Outcomes of an intensive exercise-based swallowing program for persons with Parkinson's disease: A single-case experiment

Jocelyn Jenks
University of Northern Iowa

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OUTCOMES OF AN INTENSIVE EXERCISE-BASED SWALLOWING PROGRAM
FOR PERSONS WITH PARKINSON’S DISEASE: A SINGLE-CASE EXPERIMENT

An Abstract of a Thesis

Submitted

in Partial Fulfillment

of the Requirements for the Degree

Master of Arts

Jocelyn Jenks

University of Northern Iowa

May 2019
ABSTRACT

The leading cause of death in Parkinson’s disease (PD) is aspiration pneumonia, a negative consequence of swallowing impairment. Approximately 80%, and upward of 95%, of persons with Parkinson’s disease (PwPD) will present with progressive dysphagia, or swallowing impairment. Dysphagia may also lead to dehydration, malnutrition, weight loss, reduced quality of life, hospital readmissions, and high financial burden. Recent evidence suggests swallowing exercise is beneficial for PwPD and there is growing evidence that supports intense programs combining exercises, which may provide sufficient treatment intensity to improve swallowing. The present study investigated whether a four-week Intensive exercise-based Swallowing Program (ISP) combining lingual and respiratory exercises for two PwPD would improve outcomes on multiple probe and endpoint measures of swallowing, respiratory, and vocal functions. Specifically, probes included maximum isometric pressures (MIP) of the tongue, maximum expiratory pressure (MEP), maximum phonation time (MPT), and maximum phonation intensity (MPI). Endpoint measurements consisted of the Mann Assessment of Swallowing Ability (MASA), Timed Water Test (TWT), Repetitive Saliva Swallow Test (RSST), and Swallowing Quality of Life Questionnaire (SWAL-QOL). Post-treatment gains occurred in lingual strength, MEP, and MPI ($p < .003$) with moderate-to-strong effect sizes. Both MASA scores and TWT swallowing capacity increased, and RSST performance improved to or was maintained within healthy ranges. Neither MPT, an untrained task, nor SWAL-QOL scores significantly changed. Overall, results suggest a positive, synergistic effect of combined treatment modalities evidenced by gains in both
swallowing and respiratory systems. Future investigation is warranted to further develop efficacious ISPs for PwPD and to determine their effectiveness to ameliorate the negative consequences of dysphagia.
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This Study by: Jocelyn Jenks, B.A.
Entitled: Outcomes of an Intensive Exercise-Based Swallowing Program for Persons with Parkinson’s Disease: A Single-Case Experiment

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CHAPTER 1
INTRODUCTION

Swallowing is a critical, highly complex event that is planned, monitored, and executed by multiple central and peripheral nervous system pathways. It is enacted through over 30 muscles of the head and neck (Dodds, Stewart, & Logemann, 1990). Damage to any part of the neurological and/or anatomical structures of the digestive or respiratory systems may result in disrupted or disordered swallowing, known as dysphagia (Murry, Carrau, & Chan, 2018). The consequences of dysphagia are serious and may include: malnutrition; unintended weight loss; reduced quality of life; and/or an infection from foreign material, such as food and pathogens invading the lower airways, known as aspiration pneumonia (Murry et al., 2018).

Aspiration pneumonia is the leading cause of death for individuals with Parkinson’s disease (PD; Akbar et al., 2015), and is related to progressive swallowing and respiratory function seen over the course of the disease (Kalf, De Swart, Bloem, & Munneke, 2012; Pflug et al., 2018). In some individuals with PD, dysphagia will occur even in early disease stages; however, in the literature it is generally described as a symptom of later disease stages (Pflug et al., 2018). Specifically, persons with PD (PwPD) often exhibit physiological disruptions to oral and pharyngeal swallowing function, which frequently manifests as the deterioration of lingual function and of airway protection (Ali et al., 1996; Argolo, Sampaio, Pinho, Melo, & Nóbrega, 2015; Bushmann, Dobmeyer, Leeker, & Perlmutter, 1989; Ebihara et al., 2003; Ellerston, Heller, Houtz, & Kendall, 2016; Fontana, Pantaleo, Lavorini, Benvenuti, & Gangemi,
1998; Logemann, Boshes, Blonsky, & Fisher, 1975; Miller, Noble, Jones, & Burn, 2006; T. Pitts, Bolser, Rosenbek, Troche, & Sapienza, 2008; L. L. Pitts, Morales, & Stierwalt, 2018; Robbins, Logemann, & Kirshner, 1986; Solomon, Lorell, Robin, Rodnitzky, & Luschei, 1995; Solomon, Robin, & Luschei, 2000). Therefore, the need to establish restorative treatments to ameliorate lingual and respiratory impairment in PD is great; however, the number of such evidence-based treatment programs is few (Troche & Mishra, 2017).

The use of targeted (i.e., task-specific) exercise to address cranial sensorimotor deficits in PD is receiving increased attention in rehabilitation literature (Russell, Ciucci, Connor, & Schallert, 2010). Specific evidence that exercise may improve swallowing function for PwPD was noted by researchers of the Lee Silverman Voice Treatment (LSVT ®) program, an intensive voice treatment that has also shown positive carryover effects to swallowing (El Sharkawi et al., 2002; Miles et al., 2017). More recently, both strengthening and skill training exercise regimens have also yielded improvements in various clinical and instrumental measures of swallowing and patient-reported quality of life (Athukorala, Jones, Sella, & Huckabee, 2014; Miles et al., 2017; L. L. Pitts et al., 2014; T. Pitts et al., 2009; Troche et al., 2010). In particular, the growing evidence of therapeutic benefits from both strengthening and skill training of swallowing and expiratory muscles, may warrant investigation as a combined exercise program.

Evidence for Intensive exercise-based Swallowing Programs (ISPs) that combine exercise protocols to treat neurogenic dysphagia is emerging (Malandraki et al., 2016). Despite evidence that swallowing function in PD is responsive to intensive programs of a
single exercise, the literature lacks applications of ISPs combining exercise modalities specifically for PD. Therefore, it is both timely and necessary to develop and test optimal exercise combinations within intensive treatment programs to effectively manage dysphagia in PD and thus, reduce its health-related consequences.
CHAPTER 2
LITERATURE REVIEW

Healthy Swallowing

Swallowing is the process of receiving, processing, and moving food or liquid from the oral cavity to the stomach and while it may seem effortless for healthy adults, in reality it may be the most complex neurological reflex (Doty, 1951; Steele & Miller, 2010). This highly coordinated neurologic and physiologic pattern ensures efficient transportation of food and liquid without permitting ingested material to enter the airway and travel below the level of the vocal folds (i.e., aspiration; Jean, 2001; Ludlow, 2005). The complexity of this life-sustaining function is rooted in the necessary integration of: many neurological networks of volitional and reflexive control, sensory processing, salivation, and of digestive and respiratory systems to ensure successful alimentation (Dodds et al., 1990; Zald & Pardo, 1999).

Specifically, the neurological pattern for a swallow must be skillfully adapted to the specific texture, consistency, and size of the food or liquid to be ingested (Steele & Miller, 2010). That pattern must then be rapidly executed through 31 pairs of striated muscles upheld by various bony and cartilaginous supports (Dodds et al., 1990). Despite the intricate and overlapping sensorimotor functions within one swallow, the process of swallowing liquids is generally simplified into four phases: oral preparatory, oral transit, pharyngeal, and esophageal (Dodds et al., 1990). Slight adaptation of the above phases has been described for solid consistencies within the Process Model of Feeding (Palmer, Rudin, Lara, & Crompton, 1992). The Process Model of Feeding, to be further described
within the four-phase model, includes stage I transport, food processing, stage II transport, pharyngeal, and esophageal stages (Hiemae & Palmer, 1999; Matsuo & Palmer, 2008; Palmer et al., 1992).

During the oral preparatory phase (i.e., stage I transport and food processing for solids), food or liquid enters the oral cavity and is manipulated (Matsuo & Palmer, 2008). This phase of swallowing is predominantly voluntary and involves the formation of the bolus, which is the collective unit of ingested food or liquid combined with saliva (Jean, 2001). The bolus is prepared in the oral cavity as it is masticated and/or mixed with saliva (Sasegbon & Hamdy, 2017). Mastication is directly supported by the teeth and indirectly by the tongue and muscles of mastication (Matsuo & Palmer, 2008; Sasegbon & Hamdy, 2017). Muscles of mastication contract and relax to perform this function, and are innervated by the mandibular portion of cranial nerve (CN) V, trigeminal (Jean, 2001; Moore & Dalley, 2006). These muscles include the temporalis, masseter, and lateral and medial pterygoids (Moore & Dalley, 2006). The temporalis, masseter, and medial pterygoids close the jaw; the lateral pterygoids depress the chin through bilateral contraction and move the jaw contralaterally by unilateral contraction (Moore & Dalley, 2006). The mandible and temporal bone connect via the temporomandibular joint and allow the opening and closing of the jaw during mastication (Moore & Dalley, 2006).

To contain the bolus within the oral cavity during mastication, the lips are closed by contraction of muscles innervated by CN VII, facial (Moore & Dalley, 2006). These muscles include the orbicularis oris, buccinator, risorius, and the elevators and depressors of the lips (i.e., levator labii superioris, levator anguli oris, levator labii superioris,
alaeque nasi, zygomatics major and minor, depressor anguli oris, depressor labii inferioris, mentalis, and platysma; Moore & Dalley, 2006). Bolus formation is also assisted by sensory information from the oral cavity and tongue, with specific input from CNs V, VII, and IX (Jean, 2001; Moore & Dalley, 2006; Steele & Miller, 2010). The bolus is shaped by the tongue, an exceptionally flexible muscular organ, and consists of intrinsic and extrinsic muscles innervated primarily by CN XII, hypoglossal, with one muscle (palatoglossus) innervated by CN X, vagus (Jean, 2001; Moore & Dalley, 2006). Three glands (i.e., parotid, submandibular, and sublingual) produce saliva to soften and dissolve food and to assist in the formation of a cohesive unit of food to be swallowed, known as a bolus (Amano, Mizobe, Bando, & Sakiyama, 2012; Pedersen, Bardow, Jensen, & Nauntofte, 2002; Sasegbon & Hamdy, 2017).

Once the bolus has been adequately formed, it is transported toward the pharynx within the oral transit phase, also referred to as Stage II transport for solid foods. This phase, which is also largely under volitional control, begins as the tip of the tongue contacts the anterior alveolar ridge and continues to compress against the hard palate from anterior to posterior in a peristaltic-like wave within the oral cavity. This peristaltic motion pushes the bolus back toward the pharynx, as the posterior tongue depresses to allow the bolus to enter the pharynx. The velum also plays an important role in this phase, as it elevates to close the nasopharynx in conjunction with the superior pharyngeal constrictor and prevents the bolus from entering the nasal cavity (Matsuo & Palmer, 2008).
As the bolus enters the pharynx, the initiation of the pharyngeal phase must be well-timed and is marked by beginning of the swallowing reflex (Jean, 2001). The swallowing reflex, an involuntary and irreversible event, consists of muscle contraction of the pharynx and larynx as the spaces reconfigure to allow cessation of respiration and protection of the airway (Leonard & Kendall, 2014). The swallowing reflex begins with contraction of the mylohyoid which initiates a sequence of muscle contraction and relaxation known altogether as the leading complex that contributes to successful bolus transportation to and through the upper esophageal sphincter (UES; Jean, 2001; Leonard & Kendall, 2014). The leading complex muscles include the: mylohyoid, anterior digastric, geniohyoid, stylohyoid, thyrohyoid, styloglossus, posterior portion of the tongue, superior/middle/inferior constrictors, palatoglossus, and palatopharyngeus (Jean, 2001; Pearson, Langmore, Yu, & Zumwalt, 2012). Initial contraction of these submental, supralaryngeal, and thyroid muscles results in superior and anterior movement of the hyoid bone and larynx, known as hyolaryngeal excursion and precedes a temporary closure of the larynx and pause in respiration (Leonard & Kendall, 2014).

Swallowing and breathing must be highly coordinated during the swallowing reflex as the pharynx and larynx are involved in not only swallowing, but also respiration (Leonard & Kendall, 2014). During the oral phases of swallowing prior to the initiation of the pharyngeal phase, breathing may continue while food collects in the pharynx (Matsuo & Palmer, 2009); however, during the pharyngeal phase breathing temporarily ceases. This brief period of apnea is caused by not only upper airway closure but also inhibitory signals sent from neural control centers in the brainstem, including
neurological structures also involved in swallowing, such as CNs IX and X (Matsuo & Palmer, 2009). Upper airway, or laryngeal vestibule closure, consists of three levels of protection (Sasaki & Isaacson, 1988). These levels include: true and ventricular vocal folds and the laryngeal additus, closure of which is completed by epiglottal inversion and aryepiglottic fold closure and is driven by thyroarytenoid contraction (Doty & Bosma, 1956; Kidder, 1995; Sasaki & Isaacson, 1988).

The respiratory pause during swallowing may be longer than the actual swallow itself and requires adequate subglottal pressurization to be maintained (Matsuo & Palmer, 2009). Temporal coordination of mastication, breathing, and swallowing is critical to ensure nutrition and to prevent pulmonary aspiration (Matsuo & Palmer, 2009). Structures critical to respiration during swallowing include the tongue, velum, pharyngeal wall, and chest wall kinematics (i.e., rib cage and abdominal contraction and relaxation), all essential to keeping the pharyngeal airway open to ensure proper ventilation and gas exchange (Matsuo & Palmer, 2009; McFarland et al., 2016). A typical respiratory pattern during swallowing is known as an “exhale, swallow, exhale” pattern, common for 67-79% of adults, as noted using inductive plethysmography and/or nasal manometry (Matsuo & Palmer, 2009).

During the pharyngeal phase of swallowing, various pharyngeal muscles contract and relax for successful propulsion of the bolus. Primary pharyngeal muscles that are active during this phase include those within the anatomical triangles of the neck, pharyngeal constrictors, and inner longitudinal muscles (i.e., stylopharyngeus, palatopharyngeus, and salpingopharyngeus). In addition, the nasal cavity must also seal
off completely to contain the bolus in the pharynx during the pharyngeal phase. To do so, the soft palate (velum) elevates and tenses against the posterior pharyngeal wall to prevent nasopharyngeal reflux; this is enacted by the tensor veli palatini, levator veli palatini, and musculus uvulae (Matsuo & Palmer, 2008; Moore & Dalley, 2006).

Together, these muscles alter the shape of the pharynx and generate necessary positive and negative pressures for safe and efficient bolus transport (Leonard & Kendall, 2014; Pearson et al., 2012).

The contraction of these various pharyngeal and laryngeal muscles are coordinated during the pharyngeal phase to generative propulsive and negative pressures to force the bolus toward and into the esophagus (Leonard & Kendall, 2014). The superior, middle, and inferior pharyngeal constrictors contract and push the bolus downward toward the UES; meanwhile, the base of tongue retracts and meets the posterior pharyngeal wall (Leonard & Kendall, 2014). Contraction of the pharyngeal constrictors is important for a successful swallow; however, manometric studies indicate the tongue’s driving pressure and the negative pressure of the UES relaxing and opening are particularly critical to successful deglutition (McConnel, 1988a; McConnel, 1988b).

**Neurological Control of Swallowing**

The phases of swallowing are carefully planned, executed, and monitored by the central nervous system (CNS) and peripheral nervous system (PNS). Peripheral neural control of swallowing heavily relies on cranial and peripheral nerves (Sasegbon & Hamdy, 2017). Cranial nerves (CNs) are predominantly mixed motor and sensory, however, a select few are sensory only (e.g., CN I, olfactory) or motor only (e.g., CN XI,
spinal accessory or CN XII, hypoglossal; Sasegbon & Hamdy, 2017). CNS structures actively involved in swallowing are both cortical and subcortical and include: portions of the precentral and postcentral gyri, midbrain, pontomedullary brainstem, basal ganglia, thalamus, cerebellum, insula, and portions of the right temporal lobe (Dodds et al., 1990; Hamdy et al., 1997; Zald & Pardo, 1999). Overall, nervous system control of swallowing is grossly divided into three subsystems: the afferent (sensory) system, the efferent (motor) system, and the interneurons.

The first of these systems, the afferent system, initiates the pharyngeal phase and modulates swallowing physiology by providing feedback to the neural control centers for swallowing (Dodds et al., 1990; Steele & Miller, 2010). Feedback during swallowing includes the relay of sensory information regarding pressure via mechanoreceptors and temperature via thermoreceptors of the aerodigestive tract. Additional special sensory information, such as taste and smell, is gathered and relayed via chemoreceptors (Steele & Miller, 2010). Ascending afferent fibers provide feedback loops to the brainstem, esophageal ganglia, thalami, subcortical, and cortical networks (Steele & Miller, 2010).

The efferent system consists of motor nerve fibers. Bundles of efferent nerves compose the upper motor neurons (UMNs) and the lower motor neurons (LMNs) for each cranial nerve and spinal nerve. Cranial nerves may receive input from bilateral UMNs depending on the muscles they innervate. The cell bodies of the UMNs originate in the primary motor cortex and synapse with the cell bodies of the LMNs within the brainstem/spinal cord. The LMNs exit the brainstem or spinal cord and synapse directly on the muscles. Therefore, an electrochemical signal sent from the primary motor cortex
through an UMN travels to the brainstem, down the LMN, and finally synapses on the muscle, instructing the muscle to contract (Dodds et al., 1990). The efferent system is integrated with the afferent system, both of which are critical for the successful execution of planned swallowing events.

To achieve integration of the efferent and afferent systems, interneurons form dense connections within swallowing networks to plan, communicate, monitor, and execute a swallow (Sasegbon & Hamdy, 2017). One group of interneurons that are critical for successful deglutition are the central pattern generator (CPG) of swallowing (Hamdy et al., 1999; Jean, 2001). The CPG consists of interneurons located bilaterally within the pontomedullary brainstem and permit precise communication between components of the CPG. Two components of the CPG include: the ventral swallowing group (VSG), consisting of motor nuclei in the brainstem, and the dorsal swallowing group (DSG), consisting of sensory nuclei in the brainstem (Ludlow, 2005). The VSG includes the nucleus ambiguus and the DSG the nucleus tractus solitarius (NTS; Hamdy et al., 1999; Jean, Kessler, & Tell, 1994; Jean, 2001). These nuclei are associated with CNs IX, X, and XI (Ludlow, 2005). The CPG of swallowing allows for successful communication and integration of swallowing with other vital subsystems, such as respiration and cough in healthy adults.

**Dysphagia**

Disruption to healthy swallowing that results in difficulty in either the oral and/or pharyngeal phases of swallowing is known as oropharyngeal dysphagia (Hamdy et al., 1997; Sasegbon & Hamdy, 2017). One of the primary functions of normal swallowing is
the adequate protection of the airway and prevention of foreign materials (e.g., food, liquid, saliva, or pathogens) from entering the lower airway. When material enters the larynx, it is clinically classified as either penetration or aspiration. Penetration occurs when material enters the larynx above the level of the vocal folds; aspiration involves material entering the larynx past the vocal folds, and subsequently into the lower airways (Rosenbek, Robbins, Roecker, Coyle, & Woods, 1996). Aspiration may be detected through overt clinical signs, such as coughing and throat clearing, or may not be apparent in an outward response, commonly referred to as silent aspiration (Linden & Siebens, 1983). Penetration and aspiration are two of the most commonly discussed negative outcomes of oropharyngeal dysphagia. Although oropharyngeal dysphagia may be a symptom of direct damage to the structures of the head and neck, its etiology is most frequently neurogenic. Neurogenic dysphagia may result from various disorders including: cerebrovascular accidents, cerebral palsy, myasthenia gravis, multiple sclerosis, or Parkinson’s disease (PD; Zald & Pardo, 1999).

Dysphagia in Persons with Parkinson’s Disease

PD is one of the most common neurodegenerative diseases and is anticipated to increase by 56% from 2005 to 2040 to reach a projected total of 770,000 PwPD in the United States (Rossi et al., 2017). This growing population is of considerable interest for the field of rehabilitation since PwPD are highly likely to experience dysphagia and subsequent aspiration pneumonia, the leading cause of death in PD (Akbar et al., 2015; Pflug et al., 2018). PwPD who also present with dysphagia are at increased risk for malnutrition, fluid imbalances, weight loss, and reduced quality of life; all of which may
contribute to increasing burden on the individual and healthcare system (Akbar et al., 2015; Barichella, Cereda, & Pezzoli, 2009; Leow, Huckabee, Anderson, & Beckert, 2010; T. Pitts et al., 2008; Plowman-Prine et al., 2009).

Current literature estimates that more than 80-95% of PwPD will present with dysphagia at some point of the disease (Pflug et al., 2018); however, Kalf and colleagues (2012) suggest the actual prevalence of dysphagia is higher in PwPD due to underreporting and/or lack of awareness of impairment. Complaints regarding swallowing difficulties by PwPD poorly correlate with current gold standard evaluations of swallowing (i.e., videofluoroscopic (VFSS) and fiberoptic endoscopic evaluation of swallowing (FEES). In general, PwPD may be either unaware of or unable to identify specific swallowing deficits, which may contribute to the trend that PD-related dysphagia is often not diagnosed until later disease stages (Bushman et al., 1989; Pflug et al., 2018).

The identification of swallowing dysfunction in PwPD does not clearly correlate with disease duration or severity. Swallowing disruptions have been documented across all stages of PD, even in the earliest disease stages and clinically asymptomatic cases (Ali et al., 1996; Jones & Ciucci, 2016; Miller et al., 2006; Pflug et al., 2018). PwPD, regardless of a confirmed diagnosis of dysphagia, often exhibit physiological disruptions of the oral and pharyngeal swallowing phases (Ali et al., 1996; Jones & Ciucci, 2016; Miller et al., 2006; Pflug et al., 2018).

Physiological deficits in swallowing secondary to PD result in a variety of aberrant behaviors. Deficits in the oral preparatory and oral transit phases of swallowing
in PwPD may include pathological lingual movements (i.e., lingual pumping and lingual tremor; Ali et al., 1996; Argolo et al., 2015). Lingual pumping is a characteristic deficit of PD, found in approximately 75% of patients (Argolo et al., 2015). Specifically, lingual pumping is an involuntary, repetitive, anteroposterior movement of the tongue on the palate executed prior to the transference of the bolus to the pharynx (Argolo et al., 2015). This phenomenon may be associated with lingual rigidity and bradykinesia, but its underlying pathophysiology remains unclear (Argolo et al., 2015). Additional oral phase disturbances may also include delayed onset or weak lingual propulsion of the bolus into the pharynx, delayed velar elevation, repetitive swallows that move only small portions of the bolus to the pharynx with each attempt (i.e., piecemeal deglutition), premature spillage of the bolus into the pharynx, inadequate bolus formation, and/or residue in the oral cavity (Ali et al., 1996; Bushmann et al., 1989; Fukuoka et al., 2018; Minagi et al., 2018). Poor bolus formation and residue in the oral cavity may be attributed to fewer and slower movements of the tongue secondary to bradykinesia (Argolo et al., 2015; Fukuoka et al., 2018). Overall, lingual dysfunction has long been recognized as a characteristic of PD-related dysphagia (Logemann et al., 1975).

In addition to disordered lingual movement and timing, prior research suggests that lingual strength or endurance may also be affected in PD (O’Day, Montgomery, Nichols, & McDade, 2005; L. L. Pitts et al., 2018; Solomon et al., 1995, 2000). The combined data of Solomon and colleagues (1995; 2000) indicated that both lingual strength and lingual endurance are impaired in PwPD in comparison to age- and gender-matched controls. On average, lingual strength in PD is 8.3 kilopascals (kPa) lower and
endurance is 8.2 seconds shorter than healthy controls (Solomon et al., 1995, 2000). L. L. Pitts and colleagues (2018) corroborate lingual strength is significantly reduced in PwPD and suggest lingual weakness is greater in PwPD and symptomatic dysphagia as compared to healthy controls and PwPD with no complaints of dysphagia. Therefore, lingual strength or lingual pressure generation may serve as an early diagnostic tool and/or viable therapy target for rehabilitation of swallowing in PwPD.

The pharyngeal phase of swallowing may also be impaired in Parkinson’s disease. Recent videofluoroscopic results from 34 PwPD indicated that one of the most common deficits in this phase is delayed airway closure (62% of PwPD; Ellerston et al., 2016). Other pharyngeal phase deficits can include reduced pharyngeal constriction (Ellerston et al., 2016), delayed swallowing reflex (Logemann et al., 1975; Robbins et al., 1986), decreased hyolaryngeal excursion, residue in the valleculae and/or piriform sinuses, reflux from the valleculae and piriform sinuses into the oral cavity (Bushman et al., 1989), and/or airway invasion (i.e., laryngeal penetration and/or aspiration; Argolo et al., 2015; Miller et al., 2006; T. Pitts et al., 2008). These deficits may be due to either motor or sensory disruption or both; specifically, pharyngeal muscles in PwPD have shown increased atrophy, decrease in fast type II muscles fibers and increase in slow type I fibers, and reduced somatosensory function exhibited by the need to reach a higher sensory threshold to trigger the swallow reflex when compared to healthy controls (Mu et al., 2012, 2013; Hammer, Murphy, & Abrams, 2013). Although penetration and aspiration are most commonly associated with deficits in the pharyngeal phase,
Bushmann and colleagues (1989) reported that aspiration can occur during any phase of swallowing in PwPD.

When penetration or aspiration occurs, a health respiratory system generates a strong reflexive cough to clear the airway through pressurized expiratory airflow (Bolser & Davenport, 2002; Fontana & Lavorini, 2006; Smith Hammond et al., 2001). However, the respiratory system may be compromised by PD (Ebihara et al., 2003; Fontana et al., 1998). Respiratory mechanics and pulmonary function may be negatively impacted in PwPD by rigidity and bradykinesia, which may impair the tone, contractility, and coordination of thoracic musculature (Sabaté, Rodríguez, Méndez, Enríquez, & González, 1996). PwPD have shown decreases in forced vital capacity, forced expiratory volume, maximum inspiratory pressure (MIP), and maximum expiratory pressure (MEP), and cough functions (Hegland, Troche, & Davenport, 2013; Sabaté et al., 1996; Silverman et al., 2006). Both reflexive and voluntary cough functions are reduced in PwPD compared to healthy controls (Ebihara et al., 2003; Fontana et al., 1998). Deficits in cough production (i.e., the inability to generate sufficient expiratory airflow) increase the risk for aspiration pneumonia (Bach, 1993; Fontana & Lavorini, 2006). Since aspiration pneumonia is the leading cause of death in PwPD (Akbar et al., 2015), research that aims to improve oropharyngeal and respiratory functions in PD are critical.

**Dysphagia Rehabilitation for PwPD**

The management of dysphagia has two general foci: compensatory strategies (e.g., chin tuck, modified diet and thickened liquids, or postural changes), all of which may improve swallowing safety while the technique is applied; or restorative
interventions, which aim to maintain or improve swallowing function long after the exercises have concluded (Baker, 2012). An expanding body of literature regarding restorative interventions incorporates the principles of exercise/sports training and the principles of neuroplasticity to develop exercise-based dysphagia interventions (Kleim & Jones, 2008; Morgan, 2017). Historically, it was widely accepted that dysphagia in neurodegenerative diseases would not respond positively to exercise; however, a recent meta-analysis suggests swallowing exercises demonstrate promise to rehabilitate swallowing for these populations (Troche & Mishra, 2017). To date, the application of restorative, swallowing exercise to PwPD has been limited (Troche & Mishra, 2017). Due to the paucity of literature regarding exercise-based dysphagia rehabilitation for PwPD, this area lacks a set of established best care practices (Troche & Mishra, 2017).

Exercise-based rehabilitation for dysphagia may treat the deconditioning of muscles in neurodegenerative disorders to improve strength and efficiency of swallowing musculature. Swallowing musculature primarily consists of type II muscle fibers, such as those found in the anterior tongue, which are better suited for quick, forceful movements necessary for safe and efficient bolus propulsion and transportation. Type II muscle fibers may be most susceptible to deconditioning, which may occur due to disease, injury, disuse, or sarcopenia. Deconditioning of the muscles involved in swallowing results in weaker, less precise, and slower movements as well as muscle atrophy and decreased number of motor units and reduced efficiency. Muscle deconditioning or weakness may be common secondary to neurodegenerative disorders such as PD (Ali et al., 1996; Argolo et al., 2015; Ellerston et al., 2016; O’Day et al., 2005; L. L. Pitts et al., 2018;
Solomon et al., 1995, 2000). Despite previous belief that persons with neurodegenerative disease would not positively respond to restorative exercise-based dysphagia interventions, a recent meta-analysis concluded that sufficiently specific and intense exercise regimens can result in improved swallowing function for these populations (Morgan, 2017).

Since weakness of the aerodigestive tract has been noted in PD (Ali et al., 1996; Argolo et al., 2015; Ebihara et al., 2003; Fontana et al., 1998; O’Day et al., 2005; L. L. Pitts et al., 2018; Solomon et al., 1995, 2000) and preliminary evidence suggests such weakness may be responsive to exercise (Morgan, 2017; Russell et al., 2010; Troche & Mishra, 2017), intensive exercise training programs may be of value to address dysphagia in PD. Nevertheless, exercise targeting swallowing impairments in PwPD is an understudied area with few developed programs targeting PD-related cranial sensorimotor deficits (Russell et al., 2010; Troche & Mishra, 2017). Targeted exercise is specific to the group of muscles activated for a certain task, and must closely represent the desired movement (Athukorala et al., 2014; Russell et al., 2010; Steele et al., 2013). Theoretically, when the therapy closely resembles the desired behavioral outcomes, there is a greater chance of successful acquisition and improvement of the direct behavior (Russell et al., 2010); however, some indirect exercise may also produce generalized effects.

Lee Silverman Voice Treatment (LSVT®). One of the first exercise-based training regimens to improve aerodigestive functions for PwPD, the Lee Silverman Voice Treatment (LSVT®), has demonstrated efficacy in improving measures of voice,
swallowing, and cough production (Countryman, Ramig, & Pawlas, 1994; Dromey, Ramig, & Johnson, 1995; El Sharkawi et al., 2002; Mead, Ramig, & Beck, 1989; Miles et al., 2017; Ramig, Countryman, O’Brien, Hoehn, & Thompson, 1996). Reduced vocal intensity is an early sign of PD and may reduce the ability of PwPD to produce intelligible speech (Darley, Aronson, & Brown, 1969; Logemann, Fisher, Boshes, & Blonsky, 1978; Streifler & Hofman, 1984). Therefore, LSVT® trains participants to use high effort during voicing tasks to alleviate the effect of hypokinesia on the respiratory and phonatory systems (Hallett & Khosbin, 1980; Ramig, 1995).

The LVST® treatment protocol is intensive and consists of 16 sessions that last 50-60 minutes, with four sessions being conducted per week for four weeks total (El Sharkawi et al., 2002; Ramig, 1995). Three tasks are conducted during each session, including maximum duration of sustained vowel phonation, maximum fundamental frequency range, and maximum functional speech loudness drills (El Sharkawi et al., 2002). The overall focus of LSVT® is to challenge participants to “think loud” (El Sharkawi et al., 2002; Ramig, 1995). Additionally, participants of LSVT® are given daily exercise homework to encourage generalization of practice to daily activities (El Sharkawi et al., 2002).

Although LSVT® has predominantly shown efficacy as a treatment for voice in PwPD, El Sharkawi and colleagues (2002) conducted a seminal investigation of LSVT® as a treatment for both voice and swallowing deficits. Researchers reported that LSVT® may be an effective treatment for upper aerodigestive tract dysfunction. Specific swallow-related improvements included decreased incidence of swallowing motility
disorders (e.g., reduced tongue coordination and lateralization, delayed laryngeal vestibule closure, and reduced tongue base retraction) and some temporal swallowing measures reduced significantly (e.g., oral transit time). Improvements in both lingual function and decreased oral transit time were hypothesized to occur secondary to greater initiation of neuromuscular control of the entire aerodigestive tract following training (El Sharkawi et al., 2002).

A follow-up investigation conducted by Miles and colleagues (2017) corroborated the efficacy of LSVT LOUD® as a treatment for both voice and swallowing in PwPD. Participants included 20 PwPD with and without dysphagia who participated in the LSVT LOUD® protocol (i.e., 16 sessions across 4 weeks plus homework). Participants were assessed at pre-treatment and both 1 week and 6 months post-treatment. Outcome measures included patient-reported swallowing-related quality of life, voice measures, instrumental swallowing assessments (i.e., videofluroscopy), and reflexive cough strength. Participants made significant gains in maximum phonation time (MPT) and in average sound pressure level (SPL) during reading and conversation; both increases were maintained at 6 months post-treatment. Additionally, pharyngeal residue and pharyngeal area at rest were reduced, while both maximal opening and duration of opening of the pharyngoesophageal segment (PES) increased at 1 week and 6 months post-treatment. No aspiration was noted at pre- or post-treatment on videofluoroscopy. Furthermore, significant improvements occurred for peak expiratory flow rate and peak expiratory flow rise time during involuntary cough at 1 week post-treatment, which was maintained at 6 months. Researchers interpreted their findings to indicate LSVT LOUD® may cause
indirect, spread effects to both swallowing and cough function in addition to phonatory gains (Miles et al., 2017).

**Expiratory Muscle Strength Training (EMST).** Researchers introduced another exercise-based therapy for PwPD that included intensive respiratory muscle strength training protocol known as Expiratory Muscle Strength Training (EMST; Saleem, Sapienza, & Okun, 2005). EMST was originally investigated for musicians (Sapienza, Davenport, & Martin, 2002) and was designed as a 4-week treatment of a single respiratory exercise. One exercise, or trial, in EMST consists of the patient inhaling deeply and exhaling forcefully through a handheld device until air rushes out of the device. This device, known as the *EMST150™*, was designed to provide adjustable resistance as goals are recalibrated following increases in respiratory function. The *EMST150™* is also used to measure a patient’s maximum expiratory pressure (MEP) in order to set the level of resistance for training (i.e., trials are set at 75% of MEP). The treatment schedule is typically implemented 5 days a week with 5 sets of 5 repetitions of exercises being conducted per day (Saleem, 2005; Saleem et al., 2005).

Saleem and colleagues (2005) were amongst the principal researchers to implement EMST for PD within a single case experiment (Saleem et al., 2005). One patient with idiopathic PD participated in EMST for 20 weeks for the standard protocol of 5 days a week, 5 sets of 5 exercises a day. The participant was trained to identify a successful completion of the exercise, which was complete when the participant produced enough expiratory strength to hear a rush of air through the device. Researchers did not consistently monitor the participant as she took the device home and kept track of her
compliance on a log sheet. Maximum expiratory pressure (MEP) was the only outcome measure, taken at both pre- and post-treatment. Two baseline evaluations were conducted, separated by one week. The participant’s compliance was monitored weekly on the day she met with researchers to measure MEP and recalibrate the device. Upon completion of the program, researchers reported a 158% increase in the patient’s MEP from baseline to post-testing, which decreased by 16% after a 4-week latent period. Therefore, researchers concluded that the individual’s preliminary improvements in measures of respiration warranted further investigation of EMST applications for PwPD (Saleem et al., 2005).

In addition to improvements in MEP, T. Pitts and colleagues (2009) hypothesized that EMST may also improve swallowing outcomes for PwPD. Therefore, researchers applied videofluoroscopy as an outcome measure for a 4-week study assessing the efficacy of EMST in improving cough and/or swallow function in 10 PwPD who demonstrated airway invasion on videofluoroscopy at baseline. The participants were given an EMST150 device to take home and were instructed to perform 5 sets of 5 exercises per day for 5 days a week. The devices were set at 75% of participants’ MEP, taken at baseline. Participants returned 1-week post-treatment for evaluation. At that time, a significant increase in cough volume acceleration as well as a significant decrease in penetration/aspiration scores were observed on videofluoroscopy. These results support EMST as a feasible and efficacious treatment modality for PwPD at risk for aspiration (T. Pitts et al., 2009).
Subsequently, Troche and colleagues (2010) built upon the literature by conducting a randomized, blinded, sham-controlled 4-week EMST trial with 60 participants, half of which were in a control group and half of which were in a treatment group. Consistent with previous research, participants were instructed to perform home practice of 5 sets of 5 exercises at 75% of participants’ MEP for 5 days across the 4 weeks. Compliance was monitored using performance charts completed by the participants. Clinicians visited the participants weekly to re-instruct procedure for exercises, including how to identify successful completion of an exercise. Overall, the treatment group performed superiorly in functional and physiologic measures of swallowing. Both mean penetration/aspiration scores and hyoid displacement measures improved for the treatment group but not the sham group. Researchers concluded that EMST is a relatively straightforward and cost-effective therapy for reducing airway invasion during swallowing in PD (Troche et al., 2010).

Skill Training and Strengthening of Swallowing Structures. While EMST and LSVT® were implemented as rehabilitative treatments indirectly targeting swallowing structures (i.e., through vocal intensity exercises/strengthening of respiratory muscles), researchers recently began to address the lack swallowing-specific exercise programs for PwPD (Athukorala et al., 2014; L. L. Pitts et al., 2014). Few studies have systematically applied strengthening and/or skill training of the swallow across multiple weeks of intervention in this population (Athukorala et al., 2014; L. L. Pitts et al., 2014). Recent investigations have piloted a computer-based intervention targeting submental muscle
contraction during swallowing (Athukorala et al., 2014) or Lingual Strengthening and Skill Training (LSST) of swallowing pressures for PwPD (L. L. Pitts et al., 2014).

Athukorala and colleagues (2014) expanded on the concept of skill training for swallowing rehabilitation in PwPD by implementing a novel training to increase the precision of swallowing using visual biofeedback from surface electromyography (sEMG) signals, Biofeedback in Swallowing Skill Training (BiSSkiT). Outcome measures included: the Timed Water swallowing Test (TWT; Hughes & Wiles, 1996), sEMG signals (taken during dry swallows and 10-mL water swallows), the Test of Mastication and Swallowing Solids (ToMASS), and the Swallowing Quality of Life Questionnaire (SWAL-QOL). Three aspects of the Timed Water swallowing Test were used as outcome measures: time per swallow, volume per swallow, and swallowing capacity (i.e., volume per second). Outcomes were compared between two baseline sessions and at two sessions post-treatment (i.e., following 10, 1-hour treatment sessions completed within a two-week period), with each evaluation session two weeks apart. The treatment protocol incorporated varying targets of force and duration of submental muscle activity to imitate changing motor patterns while eating necessary to meet the demand of varying textures, volumes, and tastes. Biofeedback was provided on a computer monitor immediately following each swallow, and proficiency at a specific level was required to advance to the next level. Overall, 100 swallow trials were conducted during each hour session, consisting of 5 blocks of 20 swallow trials separated by a short break between blocks. Results indicated time per swallow and volume per swallow increased within the timed water swallow test whereas swallowing capacity did
not. Improvements were also noted in both premotor time and preswallow time on sEMG for both saliva and 10-mL water swallows. Participants’ SWAL-QOL scores from pre- to post-treatment also improved. Thus, researchers suggested the intensive nature of the swallowing training and the skill-based swallowing tasks, rather than gross strengthening alone, may have induced positive changes in swallowing movement patterns and improved the quality of life for PwPD (Athukorala et al., 2014).

Despite growing evidence that both lingual strength and endurance are impaired in PwPD (Solomon et al., 1995, 2000; L. L. Pitts et al., 2018) and demonstrated benefit of lingual strengthening in neurogenic and elderly populations (McKenna, Zhang, Haines, & Kelchner, 2017; Rogus-Pulia et al., 2016), swallowing programs incorporating tongue exercises have only recently been applied to PwPD and dysphagia. Isometric lingual strength training consists of isometric exercise, which involves sustained contractions against a set resistance, with the goal being to improve overall lingual strength, which is thought to directly relate to lingual-palatal pressure generation and bolus propulsion during swallowing (Kays & Robbins, 2006; McKenna et al., 2017; Robbins et al., 2005, 2007; Yeates, Molfenter, & Steele, 2008). However, isometric exercise was first developed to improve limb (i.e., skeletal) muscle strength, which differs from lingual muscle in terms of type, density, and organization of muscle fibers (Kays & Robbins, 2006; Miller, Watkin, & Chen, 2002; Stål, Marklund, Thornell, De Paul, & Eriksson, 2003). McKenna and colleagues (2017) conducted a meta-analysis of ten studies, which concluded lingual muscles may be strengthened in the same manner. Each study implemented lingual strength training tasks; however, only some measured pressures
generated by the tongue during swallowing (a skilled performance). In comparison to strength training for swallowing muscles, skilled training is a more recent area of interest in PD-related dysphagia rehabilitation (McKenna et al., 2017).

Considering that there is research support for both strengthening (i.e., lingual weakness beyond natural aging in PD) and skill training benefits in persons with neurogenic dysphagia, applications of combined strengthening and skill training may be of interest in the management of PD. L. L. Pitts and colleagues (2014) recently completed a pilot study of Lingual Strengthening and Skill Training (LSST) using the Iowa Oral Performance Instrument (IOPI) with five PwPD. The LSST program incorporated both skill-based tasks (swallowing with specific lingual pressure targets) and strength-based tasks (lingual presses with effort to reach a calibrated level; L. L. Pitts et al., 2014). The investigation explored direct tongue and saliva swallowing exercises for PwPD using both clinical and instrumental swallowing evaluations as outcome measures (L. L. Pitts et al., 2014). Overall, 4 PwPD completed 16 intervention sessions over 4 weeks, with each session lasting approximately 1 to 2 hours. At post-treatment and following a 4-week latency period, all 4 participants demonstrating both improved tongue function and increased swallowing efficiency on videofluoroscopy (L. L. Pitts et al., 2014). Overall, across studies, both strength- and skill-based exercises demonstrate promise in swallowing rehabilitation, especially for populations with neurogenic dysphagia (Easterling, 2017). However, more research is needed to explore optimal delivery methods (i.e., for both levels of intensity and prescribed tasks) and efficacy of swallowing rehabilitation in PwPD.
Intensity of Treatment

Research on the optimal intensity of dysphagia rehabilitation is sparse (Baker, 2012). Treatment intensity largely depends on medical diagnosis (e.g., neurodegenerative diseases, CVA, head and neck cancer, etc.), patient’s age, reason for referral, and patient’s environment (Baker, 2012). Overall, research indicates successful exercise-based therapies for swallowing have task-specific training, where exercises reflect the intended end goal, applied in an intensive manner (Athukorala et al., 2014; Russell et al., 2010; Steele et al., 2013). Intensity is supported by the overload principle: when a demand placed upon an individual is consistently greater than one’s capacity, adaptations occur to meet the demand (Morgan, 2017). Intensity of exercise-based rehabilitation should be increased within a treatment session (i.e., total number or repetitions and sets) and over time (i.e., length of individual sessions and continued duration of therapy; Morgan, 2017). However, there is no consensus on how many sets, repetitions, or minutes of exercise should be done for optimal outcomes (Morgan, 2017). Furthermore, it has been noted that the frequency with which an exercise regimen is conducted may be equally important to the intensity of the exercise within the session (Morgan, 2017). More research is needed to determine sufficient intensity levels of swallowing exercise for clinically-relevant improvements in populations with neurodegenerative disorders.

Intensive swallowing programs (ISPs) specifically combine interventions and are relatively new to neurorehabilitation, despite widespread application of intensive aphasia programs that also combined therapy regimens (Babbitt, Worrall, & Cherney, 2015; Malandraki et al., 2016). Yet, no restorative behavioral programs incorporating intensive,
multi-modal (i.e., combined respiratory and lingual exercises) treatments have been explored to remediate dysphagia in PwPD. Currently, no intensive multi-modal dysphagia rehabilitation program exists for PwPD that incorporates both skill and strength training targeting underlying lingual and airway deficits.

**Present Investigation**

The present study is an application of an ISP that incorporates both skill and strength training for the tongue and airway within a single-subject experiment for PwPD. Specific research questions for this pilot study include: (1) Does lingual strength (maximum isometric pressure; MIP), maximum expiratory pressure (MEP), maximum phonation time (MPT), and/or maximum phonation intensity (MPI) change in PwPD after completing a 4-week ISP that combines EMST and LSST interventions?, and (2) Does swallowing function (i.e., clinical outcome measures and patient-report) change in PwPD after completing a 4-week ISP that combines EMST and LSST interventions?
CHAPTER 3

METHODS

Institutional Review Board approval was obtained from the University of Northern Iowa (#17-0218). All researchers completed human subjects training prior to conducting this study.

Participants

Two participants with diagnosed idiopathic PD, confirmed by a medical release of information and neurologist report, were recruited from a local support group and provided consent for this study. Participants also both exhibited dysphagia at baseline, identified as either aspiration and/or pharyngeal residue noted on a fiberoptic endoscopic evaluation of swallowing (FEES) conducted within a week before initiation of treatment. Participants were a 61-year-old female (Participant 1) and a 67-year-old male (Participant 2). Both participants were receiving regular medical care and stable pharmacological treatment during the investigation. Additionally, both participants had taken part in prior speech or language therapy in the form of two separate LSVT® programs. Exclusionary criteria included: gastrointestinal disease or gastroesophageal surgery, head and neck structural abnormalities or surgery affecting swallowing or swallowing structures, and concurrent speech-language therapy. Current or previous medical history of temporomandibular joint disorders, significantly reduced jaw opening, and/or unmanaged seizure activity were exclusionary criteria due to risks associated with completing therapy exercises. Participants completed intake procedures including a medical history questionnaire, oral mechanism examination, height and weight measurements, and
classification of current diet via the Functional Oral Intake Scale (FOIS). The FOIS is a validated and clinically employed 7-level scale used to classify a patient’s severity of dysphagia based on diet modifications and intake of solids (Crary, Mann, & Groher, 2005). Levels 0-3 include some amount of tube feeding, while levels 4-7 reflect complete oral intake.

*Participant 1* was diagnosed with idiopathic PD 15 years prior to the current study. Her most recent Unified Parkinson’s Disease Rating Scale (UPDRS Section III) score was 9. The UPDRS is a widely utilized scale to classify PD in various aspects including motor and cognition (Martinez-Martin et al., 2018). Total scores range from 0-199, with 0 indicating no disability and 199 indicating the most severe impairment (Martinez-Martin et al., 2018). Participant 1 was classified as Stage 2 (out of 5 stages) on the Hoehn and Yahr Scale, indicating bilateral disease involvement without balance impairment (Martinez-Martin et al., 2018). Past medical history was remarkable for Deep Brain Stimulation (DBS) surgery in April 2014, a history of smoking, and usage of a Continuous Positive Airway Pressure (CPAP) for sleep apnea. Participant 1 was rated by examiners to be a Level 5 on the FOIS (i.e., complete oral intake of multiple solid consistencies, but requiring special preparation or compensation for solids during meals). Participant 1 self-reported symptoms of swallowing impairment included: infrequent “liquid going down the wrong pipe”, coughing during meals, drooling throughout the day, the feeling of food stuck in throat resulting in coughing, and special preparation for dry foods.
Participant 2 was diagnosed with idiopathic PD two years prior to participating in the study. Participant 2 was also classified as Stage 2 on the Hoehn and Yahr scale. Participant 2’s past medical history was remarkable for multiple neurological disorders (i.e., diagnosed brain tumor surgically removed in 2015 with no recurrence). He was rated by examiners at a FOIS Level 5. Patient self-reported symptoms of swallowing impairment included: infrequent “liquid going down the wrong pipe”, coughing during meals, the feeling of food stuck in throat resulting in coughing, and use of a compensatory strategy (e.g., chin tuck) while swallowing pills.

**Probe Measurements**

Probe measurements were taken five times at baseline prior to the start of treatment, twice weekly during treatment, and four times at post-treatment, which is consistent with evidence-based practice guidelines for single-subject design in the communication sciences and disorders field (Byiers, Reichle, & Symons, 2012). During the treatment phase, probe measurements were taken at the start of the first and fourth session each week. Probe measures included: Maximum Isometric Pressure (MIP), Maximum Expiratory Pressure (MEP), Maximum Phonation Time (MPT), and Maximum Phonation Intensity (MPI).

**Lingual-Palatal Pressure Measures**

The IOPI was utilized to measure pressure of the tongue pressing upward toward the palate (also known as lingual-palatal pressure), as it is a widely-used clinical tool for both children and adults. The IOPI, a handheld and portable device, can objectively measure lingual strength and endurance via an air-filled plastic bulb connected to a
plastic tube that measures peak pressure applied by the tongue to the bulb (Adams, Mathisen, Baines, Lazarus, & Callister, 2013). The IOPI demonstrates excellent test-retest reliability (Youmans & Stierwalt, 2006; Youmans, Youmans, & Stierwalt, 2009) and captures a continuous readout of lingual pressure in kilopascals (kPa) using DI-155 Data Acquisition Software on a connected computer.

To obtain lingual-palatal pressures, a trained examiner instructed the participant to complete the following three tasks: 1. Maximum Isometric Pressure (MIP or lingual strength), 2. saliva swallow, and 3. Swallowing Target Trial (STT). All tasks were completed at two standard evaluation positions: the anterior tongue (ie., the bulb directly behind the front teeth) and the posterior tongue (ie., the bulb in line with the molars; L. L. Pitts, Stierwalt, Hageman, & LaPointe, 2017).

MIP was defined as the greatest positive pressure in kPa exerted on the IOPI tongue bulb across three trials. MIP trials were encouraged by the examiner and were the only encouraged trials. Prior to conducting an MIP, the examiner instructed the patient by saying, “When I say ‘go’, push the bulb against the roof of your mouth as hard as you can with the front/middle of your tongue without biting the plastic tube. Make sure to flatten the entire bulb.” Researchers have reported that normative data for healthy adults includes performance of 56 kPa or greater for MIP in the anterior tongue position (MIPA; Iowa Oral Performance Instrument Medical, 2018). MIP in the posterior tongue position (MIPP) should be 54 kPa or above for males and 58 kPa or above for females (Gingrich, Stierwalt, Hageman, & LaPointe, 2012).
After MIP measures, the examiner calculated 30% and 50% of the MIP for both the anterior and posterior tongue known as the Percentage of Maximum Tongue Pressure (PMTP). Participants were then asked to swallow their saliva across five trials (5 STTs) at each of the following pressure targets: PMTP at 30% in the anterior position (PMTPA 30); PMTP at 30% in the posterior position (PMTPP 30); PMTP at 50% in the anterior position (PMTPA 50); and PMTP at 50% in the posterior position (PMTPP 50). Participants were instructed, “Please do not swallow until I tell you to. When you do swallow your saliva, try to swallow with a pressure of (examiner inserted target #), using just one swallow. Do not slow your swallow but swallow at your normal speed. Remember do not bite the tube. Ok, whenever you are ready, swallow”. These targets for saliva swallows were selected based on preliminary analysis of lingual pressures in healthy adults, which indicated mean swallowing pressures across consistencies typically range between 30% and 50% of MIP (Gingrich et al., 2012).

**Maximum Expiratory Pressure (MEP)**

MEP was obtained using the CareFusion Micro Respiratory Pressure Meter (MicroRPM). The patient was instructed by the examiner, “When I say ‘go’, inhale as deeply as you can and then exhale as quickly and forcefully as possible for two seconds.” Consistent with previous studies of EMST in PD (Saleem et al., 2005; T. Pitts et al., 2009; Troche et al., 2010), this measure was conducted until three numerical values within 5% of each other were obtained. These three values were averaged to find the participants’ MEP. Researchers have reported that a healthy male adult (60-69 years) should exhibit an MEP of 111 cmH₂O (SD = 11) or greater, while a healthy female adult
(60-69 years) should achieve a score of 76 cmH₂O (SD = 11) or greater (Neder, Andreoni, Lerario, & Nery, 1999).

Maximum Phonation Time (MPT)

The first probe measurement of phonatory-respiratory control taken was MPT. Participants were asked to sit upright in a chair. They were then instructed, “When I say ‘go’, inhale as deeply as possible and say ‘ah’, holding it for as long as you can. Focus on keeping your voice strong, rather than the quality of the sound. Okay, whenever you are ready, go.” The examiner recorded the number of seconds participants sustained “ah”. MPT was recorded three times at each probe measurement (Maslan, Leng, Rees, Blalock, & Butler, 2011; Miles et al., 2017). Previous literature indicates that two to three trials of MPT is a common, stable measure; after the third trial, subsequent trials do not significantly differ (Maslan et al., 2011; Shanks & Mast, 1977).

MPT is a gross outcome measure that has been taken in addition to similar measures (i.e., sustained vowel phonation) in previous swallowing intervention studies that targeted voice and swallowing in PD (El Sharkawi et al., 2002; Miles et al., 2017). Researchers have reported that abnormal MPT performance is less than 15 seconds for males and less than 14.3 seconds for females (Maslan et al., 2011). In the present study, MPT served as a control measure as voice was not targeted in therapy. In addition to being a gross outcome measure, MPT may also depend on phonation intensity, which was measured as a separate outcome and described below.
**Maximum Phonation Intensity (MPI)**

The second probe measure of voice taken was MPI, which was the maximum sound pressure level produced in dB, obtained during the MPT trials. MPI was conducted with an A Tenma 72-945 sound level meter with a ½ inch electret condenser microphone and a ruler. Participants were asked to sit upright in a chair. The distance from their mouth to the sound pressure level meter was measured to be 30 cm at each probe. The examiner programmed the sound pressure level meter to record the maximum sound level in dB during the MPT trials. Participants were instructed, “When I say ‘go’, inhale as deeply as possible and say ‘ah’, holding it for as long as you can. Focus on keeping your voice strong, rather than the quality of the sound. Okay, whenever you are ready, go.”

**Endpoint Measurements**

Endpoint measurements were taken before treatment, across two evaluation sessions and at post-treatment, across two evaluation sessions. These measures included the Mann Assessment of Swallowing Ability (MASA; Mann, 2002), the Timed Water Test (TWT; Hughes & Wiles, 1996), the Repetitive Saliva Swallowing Test (RSST; Oguchi et al., 2000), and the Swallowing Quality of Life Questionnaire (SWAL-QOL; McHorney et al., 2002).

**The Mann Assessment of Swallowing Ability (MASA)**

The MASA is a standard clinical evaluation of swallowing which assesses oral structures, oral motor functions, and bolus trials (Mann, 2002). It consists of 24 ratings that are summed into an overall rating score of swallowing ability, with the highest possible score being 200 (Mann, 2002). Normative data for the MASA was obtained
from a sample of 150 acute stroke patients which established that a score equal to or less than 178 may suggest dysphagia (Antonios et al., 2010). The MASA rates the participant on: alertness, motor speech, auditory comprehension, respiration, lingual strength, coordination and range of motion (ROM), appearance and function of the velum, voluntary and reflexive cough, bolus clearance, and oropharyngeal phases of swallow during bolus trials (Mann, 2002).

**Timed Water Test (TWT)**

The Timed Water Test (Hughes & Wiles, 1996) was chosen as an endpoint measure as it has previously demonstrated improvement following swallowing training in time per swallow and volume per swallow in PwPD (Athukorala et al., 2014). The TWT was completed with the participant sitting upright in a chair. The participant was given a cup of 150 mL of water and instructed, “When I say ‘go’, please drink the water as quickly as possible but be careful and stop if you begin to have difficulty. I’ll ask you to hold the cup to your lips and get ready to start. ‘Go’”. The time to complete the task was recorded in mL/second, also known as swallowing capacity (Hughes & Wiles, 1996). Swallowing capacity greater than or equal to 10 mL/sec is considered healthy performance (Hughes & Wiles, 1996).

**Repetitive Saliva Swallowing Test (RSST)**

The Repetitive Saliva Swallowing Test (RSST) was also selected as an outcome measure as it was designed to be a functional and efficient way to screen individuals for dysphagia (Oguchi et al., 2000). To complete the RSST, participants were instructed to swallow their saliva as quickly and as many times as possible. The examiner lightly
placed four fingers on the participants’ anterior neck to monitor the elevation of the hyoid and larynx and count the number of completed swallows (i.e., four-finger method of laryngeal palpation; Logemann, 1998). The total number of saliva swallows completed by each participation was recorded across 30 seconds. Three or more dry swallows within the 30 second time period is considered normal performance (Horiguchi & Suzuki, 2011).

**Swallowing Quality of Life Questionnaire (SWAL-QOL)**

Participants were given a copy of the SWAL-QOL assessment to independently complete at home and return to examiners at the second of two baseline visits. The SWAL-QOL is a 44-item tool that assesses quality-of-life concepts relating to swallowing. Self-reported scores range from 1-100 with higher scores indicating greater quality of life (McHorney et al., 2002).

**Design of the Intensive Exercise-based Swallowing Program (ISP)**

Participants in this study participated in 16 sessions of two counterbalanced treatments (EMST and LSST; Figure 1). Each treatment session lasted between 1.5-2 hours. All treatment sessions were conducted by a trained, supervised graduate student researcher (Jenks).
### Figure 1.
One weekly treatment regimen of counterbalanced Expiratory Muscle Strength Training (EMST) and Lingual Strength and Skill Training (LSST) with two probe days.

<table>
<thead>
<tr>
<th>Weekly Regimen</th>
<th>Day 1</th>
<th>Day 2</th>
<th>Day 3</th>
<th>Day 4</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Probes</strong></td>
<td><strong>Collected</strong></td>
<td><strong>Collected</strong></td>
<td><strong>Collected</strong></td>
<td><strong>Collected</strong></td>
</tr>
<tr>
<td><strong>1 Session</strong></td>
<td><strong>EMST Recalibration</strong></td>
<td><strong>LSST</strong></td>
<td><strong>EMST</strong></td>
<td><strong>LSST</strong></td>
</tr>
<tr>
<td></td>
<td>5 Sets of 5 Exercises</td>
<td>Posterior: 25 MT &amp; 10 SST</td>
<td>5 Sets of 5 Exercises</td>
<td>Posterior: 10 SST &amp; 25 MT</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Anterior: 25 MT &amp; 10 SST</td>
<td></td>
<td>Anterior: 10 SST &amp; 25 MT</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Posterior: 25 MT &amp; 10 SST</td>
<td></td>
<td>Posterior: 10 SST &amp; 25 MT</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Anterior: 25 MT &amp; 10 SST</td>
<td></td>
<td>Anterior: 10 SST &amp; 25 MT</td>
</tr>
<tr>
<td><strong>LSST</strong></td>
<td><strong>Recalibration</strong></td>
<td><strong>EMST</strong></td>
<td><strong>LSST</strong></td>
<td><strong>EMST</strong></td>
</tr>
<tr>
<td>Anterior: 25 MT &amp; 10 SST</td>
<td>5 Sets of 5 Exercises</td>
<td>Anterior: 10 SST &amp; 25 MT</td>
<td>Posterior: 10 SST &amp; 25 MT</td>
<td></td>
</tr>
<tr>
<td>Posterior: 25 MT &amp; 10 SST</td>
<td></td>
<td>Anterior: 10 SST &amp; 25 MT</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

**Expiratory Muscle Strength Training (EMST)**

During EMST therapy, participants strongly exhaled through a handheld mouthpiece (*EMST150™*) to reach individualized, calibrated goals. Past EMST protocol established for PwPD was followed; however, the protocol was adapted from 5 days per week to 4 days (exercises within each day stayed the same) and the weekly goals were set based upon MEP obtained with the *EMST150™* device. Adapting the program to 4 days a week increased feasibility as all sessions were not completed as a home program, but conducted by a treating clinician that monitored the completion and accuracy of exercises.
In the present protocol, participants were not instructed to re-attempt an exercise if their first attempt was not successful (i.e., the participant was not able to force air through the EMST150™ device), therefore, all participants completed the same number of exercise attempts.

Before beginning treatment, participants’ MEP was also measured using the EMST150™ device in addition to probe measurements of MEP using the CareFusion MicroRPM. To measure MEP using the EMST150™, participants were instructed to place a nose clip over their nose to prevent air leak. Then, participants were instructed to “inhale as deeply as possible and exhale quickly and forcefully through the device until the air rushes out”. If the participant was able to do so, the dial on the device was turned ¼ turn (i.e., 7.5 cmH2O). This procedure continued until participant could no longer force air through the device. MEP was defined as the highest pressure (cmH2O) at which the participant could successful force air through the EMST150™ device. The MEP value obtained using the EMST150™ at the beginning of each week was used to determine the treatment targets for the subsequent week (Saleem et al., 2005; T. Pitts et al., 2009; Troche et al., 2010). To determine treatment intensity level, MEP was multiplied by .75 to set a submaximal EMST target.

EMST therapy was conducted after the probes on Day 1 of each week of treatment and immediately at the start of Day 3 of treatment each week. On Day 2, EMST was conducted after LSST and on Day 4, EMST was conducted after probes and LSST. At the beginning of therapy, participants were instructed to “blow hard and fast through the device until air rushes through, then stop”. Participants were given a 30 second break
between each trial and completed a total of 5 repetitions in a set. Following the fifth trial, participants were given a 1 minute break before commencing the next set of 5 exercises. In total, each participant completed 5 sets of 5 repetitions each session.

**Lingual Strength and Skill Training (LSST)**

LSST was implemented as outlined by L. L. Pitts and colleagues (2014) using the IOPI. LSST consisted of two targeted exercises using the IOPI device: MIP and Swallowing Target Trials (STTs). Both exercises were targeted in the anterior and posterior tongue position. LSST was counterbalanced with EMST, by LSST exercise (i.e., MIP versus STT), and between the anterior and posterior lingual regions (Figure 1).

All MIPs and STTs were recorded using DI-155 Data Acquisition Software on a connected computer.

For one LSST cycle, 5 sets of 5 repetitions of MIPs were conducted with a minimum of 15 second break between each set. Goals for MIP trials during therapy were set using the MIPAs and MIPPs taken during probe measurements. The highest MIP probe set the goal for the MIP therapy trials. During the first week of therapy, MIP goals were set at 60% of the highest MIP recorded, while during weeks 2-4 the goals were set at 80% of the highest MIP.

Additionally, 10 STTs were conducted, 5 at 30% of the participants’ MIP and 5 at 50% MIP. The 10 STTs were randomized. Each day, 4 cycles of LSST were run (2 anterior and 2 posterior). Prior to beginning the MIPs, participants were instructed “When I say go, push the bulb against the roof of your mouth as hard as you can with the (front/middle) of your tongue without biting the plastic tube. Make sure to flatten the
entire bulb.” During MIP exercises, participants were motivated and given encouragement to hit their goals. Researchers gave both knowledge of performance and knowledge of results to participants. Visual biofeedback was given on the IOPI device and on the computer screen graphing the participant’s waveforms for MIP. Prior to a STT trial, participants were instructed, “please do not swallow until I tell you to. When you do swallow your saliva, try to swallow with a pressure of (insert target #), using just one swallow. Do not slow your swallow but swallow at your normal speed. Remember do not bite the tube. Ok, whenever you are ready, swallow”.

**Treatment Fidelity**

Twenty-five percent of treatment sessions (i.e., a total of 8 sessions) were monitored for treatment fidelity by an outside, trained rater (Hildebrand et al., 2012). The trained rater collected frequency counts of completed exercises within sessions. All monitored sessions exhibited 100% accuracy in adherence to protocol descriptions of the type and amount of exercises to be completed by participants.

**Analyses**

Descriptive analyses using SPSS 22.0 and Tau-\(U\) calculations which account for potential baseline trend and provide 90% confidence intervals were conducted for all probe measures (MIP, MEP, MPT, and MPI; Vannest, Parker, Gonen, & Adiguzel, 2016). Specifically, MIP waveforms were obtained from the IOPI and were analyzed for peak pressure using Windaq software automated analysis (DI-155). Endpoint measures (i.e., MASA, TWT, RSST, and SWAL-QOL) were descriptively analyzed for change between baseline and post-treatment testing.
CHAPTER 4

RESULTS

Probe Measurements

Lingual-Palatal Pressure Measures

Overall, at post-treatment, MIP scores descriptively increased for both the anterior and posterior tongue within each participant. Highest MIP scores across probe measures obtained at both phases (i.e., baseline and post-treatment) are reported by participant in Table 1. Probe results for both MIPA and MIPP are presented in line graph format across phases (baseline, treatment, and post-treatment) for Participant 1 (top) and Participant 2 (bottom) in both Figure 2 (MIPA) and Figure 3 (MIPP). Individual and combined Tau-\(U\) calculations are reported for MIPA in Table 2 and for MIPP in Table 3.

No significant trends in MIPA and MIPP were found across the five baseline probes for either participant. Significant gains in MIPA did not occur at post-treatment for Participant 1 (TAU \(U = 0.436, p = 0.174\) with 90% confidence interval (CI) \([-0.092, 0.964]\)); however, Participant 2 demonstrated significant gains in MIPA (TAU \(U = 1.000, p = 0.002\) with a 90% CI \([0.472, 1.000]\)). Data combined across the two participants indicated a significant effect on MIPA (TAU \(U = 0.718, p = 0.002\) with 90% CI \([0.3448, 1.000]\)). Significant gains in MIPP occurred for Participant 1 (TAU \(U = 0.673, p = 0.036\) with 90% CI \([0.145, 1.000]\)), Participant 2 (TAU \(U = 0.873, p = 0.007\) with 90% CI \([0.345, 1.000]\)), and when data were combined (TAU \(U = 0.773, p = 0.001\) with 90% CI \([0.3994, 1.000]\)). Effect sizes were moderate-strong (0.67 < \(U\)) for significant MIPA and MIPP gains.
Table 1

Maximum Isometric Pressure (MIP) at Baseline, Post-Treatment, and Calculated Gains in kilopascals (kPa).

<table>
<thead>
<tr>
<th>Participant</th>
<th>MIPA B</th>
<th>MIPA PT</th>
<th>MIPA Gain</th>
<th>MIPP B</th>
<th>MIPP PT</th>
<th>MIPP Gain</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>60.9</td>
<td>62.5</td>
<td>1.6</td>
<td>53.9</td>
<td>60.3</td>
<td>6.4</td>
</tr>
<tr>
<td>2</td>
<td>48.7</td>
<td>56.1</td>
<td>7.4</td>
<td>51.3</td>
<td>56.4</td>
<td>5.1</td>
</tr>
</tbody>
</table>

Note. MIPA = Maximum Isometric Pressure Anterior; MIPP = Maximum Isometric Pressure Posterior; B = Baseline; PT = Posttreatment; Gain = PT-B performance.

Figure 2. Line graphs of probe measures by participant for maximum isometric pressure of the anterior tongue.
Figure 3. Line graphs of probe measures by participant for maximum isometric pressure of the posterior tongue.
Table 2

TAU-Calculations of Trend, Phase, and Combined Effects for Maximum Isometric Pressure of the Anterior Tongue (MIPA).

<table>
<thead>
<tr>
<th>Effect</th>
<th>Participant</th>
<th>Tau-U</th>
<th>Z score</th>
<th>p-value</th>
<th>90% Confidence Interval</th>
</tr>
</thead>
<tbody>
<tr>
<td>Trend</td>
<td>1</td>
<td>-0.600</td>
<td>-1.470</td>
<td>0.142</td>
<td>-1.000&lt; &gt;0.072</td>
</tr>
<tr>
<td></td>
<td>2</td>
<td>0.400</td>
<td>0.980</td>
<td>0.327</td>
<td>-0.272&lt; &gt;1.000</td>
</tr>
<tr>
<td>Phase</td>
<td>1</td>
<td>0.436</td>
<td>1.360</td>
<td>0.174</td>
<td>-0.092&lt; &gt;0.964</td>
</tr>
<tr>
<td></td>
<td>2</td>
<td>1.000</td>
<td>3.115</td>
<td>0.002**</td>
<td>0.472&lt; &gt;1.000</td>
</tr>
<tr>
<td>Combined</td>
<td></td>
<td>0.718</td>
<td>3.164</td>
<td>0.002**</td>
<td>0.3448&lt; &gt;1.000</td>
</tr>
</tbody>
</table>

Note. *p < .05, **p < .01, ***p < .001.

Table 3

TAU-Calculations of Trend, Phase, and Combined Effects for Maximum Isometric Pressure of the Posterior Tongue (MIPP).

<table>
<thead>
<tr>
<th>Effect</th>
<th>Participant</th>
<th>Tau-U</th>
<th>Z score</th>
<th>p-value</th>
<th>90% Confidence Interval</th>
</tr>
</thead>
<tbody>
<tr>
<td>Trend</td>
<td>1</td>
<td>0.000</td>
<td>0.000</td>
<td>1.000</td>
<td>-0.672&lt; &gt;0.672</td>
</tr>
<tr>
<td></td>
<td>2</td>
<td>0.000</td>
<td>0.000</td>
<td>1.000</td>
<td>-0.672&lt; &gt;0.672</td>
</tr>
<tr>
<td>Phase</td>
<td>1</td>
<td>0.673</td>
<td>2.096</td>
<td>0.036*</td>
<td>0.145&lt; &gt;1.000</td>
</tr>
<tr>
<td></td>
<td>2</td>
<td>0.873</td>
<td>2.719</td>
<td>0.007**</td>
<td>0.345&lt; &gt;1.000</td>
</tr>
<tr>
<td>Combined</td>
<td></td>
<td>0.873</td>
<td>3.405</td>
<td>&lt;0.001***</td>
<td>0.3994&lt; &gt;1.000</td>
</tr>
</tbody>
</table>

Note. *p < .05, **p < .01, ***p < .001.

Maximum Expiratory Pressure (MEP)

Overall, at post-treatment, MEP scores descriptively increased for each participant. Highest MEP scores across probe measures obtained at both phases (i.e., baseline and post-treatment) are reported by participant in Table 4. Probes results for both MEP are presented in line graph format across phases (baseline, treatment, and post-treatment) for Participant 1 (top) and Participant 2 (bottom) in Figure 4. Individual and combined Tau-U calculations are reported for MEP in Table 5.
No significant trends in MEP were found across the five baseline probes for either participant. Significant gains in MEP occurred for Participant 1 (TAU $U = 0.800$, $p = 0.013$ with 90% CI [0.272, 1.000]) and Participant 2 (TAU $U = 1.000$, $p = 0.002$ with 90% CI [0.472, 1.000]). Data combined across the two participants indicated a significant effect on MEP (TAU $U = 0.900$, $p = 0.000$ with 90% CI [0.527, 1.000]). Effect sizes were moderate-strong ($U > 0.799$) for MEP gains.

Table 4

*Maximum Expiratory Pressure (MEP) at Baseline and Post-Treatment in cmH$_2$O.*

<table>
<thead>
<tr>
<th>Participant</th>
<th>MEP B</th>
<th>MEP PT</th>
<th>MEP Gain</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>92.0</td>
<td>99.0</td>
<td>7.0</td>
</tr>
<tr>
<td>2</td>
<td>66.3</td>
<td>79.3</td>
<td>13.0</td>
</tr>
</tbody>
</table>

*Note.* MEP= maximum expiratory pressure, B= baseline, PT= posttreatment, Gain= PT-B performance.
Figure 4. Line graphs of probe measures by participant for maximum expiratory pressure.
Table 5

TAU-Calculations of Trend, Phase, and Combined Effects for Maximum Expiratory Pressure (MEP).

<table>
<thead>
<tr>
<th>Effect</th>
<th>Participant</th>
<th>Tau-U</th>
<th>Z score</th>
<th>p-value</th>
<th>90% Confidence Interval</th>
</tr>
</thead>
<tbody>
<tr>
<td>Trend</td>
<td>1</td>
<td>0.600</td>
<td>1.470</td>
<td>0.142</td>
<td>-0.072&lt; &gt;1.000</td>
</tr>
<tr>
<td></td>
<td>2</td>
<td>-0.400</td>
<td>-0.980</td>
<td>0.327</td>
<td>-1.000&lt; &gt;0.272</td>
</tr>
<tr>
<td>Phase</td>
<td>1</td>
<td>0.800</td>
<td>2.492</td>
<td>0.013*</td>
<td>0.272&lt; &gt;1.000</td>
</tr>
<tr>
<td></td>
<td>2</td>
<td>1.000</td>
<td>3.115</td>
<td>0.002**</td>
<td>0.472&lt; &gt;1.000</td>
</tr>
<tr>
<td>Combined</td>
<td></td>
<td>0.900</td>
<td>3.965</td>
<td>&lt; .001***</td>
<td>0.527&lt; &gt;1.000</td>
</tr>
</tbody>
</table>

Note. *p < .05, **p < .01, ***p < .001.

Maximum Phonation Time (MPT)

Highest maximum phonation time (MPT) scores across probe measures obtained at both phases (i.e., baseline and post-treatment) are reported by participant in Table 6. Overall, at post-treatment, highest MPT scores in seconds increased for participant 1 while they decreased for participant 2. Probes results for MPT in seconds are presented in line graph format across phases (baseline, treatment, and post-treatment) for Participant 1 (top) and Participant 2 (bottom) in Figure 5. Individual and combined Tau-U calculations are reported for MPT in seconds in Table 7.

No significant trends in MPT were found across the five baseline probes for either participant. Significant gains in MPT in seconds occurred for Participant 1 (TAU U = 0.964, p = 0.003 with 90% CI [0.436, 1.000]); however, Participant 2 demonstrated reduced MPT at post-treatment (TAU U = -0.818, p = 0.011 with 90% CI [-1.000, 0.290]). Data combined across the two participants indicated no overall significant change in MPT between baseline and post-treatment phases (TAU U = 0.073, p = 0.749 with 90% CI [-0.301, 0.446]).
Table 6

Maximum Phonation Time (MPT) in seconds at Baseline and Post-Treatment.

<table>
<thead>
<tr>
<th>Participant</th>
<th>MPT B</th>
<th>MPT PT</th>
<th>MPT Gain</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>23.65</td>
<td>27.47</td>
<td>3.82</td>
</tr>
<tr>
<td>2</td>
<td>11.34</td>
<td>8.66</td>
<td>-2.68</td>
</tr>
</tbody>
</table>

Note. B = baseline, PT = posttreatment, Gain = PT-B performance.

Figure 5. Line graphs of probe measures by participant for maximum phonation time in seconds.
Table 7
TAU-Calculations of Trend, Phase, and Combined Effects for Maximum Phonation Time (MPT) in seconds.

<table>
<thead>
<tr>
<th>Effect</th>
<th>Participant</th>
<th>Tau-U</th>
<th>Z score</th>
<th>p-value</th>
<th>90% Confidence Interval</th>
</tr>
</thead>
<tbody>
<tr>
<td>Trend</td>
<td>1</td>
<td>-0.400</td>
<td>-0.980</td>
<td>0.327</td>
<td>-1.000&lt; &gt; 0.272</td>
</tr>
<tr>
<td></td>
<td>2</td>
<td>-0.800</td>
<td>-1.960</td>
<td>0.050</td>
<td>-1.000&lt; &gt;-0.128</td>
</tr>
<tr>
<td>Phase</td>
<td>1</td>
<td>0.964</td>
<td>3.002</td>
<td>0.003**</td>
<td>0.436&lt; &gt;1.000</td>
</tr>
<tr>
<td></td>
<td>2</td>
<td>-0.818</td>
<td>-2.55</td>
<td>0.011*</td>
<td>-1.000&lt; &gt;-0.290</td>
</tr>
<tr>
<td>Combined</td>
<td></td>
<td>0.073</td>
<td>0.320</td>
<td>0.749</td>
<td>-0.301&lt; &gt;0.446</td>
</tr>
</tbody>
</table>

Note. *p < .05, **p < .01, ***p < .001.

Maximum Phonation Intensity (MPI)

Highest maximum phonation intensity (MPI) obtained during MPT probe measures across both phases (i.e., baseline and post-treatment) are reported by participant in Table 8. Overall, at post-treatment, MPI increased across both participants. Probes results for MPI in decibels (dB) are presented in line graph format across phases (baseline, treatment, and post-treatment) for Participant 1 (top) and Participant 2 (bottom) in Figure 6.

Individual and combined Tau-U calculations are reported for maximum phonation intensity (MPI) in Table 9. No significant trends in MPI were found across the five baseline probes for either participant. MPI was maintained for Participant 1 (TAU U = 0.509, p = 0.113 with 90% CI [-0.019, 1.000]); however, Participant 2 demonstrated significant increase in MPI at post-treatment (TAU U = 0.891, p = 0.006 with 90% CI [0.363, 1.000]). Data combined across the two participants indicated a significant post-treatment gain in MPI (TAU U = 0.700, p = 0.002 with 90% CI [0.327, 1.000]).
Table 8

*Maximum Phonation Intensity (MPI) during Maximum Phonation Time (MPT) in decibels (dB) at Baseline and Post-Treatment.*

<table>
<thead>
<tr>
<th>Participant</th>
<th>MSL B</th>
<th>MSL PT</th>
<th>MSL dB Gain</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>80.00</td>
<td>83.00</td>
<td>3.00</td>
</tr>
<tr>
<td>2</td>
<td>88.00</td>
<td>92.00</td>
<td>4.00</td>
</tr>
</tbody>
</table>

*Note.* B = baseline, PT = posttreatment, Gain = PT-B performance.

*Figure 6.* Line graphs of probe measures by participant for maximum phonation intensity during maximum phonation time in decibels.
Table 9

TAU-Calculations of Trend, Phase, and Combined Effects for Maximum Phonation Intensity (MPI) during Maximum Phonation Time (MPT) in decibels (dB).

<table>
<thead>
<tr>
<th>Effect</th>
<th>Participant</th>
<th>Tau-U</th>
<th>Z score</th>
<th>p-value</th>
<th>90% Confidence Interval</th>
</tr>
</thead>
<tbody>
<tr>
<td>Trend</td>
<td>1</td>
<td>0.300</td>
<td>0.735</td>
<td>0.462</td>
<td>-0.372&lt; &gt;0.972</td>
</tr>
<tr>
<td></td>
<td>2</td>
<td>0.500</td>
<td>1.225</td>
<td>0.221</td>
<td>-0.172&lt; &gt;1.000</td>
</tr>
<tr>
<td>Phase</td>
<td>1</td>
<td>0.509</td>
<td>1.586</td>
<td>0.113</td>
<td>-0.019&lt; &gt;1.000</td>
</tr>
<tr>
<td></td>
<td>2</td>
<td>0.891</td>
<td>2.776</td>
<td>0.006**</td>
<td>0.363&lt; &gt;1.000</td>
</tr>
<tr>
<td>Combined</td>
<td></td>
<td>0.700</td>
<td>3.084</td>
<td>0.002**</td>
<td>0.327&lt; &gt;1.000</td>
</tr>
</tbody>
</table>

Note. *p < .05, ** p < .01, ***p < .001.

Endpoint Measurements

Endpoint measures are reported from baseline and post-treatment testing for each participant in Table 10. Overall, at post-treatment, MASA and TWT scores descriptively increased for both participants. RSST measures dropped by one swallow for Participant 1 at post-treatment and increased at post-treatment for Participant 2. SWAL-QOL scores for both participants remained consistent.

Table 10

Baseline and Post-Treatment Performance on Endpoint Measures of Swallowing Function by Participant.

<table>
<thead>
<tr>
<th>Endpoint Measure</th>
<th>Participant</th>
<th>Baseline</th>
<th>Post-Treatment</th>
<th>Gain</th>
</tr>
</thead>
<tbody>
<tr>
<td>MASA (total score)</td>
<td>1</td>
<td>173.00</td>
<td>177.00</td>
<td>4.00</td>
</tr>
<tr>
<td></td>
<td>2</td>
<td>164.00</td>
<td>184.00</td>
<td>20.00</td>
</tr>
<tr>
<td>TWT (mL/sec)</td>
<td>1</td>
<td>12.71</td>
<td>14.13</td>
<td>1.42</td>
</tr>
<tr>
<td></td>
<td>2</td>
<td>6.55</td>
<td>7.73</td>
<td>1.18</td>
</tr>
<tr>
<td>RSST (frequency count)</td>
<td>1</td>
<td>6.00</td>
<td>5.00</td>
<td>-1.00</td>
</tr>
<tr>
<td></td>
<td>2</td>
<td>2.00</td>
<td>4.00</td>
<td>2.00</td>
</tr>
<tr>
<td>SWAL-QOL (total score)</td>
<td>1</td>
<td>92.12</td>
<td>92.12</td>
<td>0.00</td>
</tr>
<tr>
<td></td>
<td>2</td>
<td>71.19</td>
<td>72.83</td>
<td>1.64</td>
</tr>
</tbody>
</table>

Note. MASA = Mann Assessment of Swallowing Ability, TWT = Timed Water Test, RSST = Repetitive Saliva Swallowing Test, SWAL-QOL = Swallowing Quality of Life Questionnaire, Gain = Post-Treatment—Baseline performance.
CHAPTER 5

DISCUSSION

Recent evidence has demonstrated that combining rehabilitative interventions targeting multiple subsystems of swallowing may improve swallowing outcomes in persons with neurogenic dysphagia (Malandraki et al., 2016). To date, such an intensive swallowing program has not been trialed for PwPD (e.g., targeting both disordered respiratory and lingual function across multiple weeks of intervention). The present investigation provides preliminary evidence that a 4-week intensive exercise-based swallowing program may improve tongue strength, maximal expiratory pressure, and clinical measures of swallowing function for PwPD.

Influence of ISP on Lingual Strength

Despite the participants’ diverse, complex medical histories, both participants demonstrated descriptive gains in both anterior and posterior lingual strength at post-treatment. While the increase in anterior tongue strength for Participant 1 was not statistically significant at post-treatment, significant gain in MIPA was found for Participant 2 and significant gain in MIPP occurred at post-treatment for both participants. Normative data for MIPA in healthy adults is 56 kPa or greater for MIPA (Iowa Oral Performance Instrument Medical, 2018), indicating that Participant 2 was below average at baseline and Participant 1 was within normal limits. Normative MIPP performance is suggested as 54 kPa or above for males and 58 kPa or above for females (Gingrich et al., 2012). Therefore, both participants were below average at baseline and both increased their posterior lingual strength to within normal limits at post-treatment.
Furthermore, combined data showed that overall significant improvements were evident in both MIPA and MIPP following intervention. These results are consistent with the work of L. L. Pitts and colleagues (2014) in which PwPD demonstrated improved lingual strength scores at a 4-week follow-up evaluation after completing a 4-week LSST program.

One possible explanation for increases in lingual strength following ISP is the implementation of principles of muscle strength training during LSST. Previous research has shown that the tongue is responsive to strengthening exercises applied in an intensive manner, even gaining approximately 5% in muscle mass over 8 weeks of training, likely secondary to both overload and progressive resistance principles (Morgan, 2017; Robbins et al., 2005, 2007). The overload principle dictates that, in order to improve muscle strength, a resistance of no less than 60% of one’s maximum ability should be set as a targeted goal (Morgan, 2017). Furthermore, as one’s maximum ability increases, the resistance or targeted goals should also increase (e.g., resistance may be set at 75 or 80% of maximum capacity), known as progressive resistance (Morgan, 2017; Robbins et al., 2005, 2007). These principles were applied during LSST training in the present investigation. The overload and progressive resistance principles have shown efficacy in previous research aiming to improve lingual strength across various etiologies of neurogenic dysphagia (McKenna et al., 2017; L. L. Pitts et al., 2014; Robbins et al., 2005, 2007), consistent with the results of the present study.

Additionally, gains in MIPA and MIPP scores may be due to the specificity of the training. Research has shown that targeting lingual strength via maximum and sub-
maximal isometric pressure tasks leads to increased maximal lingual strength (McKenna et al., 2017; L. L. Pitts et al., 2014; Rogus-Pulia et al., 2016; Steele et al., 2013; Robbins et al., 2005, 2007). These studies have implemented principles of specificity, in that the trained task directly practices the end goal or desired behavior (Morgan, 2017). Therefore, the tasks that were trained (e.g., isometric lingual presses) led to gains in the related outcome measures (e.g., maximum isometric pressures). Furthermore, the increased lingual strength noted in the present study is of clinical significance, as various studies have documented lingual weakness in PwPD and lingual strength is critical for successful bolus propulsion during swallowing (O’Day et al., 2005; L. L. Pitts et al., 2018; Solomon et al., 1995, 2000). The improved lingual strength of the anterior and/or posterior tongue noted in this study may have translated to more efficient bolus propulsion and thus faster swallowing as seen on TWT and RSST performance at post-treatment.

Finally, while Participant 1’s MIPA scores did not significantly increase following the ISP, they were within normal limits at baseline and demonstrated a small descriptive gain over the course of the study. However, previously investigated protocols consist of approximately 8 or more weeks of lingual strength training, therefore a longer program may be necessary to observe an increase in tongue strength (Robbins et al., 2005, 2007; Yeates et al., 2008).

Influence of ISP on Expiratory Strength

Significant improvements in maximum expiratory pressure (MEP) generation were evident for both participants individually and when combined at post-treatment.
Results were consistent with the stated hypotheses and with prior literature, specifically that Expiratory Muscle Strength Training (EMST) improves MEP and cough parameters in PwPD (Saleem et al., 2005; T. Pitts et al., 2009; Troche et al., 2010). Prior applications of EMST have included 20 sessions across 4 weeks; however, the present study found significant benefit in PwPD across 16 sessions within 4 weeks. It remains to be determined if the extent of change in MEP significantly differs between these two prescribed regimens and what the intensity of treatment should be.

At initial baseline testing, both participants were below MEP norms for both healthy adults (Neder et al., 1999). Gains in MEP may be due to the demonstrated efficacy of EMST as a strength training protocol. EMST applies exercise principles from limb studies to abdominal and internal intercostal muscles. Abdominal and internal intercostal muscles are both composed of an approximately equal distribution of Type I and Type II muscle fibers. This ratio is similar to the composition of limb muscles (Mizuno, 1991), and may contribute to the responsiveness of expiratory muscles to exercise protocol. Furthermore, EMST trials reflect the exercise principle of specificity. Trials are specific as they include forced expiration, which exactly matches the goal behavior (Morgan, 2017). By specifically practicing expiratory movements, EMST may be effective in promoting optimal muscle recruitment, decruitment of antagonistic muscles, and neural adaptation (Saleem, 2005). PwPD often attempt to compensate for rigidity, bradykinesia, and deconditioning of muscles by employing antagonist muscles, which may unintentionally interfere with the intended movement (Glendinning & Enoka, 1994).
EMST also incorporates the exercise principles of: intensity, overload, and progressive resistance; which may have also contributed to overall MEP gains. First, 16 sessions delivered across four weeks is greater intensity than traditional rehabilitation schedules (El Sharkawi et al., 2002). In regard to overload, exercise trials for muscle strengthening should be no less than 60% of the maximum ability (Morgan, 2017) and, consistent with previous research, the EMST practice levels were set at 75% of each participant’s MEP in the present study (Saleem, 2005; Saleem et al., 2005; T. Pitts et al., 2009; Troche et al., 2010). Finally, EMST utilizes the principle of progressive resistance to encourage gains in MEP (Morgan, 2017), and like prior studies, the level of resistance in the present study were recalibrated at the beginning of each week as MEP performance increased.

**Influence of ISP on Voicing Measures**

Overall, MPT and MPI measures were not uniformly responsive to ISP training which was not surprising considering voicing measures were untrained probes. Participant 2 exhibited abnormal baseline performance that did not improve to within healthy ranges at post-treatment while Participant 1 exhibited above average performance throughout baseline, treatment, and post-treatment phases. Both participants demonstrated a tradeoff between duration of sustained phonation and intensity between baseline and post-treatment testing. While Participant 1 demonstrated significant gains in MPT without change in intensity, Participant 2 demonstrated decreased MPT with significant gains in intensity at post-treatment. This lack of uniform responsiveness in the untrained, referent tasks of voicing duration and intensity suggests that there was not a
placebo or learning effect influencing participant performance on probe measures. In prior studies, LSVT® has demonstrated improved MPT and MPI (El Sharkawi et al., 2002; Miles et al., 2017); however, isolated applications of LSST or EMST have not investigated measures of phonation as a secondary or generalized outcome.

**Influence of ISP on Clinical Measures of Swallowing Function**

Both participants improved or maintained a healthy performance on all three of the clinical assessments of swallowing at post-treatment: the MASA, TWT, and RSST. The present findings are in agreement with previous research that has shown that both EMST and LSST programs in isolation result in improved swallowing on instrumental evaluations of swallowing. None of the prior studies of EMST or LSST have applied clinical measures of swallowing such as the MASA, TWT, and RSS (T. Pitts et al., 2009; L. L. Pitts et al., 2014; Troche et al., 2010).

The first of the clinical measures, the MASA, demonstrated increased scores posttreatment: Participant 2 gained 20 points, while Participant 1 gained 4 points. Gains in MASA scores were related to improved scores on the tongue strength section of the MASA for both participants. Participant 1 also exhibited an improved tongue coordination score on the MASA; whereas Participant 2’s scores also increased on both the pharyngeal phase and pharyngeal response sections. Increased tongue strength may be related to increased MIP; Participant 2’s increased pharyngeal response scores on the MASA may be related to increased MEP and thus improved airway protection during the pharyngeal phase (T. Pitts et al., 2009).
In addition to the MASA, both participants’ performance improved on the TWT, a swallow screening in which participants must drink 150mL as quickly as possible (Horiguchi & Suzuki, 2011; Oguchi et al., 2000). Both participants reduced the length of time needed to complete the swallowing task, as their average swallowing capacity (i.e., mL/second) increased by at least 1 mL/second each. These results differ slightly from those of Athukorala and colleagues (2014) in which post-hoc analysis revealed that swallowing capacity (i.e., volume over time) did not significantly improve for the 10 participants with PD upon completion of a 4-week swallow skill training. However, both time per swallow and volume per swallow did increase following the BiSSKiT training intervention (Athukorala et al., 2014). Outcome measures of time per swallow and volume per swallow during TWT were not taken during the present investigation; therefore, it is not known whether they changed at posttreatment. Participant 1’s TWT scores remained in an abnormal category (i.e., 7.73 mL/second) upon completion of the program; however, both participant’s scores increased overall with Participant 2’s scores remaining within normal capacity (i.e., 10 mL/second).

The final clinical outcome measure was the Repetitive Saliva Swallowing Test (RSST), for which 3 dry swallows completed within 30 seconds is considered normal performance (Horiguchi & Suzuki, 2011). Although Participant 1’s performance on the RSST decreased by 1 swallow, performance at both baseline and post-treatment remained within healthy performance. Additionally, Participant 2’s performance increased by 2 saliva swallows, suggesting improvement from abnormal swallowing performance to within normal limits at post-treatment.
Theoretically, the improvements noted in clinical measures of swallowing due to its multi-modal nature. This was a preliminary investigation of an ISP incorporating interventions that target more than one subsystem related to swallowing, thereby utilizing a more holistic approach to dysphagia intervention. As noted in previous literature, single modality gains, which also were seen in isolated applications of EMST and LSST (i.e., MEP gains following EMST, and increased tongue strength following LSST; Saleem et al., 2005; T. Pitts et al., 2009; L. L. Pitts et al., 2014; Troche et al., 2010), were both present following the ISP. The finding of multiple areas of gain following an ISP agree with the findings of Malandraki and colleagues (2016), that suggest an Intensive Dysphagia Rehabilitation approach that combines exercise modalities improves physiological and functional swallowing outcome measures for persons with dysphagia secondary to neurogenic etiologies. The present ISP specifically demonstrated preliminary efficacy of an ISP specifically for PwPD, a population at risk for both lingual and respiratory dysfunction.

**Impact of ISP on Self-Reported Swallowing-Quality of Life**

Dysphagia interventions may increase swallowing-related quality of life outcomes in PwPD and dysphagia; however, in the present investigation, both participants largely maintained their SWAL-QOL scores from pre-treatment to post-treatment without significant gains. A possible explanation for maintained, rather than increased, SWAL-QOL scores may be an increased awareness in swallowing impairment posttreatment (Kalf et al., 2012). During the process of assessing and treating dysphagia, participants may have come to better realize the extent of their swallowing impairment and its effect
on their lives. Furthermore, the participants in this study presented with mild dysphagia (e.g., FOIS ≥ 5 and MASA = 168-177; Crary et al., 2005; Mann, 2002), which may have masked gains due to a ceiling effect (i.e., already high scores at baseline) and an increased awareness throughout treatment.

Limitations

There are several limitations to note. First, this investigation did not corroborate probe measurements or clinical outcome measures with instrumental assessment of swallowing. Implementing either fiberoptic endoscopic evaluation of swallowing (FEES) or videofluoroscopic swallow studies (VFSS) in future studies would assist in confirming positive outcomes especially for changes in pharyngeal phase physiology and/or airway protection. Instrumental assessment would also serve as a record of physiological performance in comparison to self-report of swallowing function. Another limitation of this study is that it does not include participants with a variety of PD severity (e.g., Hoehn & Yahr Stage 3 or greater). Including participants with a greater range of disease severity would provide insight on how patients with more severe PD and possibly more severe dysphagia would respond to a similar ISP. One final limitation to this study is that the evaluator of the MASA was not blinded to the evaluation timepoint (e.g., baseline vs. post-treatment). This may have contributed to potential examiner bias in the subjective MASA ratings, and future research may benefit from recorded evaluations reviewed by a blinded examiner.
Clinical Implications and Future Research

The results of this study indicate that an ISP may have varying, but overall positive outcomes for PwPD in the areas of lingual strength, maximum expiratory pressure, and clinical measures of swallowing. Differential effects in this study may be attributed to diverse past medical histories or individual differences in baseline performance across probe and endpoint measures (e.g., if the participant performed within normal limits or demonstrated impairment at baseline). In total, the results are supportive of development of ISPs to treat PD-related dysphagia, even for patients with complex medical histories. It is critical for clinicians to consider the growing literature that supports intensive dysphagia rehabilitation for neurodegenerative populations, such as those with PD.

Overall, the promising results of the current investigation coupled with positive outcomes reported in previous research warrants further investigations of applications of ISPs in PwPD. To expand upon these preliminary results and their limitations, future research studies could be strengthened in several ways. First, future research may employ an extended multiple baseline approach, which would validate findings by providing evidence that gains are due to the treatment rather than a chance outcome. In the future, it would also be beneficial to include participants with varying levels of disease severity and with varying levels of swallowing impairment to assess outcomes of ISPs across a more heterogeneous group with PD. Finally, it would be beneficial for future research to implement instrumental examination of swallowing (e.g., FEES or VFSS) to explore
specific changes in timing or in the extent of displacement of structures within the oropharyngeal phases of swallowing.

**Conclusion**

As the prevalence of PwPD is rapidly increasing and the vast majority of PwPD present with dysphagia, there is a great need for evidence-based swallowing interventions to address dysphagia in PD. Overall, our preliminary results from a 4-week Intensive Swallowing Program (ISP) for PwPD and dysphagia are positive for participants with PD and medical comorbidities. Participants demonstrated gains in measures of lingual strength and maximal expiratory pressure, as well as generalization to clinical measures of swallowing safety and efficiency. To establish best care practices and optimize ISPs, further investigation of ISPs to manage PD-related dysphagia is warranted and timely.
REFERENCES


