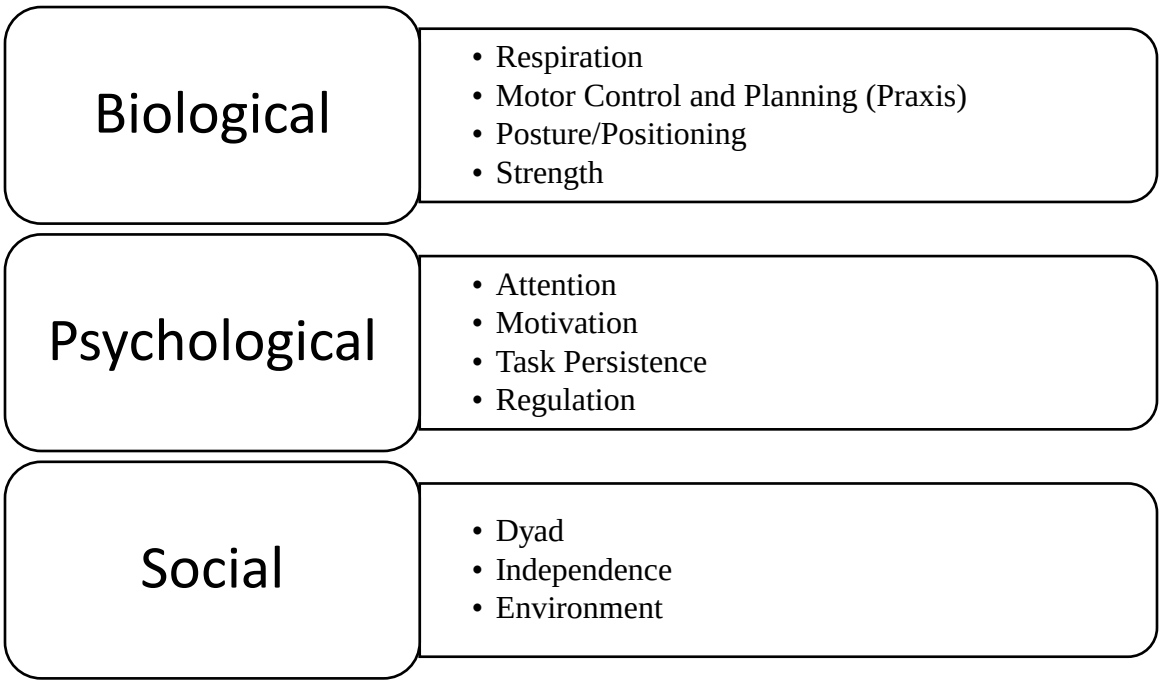
Feeding and swallowing require numerous underlying biological, psychological and social competencies. Figure 1 displays the competencies as drawn from the biopsychosocial

model of feeding and swallowing (Berlin et al., 2009; Sheppard, 2011). Oropharyngeal function and respiratory-swallow coordination were selected for inclusion in the present investigation based on physiological and clinical rationales. Physiologically, these competencies are required for accurate and precise movement of the oropharyngeal structures during the preparatory, oral and pharyngeal stages of swallowing. These competencies are vital to the gradual and safe development of feeding and swallowing. In addition, these competencies provide insight into neuroanatomical and neurophysiological functioning as it relates to feeding and swallowing (Martin, Logemann, Shaker, & Dodds,1994; Rogers & Arvedson, 2005; Leder et al., 2013). Oropharyngeal function and respiratory-swallow coordination are relatively easy to assess through non-invasive instrumentation and can provide objective data in this population. Furthermore, despite the importance of these competencies, data on these measures in children with neurodevelopmental disabilities are extremely sparse. Data on these competencies are important when considering the airway protective deficits experienced by children with CP.

Table 1. Development of feeding and swallowing from 0-24 months of life.

| **Months of Life** | **Recommended Caloric Intake** | **Modality** | **Bolus** | **Milestone** | **Emerging Skill** |
|---|---|---|---|---|---|
| Birth – 6 | 40-45 calories/pound/day 520-270 calories/day | Nipple | Liquid | Dissociated voluntary movements | SSB Pattern |
| 6 – 8 | 35-40 calories/pound/day 646-743 calories/day | Spoon / Cup | Puree | Coordinated movements | Munch chew |
| 8 – 12 | 35-40 calories/pound/day 646-743 | Utensils / Cup | Puree and Chopped Solids | Adequate lip seal + sustained bite | Lingual lateralization |
| 12 - 15 | Dependent on BMI and weight/height growth charts | Utensils / Cup | Solids and Liquids | Oral functioning for all bolus types | Diagonal rotary chew |
| 15 - 24 | Dependent on BMI and weight/height growth charts | Utensils / Cup (straw) | Solids and Liquids | Complete self-feeding with utensils | Rotary chew + sequential sips |

Figure 1. Biopsychosocial model of feeding and swallowing competencies.



### v. Cerebral Palsy: Feeding and Swallowing Dysfunction

Children across CP types are prone to having feeding and swallowing disorders (Adams et al., 2012; Arvedson, 2013; Benfer et al., 2015; Dahlseng et al., 2012; Gangil et al., 2001; Rogers et al., 1994). Feeding and swallowing difficulties may be the first identifiable symptom of cerebral palsy and are related to gross motor function (Aisen et al., 2011; Kim et al., 2013; Weir et al., 2013). In contrast to typical development, children with CP are developing on an abnormal motor system, usually involving pathological reflexes and disrupted underlying competencies (Santos & Nogueria, 2005; Sheppard, 1964). The abnormal system arising from CP negatively affects all phases of swallowing (Calis et al., 2008; Jones et al., 2007). Specific abnormalities by phase of swallowing are presented in Table 2, as determined through radiographic studies (Rogers et al., 1994; Mirrett et al., 1994; Arvedson et al., 1994; Del Giudice et al., 1999). As can be seen, there are numerous disorders that may disrupt the oral preparatory, oral transport, pharyngeal and esophageal phases of swallowing. These disorders impact the safety and efficiency of oral intake required for adequate nutrition and hydration.

A comprehensive literature review spanning the years 1965-2004 reviewed the prevalence of signs and symptoms of dysphagia in children across CP types and showed that 56% of children exhibited choking, 28% had extended feeding time, 59% experienced constipation, and 22% demonstrated emesis (Odding et al, 2006). In children presenting primarily with spastic CP, 77.3% had aspiration with 68.2% having silent aspiration (food or liquid entering the airway without a response from the individual) and 68.2% displayed gastroesophageal reflux (Mirret et al., 1994). These feeding and swallowing deficits in conjunction with an abnormal cough response may result in malnutrition and reduced hydration leading to abnormal growth and poor outcomes (Feng et al., 2002).

Literature has generally focused on children with a greater degree of motor involvement such as those with BSCP as these children are known to have more severe feeding and swallowing difficulties. Yet, current evidence shows that children with milder involvement, as seen in USCP, also demonstrate feeding and swallowing difficulties (Benfer et al., 2013; Malandraki et al., 2014; Malandraki et al., 2015; Mishra et al., 2014). Children with USCP are found to exhibit difficulties in bolus reception, containment, oral transport, mastication, and the pharyngeal swallow in addition to showing signs and symptoms of aspiration risk (coughing and wet voice) during meals (Malandraki et al., 2014; Mishra et al., 2014). Though authors have described the behavioral symptoms of dysphagia seen in these children, information on the underlying coordinative competencies that lead to these deficits have not yet been investigated.

Table 2. Abnormalities by swallow phase in children with cerebral palsy.

| **Oral Preparatory** | **Oral Transport** | **Pharyngeal** | **Esophageal** |
|---|---|---|---|
| Prolonged oral preparatory time | Prolonged oral transit time | Prolonged pharyngeal transit time | Esophageal distention |
| Tongue rest position not at midline | Piecemeal deglutition | Delayed triggering of the pharyngeal swallow | Esophageal backflow |
| Lingual Discoordination | Lingual Discoordination | Nasal regurgitation | |
| | | Pharyngeal dysmotility | |
| | | Reduced upper esophageal sphincter opening | |
| | | Overt and/or Silent Aspiration | |

### vi. Oral Praxis

Praxis has been defined as "the ability to perform skilled movements on command or demonstration" (Kools & Tweedy, 1975, p. 11). Praxis, viewed as an umbrella term, has also been equated to motor control, planning and coordination (Murray et al., 1990). The skilled movements that define praxis should be meaningful and goal oriented. In the context of swallowing, skilled movements of the oropharyngeal musculature are required for a variety of components including bolus reception, containment in the oral cavity, lingual propulsion during the oral phase of swallowing, and timely triggering of the pharyngeal swallow (Rothi & Heilman, 2014). Typical oral praxis tasks are presented in Table 3 (Kaufman, 1995; Kools & Tweedy, 1975). These tasks often involve lingual and labial musculature. For functional feeding and swallowing, age appropriate praxis of the oral system is necessary. The development of a competent SSB pattern relies on motor planning and coordination of the lingual and labial musculature as well as intact airway protective mechanisms to ensure adequate and safe nutritional intake. As the child ages, a mature rotary chew develops and involves numerous praxis components including lingual lateralization and biomechanical stability of the mandible along with timely labial seal to prevent anterior spillage of the masticated bolus (Wilson & Green, 2009). With maturation, the developing child must become an independent feeder and must have the praxis skills required for the intake of various bolus volumes and viscosities.

Clinicians have noted the interaction between feeding and swallowing and the praxis system. This has led to the development of clinical tools that assess praxis-related skills (Palmer, 2002; Palmer et al., 1992; Reilly et al., 1995; Skuse et al., 1995). Despite the clinical importance of the praxis system to feeding and swallowing, research on the development of oral praxis in healthy children is limited. Robbins and Klee (1987) assessed the development of oral praxis in

90 typically developing children across nine age groups and found maturation of skills to occur by 4 years of age. However, decreases in variability of performance were observed through 6;11 years of age suggesting a process of praxis refinement from 4 to 7 years of age. Similarly, Kools and Tweedy (1975) used a 20-item test to study oral and limb praxis in 87 healthy male children and found that praxis competencies developed over time and reached maturation near 6 years of age. Variability of praxis tasks was reduced as the child aged. It appears that oral praxis development demonstrates a steady progression of development until 4 years of age, with refinement processes occurring at approximately 6 years of age. This is in agreement with Bearzotti and Fabbro (2007), who studied 93 healthy children and noted that an objective assessment of oral praxis, including activities such as coughing, blowing, biting, lingual lateralization, and yawning, could yield insight into motor skill level and possible disorders. Following maturation of oral praxis skills, Fucile et al. (1998) demonstrated that these skills remain intact throughout the healthy adult lifespan.

Table 3. Common oral praxis tasks by structure.

| **Mandible** | **Tongue** | **Lips** | **Larynx** |
|---|---|---|---|
| Depression | Protrusion | Protrusion | Voicing |
| Lateralization | Lateralization | Retraction | Cough |
| Elevation | Elevation | Rounding | Pitch Variation |

### vii. Abnormal Oral Praxis

Information about the praxis system in cerebral palsy is sparse. The motor programming deficits secondary to neurological insult overlaid on development provide support for the abnormal praxis that is frequently observed in CP. Abnormal praxis *(*i.e., apraxia) is broad and describes numerous "neurologically induced, acquired, and developmental disorders including buccofacial apraxia, constructional apraxia, dressing apraxia, gait apraxia, gaze apraxia, limb apraxia, speech apraxia, truncal apraxia, and swallowing apraxia" (Rothi & Heilman, 2014, p. 1). Buccofacial apraxia is often used synonymously with oral apraxia (Watson et al., 1986). The apraxia types are named according to the structures or functions that are motorically disrupted. It is best to define apraxia with reference to normal function. Given that oral praxis is defined as the ability to perform movements of the oral structures (e.g., lips, tongue, mandible, cheeks) by imitation or on command (Bodison & Mailloux, 2006), oral apraxia can be defined as any disruption or inability of oral praxis. The etiology and characteristics of oral apraxia are herein presented.

The etiology of oral apraxia has been linked to reduced cerebellar volume and cerebellar damage, specifically involving the vermis (Daily et al., 1995). It has also been linked to reduced basal ganglia volume, particularly in the striatum (i.e., caudate nucleus and putamen), as well as disruption in thalamic function (Berger, 2012; Krishnan et al., 2015). These neurological deficits result in abnormal oral movements characterized by "disturbances in initiation of movement, spatial targeting coordination of motor subsystems, rate of movement, additive motor behaviors, disturbances of sequencing, and perseverative behavior" (Daniels, 2000, p. 161; Daniels et al., 1999). Research on the development of oral praxis in children with cerebral palsy is sparse. It is known that the primitive reflexes should be considered in identifying children with cerebral

palsy (Blasco, 1994). The persistence of primitive reflexes, thereby making them pathological in nature, is one of many signs observed in children with CP (Jones et al., 2007). These reflexes negatively affect praxis by disrupting volitional movement (Blasco, 1994). Pathological reflexes have often been linked to frontal lobe damage (Isakov et al., 1984). They disrupt gross and fine motor development and control (Berker & Yalçın, 2008). The infantile patterned responses to stimuli, as they pertain to oral function, have been termed cranio-oral motor patterns; they have been observed across CP types and have been found to interfere with volitional movements (i.e., praxis) required for oral hygiene, feeding and swallowing (Santos & Nogueria, 2005; Sheppard, 1964). Reilly et al (1996) observed disorders of oral motor control in > 90% of preschool children with CP leading to difficulty in sucking, swallowing, and overall coordination. Interestingly, the observed deficits were made prior to the diagnosis of CP in over half of the children. Research regarding oral praxis development and disorders over the past decade has demonstrated that approximately 80% of children with CP have some degree of oral involvement (Wilson & Hustad, 2009). The difficulties have been associated with lingual movements, oral sequencing and repetition of movement. These deficits have manifested in functional deficiencies in biting and mastication (Gisel et al., 2000; Benfer, 2015), which have further been associated with reduced nutrition and a reduction in eating efficiency (Santos et al., 2012).

### viii. Submental Amplitude

The pharyngeal component of swallowing is critical in protecting the airway. The primary muscle group that is of significance to swallowing is the submental musculature (i.e., mylohyoid, geniohyoid, anterior belly of the digastric). Current data assessing submental amplitude utilizes surface electromyography (sEMG). These muscles are known to be active in infants and children during many components of feeding and swallowing such as labial seal,

mastication, lingual propulsion, and the pharyngeal swallow (Gomes et al., 2006; Green et al., 1997; Tamura et al., 1996; Inoque et al., 1995). The submental region is highly active during triggering of the pharyngeal swallow as it maintains laryngeal elevation in order to protect the airway (Perlman et al., 1999; Ertekin & Aydogdu, 2003). Subsequently, activation of the submental muscle group during swallowing also contributes to opening of the upper esophageal sphincter to allow the bolus to pass through the esophagus (Ertekin & Aydogdu, 2003). Submental swallow amplitude in typically developing children and adults has been assessed through sEMG. This technique is based on the summation of motor unit action potentials (MUAPs) caused by electrophysiological activity (Sommerich et al., 2000). Amplitude is best assessed through the integral of the smoothed sEMG corresponding to the pharyngeal swallow. In terms of submental amplitudes during saliva and water swallows, children between 4 – 12 years of age produce approximately 20 – 40 microvolts per swallow. Notably, these values are not significantly different from healthy adults between 18 – 30 years of age (Vaiman et al., 2004b). However, it must be noted that studies often fail to report normalized sEMG values which are highly valid and allow for individual and group comparisons.

### ix. Abnormal Amplitude

A large body of evidence reporting reduced and discoordinated muscle activity in cerebral palsy is derived from literature regarding skeletal limb musculature (Damiano et al., 1995; Stackhouse et al., 2005). In children with spastic CP, reduced activation and recruitment of MUAPs and reduced contraction of the upper (i.e., wrist flexors, wrist extensors) and lower (i.e., iliopsoas, rectus femoris, gluteus maximus, hip abductors, hip adductors, hamstrings, quadriceps, gastrocnemius, soleus, anterior tibialis) limbs have been documented (Rose and McGill, 2005; Wiley & Damiano, 1998; Vaz et al., 2006). Children with bilateral CP were also found to have

significantly reduced voluntary muscle activation (Stackhouse et al., 2005). Muscle weakness in children with spastic CP has been attributed to the failure of muscle activation coupled with co-activation of antagonist muscles (Elder et al., 2003).

Data on specific submental muscle amplitudes during swallowing in children with CP is limited. In this population, sEMG has been used in the analysis of chewing or has been coupled with other instrumentation to assist in determining respiratory-swallow interactions (Briesemeister, Schmidt, & Ries, 2013; Casas, Kenny, & McPherson, 1994). Given the observed pharyngeal phase deficits in CP, research is needed to determine submental activation and variability during the swallow across bolus consistencies in order to determine the level of impairment in these children and its impact on clinical feeding and swallowing severity.

### x. Airway Protective Behaviors

The study of airway protective behaviors is a rapidly evolving area of research. Troche et al. (2014) has outlined the framework for airway protection which involves a continuum of behaviors from swallow to cough that require reconfiguration of the respiratory cycle (Troche et al., 2014). Swallowing is one airway protective behavior and much research on its phases, temporal sequence, and neural components has been previously described in the literature (Ertekin & Aydogdu, 2003, Jean, 2001; Malandraki, Sutton, Perlman, Karampinos, & Conway, 2009). During normal swallowing, there is a period of breathing cessation known as the swallow apnea duration (SAD). It is an airway protective mechanism that allows for the safe temporal cascade of swallowing events to occur. Of relation to SAD is the respiratory swallow pattern (RSP), which is another airway protective behavior. The RSP can be one of the following four types: Exhale-Swallow-Exhale, Inhale-Swallow-Exhale, Exhale-Swallow-Inhale, or Inhale-Swallow-Inhale. There are developmental changes in both SAD and RSP. In infants,

the apneic period is more pronounced as a means to prevent aspiration. With maturation there is a shift to coughing as the primary behavior to prevent aspiration (Loughlin, 1989). In children with feeding and swallowing dysfunction, the cough was determined to be a more significant predictor of aspiration than gagging or changes in vocal quality (DeMatteo, Matovich, & Hjartarson, 2005). Additionally, the infant RSP varies early in life, but the main pattern is for the swallow to occur following the inhalation, which is likely associated with the infant oropharyngeal anatomy (Bamdord, Taciak, & Gewolb, 1992; Kelly, Huckabee, Jones & Frampton, 2007). Typically developing children over one year of age predominantly swallow at the peak of inspiration and exhibit a post-swallow expiration (McPherson et al., 1992). In healthy adults, the RSP of exhale-swallow-exhale is most frequently demonstrated, occurring between 71-100% of the time, and is believed to serve as an airway protective behavior (Klahn & Perlman, 1999; Martin-Harris et al., 2005; Martin et al., 1994; Preiksaitis, Mayrand, Robins, & Diamant, 1992; Preiksaitis & Mills, 1996).

Research on airway protective behaviors in children with CP is necessary given that respiratory dysfunction is the leading cause of death in individuals with CP (Strauss et al., 1999). Research regarding RSP and SAD has been conducted in children with CP. Specifically, McPherson et al. (1992) found that children with CP (5-12 years of age) frequently exhibited a post-swallow inspiration following liquid swallows more often than solid swallows. Similarly, Rempel and Moussavi (2005) found that individuals with CP (13-30 years of age) exhibited post-swallow inspiration 50% of the time following liquid consumption and had a range of SAD between .84-1.03 seconds across all liquids and puree boluses. This was significantly longer than TDCs, whose average SAD was .63 seconds. These findings point to the need to analyze airway protective behaviors and their relationship with clinical feeding and swallowing severity in

children with cerebral palsy.

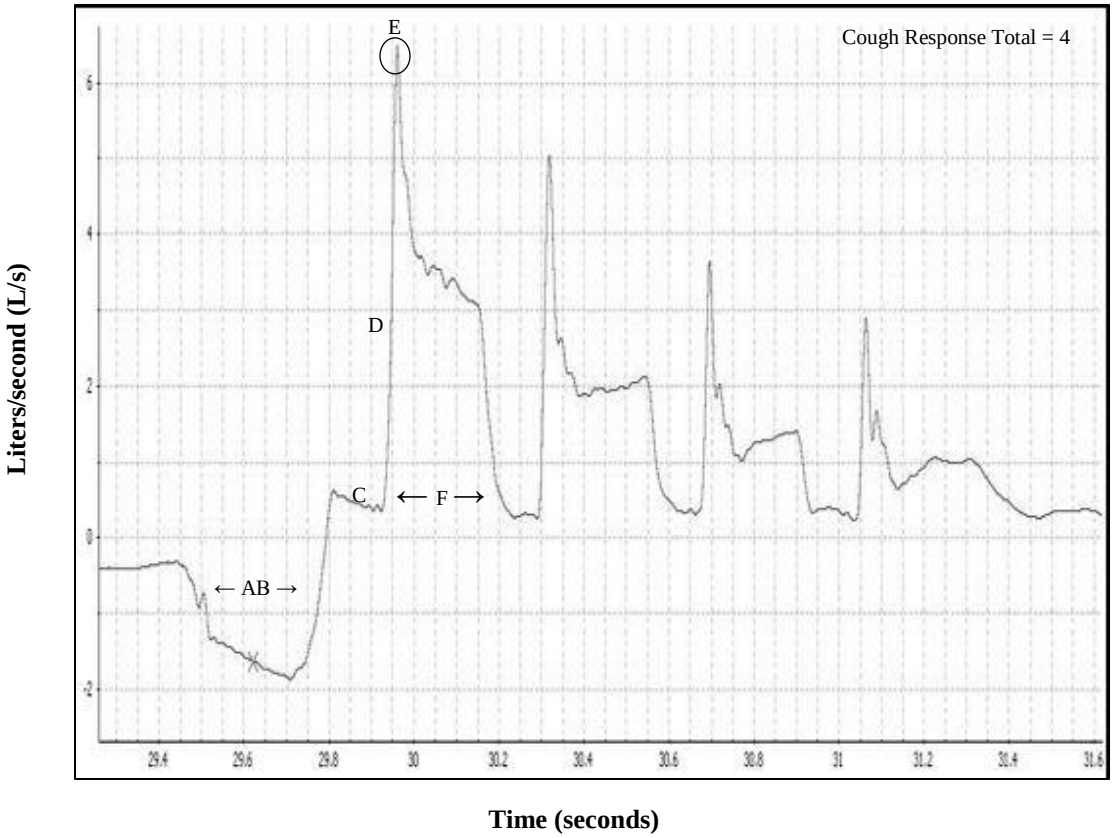
It is known that children with SCP present with a high frequency of aspiration events (Mirret et al. 2994; Odding et al., 2006). The primary airway protective behavior in response to aspiration is the cough. It is known that cough and swallow have shared neural substrates, and that both responses involve exertion over their respective central pattern generators by the behavioral control assembly in order to reconfigure the respiratory cycle and allow for sufficient airway protection (Troche et al., 2014). Cough is necessary to protect and clear the airways, especially following aspiration. While this has been appreciated clinically in measures such as the penetration-aspiration scale (Rosenbek, Robbins, Roecker, Coyle, & Wood, 1996), objective research on cough is still emerging. Cough can be analyzed in its reflexive form or in its voluntary form through the cough waveform (Figure 2) (Hegland, et al., 2014; Pitts et al., 2010). In our population of interest, experimentally inducing the vagally mediated reflex cough would require the use of a tussigenic stimulus such as capsaicin, citric acid, and/or ultrasonically nebulized distilled water. Children with CP may have a history of respiratory tract infections and reduced ability to clear the airways due to purportedly diminished function of sensory receptors in the airway (Seddon & Khan, 2003; Sullivan, 2013). Therefore, voluntary cough function was determined to be the most appropriate measure for this study.

Voluntary cough is modulated by the cerebral cortex. Areas of activation may be similar to that of volitional inspiration and can include: premotor cortex, superior motor cortex, supplementary motor area, inferolateral sensorimotor cortex, prefrontal cortex, and the striatum (Evans, Shea, & Saykin, 1999). Voluntary cough has been delineated into the following three phases: inspiration, compression, and expiration (Simonyan, Saad, Loucks, Poletto, & Ludlow, 2007). The inspiratory phase occurs when alveolar pressure is lower than the atmospheric

pressure. It involves the rapid influx of air through the abducted vocal folds along with contraction of inspiratory musculature, including the diaphragm and the external intercostals (Fontana, 2003). The compression phase involves adduction of the vocal folds causing a brief cessation of breathing necessary to build subglottic pressure required for the final expiratory phase. During the expiratory phase of the cough, the vocal folds rapidly abduct along with contraction of the abdominal and thoracic musculature to generate a burst of airflow that is necessary to clear material from the airways (Fontana, 2003). The main expiratory muscles include the internal intercostals, external oblique, internal oblique, transversus abdominis and rectus abdominis. As with swallowing, cough must be accurately and motorically sequenced to maintain airway protection. Common measures of the cough motor response are listed in Table 4.

Voluntary cough has been found to be associated with penetration/aspiration in diseased populations (Hegland, Okun, & Troche, 2014; Pitts, Bolser, Rosenbek, Troche, & Sapienza, 2008; Pitts et al., 2010). While a single voluntary cough is often elicited by the SLP during a clinical examination, it is typically observed that individuals who cough in response to aspiration do so multiple times. Additionally, voluntary sequential cough measures including compression phase duration, peak expiratory flow rate, and cough expired volume were found to be different between individuals with and without dysphagia. Therefore, the assessment of voluntary sequential cough is ideal in children with SCP as it reflects the ecological validity of the cough motor response without requiring exposure to tussigenic stimuli. Additionally, assessment of voluntary sequential cough is especially pertinent in individuals with motor impairment, as there may be differences between the first and subsequent cough responses in a cough epoch.

Figure 2. Cough waveform.




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

Table 4. 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 |

### xi. Aims and Predictions

It has clinically recognized that children with SCP exhibit dysfunctional feeding and swallowing as well as deficits in airway protection. Given the importance of these behaviors in maintaining safe and efficient nutrition and hydration, it is necessary to obtain objective data regarding the underlying deficiencies as related to functional outcomes. Literature that exists often contains samples of children with mixed CP-types without controlling for age or gender. Furthermore, when typically developing controls (TDC) are included they are often not age-matched nor equivalent in size to the cerebral palsy group. This makes it difficult to draw conclusions and to confidently translate results of these studies to clinical domains. Empirical evidence is sparse and, at times, non-existent in regards to the underlying competencies of feeding and swallowing as well as airway protective behaviors. These areas need to be explored in order to further identify the impact and consequences of impaired neuromuscular development and function in this population. Objective data will allow clinicians to rely less on subjective, clinical judgment. The knowledge gained from the present investigation would allow for a better understanding of the etiology of deficits in this understudied population and help guide evidence-based assessment. To that end, the overarching aim of the present investigation was to objectively examine oropharyngeal function and airway protective behaviors, and clinical feeding and swallowing severity in children with SCP and TDCs.

### xii. Specific Aim 1

a) To test the differences in measures of oropharyngeal function and clinical feeding and swallowing severity in children with SCP and TDCs

- It is predicted that children with SCP would have reduced oral praxis and abnormal submental activation compared to TDCs.

b) To test the relationship between oropharyngeal function and clinical feeding and swallowing severity in children with SCP and TDCs

- It is predicted that oral praxis would be more significantly correlated to clinical feeding and swallowing severity than submental activation in children with SCP and TDCs.

### xiii. Specific Aim 2

a) To test the differences in measures of respiratory-swallow coordination in children with SCP and TDCs.

- It is predicted that children with SCP would demonstrate prolonged SAD and abnormal RSPs compared to TDCs.

b) To test the relationship between respiratory-swallow coordination and clinical feeding and swallowing severity in children with SCP and TDCs

- It is predicted that respiratory-swallow pattern would be more significantly correlated to clinical feeding and swallowing severity than swallow apnea duration in children with SCP and TDCs.

### xiv. Specific Aim 3

To test the differences between children with SCP and TDCs on measures of cough effectiveness and their relationship on clinical feeding and swallowing severity.

- It is predicted that children with SCP would demonstrate reduced cough effectiveness compared to TDCs as indicated by increased IPD, PEFRT and CPD, and decreased CIV, PEFR, CVA, and CEV.
- Cough effectiveness, as measured by PEFRT, PEFR, and CVA, would be significantly correlated with clinical feeding and swallowing severity.

## II. METHODS

### i. Study Design

This prospective investigation examined a cohort including 11 children with spastic CP and 10 age-matched controls. All participants participated in a single session of data collection, which comprised a comprehensive and non-invasive evaluation of swallow and cough function.

### ii. Participants

Participants were recruited from the annual cerebral palsy camp at Teachers College, Columbia University and organizations in NYC through flyers, phone calls, emails and site visits. For children with SCP, inclusion criteria included:

- Diagnosis of SCP by a physician as documented by caregiver-report and pediatric intake form
- Elementary school-age (4-11 years)
- Caregiver consent
- Child consent (if $\geq$ 8 years of age)

Exclusion criteria included:

- Neurological conditions unrelated to CP
- Current respiratory infections
- Asthma
- Unaided auditory-visual impairments
- History of tracheostomy and/or head-neck surgery

For the control group, inclusion criteria included:

- Typically developing as documented by caregiver-report and pediatric intake form
- Elementary school-age (4-11 years)
- Caregiver consent
- Child consent (if $\geq$ 8 years of age)

Exclusion criteria included:

- Neurological condition
- Respiratory condition
- Unaided auditory-visual impairment
- History of feeding and/or swallowing disorders
- History of tracheostomy and/or head-neck surgery

### iii. Screening Procedures

Participants were required to meet all of the inclusion criteria and none of the exclusion criteria expounded above. An IRB approved screening form consisting of questions relating to all criteria were presented to caregivers and were specific to the assigned group (SCP or TDC). All participants were administered Subtest I (Vocabulary) of the Test for Auditory Comprehension of Language (TACL) (Carrow-Woolfolk, 1998), which has been used in children with CP and provides insight into participants' comprehension skills (Levy et al., 2015).

### iv. Data Collection

Data was collected in the Upper Airway Dysfunction Laboratory (955 Thorndike Hall) by the primary investigator and a trained graduate research assistant. The primary investigator provided the research assistant with detailed training in data acquisition and analysis for all measured variables. Given investigator guidance, caregivers completed a thorough intake form documenting previous/ongoing evaluations/treatments, medical history, neurological reports, developmental history, feeding/swallowing development, current feeding/swallowing function, and mealtime behaviors. Additionally, the investigator assigned each participant a level on the gross motor function classification system (GMFCS) (Rosembaum et al., 1997), manual ability classification system (MACS) (Eliasson et al., 2006), and the eating and drinking ability classification system for individuals with cerebral palsy (EDACS) (Sellers et al., 2014). The assigned levels were based on careful review of the classification system guidelines and were assigned using live and video-recorded observations of each participant. Furthermore, given that the data collection phase provided a snapshot of each child's performance, communication with caregivers was done to ensure accurate documentation of function. Data collection occurred in two blocks. The first block consisted of completion of the intake form as well as measures of receptive language, oral praxis and clinical feeding and swallowing severity. Tasks within the second block consisted of the simultaneous measuring of submental amplitude, SAD, and RSP across bolus consistencies. This was then followed by measures of voluntary sequential cough airflow: CIV, IPD, CPD, PEFRT, PEFR, CVA, and CEV. A detailed overview of each variable addressed in aims 1-3 is provided below.

*Oral Praxis*: This was measured through the Oral Movement portion of the Kaufman Speech Praxis Test (KSPT) (Kaufman, 1995). This standardized assessment has been used

extensively in clinical settings to assess the oral praxis system in children between 2.6 – 5.11 years of age and was administered within 5-15 minutes. Given that the participants in the present investigation were between 4-11 years of age, the KSPT was used to obtain descriptive information regarding praxis.

The Oral Movement portion of the KSPT contained 11 items and has shown high reliability of measurement (Cronbach's alpha = .84) and high test-retest reliability (Cronbach's alpha =.93) (Kaufman, 1995). Construct, convergent and discriminant validity was measured on the KSPT-A, the original test model prior to slight revisions in developing the final KSPT. Strong validity was evidenced by unidimensionality measures such as principal component analysis, Guttman scale analysis, and correlational modeling (Kaufman, 1995).

Per the KSPT guidelines, a complete oral mechanism examination was first conducted on all participants in order to rule out structural abnormalities related to performance. According to the KSPT administration manual, all participants were asked to imitate the investigator ("Do what I do") as the 11 components of the KSPT were performed and scored in a binary manner.

*Submental amplitude, Swallow Apnea Duration, and Respiratory Swallow Pattern*: These were measured simultaneously as participants consumed 2 trials of 5ml water, 2 trials of 5cc pudding/yogurt, and 2 trials of ¼ cookie. The trials were presented using a randomized block design. The chosen volumes of consistencies and number of trials has been typically used during clinical, research and instrumental protocols involving pediatric patients (Casas, McPherson, & Kenny, 1995; Leder & Karas, 2000; Morgan, Ward, Murdoch, & Bilbie, 2002). Figure 3 displays the configuration of the equipment and participant.

Surface electromyography (sEMG), which has been used in children with cerebral palsy, was selected as the optimal measure of submental amplitude in our pediatric population (Casas et

al., 1994). The parameters were assessed with a bio-amp system and LabChart 7 Pro software (PowerLab 16/30, ADInstruments, Inc., Colorado Springs, CO). Surface electromyography is an objective and noninvasive tool that has been used extensively to study swallow amplitude across pediatric and adult populations for both clinical and research purposes (Vaiman et al., 2004a; Vaiman et al., 2004b; Perlman, 1993; Gupta et al., 1996; Crary & Baldwin, 1997; Spiro et al., 1994; Palmer, 1989). In addition, the validity (kappa = .80) and inter-rater reliability (kappa = .78) of sEMG in the identification of swallow related events have been well documented (Crary et al., 2007).

The procedure for sEMG data acquisition was consistent with commonly employed techniques (Ferdjallah et al., 2000; Reaz et al., 2006; Vaiman et al., 2004b; Stepp, 2012). Participants were seated comfortably with a strong base of support. Bipolar pediatric electrodes were placed bilaterally over the submental 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. Following a one-minute rest period needed to establish a clean baseline signal, participants were instructed to perform maximum submental contraction tasks for normalization and the establishment of a maximum reference value (Vaiman et al., 2004b; Stepp, 2012). Three maximum tasks were conducted as follows:

1. Participants performed lingual elevation against the Iowa Oral Performance Instrument (IOPI) bulb (Northwest, 2005). The IOPI is a standard non-invasive tool with excellent validity and reliability that provides manometric values in kilopascals (kPa) (Adams et al., 2014; Potter & Short, 2009). The IOPI digitally displayed

pressure levels on a feedback screen. The device was connected, via a thin 2-inch tube, to the IOPI stem and bulb. The primary investigator held the IOPI stem to maintain consistency of positioning. The bulb was positioned longitudinally along the hard palate and posteriorly to the central incisors (Potter& Short, 2009). Participants were instructed to push against the air filled bulb toward the roof of their mouth as hard as possible with their tongue for three seconds. Following familiarization with the device, three trials with 30 seconds of rest between trials were obtained.

2. Participants performed three trials of maximum mandibular depression after being provided with visual and verbal cues by the investigator.
3. Participants performed three effortful swallows with one minute of rest in between trials. Visual and verbal cues were provided.

Surface EMG recordings of maximum submental contraction during these tasks were obtained. The task that yielded maximum amplitude values and was performed most efficiently by all children was maximum mandibular depression. Therefore, the signal corresponding to this task was selected as the basis for normalization across participants. Throughout the sEMG task, swallows were tagged according to trial number and bolus type.

SAD and the RSP were measured using pediatric thoracic and abdominal cotton elastic bands. Thoracic and abdominal movements during breathing have been frequently measured using elastic bands and provide valid and reliable data (Hegland, Huber, Pitts, & Sapienza, 2009; Hegland, Huber, Pitts, Davenport, & Sapienza, 2011). Consistent with Martin et al. (1994), the thoracic band was placed beneath the axilla and the abdominal band was placed at the umbilicus level. Movement of the rib cage and abdomen were detected. Observation of 20 seconds of tidal breathing was first conducted and was followed by tasks to ensure that the elastic bands were

accurately detecting movement required for the acquisition of SAD and RSP. These tasks included having each participant inhale deeply and exhale, and then maximally displace the abdomen inwardly and outwardly. The following RSP patterns were possible: Exhale-Swallow-Exhale, Inhale-Swallow-Exhale, Exhale-Swallow-Inhale, and Inhale-Swallow-Inhale.

*Clinical Feeding and Swallowing Severity*: This was evaluated using the Dysphagia Disorder Survey (DDS), a standardized mealtime assessment (Sheppard et al., 2014). This was chosen over an invasive tool such as fiberoptic endoscopic evaluation of swallowing (FEES), which would subject the child to risks that may include gagging, vasovagal syncope, epistaxis and laryngospasm (Hiss & Postma, 2003). It was also determined that videofluoroscopy would not be used as it would be clinically unethical to expose our pediatric control and non-referred patient groups to harmful ionizing radiation (Hiorns & Ryan, 2006).

The DDS has been standardized on 654 individuals (age range: 8-82 years) with developmental and intellectual disabilities including children with CP (Sheppard et al., 2014). It has been used reliably in children with CP as young as 2.1 years of age (Calis et al., 2008). Sensitivity and specificity of the DDS are as follows: Part 1 (.88 and .85) and Part 2 (.94 and .87); Kaiser-Meyer-Olkin measure (>.90) and Spearman correlations (>.85). Reliability was evidenced through moderate kappa values (.53-.71) (Sheppard et al., 2014). The DDS has two parts comprising the DDS Total score. DDS Part 1 contains seven components indirectly related to feeding and swallowing (e.g., independence, positioning). DDS Part 2 contains eight components specifically related to feeding and swallowing competencies. The eight components of Part 2 are scored using a binary system of zero to signify competence and one to signify deficiency. Five trials of liquid, semisolid and solid boluses are required to derive an accurate task analysis of feeding and swallowing using the DDS. A factor analysis of DDS components

revealed that the Body Mass Index (BMI) and the report of emesis/rumination were not of strong significance in reference to the other components (Sheppard et al., 2014). However, given their clinical importance, these items were obtained during the present investigation.

The children were seated comfortably to allow for a natural eating position. The following items were placed on the table: one cup of water, one pudding cup, and one two cookies. The investigator positioned a Canon HD video camera three feet in front of the participant, with the view that allowed for visualization of the child and food items (Figure 4). Participants were told to eat and drink in a regular manner. The participants ate at their normal pace. The children were only interrupted to facilitate eating/drinking of a particular bolus type or for safety intervention. Following five trials of each bolus type, the recording continued until meal conclusion.

*Voluntary sequential cough:* Voluntary sequential cough was elicited and measured in a manner similar to what has been described in the literature (Hegland et al., 2014). However, adaptations were made given the pediatric population of interest. Participants were seated comfortably with arms at the side. A facemask was placed over the participant's mouth and nose and was coupled to a pneumotachograph system that input differential pressure change to a digital spirometer (ADInstruments). The airflow signal was digitized at 2KHz with a range of 2V, and samples were low pass filtered at 150Hz. Recording was obtained in Liters/seconds. Following 20 seconds of tidal breathing, the investigator modeled and instructed the participants to take a deep breath and cough sequentially (or several times in a row) 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.

Figure 3. Participant seated in standard chair and connected to surface electrodes and respiratory/abdominal bands.

Figure 4. Participant seated in standard chair during clinical feeding and swallowing task.

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### v. Data Analysis

The primary investigator analyzed all data. Intra- and inter-rater reliability was assessed across all measures by the primary investigator (Rater 1) and co-investigator (Rater 2) using data sets from four randomly selected participants (2 TDCs, 2 SCP). Rater 2 was blinded to participant group.

*Demographic Information:* Information obtained from the intake form was used to characterize the sample, with emphasis on neurological function and developmental milestones. The TACL, GMFCS, MACS, and EDACS were scored according to their respective guidelines, in conjunction with parent reports, and were used to assign levels of ability related to gross motor, manual, and feeding/swallowing function. Outcomes included age equivalency on the TACL as well as a rating on functional scales (GMFCS, MACS, EDACS) that ranged from level I (mild impairment) to V (severe impairment).

*Oral Praxis:* The Oral Movement portion of the KSPT was scored according to the KSPT scoring manual, which provided detailed instructions on how to interpret and score each of the 11 components. The principal investigator has administered and scored the KSPT numerous times in clinical practice and trained the co-investigator in its usage and scoring. The raw score (total number of components scored correctly) was used for analysis.

*Submental Amplitude:* LabChart 7 Pro software (ADInstruments) was utilized for all sEMG analysis. Channel 5 was assigned to the left submental swallow signal and Channel 6 was assigned to the right submental swallow signal. Maximum functional swallows and six tagged, bolus swallow signals were analyzed by taking the average of both channels. For participants who swallowed more than one time per bolus trial, the first swallow was analyzed. Using the arithmetic feature of LabChart, raw sEMG swallow signals were rectified (transformed to a

positive polarity frequency) and smoothed through the root mean square equation for the left [RMS (CH5, .2)] and right [RMS (CH6, .2)] swallow signals. Within the channel settings feature, standard integral channels were created and corresponded to the sEMG signal. Start and end values corresponding to each swallow signal were determined based on the respitrace signal (flat slope signifying apnea) as this signal was synchornized with the sEMG signal. Using the data pad feature in LabChart, integral values for each swallow were obtained.

*Swallow Apnea Duration and Respiratory Swallow Pattern:* LabChart software (ADInstruments) was used to determine the duration of swallow apnea. The SAD and RSP demonstrated by each child were analyzed through analysis of the respitrace signal. The positive slope represents inhalation, the zero slope represents the period of swallow apnea, and the negative slope represents exhalation. Figures 5 and 6 display the exhale-swallow-exhale pattern and the inhale-swallow-inhale pattern, respectively. Further verification of apnea duration across bolus swallows was assessed with the simultaneous sEMG swallow signal.

*Clinical Feeding and Swallowing Severity:* The primary and co-investigator are certified in the administration, scoring and interpretation of the DDS. DDS scoring was done in real time during the mealtime assessment using the DDS scoring form and instruction manual. This allowed for Part 1, Part 2 and Total scores to be determined immediately following the mealtime assessment. The investigators could not be blinded to group during the live DDS assessment; however, video recording analysis using the DDS was conducted in a blinded fashion by the co-investigator. Outcomes included DDS Part 1 (0-15), DDS Part 2 (0-19), and DDS Total Score (0-34). The DDS Total Score is the sum of DDS Part 1 and DDS Part 2. A score of 34 indicates the most severe feeding and swallowing impairment.

*Voluntary sequential cough:* All measures of voluntary sequential cough airflow (CIV,

IPD, CPD, PEFRT, PEFR, CVA, and CEV) were analyzed using LabChart7 Pro software (ADInstruments). Thirty seconds of tidal breathing was recorded to ensure accurate signal detection and to establish the signal baseline (0 liters/second). The Data Pad feature was utilized to assess volume, duration, and peak value parameters corresponding to the airflow channel. The area between the point at which the cough signal fell below 0 L/s to the point at which it returned to 0 L/s was taken as the CIV. The time between those same two points was taken as the IPD. The time between the end of the inspiratory phase to the point at which the signal began to sharply increase was taken as the CPD. The time taken for the signal to travel from the end of the compression phase to the maximum value was taken as the PEFRT. The maximum signal value of the cough waveform was taken as the PEFR. The CVA was calculated by dividing the PEFR by the PEFRT. Finally, the area under the cough response, or the area between the end of the compression phase to the start of the subsequent compression phase, was taken as the CEV.

Figure 5. The ideal exhale-swallow-exhale pattern.

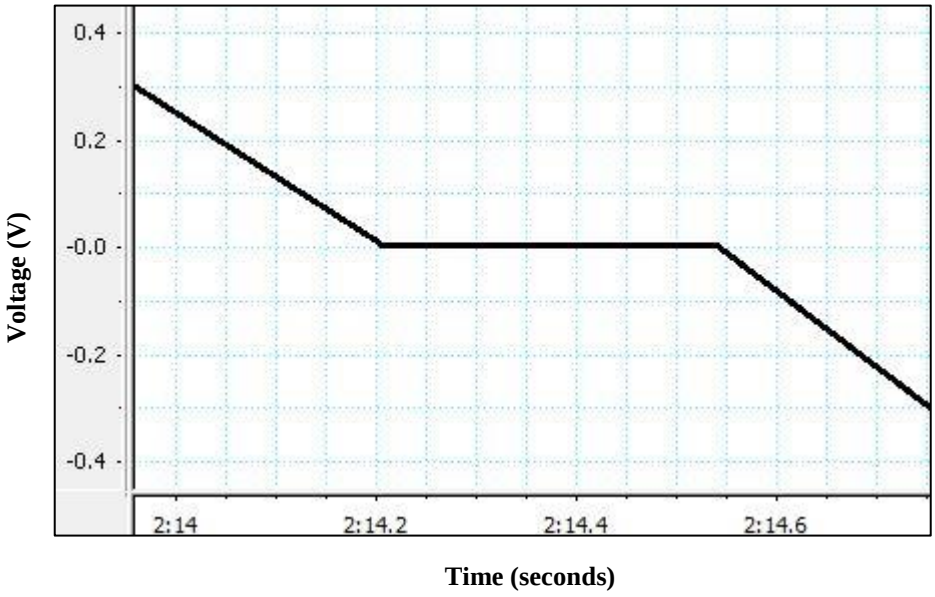
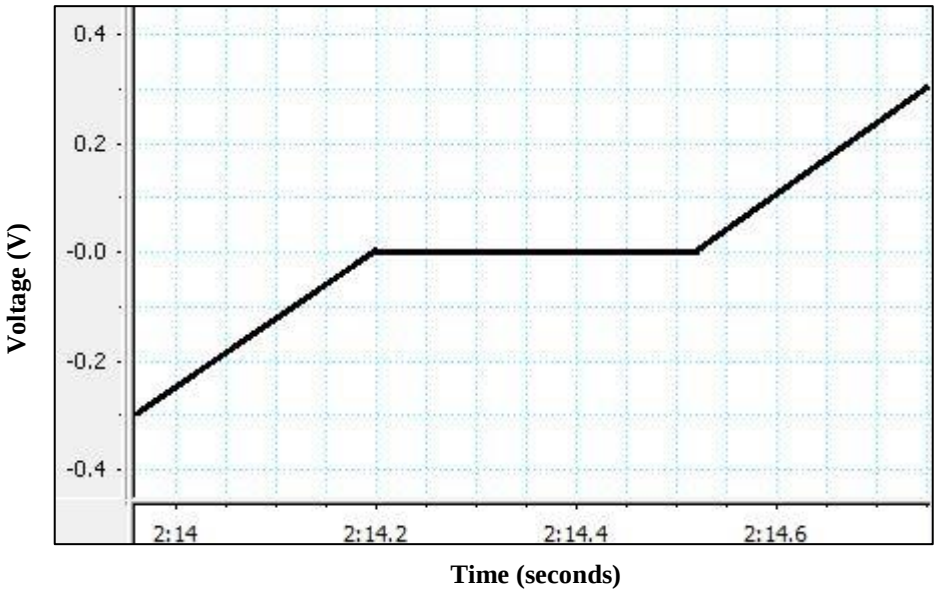



Figure 6. The abnormal inhale-swallow-inhale pattern.



### vi. Statistical Analyses

Based on the results of power analysis using G*Power 3 software (Faul, Erdfelder, Lang, & Buchner, 2007), 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). Intra-and inter-rater reliability of all variables was assessed using data sets from four randomly selected participants. Reliability statistics included intra-class correlation coefficients for continuous variables and Cohen's Kappa statistic for binary variables.

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. Kruskal-Wallis tests were used to assess between-group differences in measures of feeding/swallowing competencies and clinical feeding and swallowing severity. Specifically, group differences in oral praxis, submental amplitudes across bolus swallows, swallow apnea duration, respiratory swallow patterns, and DDS Part 1, DDS Part 2, and DDS Total scores were evaluated. Spearman's Rank Order Correlations were used to assess relationships between oral praxis and all DDS scores, post swallow inhalation pattern and all DDS scores, and submental amplitudes across bolus swallows and all DDS scores. These correlations were assessed in the total sample (n=21) and in the SCP group (n=11).

The Mann Whitney test was used to evaluate between-group differences in the number of coughs produced per cough epoch. Kruskal-Wallis tests were used to assess between-group differences in measures of cough effectiveness. Specifically, group differences in CIV, IPD, CPD, PEFRT, PEFR, CVA, and CEV were evaluated. Within the SCP group (n=11), Spearman's Rank Order Correlations were used to assess relationships between PEFR and DDS Total score, and between CVA and DDS Total score, for the first (Cr1) and second (Cr2) cough of the cough

epoch. Spearman's Rank Order Correlations were also used to determine relationships between DDS Total score and GMFCS in addition to PEFR and GMFCS, and CVA and GMFCS, for the first (Cr1) and second (Cr2) cough of the cough epoch.

Effect sizes were calculated for all applicable measures. Specifically, the effect size (r) was used with Mann Whitney tests, and the effect size ($\eta^2$) was used with Kruskal-Wallis tests. The alpha level of significance for all statistical tests was set at $p < .05$.

# III. RESULTS

## i. Reliability

Intra- and inter-rater reliability was excellent for all continuous and binary responses as evidenced by intraclass correlation coefficients and Cohen's kappa statistic (Table 5). All values exceeded .80.

## ii. Participant Characteristics

A total of 23 children were enrolled in the study, but two children withdrew as they were unable to complete the evaluation. Therefore, 21 children completed the protocol (10 controls and 11 children with SCP). Demographic information for all participants is presented in Table 6. Table 7 presents information specific to the SCP group. This group comprised 8 children with unilateral impairment and 3 children with bilateral impairment. The most common central nervous system damage in this sample was intra- and peri-ventricular hemorrhage. The mode, median, and range of functional classification scores were as follows: GMFCS (I, II, I-V), MACS (II, II, I-V), and EDACS (II, II, I-IV). This indicates mild functional impairment for this sample. The two groups were well matched with no significant between-group differences in age, sex, body mass index, or age equivalency on the TACL (Table 8).

Table 5. Intra- and inter-rater reliability results.

| Measure | Intra-rater (ICC) | Inter-rater (ICC) |
|---|---|---|
| DDS | .992 | .961 |
| KT | .980 | .935 |
| sEMG (Left) | .978 | .863 |
| sEMG (Right) | .989 | .987 |
| SAD | .989 | .906 |
| CIV | .999 | .999 |
| IPD | .996 | .918 |
| CPD | 1.00 | .841 |
| PEFRT | 1.00 | .868 |
| PEFR | 1.00 | 1.00 |
| CVA | .995 | .982 |
| CEV | .999 | .996 |

| Measure | Intra-rater (κ) | Inter-rater (κ) |
|---|---|---|
| RSP (Pre-swallow) | 1.00 | .895 |
| RSP (Post-swallow) | 1.00 | .882 |

ICC, intraclass correlation coefficient, **κ**, Cohen's kappa, DDS, Dysphagia Disorder Survey, KT, Kaufman Test, sEMG, surface electromyography, SAD, swallow apnea duration, CIV, cough inspired volume, IPD, inspiratory phase duration, CPD, compression phase duration, PEFRT, peak expiratory flow rate rise time, PEFR, peak expiratory flow rate, CVA, cough volume acceleration, CEV, cough expired volume, RSP, respiratory-swallow pattern

Table 6. Participant characteristics.

| **Variable** | **TDC group (n=10)** | **SCP group (n=11)** |
|---|---|---|
| Mean age (SD) (years) | 7.6 (2) | 7.7 (2.4) |
| Median TACL (IQR) | 11 (2.3) | 7 (2.5) |
| Mean BMI (SD) | 15.5 (3) | 18.2 (3.8) |
| Males (%) (n) | 50 (5) | 72.7 (8) |

TDC, typically developing control, SCP, spastic cerebral palsy, SD, standard deviation, TACL, test of auditory comprehension of language, IQR, interquartile range, BMI, body mass index

Table 7. Neurological insult and functional classification in children with spastic cerebral palsy.

| Participant ID | Neurological Insult | Motor Involvement | GMFCS | MACS | EDACS |
|---|---|---|---|---|---|
| SCP1 | PVL | Unilateral | 1 | 2 | 2 |
| SCP2 | PVH II | Unilateral | 2 | 2 | 2 |
| SCP3 | PVH II | Unilateral | 1 | 2 | 2 |
| SCP4 | PVL | Bilateral | 4 | 5 | 4 |
| SCP5 | PVH II | Unilateral | 2 | 2 | 2 |
| SCP6 | IVH IV | Bilateral | 5 | 3 | 2 |
| SCP7 | IVH II | Unilateral | 2 | 3 | 2 |
| SCP8 | PVL | Unilateral | 1 | 1 | 1 |
| SCP9 | PAIS | Unilateral | 1 | 1 | 2 |
| SCP10 | IVH II | Unilateral | 1 | 2 | 1 |
| SCP11 | HIE | Bilateral | 4 | 5 | 3 |

SCP, spastic cerebral palsy, GMFCS, Gross Motor Function Classification Scale, MACS, Manual Ability Classification Scale, EDACS, Eating and Drinking Ability Classification Scale, PVL, periventricular leukomalacia, PVH, periventricular hemorrhage, IVH, intraventricular hemorrhage, PAIS, perinatal arterial ischemic stroke, HIE, hypoxic-ischemic encephalopathy

Table 8. Statistical comparisons of demographic characteristics between typically developing controls and children with spastic cerebral palsy.

| | **Age** | **Sex** | **BMI** | **TACL Age Equivalency (years)** |
|---|---|---|---|---|
| Mann Whitney U | 54 | 42.5 | 32 | 28.5 |
| Z | -.071 | -1.045 | -1.621 | -1.878 |
| p-value | .973 | .378 | .114 | .061 |
| Effect size (r) | .025 | .369 | .573 | .664 |

BMI, body mass index, TACL, test of auditory comprehension of language

### iii. Between-Group Comparisons: Oropharyngeal Function and Clinical Feeding and Swallowing Severity

Table 9 shows group means and standard deviations for the Kaufman Test and all components of the DDS. Figure 7 displays oral praxis performance by group. Compared to children with SCP, the TDC group had significantly higher oral praxis scores ($\chi^2 = 8.573$, $p = .003$, $\eta^2 = .429$) as well as significantly lower 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 DDS Total ($\chi^2 = 15.421$, $p < .0001$, $\eta^2 = .771$). A group comparison of the feeding/swallowing task analysis portion of the DDS is presented in Figure 8. Compared to controls, children with SCP had greater difficulties across all feeding/swallowing tasks. No child in the TDC group displayed overt signs of aspiration, whereas three children with CP exhibited a single instance of cough.

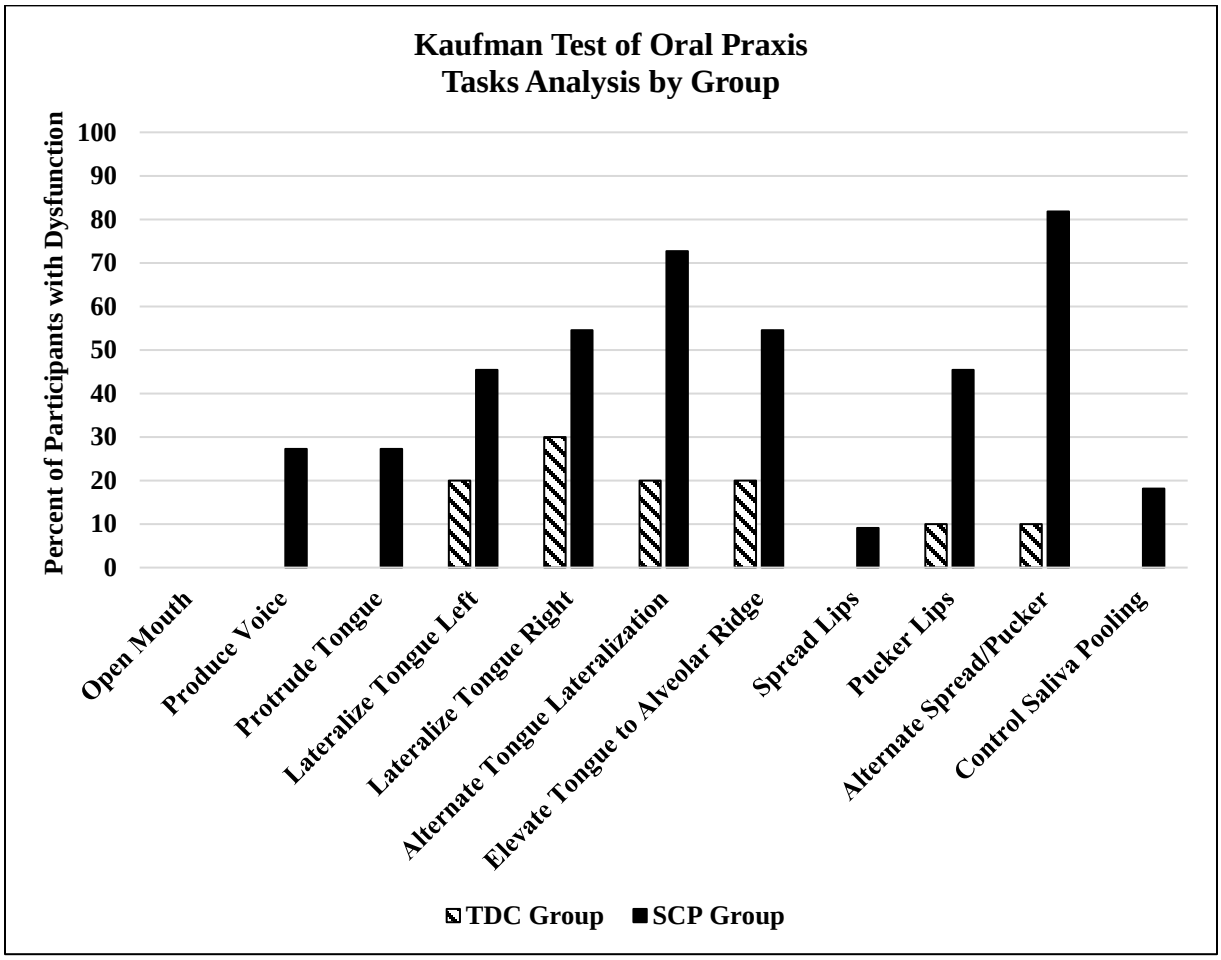
Significant between-group differences in normalized sEMG values were observed ($\chi^2 = 11.731$, $p = .001$). Specifically, children with SCP required a significantly greater percent of maximum amplitude during puree ($\chi^2 = 6.091$, $p = .014$, $\eta^2 = .305$) and solid swallows ($\chi^2 = 10.555$, $p = .001$, $\eta^2 = .528$), but not for liquid swallows ($\chi^2 = .077$, $p = .782$, $\eta^2 = .004$) (Figure 9). Further analysis within the SCP group revealed no significant differences in swallow amplitude values between the less impaired and more impaired side of function ($\chi^2 = .838$, $p = .360$, $\eta^2 = .084$).

Table 9. Results of the Kaufman Test and Dysphagia Disorder Survey for typically developing controls and children with spastic cerebral palsy.

| **Variable** | **TDC group (n=10)** | **SCP group (n=11)** |
|---|---|---|
| Mean KT (SD) (Maximum = 11) | 9.9 (1.9) | 6.6 (2.2) |
| Mean $DDS_1$ (SD) (Maximum = 15) | 0 (0) | 3.7 (2.3) |
| Mean $DDS_2$ (SD) (Maximum = 19) | 0.8 (1.3) | 7.6 (3.6) |
| Mean $DDS_T$ (SD) (Maximum = 34) | 0.8 (1.3) | 11.4 (5.7) |

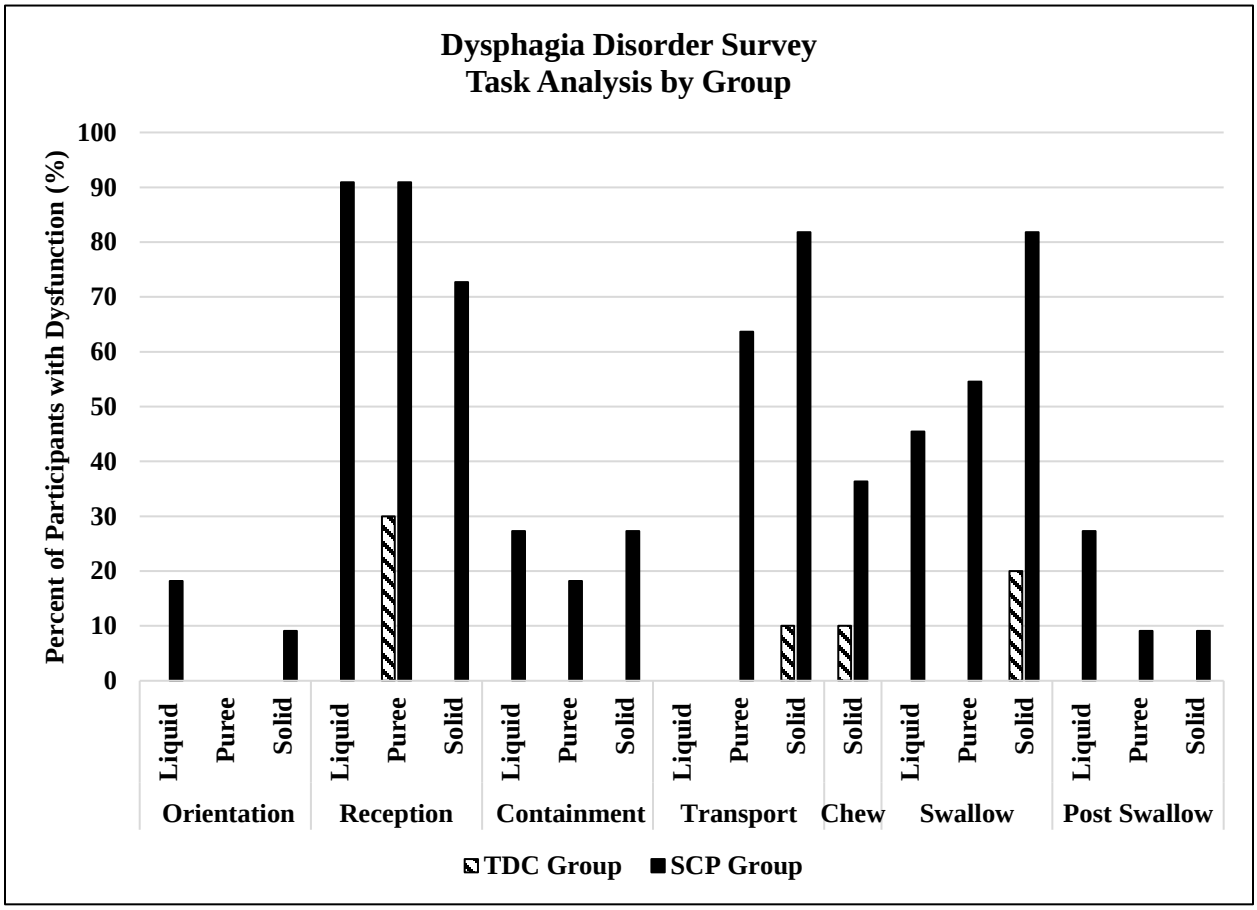
TDC, typically developing control, SCP, spastic cerebral palsy, KT, Kaufman Test, $DDS_1$, Dysphagia Disorder Survey Part 1, $DDS_2$, Dysphagia Disorder Survey Part 2, $DDS_T$, Dysphagia Disorder Survey Total

Figure 7. Differences between typically developing controls and children with spastic cerebral palsy on measures of oral praxis.



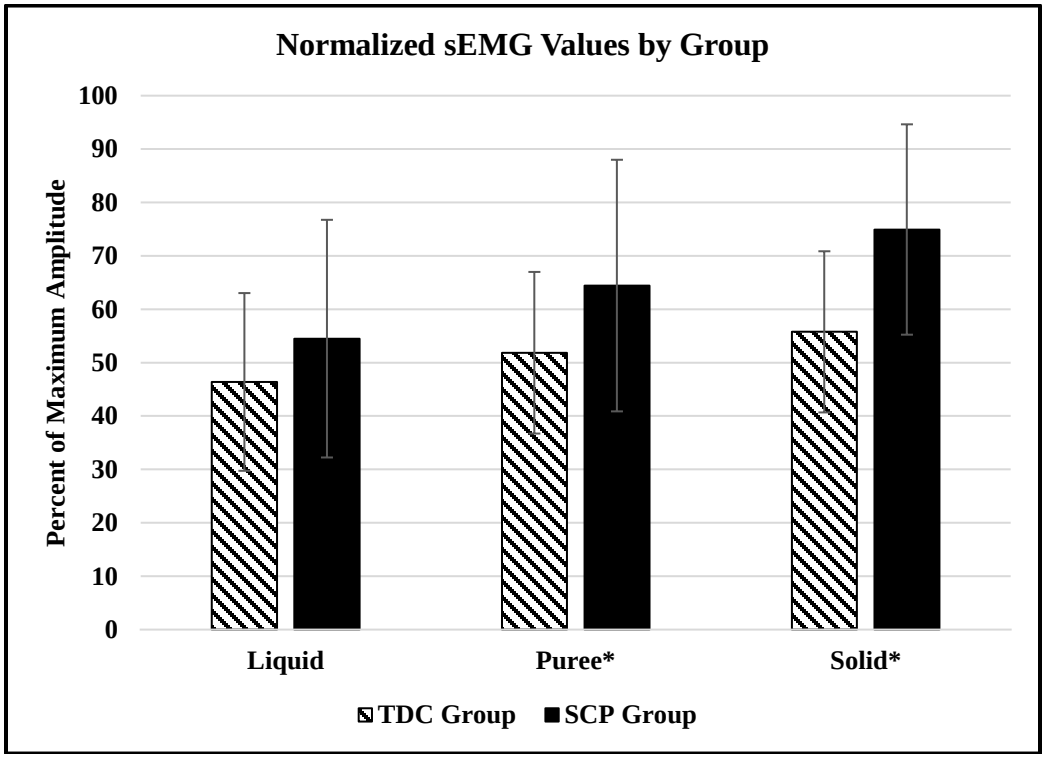

TDC, typically developing control, SCP, spastic cerebral palsy

Figure 8. Differences between for typically developing controls and children with spastic cerebral palsy on measures of clinical feeding and swallowing severity.




TDC, typically developing control, SCP, spastic cerebral palsy

Figure 9. Differences between typically developing controls and children with spastic cerebral palsy on normalized submental swallow amplitude across bolus consistency.




*p<0.05
TDC, typically developing control, SCP, spastic cerebral palsy

### iv. Relationships Between Oropharyngeal Function and Clinical Feeding and Swallowing Severity

Results of the Spearman Rank Order Correlation revealed significant, negative correlations between the Kaufman Test and the DDS (Table 10). Age was not correlated with oral praxis or any component of the DDS. Normalized sEMG values were not correlated with the DDS. When looking at the SCP group only, significant, negative correlations between the Kaufman Test and the DDS Part 1 ($r_s$=-.664, p=.026), DDS Part 2 ($r_s$=-.633, p=.036) and DDS Total ($r_s$=-.671, p=.024) were observed. Figure 10 displays the association between the Kaufman Test and the DDS Total for children with SCP. No significant correlations between normalized sEMG values and DDS scores were observed for children with SCP.

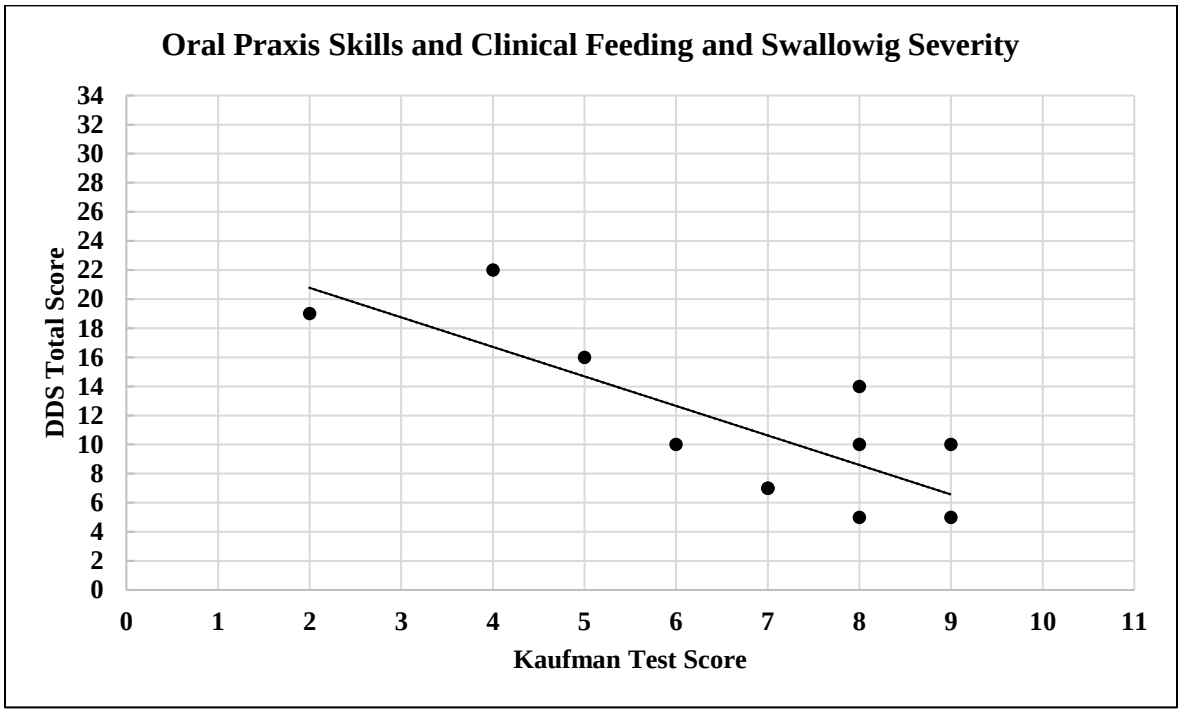
Table 10. Correlations between the Kaufman Test and the Dysphagia Disorder Survey for all participants.

| **DDS Component** | **Value** | **KT** |
|---|---|---|
| $DDS_1$ | $r_s$ | $-0.733^{***}$ |
| $DDS_2$ | $r_s$ | $-0.815^{***}$ |
| $DDS_T$ | $r_s$ | $-0.802^{***}$ |

*p<0.05, **p<0.005, ***p<0.0005

DDS, Dysphagia Disorder Survey, $DDS_1$, Dysphagia Disorder Survey Part 1, $DDS_2$, Dysphagia Disorder Survey Part 2, $DDS_T$, Dysphagia Disorder Survey Total, KT, Kaufman Test

Figure 10. Negative correlation between the Kaufman Test and the Dysphagia Disorder Survey Total score in children with spastic cerebral palsy.

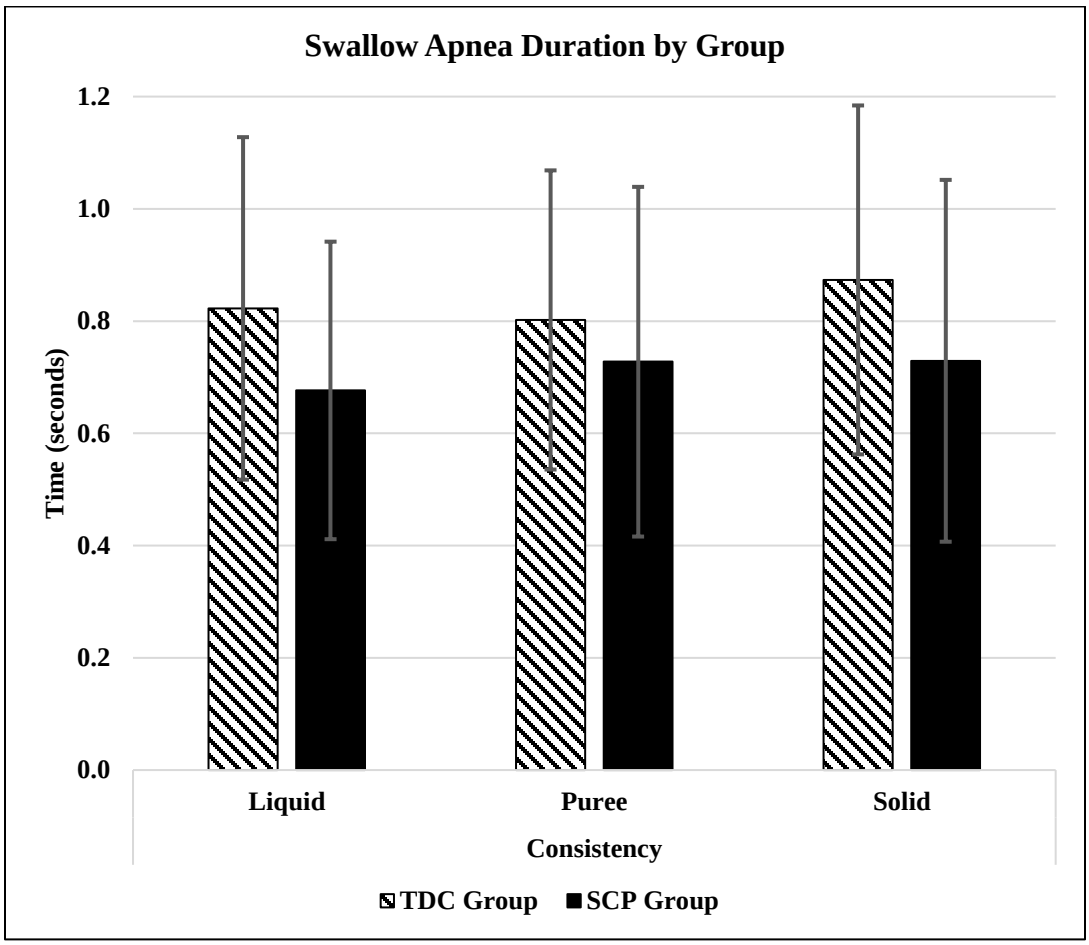



DDS, Dysphagia Disorder Survey

### v. Between-Group Comparisons: Respiratory-Swallow Coordination and Clinical Feeding and Swallowing Severity

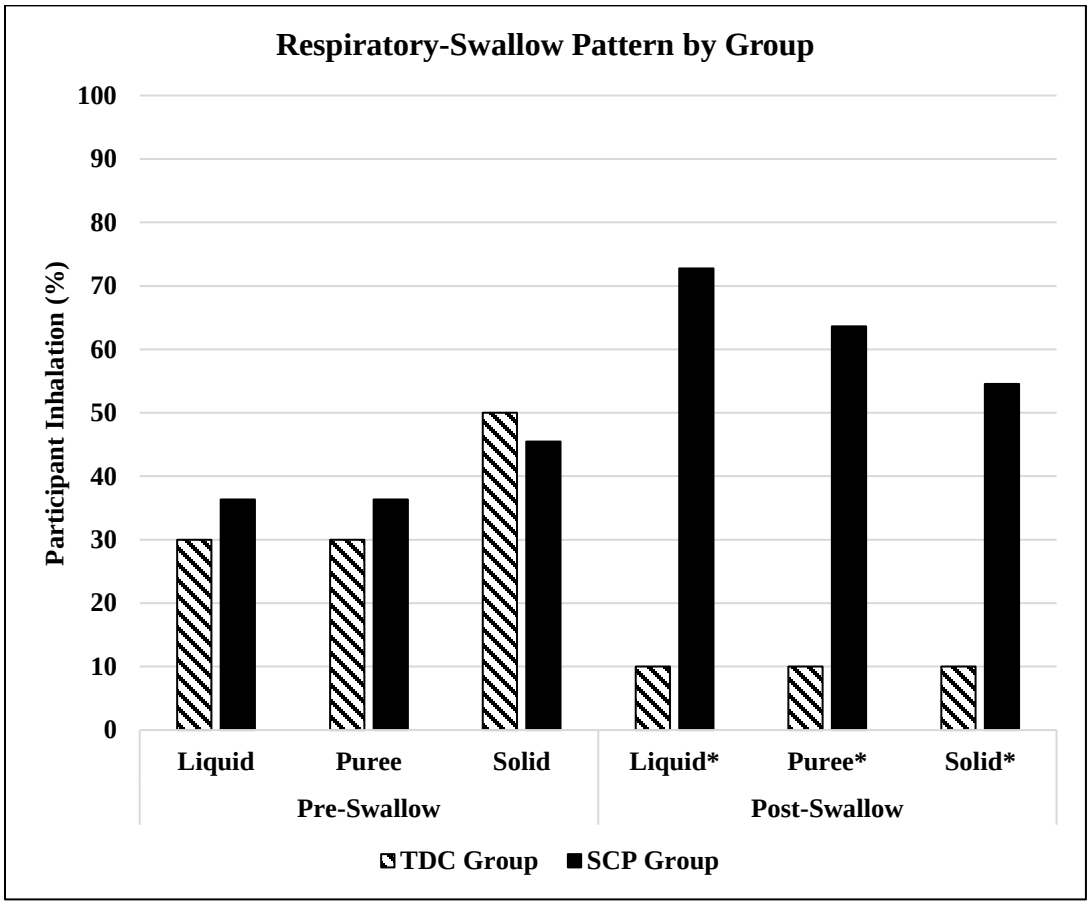
No significant between-group differences in SAD were observed across liquid ($\chi^2$ = 1.698, p = .193, $\eta^2$ = .085), puree ($\chi^2$ = .179, p = .672, $\eta^2$ = .009), or solid ($\chi^2$ = 1.983, p = .159, $\eta^2$ = .099) bolus consistencies. Figure 11 displays the group means and standard deviations across consistencies. Respiratory-swallow patterns for both groups are presented in Figure 12, and a significantly greater number of participants in the SCP group (>50%) demonstrated a post-swallow inhalation on liquids ($\chi^2$ = 8.015, p = .005, $\eta^2$ = .401), purees ($\chi^2$ = 6.086, p = .014, $\eta^2$ = .304), and solids ($\chi^2$ = 4.455, p = .035, $\eta^2$ = .223) compared to controls. No significant pre-swallow differences were observed. Representative examples of the respiratory-swallow pattern from a TDC and a child with SCP are presented in Figures 13 and 14, respectively.

Figure 11. Differences between typically developing controls and children with spastic cerebral palsy on duration of swallow apnea across bolus consistency.




TDC, typically developing control, SCP, spastic cerebral palsy

Figure 12. Differences between typically developing controls and children with spastic cerebral palsy on respiratory swallow pattern across bolus consistency.




*p<0.05
TDC, typically developing control, SCP, spastic cerebral palsy

Figure 13. An exhale-swallow-exhale pattern produced by a typically developing control.

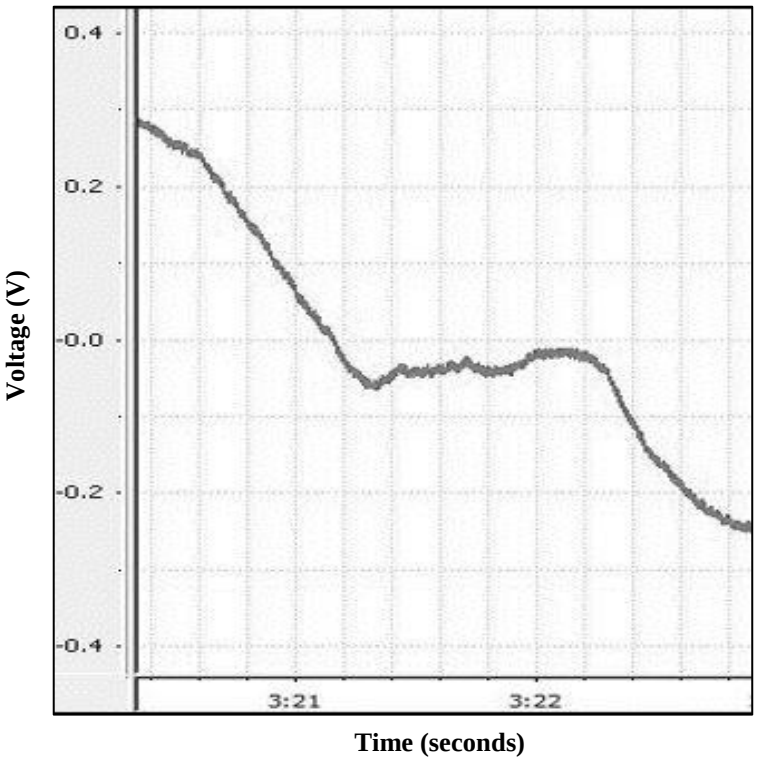
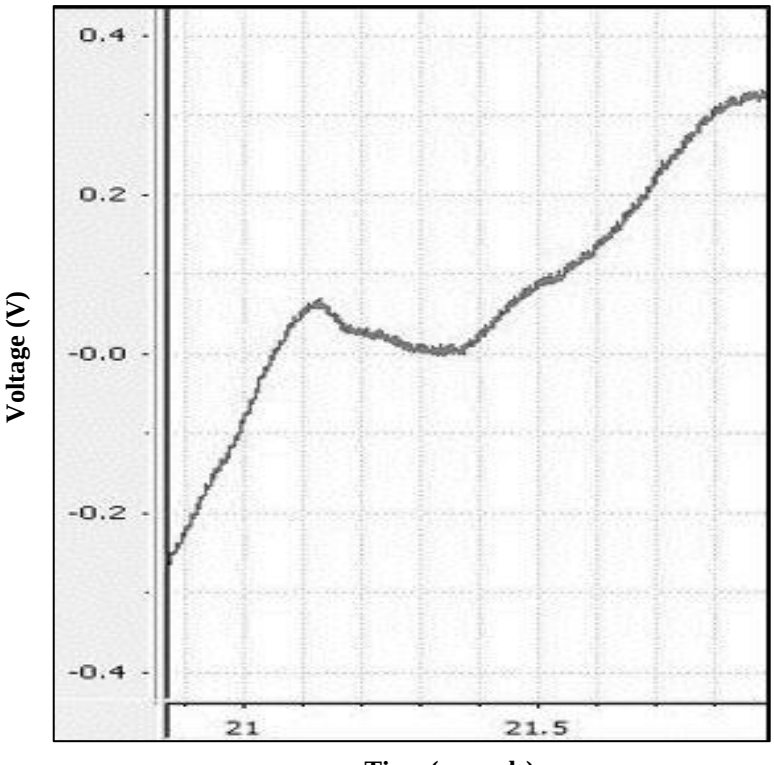


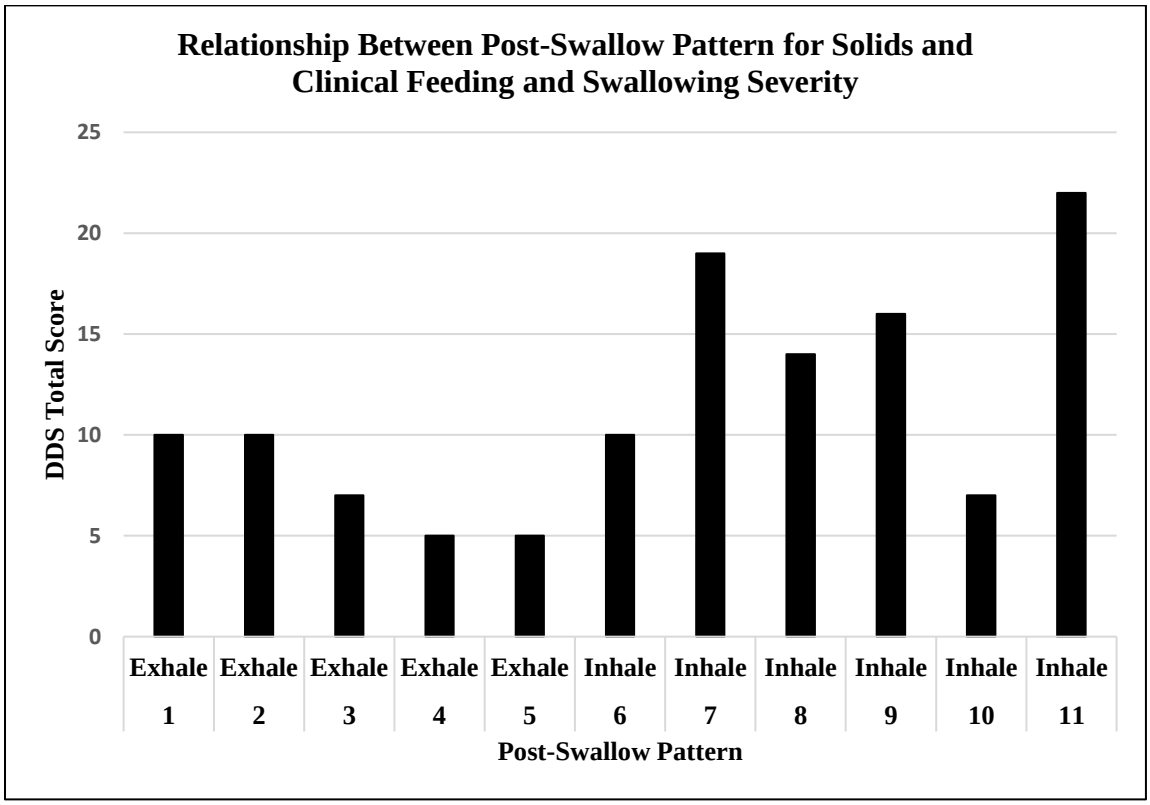

Figure 14. An inhale-swallow-inhale pattern produced by a child with spastic cerebral palsy.



### vi. Relationships Between Respiratory-Swallow Coordination and Clinical Feeding and Swallowing Severity

Results of the Spearman Rank Order Correlation revealed significant correlations between RSP and all components of the DDS (Table 11). In the SCP group, significant correlations between the post-swallow respiratory pattern for solids and the DDS Part 1 ($r_s$=.734, p=.010), DDS Part 2 ($r_s$=.610, p=.046) and DDS Total ($r_s$=.673, p=.023) were observed. Figure 15 displays the relationship between RSP for solids and DDS scores for all 11 children with SCP as grouped by post-swallow exhalation versus post-swallow inhalation. Neither age nor gross motor impairment were correlated with RSP. No correlations between swallow apnea duration and DDS components were observed.

Figure 15. Association between respiratory-swallow pattern for solids and the Dysphagia Disorder Survey in children with spastic cerebral palsy.




DDS, Dysphagia Disorder Survey

### vii. Between-Group Comparisons on Measures of Cough Effectiveness and their Relationship with Clinical Feeding and Swallowing Severity

The means and standard deviations of cough response totals across all 3 trials of cough were TDC group (3.24 ± 1.53) and SCP group (2.95 ± .701). The median number of coughs produced per epoch for both groups was three. A Mann Whitney Test revealed no significant between-group differences in number of coughs produced per epoch.

Table 12 presents group means and standard deviations of voluntary cough airflow measures (CIV, IPD, CPD, PEFRT, PEFR, CVA, and CEV) for cough response 1 and cough response 2, while Table 13 presents statistical comparisons across these measures. 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 compared to the TDC group for the first cough (Cr1) ($p=.019$) of the cough epoch. A cough epoch from a TDC and a child with SCP is presented in Figures 16 and 17, respectively.

In children with SCP, no significant correlations were observed between the DDS and PEFR for the first cough (Cr1) ($r_s=-.475$, $p=.165$) or second cough (Cr2) ($r_s=-.438$, $p=.205$) of the cough epoch. Additionally, no significant correlations were observed between the DDS and CVA for Cr1 ($r_s=-.043$, $p=.906$) or Cr2 ($r_s=.043$, $p=.906$) of the cough epoch. In the SCP group, the DDS was positively correlated with the GMFCS ($r_s=.749$, $p=.008$) (Figure 18). Correlations between measures of cough effectiveness and the GMFCS were assessed. Though no significant correlations were found between the GMFCS and CVA for Cr1 ($r_s=-.492$, $p=.149$) or Cr2 ($r_s=-.479$, $p=.162$) of the cough epoch, significant, negative correlations were observed between the GMFCS and PEFR for Cr1 ($r_s=-.702$, $p=.024$) and Cr2 ($r_s=-.715$, $p=.020$) of the cough epoch (Figures 19 and 20).

Table 12. Means and standard deviations of measures of voluntary cough airflow for typically developing controls and children with spastic cerebral palsy.

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

| **Cough Response 2** | **CIV** | | **IPD** | | **CPD** | | **PEFRT** | | **PEFR** | | **CVA** | | **CEV** | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| Group | **TDC** | **SCP** | **TDC** | **SCP** | **TDC** | **SCP** | **TDC** | **SCP** | **TDC** | **SCP** | **TDC** | **SCP** | **TDC** | **SCP** |
| Mean | NA | NA | NA | NA | .141 | .148 | .040 | .059 | 1.49 | 1.48 | 43.3 | 33.8 | .180 | .187 |
| Standard Deviation | NA | NA | NA | NA | .107 | .109 | .022 | .050 | .381 | .661 | 18.3 | 19.8 | .060 | .116 |

CIV, cough inspired volume, IPD, inspiratory phase duration, CPD, compression phase duration, PEFRT, peak expiratory flowrate rise time, PEFR, peak expiratory flow rate, CVA, cough volume acceleration, CEV, cough expired volume, TDC, typically developing control, SCP, spastic cerebral palsy, NA, not applicable

Table 13. Statistical comparisons between typically developing controls and children with spastic cerebral palsy on measures of voluntary cough airflow.



| **Cough Response 1** | **CIV** | **IPD** | **CPD** | **PEFRT** | **PEFR** | **CVA** | **CEV** |
|---|---|---|---|---|---|---|---|
| $\chi^2$ | .463 | .091 | .463 | 3.158 | .823 | 5.491 | .691 |
| p-value | .196 | .762 | .496 | .076 | .364 | .019 | .406 |
| Effect size ($\eta^2$) | .024 | .005 | .024 | .166 | .043 | .289 | .036 |

| **Cough Response 2** | **CIV** | **IPD** | **CPD** | **PEFRT** | **PEFR** | **CVA** | **CEV** |
|---|---|---|---|---|---|---|---|
| $\chi^2$ | NA | NA | .092 | 1.751 | .051 | .691 | .036 |
| p-value | NA | NA | .762 | .186 | .821 | .406 | .850 |
| Effect size ($\eta^2$) | NA | NA | .005 | .092 | .003 | .036 | .002 |

CIV, cough inspired volume, IPD, inspiratory phase duration, CPD, compression phase duration, PEFRT, peak expiratory flowrate rise time, PEFR, peak expiratory flow rate, CVA, cough volume acceleration, CEV, cough expired volume, NA, not applicable

Figure 16. Cough epoch produced by a typically developing control.

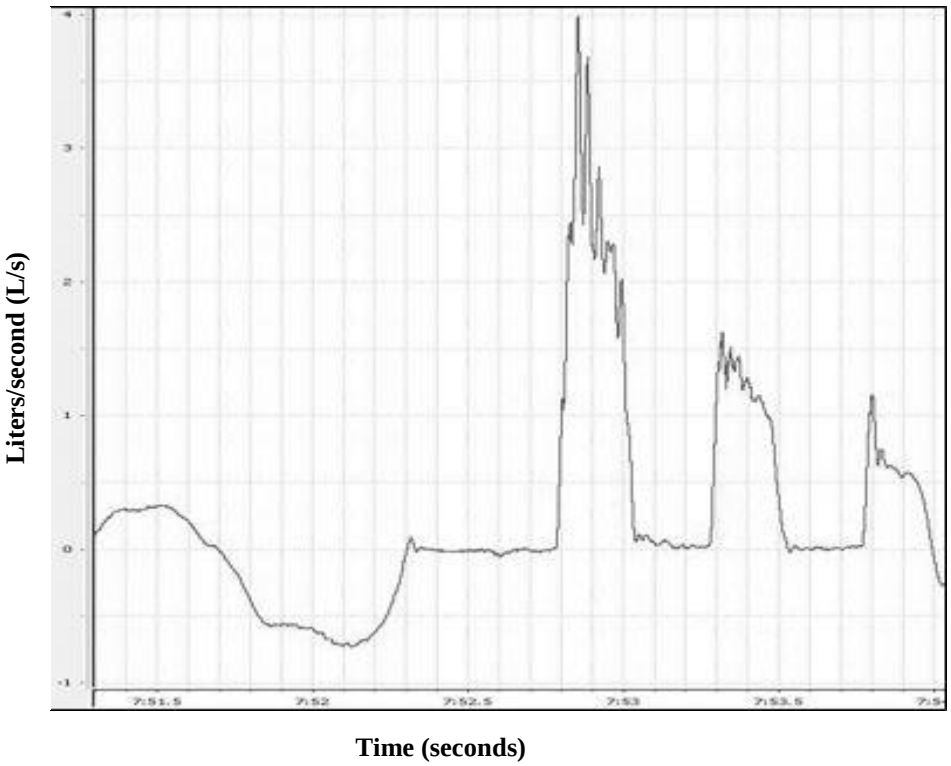



Figure 17. Cough epoch produced by a child with spastic cerebral palsy.

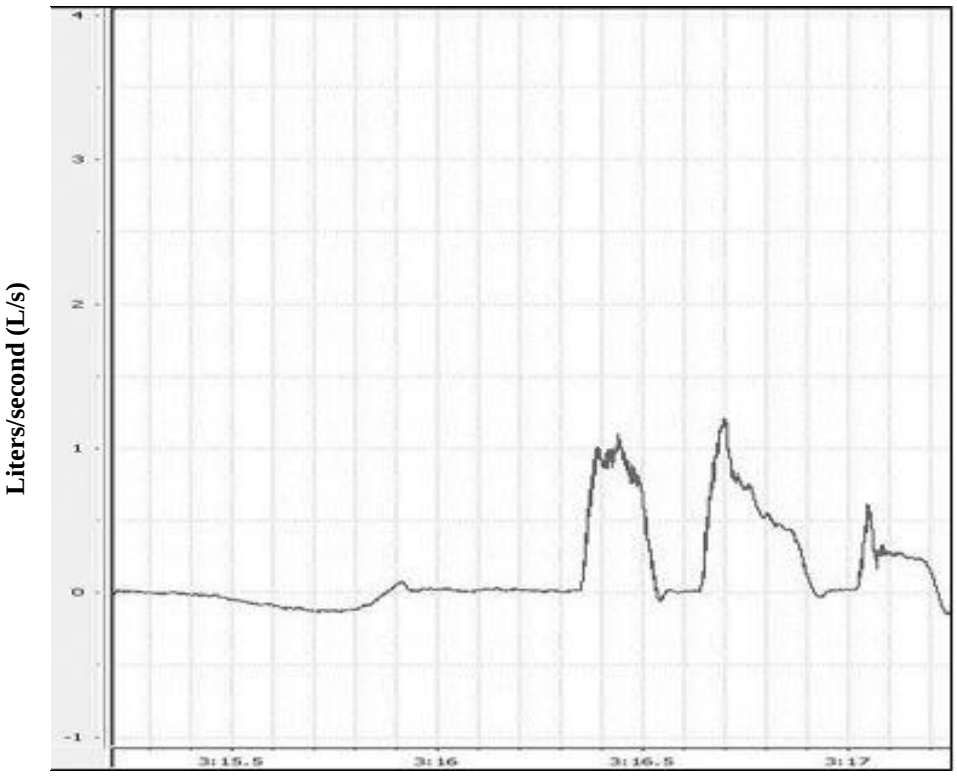


Figure 18. Positive correlation between the Dysphagia Disorder Survey and GMFCS in children with spastic cerebral palsy.

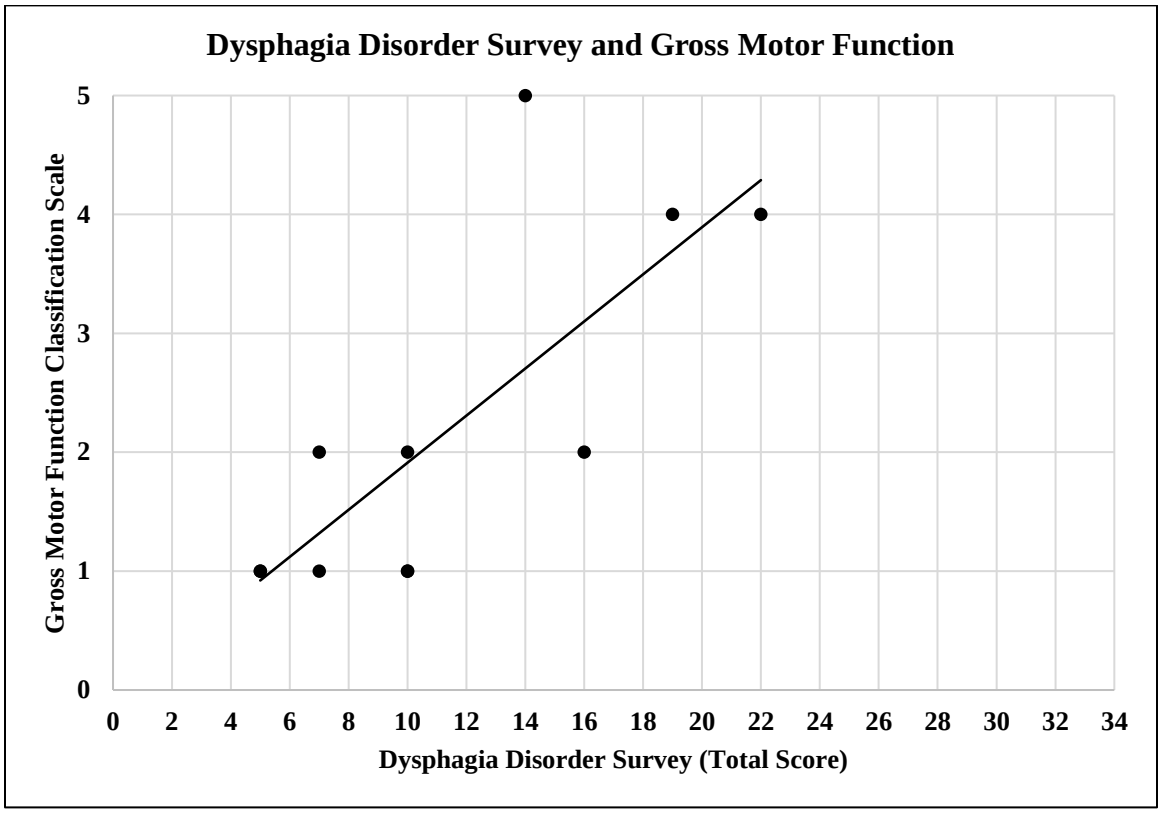


Figure 19. Negative correlation between PEFR and GMFCS for cough response 1 in children with spastic cerebral palsy.

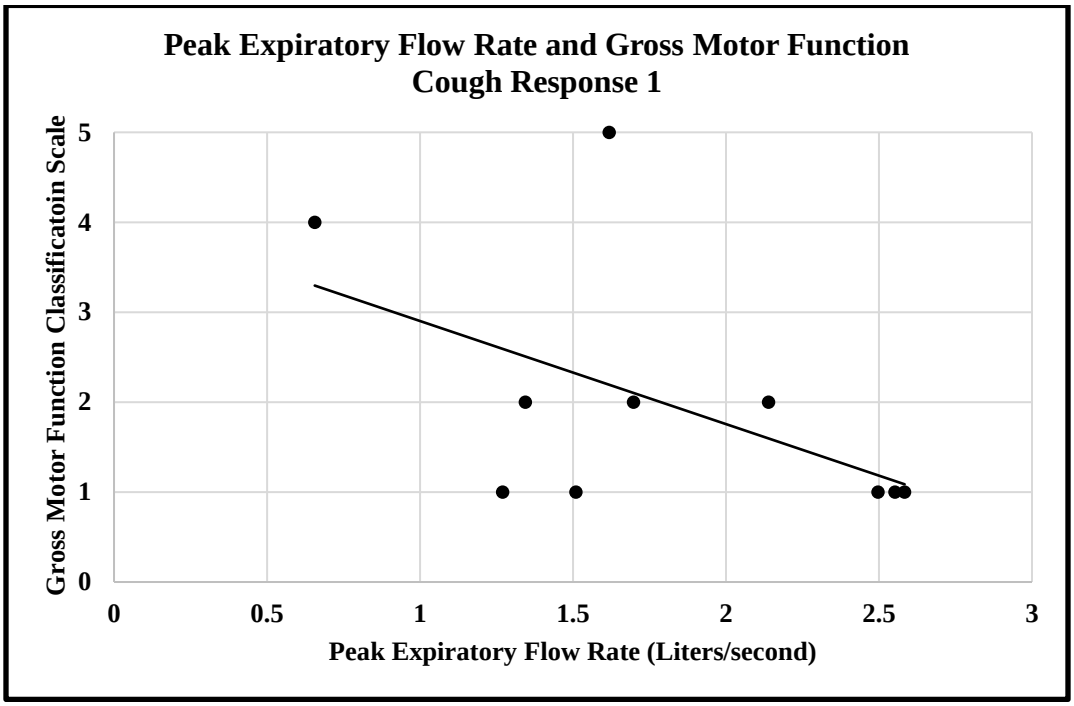
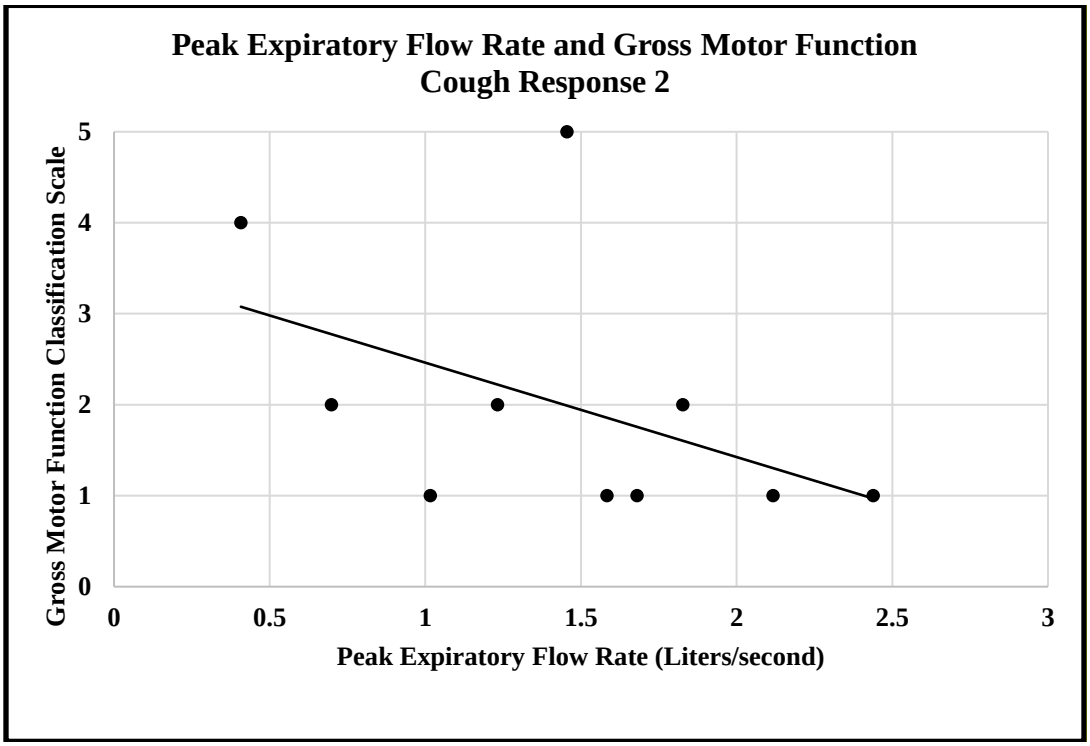



Figure 20. Negative correlation between PEFR and GMFCS for cough response 2 in children with spastic cerebral palsy.



# IV. DISCUSSION

## i. Overview of Results

This investigation aimed to objectively examine airway protective behaviors and clinical feeding and swallowing severity in children with SCP and healthy controls by testing the differences in oropharyngeal function, respiratory-swallow coordination, and measures of cough effectiveness between children with SCP and TDCs. It further aimed to identify and measure physiological factors contributing to clinical feeding and swallowing severity. Differences in the SCP and TDC groups were found on measures of clinical feeding and swallow severity, oropharyngeal function, respiratory-swallow coordination, and cough volume acceleration. Factors that correlated with clinical feeding and swallowing severity in children with SCP included oral praxis and RSP. Additionally, a positive correlation between the DDS and gross motor function was observed, and a negative correlation between PEFR and gross motor function for children with SCP was demonstrated.

## ii. Oropharyngeal Function and Clinical Feeding and Swallowing Severity

Children with SCP demonstrated reduced oral praxis and impaired clinical feeding and swallowing severity compared to controls. In a population characterized by motoric deficits, the present finding of oral motor dysfunction in the SCP group was expected. Oral praxis was significantly correlated to clinical feeding and swallowing severity in children with SCP. This is in agreement with previous literature demonstrating the high prevalence of oral-motor dysfunction in younger children with cerebral palsy and its interference with meals (Wilson & Hustad, 2009; Reilly, Skuse, & Poblete, 1996). The Kaufman Test was originally designed to assess speech praxis, though the first portion of this test (oral praxis) was selected as it

contained tasks that frequently occur during mealtime. Tasks including lingual, labial, and mandibular movement must be completed accurately and precisely in order to have safe and efficient feeding and swallowing. The oral praxis test was also deemed to be the most time efficient as compared to other tests. All children completed the tasks within 5-10 minutes, thereby reducing the possibility of fatigue during task completion.

An in-depth analysis of specific components of oral praxis revealed that children with SCP had greatest difficulty in lingual lateralization/elevation, labial protrusion, and alternating lingual and labial movements. These difficulties are translated to functional skills required by children during meals. The reduced and inaccurate lingual and labial movements seen in this sample are supported by the NGST. The planning deficits and limited range of movement hinder accuracy of production. The observed deficits were well reflected in the task analysis of the DDS. Children showed clear deficits in bolus reception, containment, oral transport, mastication, and the oropharyngeal swallow. All components of the DDS require adequate coordination of movement. This indicates that these children are exhibiting non-productive oral movement patterns. These patterns are likely voluntary attempts to manipulate and transport the bolus. However, there is also a likely sensory level deficit coupled to the oral motor dysfunction which exacerbates the problem during meals and can explain the lack of timely swallows as well as reduced clearance of post-swallow residue from the oral cavity observed in this sample through the DDS.

The majority of children with SCP in this sample were reported to have feeding difficulties early in development requiring diet or environmental modifications. However, only 3 out of the 11 children received feeding therapy, and these were bilaterally impaired (GMFCS IV or V). Children in this sample may have developed adaptive feeding patterns in order to

compensate for the oral praxis deficits. Patterns observed included placing the utensil deeper into the oral cavity while titling the head posteriorly to avoid anterior spillage, positioning solid consistencies on the molar surfaces to assist with chewing, and exhibiting multiple swallows to clear the bolus given inefficient bolus manipulation.

Oral motor dysfunction may also have contributed to increased mealtime duration. Though not a specific outcome in the present study, children with SCP had an average, parent-reported mealtime duration of 37 minutes compared to 22 minutes for the control group. As mealtimes increase, there is a presumed increase in energy expenditure and fatigue. Though children in this sample presently displayed an appropriate body mass index, they may be at risk for reduced growth in the future (Fung et al., 2002).

This is the first study to obtain measures of submental muscle activity during swallowing across bolus consistencies in children with SCP and TDCs. Children with SCP demonstrated greater normalized amplitude values across consistencies, with the puree and solid bolus swallows being significantly greater than controls. Children with cerebral palsy are known to have impaired swallowing manifesting as reduced oral preparation, delayed pharyngeal triggering of the swallow, reduced hyolaryngeal excursion and reduced pharyngeal contraction (Casas et al., 1995; Mirrett et al., 1994).

The increased amplitudes in the SCP group may, in part, be reflective of generalized postural variability (de Graaf-Peters et al., 2007). Sitting with the hips at 90º reduces extensor hypertonicity in children with SCP, while angles of 70º or 120º increase tonicity (Nwaobi, Brubaker, Cusick, & Sussman, 1983). In healthy populations, it has been shown that increased submental and infrahyoid muscle activity at rest corresponds to both neck extension and flexion (Forsberg, Hellsing, Linder-Aronson, & Sheikholeslam, 1985). In the present study, children

with SCP were provided with a base of support allowing for improved stabilization of the feet and back.

A more likely explanation for the increased swallow amplitudes for children with SCP is grounded in neuromechanistic and developmental deficits. These children present with difficulty in motor programming and in modulating volitional motor responses. They also have reduced adaptability to stimuli, which in this case consists of the varying bolus consistencies. The inability to modulate the swallow according to bolus consistency is likely reflected in the heightened amplitudes of submental musculature. Considering the developmental aspect, the amplitude values observed in this sample may also be viewed as a compensatory means of prioritizing safety and efficiency of the swallow. Children with SCP may be exerting additional effort across thicker consistencies in an attempt to improve bolus flow through the oropharynx. Increased amplitudes have been demonstrated in previous literature with respect to masseter and temporal muscle activation (Briesemeister, Schmidt, and Ries (2013). This increased effort may serve to counter balance oral muscular and mandibular dysfunction as well as compromised motor planning abilities (Ries and Berzin, 2005; Rosenbaum et al., 2007). The swallow amplitudes may have been further increased as children with SCP have difficulty inhibiting muscle activity and often present with co-activation of agonist/antagonist musculature (Dietz and Sinkjaer, 2007). Whether or not the increased muscular contraction corresponds to improved swallow physiology in this population remains to be explored.

### iii. Respiratory-Swallow Coordination and Clinical Feeding and Swallowing Severity

Adequate SAD, coinciding with airway closure, is part of the normal temporal cascade of

swallow events which occur in most safe and efficient swallows (Hiss, Treole, & Stuart, 2011). In the present study, between-group differences in SAD were not statistically significance. Rempel & Moussavi (2005) demonstrated prolonged apnea duration in adults with CP at larger bolus volumes (180ml). Certainly, exploration is warranted to determine if children with SCP are unable to regulate the onset and offset of the swallow and to see if SAD coincides with swallow safety for this population.

Pre- and post-swallow exhalation is the predominant pattern observed in healthy adults and is considered to be an effective airway protective behavior (Martin-Harris, Brodsky, Price, Michel, & Walters, 2003; Preiksaitis et al., 1992). The post-swallow exhalation is considered to facilitate the clearance of residue from the airway, thereby decreasing the likelihood and/or severity of penetration/aspiration events (Troche, Huebner, Rosenbek, Okun, & Sapienza, 2011). Children with SCP exhibited significantly abnormal RSPs compared to controls. Specifically, greater than 50% of children with SCP displayed post-swallow inhalation for all consistencies, and greater than 70% of children with SCP displayed this pattern for liquids. This is in agreement with McPherson et al. (1992) who demonstrated that children with CP frequently exhibit a post-swallow inhalation for liquids. Rempel and Moussavi (2005) also observed post-swallow inhalation in adolescents and adults with CP for liquids.

The post-swallow inhalation pattern in children with SCP was also significantly correlated with clinical feeding and swallowing severity. All three children who coughed during the DDS also exhibited a post-swallow inhalation pattern. Post-swallow inhalation of solids was highly correlated with the DDS. The post-swallow exhalation pattern serves as a safety mechanism to expel material that has erringly entered the upper airway. Children with SCP are prone to shallow and aberrant breathing patterns and may not have the required reserve to produce post-

swallow exhalations, particularly in the context of prolonged respiratory cessation (Seddon & Khan., 2003). Additionally, the coordination of breathing and swallowing is likely disrupted due to the impaired ability of children with CP to rapidly adjust their swallow according to bolus type given deficits in oropharyngeal and respiratory function (Calis et al., 2008; Seddon & Khan., 2003). Thicker consistencies require greater manipulation prior to the swallow, and these consistencies were most correlated to clinical feeding and swallowing severity in this sample. These factors may increase the risk of penetration and aspiration.

### iv. Cough Effectiveness and Gross Motor Function

The primary airway protective behavior in response to aspiration is the cough. In the current sample, voluntary sequential coughs were measured as they reflect the ecological validity of the cough motor response without requiring exposure to tussigenic stimuli. It was predicted that children with SCP would have reduced cough effectiveness, and that this reduction would be correlated with clinical feeding and swallowing severity. Children with SCP demonstrated reduced CVA compared to controls for the first cough response in the cough epoch. Cough Volume Acceleration is defined as PEFR/PEFRT. Children with SCP demonstrated lower PEFR and longer PEFRT compared to TDCs, though these differences were not statistically significant. However, the differences were large enough to yield a significant CVA difference. CVA relates to the shearing force potential of the cough to clear the airway (Pitts et al., 2009). Children in the SCP group had a range of impairment in gross motor function and mobility. Impairment in mobility with subsequent activity limitation coincides with deconditioning that can worsen over time (Vaz Fragozo et al., 2014). In addition to deconditioning, spasticity of the respiratory muscles has been shown to impair pulmonary

function (Da Silva et al., 2005). An effective CVA requires adequate strength and coordination of the respiratory musculature. It may be influenced by postural stability (upright vs. forward/recline) and lung volume at initiation of the cough. Children with SCP are known to exhibit variable posture as well as chest wall rigidity that can restrict intake of air during respiration (Seddon & Khan., 2003). The shallow inhalation and discoordination of the respiratory muscles likely contributed to the generation of intrathoracic pressure needed for adequate CVA.

The measures of cough effectiveness were not correlated with clinical feeding and swallowing severity. However, the DDS only contains a single item accounting for post-swallow signs of aspiration. It may not be sensitive enough to capture deficits in cough function. Given that Benfer et al. (2013) demonstrated a stepwise relationship between oropharyngeal dysphagia and gross motor function, and the fact that the GMFCS is a robust classification system, this scale was used as an outcome measure in relation to cough effectiveness measures. PEFRT and CVA were not significantly correlated to the GMFCS in the present sample. PEFRT was similar across level of gross motor function. While children with more impaired gross motor function generally displayed lower CVA than those with milder impairment, it was observed that one child with GMFCS I demonstrated a near identical CVA to a child with GMFCS V. Knowing that CVA is determined by dividing PEFR by PEFRT, the findings suggest that PEFR is the contributing factor to the between-group differences in CVA identified earlier. Knowing that children with SCP have reduced PEFR, it follows that their time to reach the peak flow would not be prolonged.

Significant correlations between PEFR and the GMFCS were found across cough responses, thus documenting the first reported relationship between dystussia and gross motor

function in children with SCP. In this sample, children with GMFCS I had the highest PEFR while children with GFMCS IV had the lowest PEFR. Children with more impaired gross motor function are less likely to be ambulatory, and thus experience deconditioning over time. Furthermore, the abnormal and shallow breathing that is frequently observed in this population may shorten the respiratory muscles and stiffen the costovertebral joints (Ersöz et al., 2006). Therefore, the correlation between PEFR and GMFCS was expected.

PEFR is the maximum flow rate generated during the cough and requires strength of the expiratory muscles and individual effort. Children with SCP have decreased chest mobility, expiratory muscle weakness, reduced pulmonary capacity, and postural abnormalities (Papastamelos, Panitch, & Allen, 1996). Additionally, the present study examined PEFR in the context of voluntary sequential cough which requires the precise coordination between inspiratory, compression, and expiratory phases of the cough. Cerebral palsy is a neuromotor disorder that adversely affects motor performance. Though maximum visual and verbal modeling of the cough was provided to participants in this study, the coordination deficits likely contributed, in part, to the reduced peak flow rates.

Peak expiratory flow rate is known to be lower in individuals with pulmonary dysfunction and is predictive of pulmonary morbidity in individuals with dysphagia (Bianchi, Baiardi, Khirani, & Cantarella, 2012). PEFR is also predictive of penetration/aspiration events in neurologically impaired populations (Pitts et al., 2009; Pitts et al., 2010). In children with SCP, CVA and PEFR are particularly important given the observed deficits oral motor function, respiratory-swallow pattern, and clinical swallow function. All of these deficits would require that an individual has adequate PEFR and CVA to clear the airways from endogenous or aspirated material.

### v. Challenges and Limitations

Though this study successfully acquired data from a sample of children with SCP and controls, certain challenges arose. Specifically, two children were unable to complete the protocol. This may be attributed to age as these children were 4 years old and at the youngest end of the age range for inclusion in the study. Generally, challenges in the younger-aged children included acclimating them to the equipment in a timely manner and ensuring proper comprehension of tasks. This was addressed through the use of props and reinforcers as well as frequent modeling throughout the protocol.

This study was not without limitations. The sample size consisted of 11 children with spastic cerebral palsy and 10 healthy controls. The limited power and non-normal distribution of the data required the use of non-parametric statistical analyses. This limitation may have been a factor in detecting further between-group differences across variables.

This investigation focused on a minimally impaired sample in terms of gross motor, manual, and feeding/swallow function. Three children presented with bilateral SCP and although their gross motor function was worse than the children with unilateral CP, gross motor function did not correlate with respiratory-swallow coordination. However, given the small sample size, it cannot be ruled-out that the performance of the children with BSCP influenced the oropharyngeal function and cough findings. The majority of children in the present sample consisted of independent, self-feeders on unrestricted oral diets. Children self-fed during the assessment of clinical feeding and swallowing severity, but were fed by the investigator during the sEMG-related bolus swallows. This was done to increase standardization of task procedures, though may have disrupted the respiratory-swallow coordination by reducing proprioceptive cues and control over bolus volume and rate of eating (Logemann, 2000). Though clear deficits were

observed across research variables, these deficits are likely understated with respect to more impaired children that clinicians often encounter in medical settings. This study utilized small volumes based on previous pediatric literature (Casas, McPherson, & Kenny, 1995; Leder & Karas, 2000; Morgan, Ward, Murdoch, & Bilbie, 2002). The use of a range of bolus volumes and additional trials across tasks may have allowed for improved observations, specifically as related to respiratory-swallow coordination. However, given that this was the first investigation to objectively explore these relationships, it was deemed best to begin with smaller and safer boluses.

Finally, this investigation did not incorporate an instrumental swallow evaluation. However, the use of videofluoroscopic or endoscopic swallow assessments would be unethical in the present sample (Arvedson, 2008). The participants did not present with a history of respiratory infections or frequent signs of aspiration. Assessment of feeding and swallowing severity was based on a standardized clinical assessment (DDS). The DDS is a standardized tool that has been frequently utilized in prior research to evaluate clinical swallow function in children with CP (Benfer et al., 2013; Calis et al., 2008; Sheppard et al., 2014).

### vi. Conclusions and Future Directions

This prospective investigation successfully tested children with SCP and healthy controls on objective, physiological measures of oropharyngeal function, respiratory-swallow coordination, clinical feeding and swallowing severity, and cough effectiveness. Deficits in oropharyngeal function and respiratory-swallow coordination correlated with clinical feeding and swallowing severity. Therefore, evaluation of these domains must be conducted during the clinical swallow examination. Dysfunctional clinical feeding and swallowing is compounded

by deficits in cough function. This highlights the importance of objectively assessing both dysphagia and dystussia in children with CP to allow for quantification of clinically observed deficits.

Future research in this population should utilize larger samples with greater representation and equal balance across CP severity. Further exploration of electromyographic laterality indices should be conducted to detect possible compensations and adaptations of the swallow. Exploration of cough airflow measures and cough sensitivity in the context of standard reflex-cough testing is also warranted. Finally, for children presenting with overt signs of aspiration and significant medical history, instrumental assessment is required to assess swallow physiology and to determine relationships between clinically observed deficits and swallow safety.

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