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The effects of a volitional breathing technique on swallowing and respiratory coordination in individuals with amyotrophic lateral sclerosis: A pilot investigation

by

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Abstract

Individuals with amyotrophic lateral sclerosis (ALS) often present with aberrant respiratory-swallowing relationships. In this pilot study, eight individuals with ALS learned a volitional breathing technique designed to promote safe swallowing. The primary focus of the study was to evaluate the effectiveness of the technique in (1) promoting expiratory breathing after swallowing and (2) improving patients' perceptions of swallowing. As a group, the participants showed a statistically significant increase in the average number of typical swallows (expiration after swallowing) following training. With regard to perceptions of swallowing, there were no obvious differences between participants' baseline and post-treatment quality of the life scores on a questionnaire; however, participants' responses to qualitative interview questions were generally positive and suggest that participants found the treatment technique to be beneficial and effective in improving their safety and comfort while swallowing.

Keywords: dysphagia, swallow-respiration coordination, expiration, inspiration, lung volume, amyotrophic lateral sclerosis

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List of Symbols, Nomenclature, or Abbreviations

ALS	amyotrophic lateral sclerosis
ALSFRS-R	Revised ALS Functional Rating Scale
CN	cranial nerve
CPG	central pattern generator
EMG	electromyography
ER	expiratory reflex
ex-ex	expiration-swallow apnea-expiration
ex-in	expiration-swallow apnea-inspiration
fMRI	functional magnetic resonance imaging
FVC	forced vital capacity
in-ex	inspiration-swallow apnea-expiration
in-in	inspiration-swallow apnea-inspiration
LMN	lower motor neuron
M1	primary motor
MEG	magnetoencephalography
MH	mental health
MND	motor neuron disease
RCT	randomized controlled trial
NA	nucleus ambiguous
NTS	nucleus of the tractus solitarius
O_1 X O_2	one-group pretest-posttest research design
PCF	peak cough flow
PEG	percutaneous endoscopic gastrostomy

PES	pharyngoesophageal segment
PET	positron emissions tomography
S1	primary sensory
S2	secondary sensory
SF	symptom frequency
SMA	supplementary motor area
SWAL-QOL	Swallowing Quality of Life Scale
TMS	transcranial magnetic stimulation
UMN	upper motor neuron
VRG	ventral respiratory group
VSG	ventral swallowing group
xE	swallow apnea followed by expiration
xI	swallow apnea followed by inspiration

Amyotrophic Lateral Sclerosis (ALS), sometimes referred to as Lou Gehrig's disease, is a progressive neurodegenerative disease of unknown cause. ALS is categorized under the general term, *motor neuron disease* (MND), a broad, heterogeneous group of clinical syndromes that involve disorders of motor neurons in the spinal cord, brainstem, and cerebral cortex (Mitsumoto, 1994). MNDs are typically characterized by varying combinations of muscular weakness, atrophy, and pyramidal tract signs. Among adults, ALS is the most common motor neuron disease and refers specifically to a "relentlessly progressive, generalized, and fatal wasting of skeletal muscle" (Mitsumoto, 1994, p. 1). Although the average age of onset of ALS is 55 years of age, the disease can begin as early as the late teens and as late as the 80's. Though the likelihood of developing ALS is equal across males and females older than 50 years of age, the incidence rate for males under 50 is twice that of females. Consequently, men are more frequently affected than women (Nelson & McGuire, 2006). While the incidence and prevalence of ALS are increasing (Strong, 2003) with worldwide incidence rates ranging between 1.8 and 2.1 per 100,000 individuals (Strong, 2004), there are relatively few number of people currently living with this disease. This is due to the relentless and rapidly progressive nature of the disease. Approximately 50% of patients die within three years of onset and 90% die within five years. Over 3000 people in Canada are living with ALS, with two to three Canadians dying each day (Statistics Canada, 2003).

Neuropathology

ALS attacks brain and spinal cord nerve cells that control voluntary movement, causing skeletal muscles to weaken and atrophy and leading to paralysis as the disease progresses (Yorkston, Miller, & Strand, 1995). Both upper and lower motor neurons are activated to initiate voluntary movement; however, the disease affects the upper and lower motor neuron systems to varying degrees. Lower Motor Neuron (LMN) cells are located in the spinal cord and brainstem and their axons directly contact muscle fibers (Shaw, 2000). As LMNs degenerate and die, the affected muscles weaken and atrophy, resulting in flaccid muscle tone, fasciculations, and fibrillations. LMN involvement also manifests as fatigue, diminished range of motion, loss of coordination, and clumsiness (Mitsumoto, 1994; Yorkston et al., 1995). Upper Motor Neuron (UMN) cells predominantly reside in the precentral motor cortex and their axons make contact with LMNs (Shaw, 2000). Degeneration of UMNs results in stiff and spastic muscles with increased tone, often resulting in muscle cramps and spasms (Mitsumoto, 1994). Initially, either upper or lower motor neurons may be involved; however, as ALS progresses, both UMN and LMNs become involved (Yorkston et al., 1995). In order to clinically diagnose ALS, the El Escorial criterion is typically used to rule out other neuromuscular disorders. A neurologist will test for impairment in both the upper and lower motor neurons, and Electromyography (EMG) is conducted to further support the diagnosis (Brooks et al., 2000).

ALS is also characterized by pathology in the peripheral nervous system (Yorkston et al., 1995). Nerves in the periphery show reduced numbers of large myelinated fibers, as well as axonal degeneration and atrophy. Specifically, cranial nerves V, VII, IX, X, and XII are negatively affected. The loss of large myelinated nerve axons occurs in the ventral roots and peripheral nerves in the cervical, thoracic, and lumbar regions.

Clinical Course

ALS typically begins in one muscle group and spreads to involve all skeletal muscles, including the limb, bulbar, thoracic, and abdominal muscles (Lechtzin, 2006). The clinical presentation is progressive weakness, which is evident in two-thirds of all patients with ALS (Mitsumoto, 1994). Weakness is typically focal during onset and the effects are evident in two areas – spinal and bulbar (Yorkston et al., 1995). Spinal symptoms affect upper and lower limb function, which greatly impair physical mobility. Approximately 70% of patients initially present with spinal symptoms, characterized by arm and hand or leg weakness. Bulbar symptoms are the second most common initial complaint and occur in 22% of patients (Mitsumoto, 1994). Bulbar symptoms affect the face, tongue, and throat, and manifest as speech and swallowing difficulties. Patients who present with bulbar symptoms have a poorer prognosis as the disease progresses more rapidly, with an average lifespan of only 2.2 years after the initial appearance of symptoms (Yorkston et al., 1995). Respiratory symptoms are also significant in ALS and usually occur as bulbar and spinal symptoms progress. Less than 10% of patients with ALS initially present with muscle atrophy or wasting, and fewer than 10% present with pain and muscle cramps (Mitsumoto, 1994).

Spinal symptoms. A lower limb onset typically presents as an asymmetric distal disturbance, with the most common complaints consisting of tripping while walking or running, difficulty with curbs or stairs, dragging of a foot, or a foot drop, which results in a steppage gait (Gordon & Mitsumoto, 2007). These early symptoms are usually asymmetrical and frequently involve only one limb. Following the onset of a distal weakness, patients often notice total or proximal leg weakness with progressive muscle wasting. These symptoms occur as a result of LMN degeneration and eventually occur in the unaffected leg. UMN degeneration in the lower limbs will manifest as a gait or balance disturbance and patients will complain of falling on quick turns or leg clonus, an involuntary shaking in the legs. As ALS progresses, the loss of fine and gross motor control is followed by the loss of independent ambulation and eventually quadriplegia.

The symptoms of limb-onset ALS may also be apparent in the upper extremities. The typical presentation consists of unilateral hand weakness, causing weakness of grip, and an accompanying weak wrist extension (Shaw, 2000). As a result of lost LMN innervation, the muscles in the hand atrophy and patients experience the loss of fine motor control in their fingers (Gordon & Mitsumoto, 2007). Patients may have difficulty picking up small objects, writing, turning keys and fastening buttons. Although UMN symptoms in the upper extremities are less common, they include stiffness, slowness, and involuntary tremors due to clonus. Furthermore, according to Mitsumoto (1994), although muscle cramping is not a common presenting symptom, it is one of the most frequent symptoms during the course of the disease. Numbness and pain may also occur; however, they are typically not part of the initial onset of the disease.

Bulbar symptoms. Bulbar symptoms are evident when motor neurons in the brainstem degenerate and cause weakness and atrophy of the tongue and pharyngeal muscles (Shaw, 2000). Nearly 30% of patients show the initial effects of ALS in the corticobulbar musculature, and all patients demonstrate bulbar involvement at later stages of the disease (Haverkamp, Appel, & Appel, 1995; Muller, 1952). UMN involvement in ALS results in "spastic bulbar palsy" (or pseudobulbar palsy) (Mitsumoto, 1994). The clinical characteristics of pseudobulbar palsy include spasticity of the bulbar muscles (jaw, face, soft palate, pharynx, larynx and tongue), emotional lability (pathological laughing and crying), and a pathological jaw and gag reflex. When the LMNs of the brainstem are primarily affected, the condition is called "paretic" or "flaccid bulbar palsy", and is characterized by flaccid facial muscles, muscular atrophy and fasciculations of the tongue, and an absent jaw and gag reflex. It is not uncommon for patients to have a mixed bulbar palsy, consisting of a combination of flaccid and spastic characteristics.

Bulbar involvement often manifests as dysarthria (impaired articulation) and/or dysphagia (impaired mastication and deglutition). In ALS, dysarthria can be spastic, flaccid, or mixed (Mitsumoto, 1994). It is not uncommon for patients to initially complain of a weak or hoarse voice and the inability to yell or sing, which results from paretic vocal folds. Patients may also complain of slurred speech due to articulator weakness and nasal speech due to palatal weakness. In spastic dysarthria, voice quality often sounds strained-strangled and repetitive movements of the lips, tongue and pharynx are especially difficult. As the disease progresses, articulation becomes increasingly challenging for patients. This may culminate in a complete inability to speak during advanced stages of ALS.

Following the onset of speech difficulties, patients often experience a swallowing impairment (Mitsumoto, 1994). Swallowing difficulty, or dysphagia, is a common consequence of ALS. Dysphagia is a delay in, or misdirection of, a fluid or solid food bolus as it moves from the mouth to the stomach (Crary & Groher, 2003). Approximately 9-13% of patients with ALS initially present with dysphagia (Mitsumoto, 1994). Although the clinical course and time of onset differs, dysphagia eventually occurs in all patients with ALS (Kawai et al., 2003). The swallowing deficits observed in ALS are characterized as progressive and widespread (Groher & Crary, 2010), and reflect a weakness across the muscle groups used to prepare and transport a bolus (Crary & Groher, 2003). Early in the course of the disease, dysphagia may be characterized by oral limitations resulting from lingual weakness, such as difficulty chewing solid food, loss of food or liquid from the lips, pooling of food between gum and cheek, and difficulty moving food into the throat (Crary & Groher, 2003; Mitsumoto, 1994). As weakness in the swallowing mechanism progresses, patients may experience delay in triggering the pharyngeal swallowing response, reduced pharyngeal contraction, reduced laryngeal elevation, liquid regurgitation into the nose, stasis in the valleculae and pyriform sinuses, and pharyngeal residue (Corbin-Lewis, Liss, Sciortino, 2005; Logemann, 1998). Eventually, swallowing may trigger coughing to clear the airway of bolus residue (Mitsumoto, 1994). Coughing is a protective mechanism which is induced when sensory receptors detect penetration of the laryngeal vestibule. As the disease progresses and the cough reflex is weakened, there is an increased risk of aspiration of food and saliva into the airway.

Drooling, or sialorrhea, is also a common consequence of ALS, which can be severely disabling and embarrassing for individuals with the disease (Mitsumoto, 1994). Although patients with ALS do not experience increased salivary flow, they lack spontaneous automatic swallowing to clear excessive saliva in the mouth. Individuals with ALS display lingual weakness and consequently saliva cannot be collected and propelled into the pharynx. The lack of an efficient transport mechanism and excess saliva may result in aspiration of saliva into the airway (Gordon & Mitsumoto, 1997; Mitsumoto, 1994).

Conversely, patients with ALS may experience dry mouth, or xerostomia, which also presents problems when swallowing. Chronic open-mouth posture that results from weakening of the muscles that maintain jaw closure, or the inability to keep the lips closed, leads to an increase of breathing through the mouth, which causes the saliva to thicken due to evaporation (Newall, Orser, & Hunt, 1996; Robbins, 1987). Xerostomia is problematic for swallowing as there is less watery saliva available to mix with food to assist in bolus transport, thereby increasing the risk of pharyngeal residue. (Crary & Groher, 2003). Reduced salivary flow can also contribute to compromised oral and dental health.

Respiratory symptoms. ALS patients with either bulbar or spinal symptoms will eventually suffer from a progressive decline in respiratory functioning directly related to reduced respiratory muscle strength (Vitacca et al., 1997; Yorkston et al., 1995). There is no reliable method to predict when respiratory muscle weakness will occur and the rate of progression is variable between patients (Lechtzin, 2006). In the early stages of ALS, patients often report shortness of breath during physical exertion, frequent sighing, and excessive yawning (Mitsumoto, 1994). As the disease progresses, reductions in respiratory muscle strength become sufficient to cause patients to experience symptoms of respiratory insufficiency, including shortness of breath (dyspnea) despite no physical exertion, fear and anxiety of suffocation, and orthopnea (dyspnea that occurs when lying flat) (Lechtzin, Rothstein, Clawson, Diette, & Wiener, 2002). Additionally, many patients report restless sleep, morning headaches, lethargy, and mental confusion, which likely occur as a result of nocturnal hypoxemia (Lechtzin, 2006). The use of accessory respiratory muscles and paradoxical breathing is often indicative of diaphragm weakness, which is an inevitable consequence of ALS (Mitsumoto, 1994). Routine measurements of vital capacity while sitting and in supine position are used to detect diaphragmatic weakness in ALS patients.

Bulbar- and limb-onset ALS affect respiratory functioning in different ways. Bach (1993) states that those with limb-onset ALS will develop chronic alveolar hypoventilation resulting in respiratory muscle weakness as the disease progresses. However, as respiration is an integral component of airway clearance and swallowing, patients with bulbar-onset ALS experience respiratory impairment as a result of difficulty managing their airway secretions. A common cause of death in individuals with bulbar onset ALS is related to atelectasis, which occurs when the gas exchanging areas of the lungs become occluded, resulting in the collapse of alveoli that do not recover even after the obstruction is relieved (Curtis & Langmore, 1997). Atelectasis is a significant predisposing factor for the development of pneumonia, as it impairs alveolar cellular defenses and decreases the efficiency with which secretions are cleared. Regardless of whether patients present with limb- or bulbar-onset ALS, all patients eventually develop respiratory muscle impairment and most patients die from respiratory failure or pneumonia related to respiratory muscle weakness (Shaw, 2000; Tandan & Bradley, 1985).

Airway Protection and Swallowing in Healthy Individuals

Respiration and swallowing share the upper aerodigestive tract and are linked by their neuroanatomic relations in the medulla of the brainstem (Groher & Crary, 2010). The high degree of anatomical and neurological overlap between the two systems necessitates protective mechanisms to prevent airway compromise during swallowing (Miller, 1982, 1993). The relationship between the two systems is expressed functionally as precise coordination to ensure timing and movement of the oropharyngeal structures for laryngeal closure and lower airway protection (Wheeler-Hegland, Huber, Pitts, & Sapienza, 2009).

The structures and muscles that most significantly participate in both respiration and swallowing include the tongue, epiglottis, pharynx, cartilaginous larynx, and intrinsic and extrinsic laryngeal musculature (Curtis & Langmore, 1997). These structures interact through several mechanisms to minimize the risk of laryngeal penetration and/or aspiration. The first line of defense against aspiration involves laryngeal elevation, epiglottic depression, and glottic closure, all of which are normal functions of the larynx during swallowing. Laryngeal elevation serves to mechanically move the larynx out of the path of the descending bolus (Perlman & Christensen, 1997). As the larynx elevates, the downward displacement of the epiglottis towards the arytenoids occludes the laryngeal vestibule to deflect the bolus away from the airway and into the esophagus (Groher & Crary, 2010). Swallowing also triggers a reflexive closure of the glottis to prevent the aspiration of foreign materials into the respiratory tract. Strong adduction of the true vocal folds is supplemented by the closure of the false vocal folds and approximation of the aryepiglottic folds (Hadjikoutis, Pickersgill, Dawson, & Wiles, 2000).

As the passageway through which the bolus travels is shared for respiration, breathing must cease at the moment of the swallow (Groher & Crary, 2010). The coordination of respiration and swallowing is evident when respiration is inhibited during swallowing for 0.6 to 2.0 seconds (Preiksatis, Mayrand, Robins, & Diamant, 1992) (known as *swallow-related apnea)*, as well as a respiratory phase characteristic surrounding the apneic period (Martin, Logemann, Shaker, & Dodds, 1994). Immediately prior to the swallow, the laryngeal vestibule closes and the vocal folds adduct to allow subglottal pressure to increase (Gross, Mahlmann, & Grayhack, 2003). In most healthy individuals, a short exhalation cycle precedes the period of airflow inhibition (swallow apnea), which does not occur until the bolus collects at the vallecular level (Palmer & Hiiemae, 2003). The pharyngoesophageal segment (PES) relaxes and is pulled open by traction created by hyolaryngeal elevation. All neural firing to the posterior cricoarytenoid muscle is inhibited, followed by inhibitory signals to the diaphragm and other respiratory muscles, resulting in the cessation of respiration (Langmore, 2001). The apneic period continues for the duration of the swallow and is not dependent on glottic closure. As the tail of the bolus passes through the PES, the larynx descends and expiration continues slightly before the PES closes. The build-up of subglottic pressure that occurred prior to the apneic period serves to remove any residual swallowed material that remains in the upper airway. This burst of exhalation is considered a protective feature as it prevents any swallowed material from being sucked into the larynx or lower airways (Groher & Crary, 2010).

In the event that aspirated material penetrates the glottis, it is expelled primarily by cough and expiratory reflex, and mucociliary action within the conducting airways (Curtis & Langmore, 1997; Shapiro, Harrison, Kacmarek, & Cane, 1985). The mucociliary escalator lines the tracheobronchial tree and aides in airway clearance by trapping secretions and foreign materials. Mucus and foreign particles are consolidated into a cohesive mass and propelled toward the major airways and the trachea by the beating action of the cilia at a rate of 1-2 centimeters per minute (Shapiro et al., 1985).

Coughing involves the forceful expulsion of air from the lungs through the airways (Leith, 1977). Coughing can be initiated voluntarily and mediated by the cortex or it can be triggered reflexively at the level of the brainstem by afferent stimuli in the in the oropharynx, nasopharynx, larynx, and proximal segments of the lower respiratory tract (Curtis & Langmore, 1997). Coughing consists of three phases, including an inspiratory phase, a compressive phase and an expulsive phase (Leith, 1977). Air is drawn into the lungs to augment the force of the subsequent expulsive phase, enabling the clearance of mucus and material from the airways. The strength of the expiratory muscles, especially the diaphragm and abdominal muscles, is essential for the cough to be effective.

A second defensive mechanism is the expiratory reflex (ER), which is initiated by mechanical or chemical irritation of the vocal folds or trachea (Widdicombe & Fontana, 2006). The ER response is similar to coughing as it consists of the closure of the glottis, followed by glottal opening and an expulsive phase. However, the ER is characterized by forced expiratory effort without the inspiratory phase of a cough (Bolser, Poliacek, Jakus, Fuller, & Devenport, 2006). The expiratory effort for an ER is related to the recoil forces of the lungs-thorax unit, which is dependent on the volume of air in the lungs, and the strength of the expiratory muscles. Therefore, when patients can adjust their breathing to take in slightly greater volumes of air prior to a swallow, the strength of their ER will increase in the case of aspiration.

If foreign material reaches the alveoli and respiratory bronchioles, it must be dealt with by cellular mechanisms, as mucociliary clearance and cough are not effective at that level of penetration (Curtis & Langmore, 1997). Through these cellular mechanisms, the alveolar macrophages that protect the alveoli stimulate phagocytosis, or particle ingestion, whereby inhaled or aspirated pathogens are carried to regional lymph nodes where immune responses are initiated.

Compromised Airway Protection and Swallowing in ALS

In patients with ALS, coordination between the laryngeal protective system and the bolus transport system is lost during voluntarily initiated swallowing, resulting in an increased risk of airway compromise. According to Hadjikoutis and Wiles (2001), triggering the swallowing reflex during a volitionally controlled swallow is delayed and eventually abolished in patients with ALS. Furthermore, the larynx often fails to move anteriorly and superiorly during swallowing such that complete closure of the airway entrance is impaired, increasing the risk of laryngeal penetration and aspiration (Logemann, 1998). Reduced tongue force, delayed triggering of the swallowing reflex and weak elevation of the hyoid and larynx, result in reduced opening of the upper esophageal sphincter, with retention of saliva, food and liquids in the valleculae and pyriform sinuses (Ertekin et al., 2000). In addition, the cricopharyngeal sphincter muscle can become hyperreflexive and hypotonic, especially in patients with pseudobulbar palsy.

The coordination of respiration and swallowing is often impaired in ALS. In healthy adults, the expiratory-apnea-expiratory (ex-ex) pattern of breathing is observed in 71% to 100% of individuals (Martin-Harris, Brodsky, Price, Michel, & Walters, 2003; Martin-Harris et al., 2005; Shelley, Flack, Ellis, & Brooks, 1989a; Wheeler-Hegland et al., 2009). However, patients with ALS frequently display an abnormal respiratory pattern during swallowing characterized by inspiration, rather than expiration, following the apneic period. In a study performed by Hadjikoutis et al. (2000), twenty-two normal subjects and twenty-two patients with neurological disorders were studied to determine if specific lesions resulted in abnormal breathing patterns during swallowing. The authors found that patients with neurological disorders were more likely to display an abnormal pattern of inhalation, rather than exhalation, after swallowing compared to normal subjects. Furthermore, patients with corticobulbar involvement were more likely than patients without corticobulbar involvement to display post-apnea inspiration. Hadjikoutis et al. also studied thirty-two patients with motor neuron disease and found that these patients more frequently displayed an abnormal pattern during swallowing characterized by inspiration after swallowing, prolonged swallow apnea, and multiple swallows per bolus.

Hadjikoutis et al. (2000) hypothesized that under normal conditions, swallowing will inhibit respiration; however, if there is an urgent need for both swallowing and breathing, respiration will prevail and deglutition will be interrupted to allow airway needs to be met. The maintenance of ventilation is particularly important in conditions where the ability to breathe is limited as a result of repeated swallows and prolonged swallow apneas. Furthermore, the authors hypothesize that the loss of corticolbulbar fibers may reduce the inhibition of inspiration during swallow apnea and increase the likelihood of inspiration.

Other respiratory aspects of swallowing can also be negatively affected in ALS. Nozaki et al. (2008) reported that swallow apnea was increased in patients with ALS and that those who aspirated or had severe respiratory limitations displayed the longest apnea durations. Furthermore, the progressive respiratory deficiency observed in ALS causes muscular weakness and decreased voluntary and reflexive coughing (Strand, Miller, Yorkston, & Hillel, 1996). A forceful cough is crucial to enable clearance of aspirated food or saliva. Typically, a peak cough flow greater than 160L/min is necessary to clear airway debris (Boitano, 2006). When peak cough flow decreases below 3L/s, patients with ALS are at risk for impaired airway clearance (Lechtzin, 2006).

Consequences of Dysphagia in ALS

Dysphagia results in severe medical and psychosocial consequences for individuals with ALS. As eating becomes more of a struggle and the time taken to eat meals becomes longer, overall food consumption typically drops (Carr-Davis, 1994), resulting in weight loss, dehydration, malnutrition, and aspiration pneumonia. Weight loss can occur as a result of muscle atrophy or as a result of reduced dietary intake secondary to dysphagia or loss of appetite (Mitsumoto, 1994). As the disease progresses, extensive diet modifications are required. Despite these interventions, there is a high risk of rapid weight loss (*ALS cachexia)* and further nutritional decline as patients lose 30 to 50 percent of their body weight (Norris & Denys, 1979).

Dysphagia may also lead to severe dehydration when patients with an impaired swallowing mechanism avoid fluid consumption in an effort to reduce their risk of coughing or choking (Zachary & Mills, 2000). Dehydration can cause increased mental confusion, and generalized organ system failure. When dehydration is coupled with malnutrition, a decline is muscle strength is typically accelerated (Lechtzin, 2006). Arora and Rochester (1982) found that food deprivation results in a reduction in the diameter of the diaphragm's muscle fibers, which leads to a decrease in maximum diaphragm strength, endurance, maximum inspiratory and expiratory pressures, and maximum voluntary ventilation. Malnutrition can also adversely affect energy levels, thus impacting the ability to sustain sufficient effort for safe and effective eating and swallowing. In cases of severe or chronic malnourishment, the immune system may become compromised, as evidence by impaired cell mediated immunity (McMurray, Loomis, Casazza, Rey, & Miranda, 1981) and decreased alveolar macrophage phagocytic activity (Moriguchi, Sonic, & Kishino, 1983). Consequently, patients are more susceptible to aspiration pneumonia, with contributing factors associated with poor oral hygiene and uncoordinated swallowing and respiratory systems.

Irregularities in the coordination of the two systems can lead to aspiration or inhalation of foreign material into the larynx and lower respiratory tract. Laryngeal penetration is the entry of foreign material into the larynx proximal to the true vocal folds, whereas aspiration is the passage of material into the lungs (Crary & Groher, 2003). Aspiration of large solids can lead to upper airway obstruction and cause asphyxiation and death if the obstruction is severe (Hadjikoutis & Wiles, 2001).

When smaller volumes of aspirated solids pass through the larynx and lodge in the bronchi, patients are likely to develop aspiration pneumonia. Silent aspiration may also occur and results when a patient inhales food or gastric material without any noticeable, outward signs such as coughing (Groher & Crary, 2010).

Poor oral hygiene and difficulty managing secretions can also increase a patient's likelihood of developing pneumonia. According to Hadjikoutis and Wiles (2001), a patient's poor oral hygiene is due to an inability to adequately clean his or her teeth, food residues associated with poor tongue functioning, reduced oral food and fluid intake, and the pooling of saliva in valleculae and pyriform fossae. These factors, along with a tendency to inhale after swallowing, cause patients to be more susceptible to oropharyngeal colonization by potential respiratory pathogens (Hadjikoutis et al., 2000). Consequently, patients have an increased likelihood of developing pneumonia. Aspiration pneumonia can be a severe illness that may lead to death, especially in patients with ALS who are medically compromised and have a weakened immune system.

Dysphagia can also result in significant, negative psychosocial consequences for individuals with ALS (Ekberg, Hamdy, Woisard, Wuttge-Hannig, & Ortega, 2002). Given the poor prognosis of the disease, patients may become depressed and uninterested, or unwilling to eat. Changes in dietary consistency to compensate for dysphagia may also contribute to a reluctance to eat and feelings of discontent. Furthermore, patients who experience drooling, or a fear of overt choking episodes may prefer to eat alone and avoid social eating occasions, often leading to social isolation and an accompanying depression. Spouses and family members are equally affected because of the potential social limitations dysphagia may precipitate. The loss of upper extremity function can also be frustrating for patients as they often lose the ability to feed themselves and become more dependent on their caregivers. Dependency for feeding not only contributes to reduced self-esteem, but is a key factor which predisposes individuals to aspiration pneumonia (Langmore et al., 1998).

Neuromuscular Control of Swallowing and Respiration

Traditionally, swallowing has been viewed as a rigid and fixed motor activity, mediated primarily by brainstem reflexes (Corbin-Lewis et al., 2005). Lower brain regions mediated by central pattern generators have been thought to control aspects of swallowing that are stable across people and swallowing conditions. However, even these relatively stable aspects can be influenced by peripheral sensory input or input from higher brain regions, suggesting that many of the swallow components are not as rigid and fixed as once thought. This flexibility in motor functioning is highly adaptive, as it enables any motor goal to be accomplished in variety of different ways. This ability is referred to as *motor equivalence*.

Current conceptualizations view swallowing as a dynamic process controlled through a complex organization of specific regions in the cerebral cortex and brainstem. Kennedy and Kent (1988) theorize that swallowing takes place at three different levels of the nervous system. The first level of control is responsible for the reflexive and involuntary aspects of swallowing. It includes the brainstem swallowing center, which contains the central pattern generator, and a peripheral level that is linked to afferent bolus characteristics. The subcortical structures, including the basal ganglia, hypothalamus, amygdala, and tegmental area of the midbrain, represent the second level of control. The subcortical structures organize and execute learned patterns of motor activity to accomplish the complex act of swallowing. The surpabulbar cortical swallowing centers represent the third level of control and are responsible for modifying aspects of swallowing as a result of volitional commands and/or perceived changes in the need to modify feeding behaviour.

Given the high degree of coordination and anatomical overlap between respiration and swallowing, it is not surprising that these two functions share areas of control at the level of the central nervous system (Wheeler & Sapienza, 2005). The regions responsible for coordinating respiration and swallowing lie within the brainstem (Saito, Ezure, Tanaka, & Osawa, 2002). Although swallowing and respiration are controlled by distinct neural networks, the networks share neurons. Neural transmissions that control respiration and swallowing occur in a turn-taking fashion to ensure that these processes do not overlap. Finely tuned neural transmission is essential given the high degree of anatomical overlap between respiration and swallowing (Hiss, Strauss, Treole, Stuart, & Boutilier, 2003).

Brainstem control and afferent input. The neurons in the brainstem that are involved in swallowing lie mainly in the dorsal region above the nucleus of the tractus solitarius (NTS), and in the ventral region around the nucleus ambiguus (NA) (Wheeler & Sapienza, 2005). The two regions are represented bilaterally and are highly interconnected such that either side alone can coordinate the pharyngeal and esophageal phases of swallowing (Miller, Bieger, & Conklin, 1997). The NTS, a second set of interneurons located in the NA, and several cranial nerve nuclei (V, VII, IX, X, XI, XII) compose the swallowing central pattern generator (CPG). The swallowing CPG coordinates the contraction of 25 pairs of muscles in the oropharynx, larynx, and esophagus that are responsible for laryngeal elevation/tilting, swallowing apnea, pharyngeal constriction, tongue base retraction, and relaxation of the PES. The NTS is also the location of the respiratory CPG (Wheeler & Sapienza, 2005). Although specific connections between respiratory and swallowing neurons have not been identified, researchers hypothesize that a swallow-respiratory CPG may exist that integrates sensorimotor information from both systems to generate the swallow-respiratory patterns observed during normal swallowing. Wheeler and Sapienza (2005) propose that the coordination between swallowing and respiration is the result of an integrated motor pattern which generates a respiratory rhythm that is specifically intended for execution with a swallow.

The dorsal group of neurons within the NTS are involved in the triggering, shaping, and timing of the swallow (Jean, 2001). Afferent inputs are critical to the control of normal swallowing as they assist in initiating swallowing, modify the threshold for a pharyngeal swallow, and alter the level of muscle recruitment during swallowing (Miller, Vargervik, & Phillips, 1985). For example, many areas within the aerodigestive tract can trigger a pharyngeal swallowing response when provided the appropriate type, intensity, and duration of stimulation. Tactile, pressure, or liquid stimulation of the receptors on the fauces, tonsils, soft palate, base of the tongue, posterior pharyngeal wall, and anterior surface of the epiglottis will initiate the pharyngeal swallow reflex (Miller et al., 1997); however, the most potent trigger for stimulating a swallow is carried by the superior laryngeal nerve, a branch of the vagus nerve (Mistry & Hamdy, 2008). Furthermore, the NTS receives afferent inputs from several cranial nerves regarding mechanical (tactile, proprioceptive, tension), thermal, and chemical (gustatory) information to the NTS (Miller et al., 1997).

Afferent information regarding swallowing and airway protection is collected by respiratory and laryngeal mechanoreceptors and relayed to the NTS (Wheeler & Sapienza, 2005). Pulmonary stretch receptors, J-receptors in the lungs, muscle spindle fibres and golgi tendon organs in the respiratory muscles are types of respiratory mechanoreceptors that send afferent information regarding lung volumes to the NTS via the glossopharyngeal (IX) and vagal (X) cranial nerves. Laryngeal mechanoreceptors, which are sensitive to subglottic pressure and the presence of foreign materials in the airway, relay information to the NTS through the vagus nerve. Specifically, the superior laryngeal nerve relays sensory information from receptors above the level of the true vocal folds, and the recurrent laryngeal nerve relays sensory information from the inferior surface of the true vocal folds and trachea. The NTS then integrates the afferent information from the respiratory and laryngeal mechanoreceptors (Miller, 1999).

Both the ventral swallowing group (VSG) and ventral respiratory group (VRG) are located within the NA (Wheeler & Sapienza, 2005). The dorsal neurons of the NTS activate a group of ventral-lateral neurons around the NA, which distribute swallowing and respiratory pattern commands to motor neurons. Specifically, the pre-motor plan is relayed to the NA, where a programmed motor response is generated and sent to the cranial nerves that coordinate the muscles required to generate a swallow (Miller, 1999). The efferent (motor) functions that carryout the respiratory pattern plans are accomplished via cranial nerves (CNs) IX, X, XI, and XII (Benditt, 2006) and the motor response for swallowing is carried out via the motor nuclei of CNs V, VII, IX, X, XII, which control more than forty paired muscles (Groher & Crary, 2010).

Subcortical and cortical control. Both respiration and swallowing can be brought under cortical control (Wheeler & Sapienza, 2005). Functional magnetic resonance imaging (fMRI), positron emissions tomography (PET), magnetoencephalography (MEG), and transcranial magnetic stimulation (TMS) have identified the motor and somatosensory cortices as important areas for control of respiration and swallowing. Furthermore, motor association areas, limbic structures, the thalamus, and basal ganglia have demonstrated increased activation during volitional swallowing and respiratory tasks. As a result, researchers hypothesize that connections may exist between distinct respiratory and swallowing cortical areas.

Critical brain regions involved in swallowing include the cerebral cortex, basal ganglia and thalamus, and the cerebellum (Corbin-Lewis et al., 2005). Collectively, these swallow regions are thought to be instrumental in continuing, modifying, and monitoring swallowing activity, as well as responding appropriately to sensory stimuli. The cortex plays a significant role in swallow initiation, as well as in the neuromuscular control of swallowing (Miller et al., 1997). Although the cortical representation for swallowing is multifocal and bilateral, there is hemispheric dominance for some swallowing tasks (Mosier, Liu, Maldjian, Shah, & Modi, 1999). Regions of the cerebral cortex identified as active participants during swallowing are the anterior insular cortex with connections to the primary and supplementary motor cortices, orbitofrontal operculum, and the medial and superior portion of the anterior cingulate gyrus (Mosier et al., 1999). Other cerebral loci that are associated with the volitional control of swallowing include the cingulate cortex, insula, inferior frontal gyrus, premotor cortex, anterolateral and posterior parietal cortex, basal ganglia, thalamus and cerebellum. Using fMRI during swallowing, Mosier and Berenznaya (2001) confirmed cortical activation of sensorimotor areas (primary motor, M1, and primary sensory, S1, and supplementary motor area, SMA), secondary sensory areas (S2), premotor cortex, posterior parietal cortex, cingulate gyrus, inferior frontal gyrus, cerebellum, insular cortex, auditory cortex, corpus callosum, basal ganglia and the thalamus. They identified the functional connections between these structures and reported the function of each group, which are outlined in Table 1.

Table 1

Summary of cortical swallowing modules, derived from Mosier and Bereznaya, 2001.

Module	Components	Function
1	M1, S1, SMA, cingulate gyrus	Sensorimotor integration, planning, and output
2	Premotor cortex, parietal cortex	Object (bolus) sensory integration, motor planning and implementation of movements
3	Inferior frontal gyrus, S2, corpus callosum, basal ganglia, and thalamus	Integrate internal properties of the bolus with internal representation of swallowing movements
4	Cerebellum	Online coordination of swallow muscles to align internal representation of swallowing movements
5	Insula	Reciprocal connections with other regions; control of the sequential aspects of movement

Based on fMRI findings, Mosier and Berenznaya (2001) identified two parallel loops between the five groups, which represent connections between units that integrate motor and sensory information to accomplish safe and efficient swallows. The feedback loops among these higher structures supply their information to the nuclei in the brainstem via interneurons. Loop 1, the *insular loop*, includes modules one, two and five and is responsible for planning sequential movements, as well as for parietal processing and interpretation. Loop 2, the *cerebellar loop*, consists of modules one, three and four and integrates and matches internal representations of swallowing movements with sensory properties of the bolus. Module 1 is an integral part of each loop and likely serves as a sensorimotor input/output convergence area.

Like swallowing, respiration can also be brought under volitional control which recruits multiple cerebral centers. Voluntary control of respiration, for tasks such as breath holding or speech, involves the contralateral primary motor strip, as well as the thalamus for control of inspiratory and expiratory muscles (Wheeler & Sapienza, 2005). Control of respiration related to humor, sadness, or fear recruit limbic areas of the brain, such as the insula, frontal operculum, and anterior cingulate gyrus. Sensory input also plays a role in the cortical control of respiration. For example, the conscious awareness of respiration and the sensation of breathlessness, as well as the resulting increased breathing are mediated by the amygdala, hippocampus, and orbitofrontal cortices.

Framework for Respiratory and Swallowing Interventions in ALS

In treating patients with ALS, it is important to consider three potential factors that can contribute to neuromuscular respiratory failure, including (1) inability to ventilate, (2) impaired airway clearance, and (3) impaired airways protection and dysphagia (Benditt, 2006). This triad of interrelated factors not only contributes to neuromuscular failure, but also significantly compromises the patient's quality of life.

As ALS progresses, respiratory muscle weakness and chest wall stiffness cause an increased load on the respiratory muscles, resulting in decreased pressure levels and flow during inspiration and expiration (Benditt, 2006). This imbalance leads to fatigue, restricted lung volume and impaired gas exchange. Expiratory and inspiratory muscle weakness, in addition to upper-airway muscle weakness, contributes to difficulty clearing the airway of secretions. An inability to clear secretions can encumber the upper airway, resulting in mucous plugging and atelectasis, which may cause discomfort, nausea, coughing, gagging, and a choking sensation (Andersen, Grönberg, Franzen, & Funegard, 2001). Furthermore, an inability to clear secretions may result in aspiration of saliva into the airway. Approximately 90% of instances of respiratory failure in ALS occur concurrent with pneumonia caused by poor secretion management and the inability to effectively clear the airway (Lahrmann, Wild, Zdrahal, & Grisold, 2003).

Rehabilitative treatment approaches, such as compensatory swallowing strategies and respiratory interventions, have the potential to prevent this cycle of inter-related respiratory conditions, thus helping to preserve functional abilities and prevent respiratory failure (Miller et al., 1999, 2009). For example, Crary and Groher (2003) found that individuals are capable of volitionally interrupting or modulating the swallowing motor sequence. This results from the ability of higher cortical centers to influence the motor programming swallowing circuitry of the brainstem. The combination of flexible motor programming and the ability to volitionally override and modulate aspects of swallowing and respiration, supports the role of voluntary behavior modification in restoration of swallowing function.

Management of Dysphagia in ALS

Maintaining a healthy body weight is important in patients with ALS as it helps to prevent infection, sustain respiratory health, and generally keep the body strong and at its highest functioning level (Kasarskis, Berryman, Vanderleest, Schneider, & McClain, 1996). Most therapeutic strategies for patients with dysphagia are designed to minimize the instances of aspiration, while improving quality of life (Swigert, 2007). Behavioural management to improve dysphagic symptoms includes the use of compensatory strategies such as diet modification, increasing oral stimulation, posture correction, and swallow maneuvers.

Diet modification is particularly effective when patients have mild or moderate dysphagia. Patients are often advised to avoid small, dry, crumbling food and instead eat soft and smooth food such as pureed items that are easier to consume (Carr-Davis, 1994). Liquid supplements are helpful in early stages when choking is less of a risk (Wagner-Sonntag et al., 2000). As the disease progresses, thickened fluids are better tolerated so powder thickeners are often added to drinks. Dieticians can give advice on how to enrich meals by use of foods high in calories, proteins and vitamins, and by the addition of high-energy supplements (Carr-Davis, 1994). In addition to a modified diet, patients may eat slower and use other approaches to aide in food consumption (e.g., chewing solids to mush before swallowing).

Swallowing abilities can also be improved by increasing oral stimulation through presentation of a sensory stimulus (Logemann, 1998). These techniques emphasize the taste, temperature and texture of the bolus to enhance the swallowing reflex. For example, sensation can be enhanced by keeping liquids cold, while texture enhancement is achieved through carbonated liquids (Strand et al., 1996). These techniques are designed to heighten oral awareness and provide afferent information to the cortex and brainstem to trigger the swallowing reflex more rapidly (Logemann, 1998).

Postural changes and swallowing maneuvers also aid in reducing the risk of aspiration. Tilting the head backwards can help patients with impaired tongue movements, but intact pharyngeal swallowing to guide the bolus into the pharynx (Wagner-Sonntag et al., 2000). The chin-tuck technique is effective if patients have difficulty triggering the swallowing reflex, reduced tongue base retraction, and/or premature spilling of bolus material into the pharynx (Logemann, 1998). The technique involves tilting the head forward and tucking the chin down, which forces the tongue base and epiglottis closer to the posterior pharyngeal wall, thus narrowing the airway entrance and reducing the risk of aspiration.

Like postural adjustments, swallow maneuvers bring specific aspects of swallowing under voluntary control to improve airway protection during swallowing

(Logemann, 1998). For example, the supraglottic swallow maneuver requires patients to hold their breath while swallowing, followed by a forceful exhale immediately afterwards, thereby expelling food or secretions and preventing aspiration. Exhalation after swallowing is especially important to clear the airway, yet post-swallow expiration may be impaired in individuals with ALS.

Swallowing with an expanded chest is a maneuver which enhances subglottal pressure to promote expiratory airflow following the apneic period of swallowing (Wheeler-Hegland et al., 2009). Specifically, encouraging patients to initiate their swallows with higher lung volumes (i.e., volumes above functional residual capacity) and to swallow with an expanded barrel chest posture is expected to capitalize on their ability to volitionally control and modify their swallow. Wheeler-Hegland et al. hypothesized that the mechanism by which the barrel chest maneuver works is associated with the recoil forces generated by the lungs-thorax unit. When individuals initiate the swallow apnea with a slightly expanded state of the lungs-thorax unit, the lungs-thorax unit will naturally recoil back to rest position. The presence of positive recoil forces enhances subglottal pressure and ensures expiratory airflow with a strong expulsion of any bolus material, if necessary. Thus, in patients with ALS where airway compromise is encountered during deglutition, swallowing with slightly higher lung volumes may promote post-apneic expiration and enable the patient to quickly initiate a more effective ER response and expel residual material.

Pilot Studies

Two research studies have yielded results to support the use of swallowing with an expanded chest, or the “barrel chest” technique, to promote expiratory breathing after swallowing in patients with ALS and dysphagia. Cleary, Kalra, and Johnston (2007) conducted an “N of 1 Randomized Controlled Trial (RCT)” with a

77 year old patient with bulbar-onset ALS and cognitive dysfunction. The patient participated in an experimental and placebo treatment. The experimental treatment involved explicit instruction in swallowing with an expanded “barrel” chest posture and the patient was cued to recruit accessory muscles of inspiration prior to swallowing. Placebo treatment consisted of laryngeal adduction and voicing exercises. A Vernier, 3-axis accelerometer and respiration monitor belt were used to measure respiratory cycle during swallowing. Following active treatment, the patient exhibited a more typical swallow-respiratory pattern. During the baseline phase, expiration was observed in only 13.8% of the trials; however, following active treatment, expiratory breathing occurred in 76.4% of trials on average. Following placebo treatment, expiration after swallowing occurred in 7.7% of trials on average. In addition, the patient rated his psychological distress as less severe and less frequent, as measured by a subsection of the Swallowing Quality of Life Scale (SWAL-QOL).

Similar results were obtained in an unpublished study conducted by Cleary, Costar, and Prior (2009). An 81 year old male with progressive, bulbar-onset ALS, cognitive impairment, dysarthria, respiratory compromise, and a fear of eating due to dysphagia, participated in a single-subject experimental design. Respiratory cycle during swallowing was measured using a Vernier, 3-axis accelerometer and respiration monitor belt. The subject demonstrated on average a normal swallowing pattern (exhalation post-swallow) in 27% of his swallows in the pre-treatment phase, and 22.67% of his swallows in the placebo condition. Following active treatment, the patient exhibited a normal swallow pattern in 94.33% of his swallows, and was able to maintain the pattern in 67% of his swallows in his post-treatment follow-up on the same day. The authors concluded that the patient was able to learn a volitional breathing technique that facilitated a normal swallow-respiratory pattern.

Purpose of the Current Study

Patients with ALS have been found to display alterations in the otherwise highly stable patterns of respiration during swallow, which may predispose them to penetration and/or aspiration leading to airway compromise. Although strong clinical and theoretical rationales exist for actively treating airway protection and clearance in the end-of-life symptom management of individuals with ALS (Lechtzin, 2006; Lechtzin et al., 2002; Benditt, 2006), there is limited research evidence to support behavioral swallowing therapy to improve airway protection in patients with ALS (Robbins, 2006). A few small treatment studies have been conducted in ALS (Ertekin et al., 2000; Kawai et al., 2003; Kidney, Alexander, Corr, O'toole, & Hardiman, 2004) but many swallowing treatments are largely unproven in managing this disease. Therefore, the purpose of the current study is to evaluate the effects of the barrel chest swallowing maneuver on breathing patterns and perceptions of swallowing in individuals with ALS.

Research Questions and Hypotheses

What is the effect of a behavioral swallowing technique, the barrel chest maneuver, on (a) the swallowing and respiratory phase relationship for patients with ALS, and (b) patients' perceptions of their swallowing and the treatment? We hypothesized that the barrel chest maneuver would promote expiration after swallowing, and that patients would perceive positive benefits of the maneuver on their swallowing.

Methods

Participants

The study was approved by the Health Research Ethics Board at the University of Alberta. Potential participants were identified through the University of Alberta Hospital ALS Clinic and the Misericordia Hospital.

Twenty individuals with ALS will participate in the larger study of which this thesis was a part. For this thesis, data were collected for 10 of the participants. The small number of participants in the study was appropriate for the purpose of the study and the goals of this stage of treatment outcomes research, which was to determine the specific nature of a treatment and its effects, and to engage in hypothesis testing (Robey & Schulz, 1998). Participants met the following inclusion criteria:

1. Had a diagnosis of ALS according to *The Revised El Escorial* criteria (Brooks et al., 2000) and were receiving services through the University of Alberta Hospital's ALS clinic, or the Misericordia Hospital;
2. Were fluent in English;
3. Presented with dysphagia (as diagnosed by a speech-language pathologist during routine clinic visits to the ALS clinic);
5. Had the ability to take some food and liquid by mouth even if they were using a percutaneous endoscopic gastrostomy (PEG) tube to meet their nutritional needs;
6. Had a bulbar score of 10 or less as measured by *The Revised ALS Functional Rating Scale* (ALSFRS-R);
6. Communicated using speech, handwriting or voice output assistive aid; and
7. Had the cognitive ability to follow simple directions.

Of the 10 participants with ALS recruited to participate in the study, data collected from two of the ten participants were not analyzed due to instrumentation limitations. Specifically, in the case of the first excluded participant, the respiration monitor belt had an air leak due to a faulty valve and thus we were unable to maintain sufficient pressure to accurately measure the participant's respiratory cycle. The respiratory data collected only captured a small range of the expiratory and inspiratory movements of the chest wall (see Figure 2). The truncated signal was difficult to analyze and did not yield high inter-rater reliability scores (i.e., 21% inter-rater reliability); therefore, the participant's data were excluded from the analysis. In the case of the second excluded participant, the respiration monitor belt was too short to wrap around the participant's chest. To compensate for the participant's large chest circumference, the respiration monitor belt was positioned higher on the chest (i.e., directly under the participant's armpits and across the sternum); however, an adequate fit was still not achieved and the chest strap could not be inflated to a sufficient pressure. As a result, the respiratory data collected showed minimal pressure changes during the respiratory cycle, which made it difficult to isolate the apneic periods and analyze the data (see Figure 2); therefore, the participant's data were excluded from the final analysis.

The final study sample consisted of 8 participants, 2 women and 6 men, ranging in age from 39 to 75 years (median = 64.50 = years, range = 36) with a median length of time since diagnosis of 10.0 months (ranging from 1 to 60). Four participants (50%) had the bulbar onset form of ALS and four had the limb onset form (50%). A summary of the participants' demographic characteristics are presented in Table 2.

Table 2

Sample Characteristics of the Participants: Demographic Information

ID	Age (years)	Gender	Site of onset	Time post diagnosis (months)
1	70	M	Bulbar	10
2	68	F	Bulbar	1
3	73	F	Bulbar	6
4	39	M	Limb	60
5	41	M	Limb	48
6	61	M	Limb	10
7	75	M	Bulbar	6
8	50	M	Limb	18
Median	64.50	-	-	10.0

Several measures were used to estimate disease severity of the participants, including *Revised ALS Functional Rating Scale (ALSFRS-R;* Cedarbaum et al., 1999) scores, forced vital capacity, and peak cough flow (see Table 3 for a summary). The ALSFRS-R is a questionnaire-based scale that measures a patient's ability to perform activities of daily living. The scale is used for both clinical and research purposes with patients who have ALS. The ALSFRS-R yields quantitative scores on Likert scales that are used to monitor the progression of the disease. In the current study, the participants' ALSFRS-R scores were obtained from their charts. The median ALSFRS-R score for the study sample was 31.50 ranging from 14 to 42 out of a total possible score of 48. Bulbar sub-scores of the ALSFRS-R were also included to characterize the overall degree of the participants' speech, salivation and swallowing impairment. The median ALSFRS-R bulbar sub-score was 9.00 ranging from 5 to 10 out of a possible score of 12 for this sub-section.

The participants' baseline forced vital capacity (FVC) and peak cough flow (PCF) were also used as measures of disease severity. Baseline FVC was compared to predicted normal values. Participants' FVC ranged from 30% to 89% of predicted normal values (median = 63%, range = 59) based on gender, age, height, and ethnic origin (Knudson, Slatin, Lewowitz, & Burrows, 1976). The median PCF rate was 221L/min. Overall, the majority of the participants were moderately impaired in terms of disease severity and were characterized as having mild-moderate respiratory insufficiency and mild-moderately severe bulbar symptoms at the time of the study.

Table 3

Sample Characteristics of Participants: Functional Status

ID	**FVC (% pred)**	**FVC (L)**	**PCF (L/min)**	**ALSFRS Total Score (/48)**	**ALSFRS Bulbar Score (/12)**
1	49	3.22	289	42	10
2	70	2.28	281	38	10
3	37	0.81	115	23	5
4	30	1.47	181	22	9
5	89	4.14	435	40	9
6	75	3.12	65	30	10
7	70	2.73	167	33	7
8	56	2.56	261	14	9
Median	63	2.65	221	31.5	9

The majority of the participants were living at home at that time of the study (n=7). One participant was living in a long-term care facility when the data were collected. Most participants (n=5) lived in urban settings (i.e., Edmonton and area) in Alberta (see Figure 1).

Figure 1. Geographic distribution of participants in Alberta

Research Design

A One-Group Pretest-Posttest Design (i.e., within-subjects experimental research design) was used to examine the effect of the treatment (i.e., the barrel chest maneuver) on swallowing and respiratory phase relationships as well as the participants' perceptions of their swallowing and the treatment. The primary outcome variable was the proportion of swallows that occurred during the expiratory phase of breathing (as compared to total swallows). The secondary outcomes were scores on the SWAL-QOL and the participants' responses to qualitative interview questions.

The use of a within-subjects experimental research design was considered appropriate for this thesis project as it was a Phase I treatment outcomes study. Based on work by Greenwald and Cullen (1985), Robey & Schulz (1998) adapted a five-phase model for structuring clinical outcomes research. In Phase I research, the goal is to develop and test hypotheses, establish the safety of a treatment, and determine the influence of the intervention and whether it is "active". In this phase, sample sizes are usually small and controls are not included. Appropriate research designs for

Phase I include single-subject experimental designs, case studies, small single group pre-post studies, and retrospective studies.

Procedures

Each participant completed a baseline, treatment, and post-treatment condition. Data were collected during the baseline and post-treatment phases. A summary of the data collection measures in the baseline and post-treatment conditions are outlined in Table 4. Also, the complete experimental protocol can be found in Appendix A.

Pre-treatment/baseline phase. All participants completed a series of tasks that were used to determine respiratory patterns during swallowing and their subjective impressions of their swallowing. The baseline phase included the following tasks:

1. At the beginning of the session, each participant ate a small amount of pudding (minimum of 10 swallows) and, immediately after, completed a short survey that included questions from the Mental Health subsection of the SWAL-QOL, as well as four questions from the Symptom Frequency subsection (see Appendix B for the survey data collection form in the baseline condition).
2. Following the completion of the survey, the respiration monitor belt was placed around the participant's chest.
3. To determine if a sufficient pressure in the respiration monitor belt had been obtained, 30 seconds of tidal breathing was collected from the participant. If the respiratory cycle was not deemed to be interpretable (i.e., lack of clear peaks and toughs), the pressure inside the respiration monitor belt was increased until a clear respiratory signal was obtained.

See Figure 2 for a comparison of the respiratory cycle data collected with and without adequate pressures in the respiration monitor belt.

4. Following 30 seconds of tidal breathing, the accelerometer was applied to the participant's neck (lateral to the thyroid prominence) and held in position by the experimenter. The participant was then asked to swallow boluses of tepid water presented in a plastic cup at a volume of 20 ml. Ten swallows of the 20 ml bolus volume were recorded for each subject. A 20 ml bolus size was chosen as it approximates the "typical" size bolus for healthy adults, which ranges from 16 to 26 ml (Adnerhill, Ekberg, & Groher, 1989). For each swallow, the pattern of breathing after each swallow apnea was recorded to determine the participant's baseline respiratory pattern during swallowing. Each swallow apnea was classified into one of two types depending on whether it was followed by expiration (xE) or inspiration (xI).

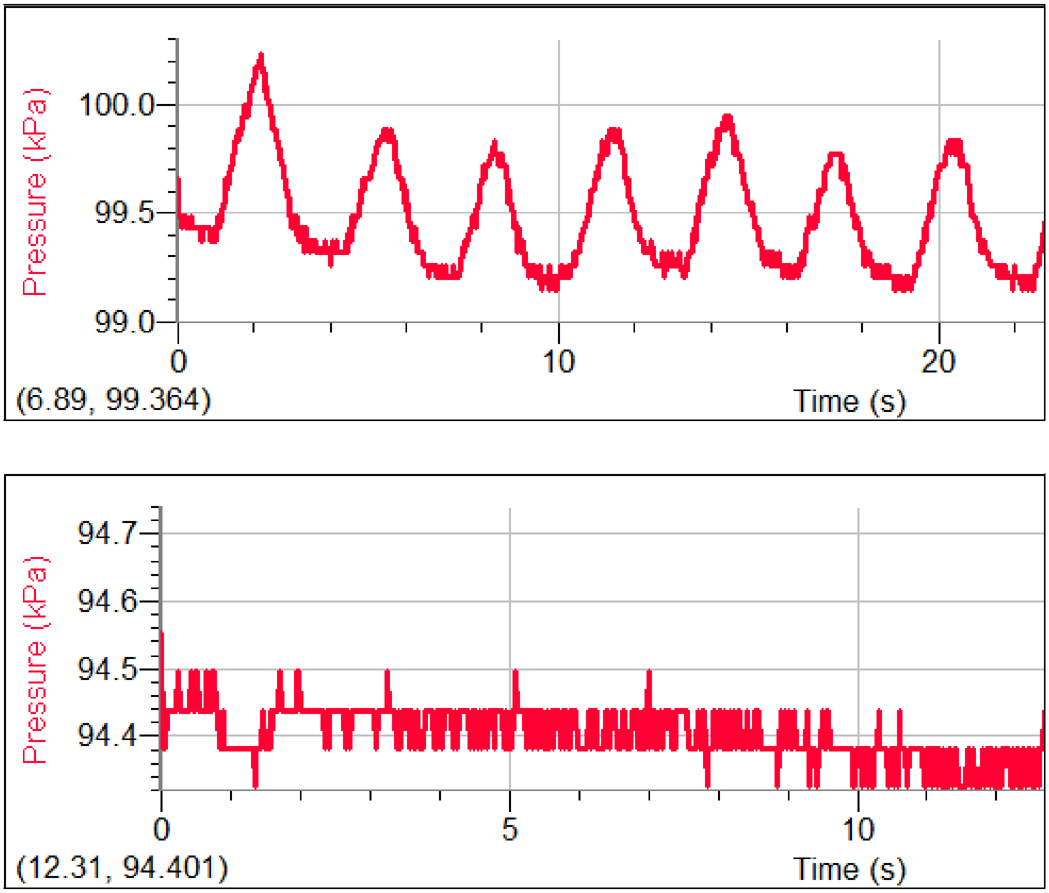

Figure 2. Respiratory phase data collected with adequate pressure (top) in the respiration monitor belt compared to inadequate pressure (bottom). Note the smaller range of pressure on the y-axis.

Treatment protocol. Participants in the treatment condition learned the behavioral swallowing strategy.

1. The patient was seated in a modified chair which included a fixed goniometer attachment and an adjustable back support to aid the participant in maintaining a 90 degree posture during the swallowing tasks.
2. The researcher first explained the technique to the participant (see Appendix A for the researcher's script). The patient was instructed to sit in an upright position, with shoulders touching the back of the chair

and feet firmly planted on the floor. They were then instructed to drink the 20 ml of water and, while holding the bolus in their mouth, they were cued to take a deep breath and hold it before swallowing. The patient was told by the clinician to swallow with an expanded “barrel” chest posture (i.e., cued to recruit accessory muscles of inspiration prior to swallow).

3. The researcher then modeled the technique for the participant three times during dry swallows.
4. The participant performed the technique with verbal cueing and physical prompts (as necessary) from the researcher. The participant practiced the technique during three dry swallows and two swallows with water.
5. When the researcher judged that the participant could correctly perform the treatment technique correctly, the participant rested for five minutes. If after 10 trials the participant was unable to perform the strategy correctly, data collection would have been discontinued with that individual.

Post-treatment test. Following the five minute rest period, the participant completed 10 liquid swallows using the technique learned during the treatment phase of the study. The participant then ate a small amount of pudding (as in the baseline phase), and answered questions from the Mental Health and Symptom Frequency subsections of the SWAL-QOL, followed by five qualitative interview questions. The participant continued to sit in the modified chair during the post-treatment phase of the study.

1. To ensure the respiration monitor belt had remained inflated to a sufficient pressure, 30 seconds of tidal breathing was collected from the participant. If the respiratory cycle was not deemed to be interpretable (i.e., lack of clear peaks and troughs), the pressure inside the respiration monitor belt was increased until a clear respiratory signal was obtained.
2. Following 30 seconds of tidal breathing, the accelerometer was applied to the participant's neck (lateral to the thyroid prominence) and held in position by the experimenter. The participant was then instructed to swallow 10 – 20 ml boluses of tepid water. The experimenter cued the participant to take a deep breath before swallowing and to swallow with a big barrel chest posture. The pattern of breathing was recorded and each swallow was categorized as followed by either expiration (xE) or inspiration (xI).
3. Following the completion of 10 swallows, the respiration monitor belt and accelerometer were removed from the participant.
4. The participant then ate a small amount of pudding (minimum of 10 swallows) using the technique demonstrated during the treatment phase of the study. They were given occasional verbal prompts and feedback regarding their posture and adherence to the treatment protocol.
5. Immediately after, the participant answered questions from the Mental Health and Symptom Frequency subsections of the SWAL-QOL, followed by five interview questions (see Appendix B for the post-treatment survey and interview data collection form).

Table 4

Data Collection Protocol

Baseline Condition	Post-Treatment Condition
1. Responses to five questions from the Mental Health subsection of the SWAL-QOL	1. Respiratory pattern during 10 swallowing trials. Each swallow was classified as either xE or xI depending on whether it was followed by expiration or inspiration, respectively
2. Responses to four questions from the Symptom Frequency subsection of the SWAL-QOL	2. Responses to five questions from the Mental Health subsection of the SWAL-QOL
3. Respiratory pattern during 10 swallowing trials. Each swallow was classified as either xE or xI depending on whether it was followed by expiration or inspiration, respectively	3. Responses to four questions from the Symptom Frequency subsection of the SWAL-QOL
	4. Responses to five qualitative interview questions

Instrumentation and Measures

To investigate participant's respiratory cycle while swallowing, a Vernier 3-Axis Accelerometer, Respiration Monitor Belt, and Logger Pro 3 Data Collection Software were used. Accelerometry is a non-invasive technique used to measure physiological vibration signals (Reddy et al., 1991). It contains three acceleration sensing integrated circuits that produce a signal on one of three outputs (X, Y, Z) while measuring acceleration (Vernier User Guide, 2006). When placed at the level of the cricoid cartilage on a patient's neck, the accelerometer can detect the epidermal vibration signals of the swallowing mechanism (Reddy et al., 1991). Using a single-axis accelerometer, Reddy et al. (2000) showed the relationship between the accelerometry signal and laryngeal elevation during swallowing. There was a significant correlation between the peak elevation of the larynx and the maximum magnitude of the acceleration signal; however, given that the larynx moves in two directions during swallowing (i.e., superior and anterior), dual-axis accelerometry is more likely to reflect the physiological movements that generate the output signals (Bech-Hansen Zoratto, 2009). Other options for measuring the onset of swallowing activity include cervical auscultation, or the use of a contact microphone, submental electromyography, or videofluoroscopy; however, the advantages of using an accelerometer to measure the movement of the hyolaryngeal complex include simplicity, cost, portability, and reliability. Furthermore, it is non-invasive and does not expose patients to harmful radiation.

A Vernier, 3-Axis Accelerometer (model # 3D-BTA) coupled with a battery operated Vernier Lab Pro transducer were used to measure the anterior and superior movement of the hyolaryngeal complex. Kinematic measures of chest wall circumference were obtained from a single nylon chest strap. A Vernier Respiration Monitor Belt (model # RMB), connected to a Vernier Gas Pressure Sensor GPS-BTA in conjunction with a Vernier LabPro transducer were used to measure participants' respiration rates and cycles. Both the accelerometer and the respiration belt interfaced with a battery operated Dell Latitude Laptop computer via LabPro software. The Logger Pro 3 Data Collection Software allowed the participants' respiratory phase (i.e., inspiration and expiration) and laryngeal elevation (i.e., swallow) to be simultaneously displayed across time (see Figure 3a and 3b). The onset of each swallow was identified by increased amplitude in the signal from the accelerometer. Once each swallow was located, the apneic period was identified as a point in the respiratory cycle that was relatively flat compared to cycles of tidal breathing. Following identification of the apneic period, the respiratory phase in which the swallow occurred was determined by visual inspection of the pressure signal from the respiration monitor belt. Post swallow inspiration and expiration were identified as upward or downward movement, respectively, of the respiratory signal following the apneic period. Subsequently, the percentage of "normal" swallows (expiration after each swallow) in relation to the total number of swallows in each condition was calculated from the data. The throat accelerometer and the chest strap were sterilized between uses with standard sterilization techniques (e.g., Metriguard Spray and alcohol swabs).

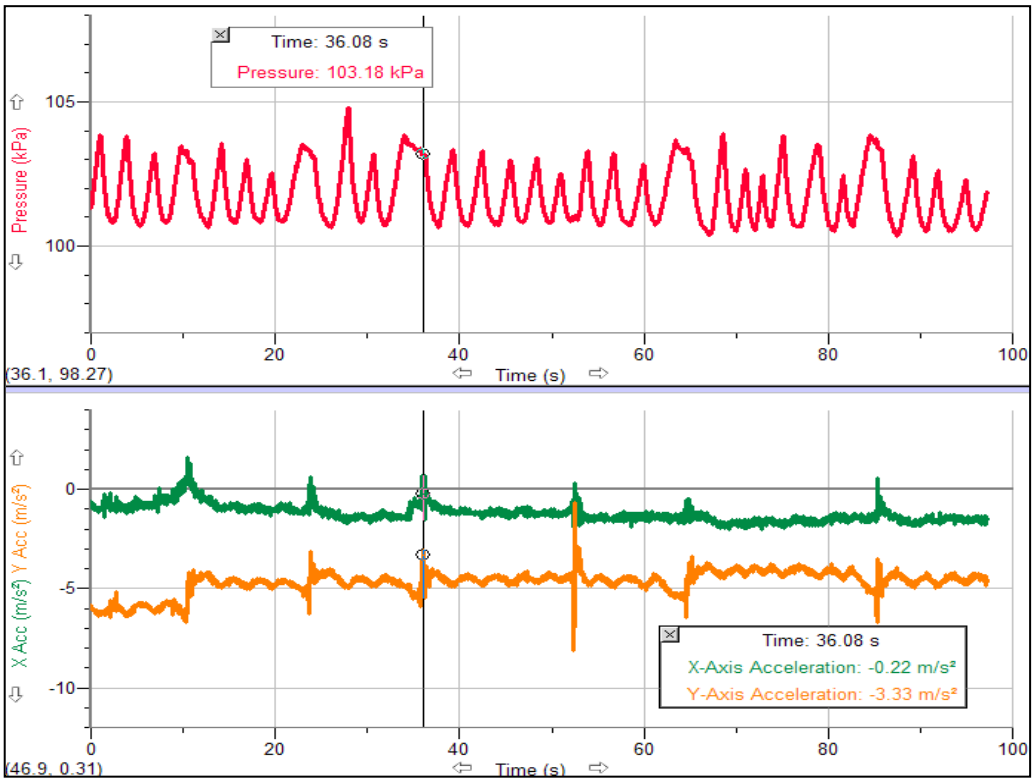

Figure 3a. Example of respiratory (top) and accelerometry (bottom) data collected simultaneously for a normal swallow pattern (expiration following the apneic period). Expiratory and inspiratory respiratory movements are represented by the downward and upward direction of the signal, respectively. The peaks in the accelerometry signal indicate hyolaryngeal movement during swallowing. The green line represents anterior movement of the hyolaryngeal complex and the orange line represents superior movement. Note the flat area in the respiratory signal (swallow apnea) that occurs shortly before the onset of hyolaryngeal elevation. The subsequent downward direction of the respiratory signal indicates post-swallow expiration.

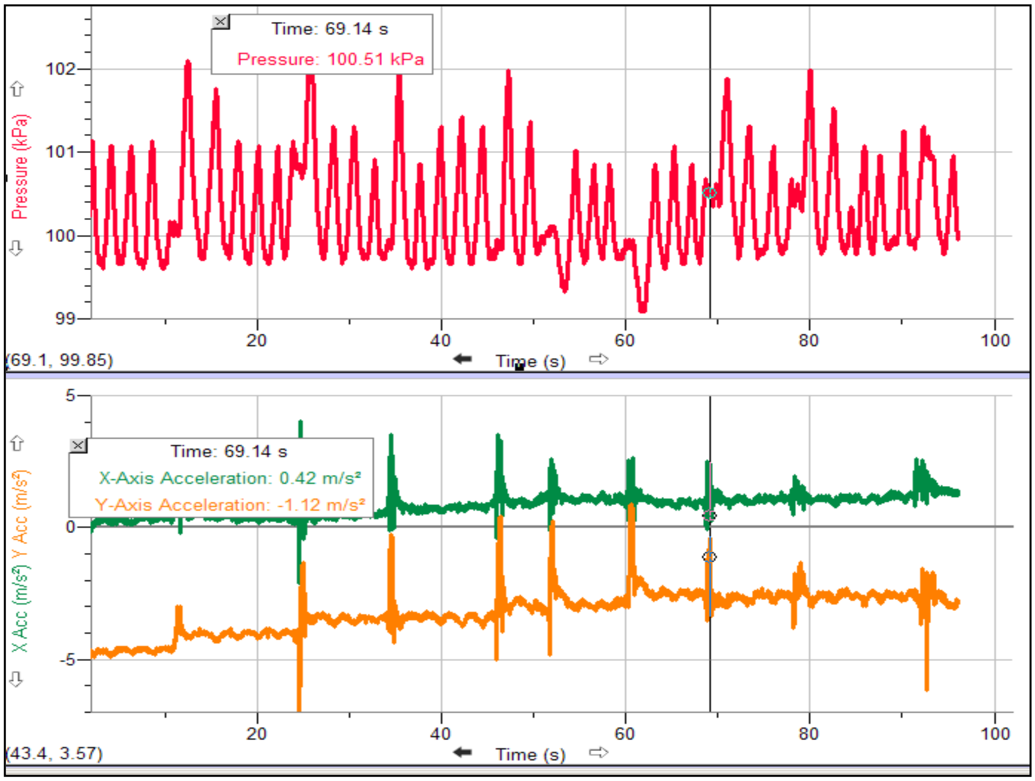

Figure 3b. Example of respiratory (top) and accelerometry (bottom) data collected simultaneously for an abnormal swallow pattern (inspiration following the apneic period). Expiratory and inspiratory respiratory movements are represented by the downward and upward direction of the signal, respectively. The peaks in the accelerometry signal indicate hyolaryngeal movement during swallowing. The green line represents anterior movement of the hyolaryngeal complex and the orange line represents superior movement. Note the flat area in the respiratory signal (swallow apnea) that occurs shortly before the onset of hyolaryngeal elevation. The subsequent upward direction of the respiratory signal indicates post-swallow inspiration.

To measure the effect of the treatment on patients' perceptions of their swallowing, five questions that comprise the Mental Health subsection and a portion of the Symptom Frequency subsection of the Swallowing Quality of Life Scale (SWAL-QOL) (McHorney et al., 2006) were administered (see Appendix B for the baseline and post-treatment survey questionnaires). The entire SWAL-QOL is a 44-item questionnaire that has been clinically validated. It assesses patient perspectives on mealtime-related quality of life across 10 domains including: Burden; Eating Duration; Eating Desire; Symptom Frequency; Food Selection; Communication;

Fear; Mental Health; Social; Fatigue; and Sleep. Perceptions relating to questions in each subsection are rated on a five-point Likert scale. A higher score indicates a better quality of life or more positive perspectives towards swallowing and eating. On the Mental Health subsection, the participants rated how often each statement was true for them during the pudding meal trial with the choices ranging from always true to never true.

A portion of a Symptom Frequency subsection was also used to assess the physical symptoms that participants' may experience as a result of their swallowing problem. The physical symptoms included coughing, choking, having to clear the throat, and food sticking in the throat. The participants rated how often they experienced each symptom during the pudding meal trial on a five-point Likert scale ranging from almost always to never.

Following the completion of the post-treatment phase of the study, the participants' also answered five qualitative interview questions to assess their perceptions of the treatment that they were instructed to use during the treatment and post-treatment phases of the study (see Appendix B for the post-treatment interview questions). The purpose of these questions was to assess the participants' comfort, confidence, and willingness to use the treatment technique during their normal eating routines, as well as their perceptions about the effectiveness of the treatment.

Results

Analysis

Research Question 1 – Effects of the technique on swallowing and respiratory phase relationships. To answer this question, the researchers counted the number of swallows that had the typical respiratory pattern (i.e., expiration following the apneic period, or xE). Participants' baseline and post-treatment respiratory phase graphs can be found in Appendix F. To analyze group data, the proportion of xE swallows in the baseline condition was compared with the proportion of xE swallows in the post-treatment condition using a Wilcoxon Signed Ranks Test (i.e., nonparametric equivalent of the paired t-test) with significance set at $p = .05$. Table 5 shows the number of swallows (out of a total of 10) with an expiratory pattern of breathing for each participant in the baseline and post-treatment conditions. Results of the Wilcoxon Signed Ranks Test indicated a significant difference between the baseline and post-treatment conditions ($z = 2.388$, N – Ties = 8, $p = .017$, two-tailed or $p = .0085$, one-tailed, $r = 0.50$) (see Appendix E for SPSS output).

Table 5

Number of swallows that were followed by the expiratory phase of breathing (xE) for each participant in the baseline and post-treatment conditions.

ID	**Baseline (# xE/10)**	**Post-treatment (# xE/10)**
1	3	6
2	10	10
3	2	10
4	9	10
5	9	10
6	7	10
7	9	10
8	4	10
Total	**53 (66%)**	**76 (95%)**

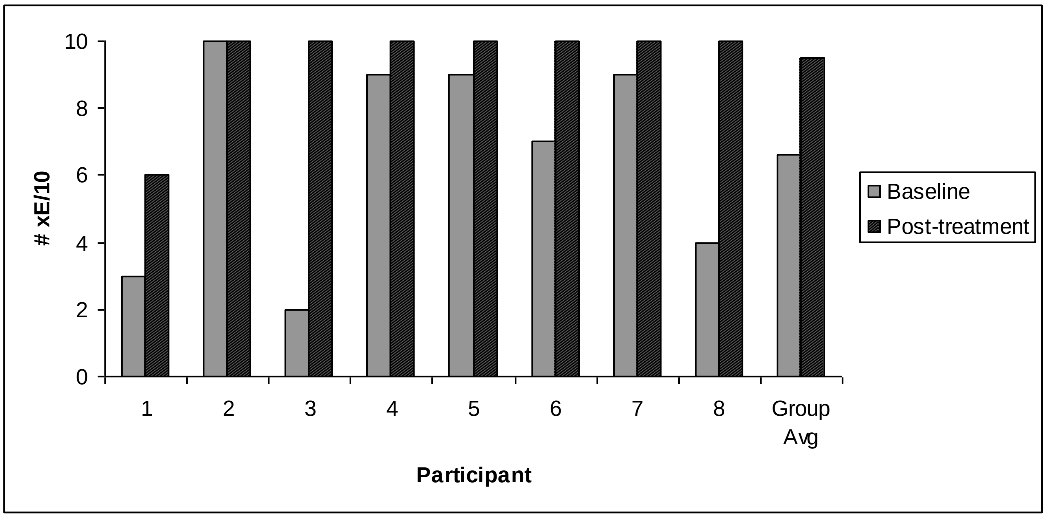

Figure 4. A comparison of the number of swallows that were followed by the expiratory phase of breathing in the baseline and post-treatment conditions for each participant.

The data (see Appendix F) were also analyzed in terms of the prevalence of each of the four possible respiratory patterns surrounding swallowing (i.e., ex-ex, ex-in, in-ex, in-in). Table 6 shows the number of swallows that occurred for each respiratory pattern in both the baseline and post-treatment conditions for each participant. Group data are summarized in Figure 5. For the majority of swallows in the baseline phase of this study, the respiratory pattern was ex-ex (53% of total swallows). The ex-in pattern was the next most frequently observed (23%), followed by the in-ex pattern (14%) and the in-in pattern (10%). In the post-treatment conditions, ex-ex was again the most common respiratory pattern observed during swallowing; however, it occurred more frequently in the post-treatment condition (74%) compared to the baseline condition (53%). Contrary to findings in the baseline conditions, in-ex was the next most frequently observed pattern (21%), followed by ex-in (4%), and in-in (1%).

Table 6.

Respiratory phase preceding and following each swallow for participants in the baseline and post-treatment conditions. There were a total of 10 swallows for each participant in both conditions.

ID	Condition	Ex-ex	In-ex	Ex-in	In-in
1	Baseline	3	0	5	2
	Post-treatment	1	5	3	1
2	Baseline	7	3	0	0
	Post-treatment	0	10	0	0
3	Baseline	0	2	2	6
	Post-treatment	10	0	0	0
4	Baseline	9	0	1	0
	Post-treatment	10	0	0	0
5	Baseline	7	2	1	0
	Post-treatment	8	2	0	0
6	Baseline	7	0	3	0
	Post-treatment	10	0	0	0
7	Baseline	5	4	1	0
	Post-treatment	10	0	0	0
8	Baseline	4	0	5	1
	Post-treatment	10	0	0	0
Total (#/80)	Baseline	42 (53%)	11 (14%)	18 (23%)	9 (10%)
	Post-treatment	59 (74%)	17 (21%)	3 (4%)	1 (1%)

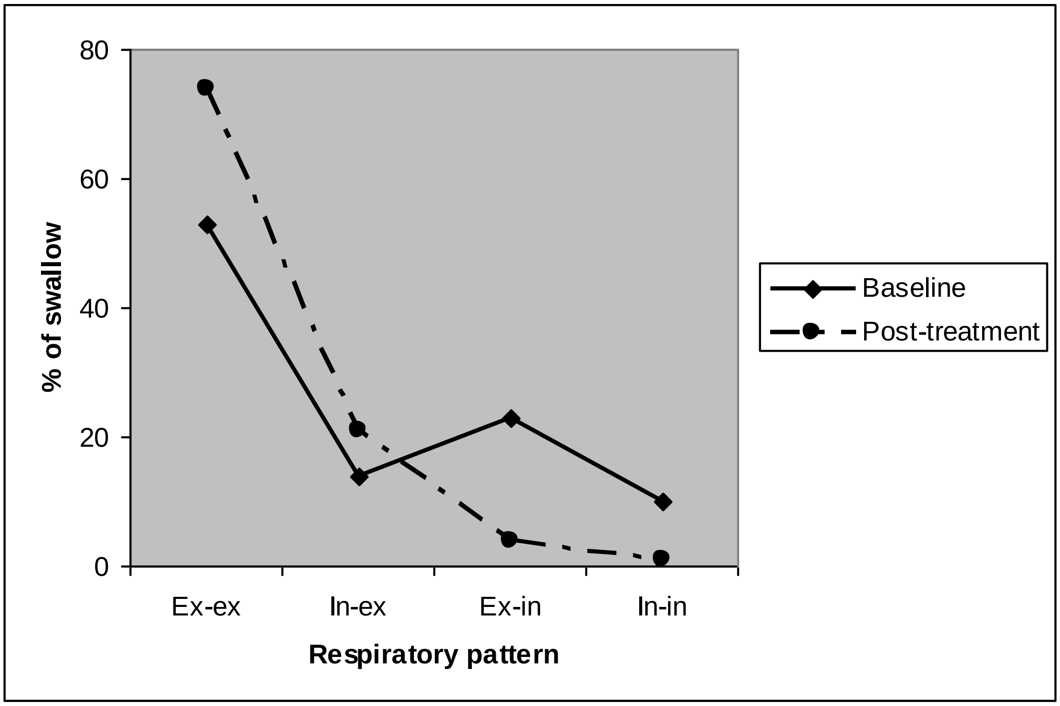

Figure 5. Comparison of the percentage of swallows that occurred for each possible respiratory pattern in the baseline and post-treatment conditions. ex-ex = expiration-apnea-expiration; in-ex = inspiration-apnea-expiration; ex-in = expiration-apnea-inspiration; in-in = inspiration-apnea-inspiration.

Because of the small number of participants, it was also appropriate to analyze results individually to assess exceptions to the overall pattern of findings. In the baseline condition, all but one participant (Participant #2) displayed at least one swallow followed by inspiration rather than expiration. Three participants had only one swallow followed by inspiration, one participant had three, and the remaining three participants had at least six swallows followed by inspiration. In the post-treatment condition, expiratory breathing followed 10 out of 10 possible swallows for all participants, with the exception of one (Participant #1), who displayed post-swallow expiration on six of ten swallows. Although the participant did not achieve post-apneic expiration on all trials, a greater number of his swallows were followed by expiration in the post-treatment condition (6/10) compared to the baseline condition (3/10). Overall, seven of the eight participants increased the number of swallows with the post-swallow expiratory pattern between the baseline and post-treatment conditions. For the participant (Participant #2) who reached ceiling in the baseline condition (i.e., 10 swallows followed by expiration), no changes were observed between conditions.

Individual differences were also observed in the prevalence of the four possible respiratory patterns that surround swallowing (i.e., ex-ex, ex-in, in-ex, in-in). In the baseline condition, four participants displayed a pattern of breathing during swallowing that was consistent with that observed in healthy adults (i.e., at least seven out of ten swallows with the ex-ex pattern). The other four participants displayed the typical respiratory-swallow pattern (i.e., ex-ex) less often than would be expected in healthy adults (i.e., fewer than seven out of ten swallows). One participant (Participant #3) had no ex-ex swallows in the baseline condition. Instead, 60% of her swallows were preceded and followed by inspiration (i.e., in-in).

In the post-treatment condition, 6/8 participants increased the number of swallows with the typical respiratory pattern. Five of the six participants had swallows preceded and followed by expiration (i.e., ex-ex) on all 10 swallowing trials and one participant displayed the ex-ex pattern on 8/10 swallows. Of the two participants who did not increase the number of swallows with the typical respiratory pattern, one (Participant #2) displayed the in-ex pattern for all 10 swallows, and the other (Participant #1) displayed each of the four patterns at least once, with the in-ex pattern occurring slightly more frequently than the other three patterns (i.e., on five of ten swallows).

Reliability. Measurement reliability refers to consistency of the judgments that comprise the outcome measures (Bailey & Burch, 2002). The most common and currently accepted manner of presenting reliability data is in terms of percentage agreement (i.e., exact agreement coefficient), which provides a percentage of instances in which two independent observers agreed in their interpretations of the data. At minimum, inter-rater reliability estimates are typically conducted on 30% of opportunities (e.g., sessions, trails, etc.) and 85% agreement is considered acceptable. For the purposes of this thesis, a second person (who was not involved in data collection) reviewed a randomly selected 30% of the data (i.e., 24 swallows from baseline condition and 24 swallows from post-treatment condition) and judged the total number of swallows that followed the typical respiratory pattern (i.e., exhalation after swallowing or xE). The inter-rater reliability was calculated to be 98%. The procedural protocol used to calculate the inter-rater reliability estimate can be found in Appendix G.

Research question 2 – The effects of the technique on participants' perceptions of their swallowing and the treatment. The participants' pre- and post-treatment scores on the Mental Health and Symptom Frequency subsections of the SWAL-QOL were presented in a table (see Table 7) and analyzed descriptively to highlight any significant trends in the data. For a summary of participants' responses to individual questions on the Mental Health and Symptom Frequency subsections, see Appendix C. Interview data were also analyzed descriptively and examples of participants' responses were included to highlight common opinions expressed by participants. See Appendix D for participants' verbatim responses to each interview question.

Table 7

Participants' baseline and post-treatment average scores (out of 5) on the Mental Health (MH) and Symptom Frequency (SF) subsections of the SWAL-QOL.

ID	Baseline MH score	Post-treatment MH score	Baseline SF score	Post-treatment SF score
1	4.20	5.00	5.00	5.00
2	4.80	4.40	4.75	4.75
3	4.60	4.80	5.00	5.00
4	4.00	5.00	5.00	5.00
5	5.00	5.00	4.75	4.75
6	4.60	5.00	4.50	4.50
7	5.00	5.00	5.00	5.00
8	3.00	3.00	3.75	4.00

The majority of participants' responses to questions from the Mental Health subsection of the SWAL-QOL were generally more positive in the post-treatment condition, resulting in slightly higher quality of life scores compared to the baseline condition. Four participants' average score on the Mental Health subsection increased from baseline to post-treatment. Three participants' average score did not change between the two conditions and one participant's score was lower in the post-treatment condition compared to the baseline condition. On the first Mental Health question (i.e., "My swallowing problem depresses me"), seven out of eight participants' responses did not change from the baseline to post-treatment condition. The participant, whose response did change, increased from a 4 (*hardly ever true*) to a 5 (*never true*). On the second Mental Health question (i.e., "Having to be so careful when I eat of drink annoys me"), four of the participants' responses increased from a 4 to a 5 between the baseline and post-treatment condition, and four participants' response did not change between conditions. In response to the third Mental Health question (i.e., “I’ve been discouraged by my swallowing problem”), three participants increased from a 4 in the baseline condition to a 5 in the post-treatment condition. Five participants’ responses on question three did not change between conditions. On the fourth (i.e., “My swallowing problem frustrates me”) and fifth (i.e., “I get impatient dealing with my swallowing problem”) Mental Health questions, two participants’ scores increased from a 4 to a 5 between the baseline and post-treatment conditions, one participant’s score decreased from a 5 to a 4, and five participants’ responses did not change.

In almost all cases, with the exception of one participant, average scores on the Symptom Frequency subsection of the SWAL-QOL did not change between the baseline and post-treatment conditions. On question one (i.e., “How often did you experience coughing?”), participants’ scores did not change between conditions, except for one participant whose response changed from a 3 (*sometimes*) in the baseline condition to a 5 (*never*) in the post-treatment condition. On Symptom Frequency question two (i.e., “How often did you experience choking when you ate?”), participants’ responses did not change between conditions. Participants’ responses also remained stable on Symptom Frequency question three (i.e., “How often did you have to clear your throat?”), with the exception of one participant, whose score changed from a 5 in the baseline condition to a 3 in the post-treatment condition. In response to the fourth question in the Symptom Frequency subsection (i.e., “How often did you experience food sticking in your throat?”), all participants selected 5 in both the baseline and post-treatment conditions, with the exception of one participant whose response changed from a 3 in the baseline condition to a 4 (*hardly ever*) in the post-treatment condition.

The interview data were analyzed in relation to three general areas: characteristics of the treatment technique, participants' comfort and confidence in using the technique, and the quality and quantity of instruction provided. The characteristics of the technique were assessed by asking each participant what they liked and disliked about the barrel chest technique. All participants expressed positive perspectives on the technique, with no negative comments. When asked what they liked, several participants felt that the technique improved the ease and safety of their swallow. For example, one participant commented that when using the technique, "it (the bolus) went down smoother", and another participant stated that "it was easier to swallow with a full breath (of air)". Two participants stated they felt the technique may reduce their risk of choking, and another participant liked that the technique gave him "more control" when he swallowed.

Participants were asked to comment on how confident they would feel using the technique independently (without help or instruction from the researcher); how they would feel about using the technique during their normal eating routines; and their level of anxiety when swallowing using the technique. In general, all eight participants felt confident that they could use the technique independently. Responses ranged from, "I feel fairly confident" to "(I feel) very confident". Examples of other responses included, "I'm good. I can do it" and, "Wouldn't bother me a bit". Two participants stated that although they felt confident in knowing how to use the technique, additional practice was necessary (e.g., "I feel confident but need more practice", "I think I'm okay, now I just need to practice"). Six participants indicated that they were planning to incorporate the technique into their normal eating routines (e.g., "I'll try it", "Going to implement it right a way", "If I thought about it, I'd use it"). One participant stated that the technique was not necessary at his particular stage of illness. Another participant's response to the question was off-topic (i.e., "Changed the rhythm of swallowing and had to get more involved"). Six of the eight participants stated that they were not anxious when using the technique (e.g., "I wasn't anxious. I felt comfortable", "No anxiety", "None at all. Felt normal"). Of the two participants who expressed some anxiety, one stated, "(I was) anxious at the beginning, but fine at the end" and the other responded, "I was somewhat anxious because I was trying something different. Getting air in isn't something I'm used to doing".

To assess the effectiveness of the training, participants were asked if they felt they needed further instruction to use the technique. Six of the participants felt the amount of training was adequate. One person indicated she would prefer more training and the other participant's response was off-topic (i.e., "Keeps you busy mentally").

Discussion

In this study, a volitional breathing technique, the barrel chest maneuver, was introduced to examine its effect on the respiratory pattern associated with swallowing in patients with ALS. Specifically, the primary focus was to evaluate the effectiveness of the technique in (1) promoting expiratory breathing after swallowing and (2) improving patients' perceptions of swallowing. The barrel chest breathing technique was based on the premise that volitionally manipulating lung volume above functional residual capacity prior to initiating a swallow may promote expiratory breathing following the apneic period. As a group, the participants showed a statistically significant increase in the average number of typical swallows (expiration after swallowing) following training. With regard to perceptions of swallowing, there were no obvious differences between participants' baseline and post-treatment quality of the life scores on the Mental Health and Symptom Frequency subsections of the SWAL-QOL. However, participants' responses to qualitative interview questions were generally positive and suggest that participants found the treatment technique to be beneficial and effective in improving their safety and comfort while swallowing.

Findings in the Context of Previous Research

This study was based on the observation that disruptions in swallowing and respiratory coordination commonly occur in the presence of neurologic diseases such as ALS. In some studies, patients with neurological disorders were more likely to display an abnormal pattern of respiration during swallowing (Butler, Stuart, & Pressman, 2007; Hadjikoutis et al., 2000; Selley et al., 1989b; Terzi et al., 2007). With regard to ALS, researchers have shown that patients are more likely to inspire, rather than expire, following the apneic period (Cleary et al., 2009; 2007; Hadjikoutis et al., 2000). Consistent with this finding, seven out of eight participants in the current study had at least one swallow apnea followed by inspiration (see Table 5). As a group, 33% of swallows in the baseline condition were followed by the inspiratory phase of breathing. Given the increased potential for aspiration of foods and liquids, and hence pneumonia, associated with post-swallow inspiration, a volitional breathing technique that promotes expiratory airflow following the apneic period may improve airway protection and clearance in patients with ALS.

There are two possible explanations, supported by existing literature, which may account for the changes in respiratory pattern observed following the use of the breathing technique. First, as proposed by Wheeler-Hegland et al. (2009), when swallows are performed with lung volumes at or above functional residual capacity, as in the current study when participants used the barrel chest technique, the lungs-thorax unit is in a slightly expanded state and will naturally recoil back to rest position, thereby promoting expiratory breathing. When the lungs-thorax unit is in an expanded state, the positive recoil forces result in greater sub-glottal pressure, allowing the expulsion of any residual material in the airways, as well as enhancing the strength of an expiratory reflex (ER) in the case of aspiration. Thus, in patients with ALS, swallowing with slightly higher lung volumes and an expanded barrel chest may capitalize on the elastic-recoil properties of the pulmonary apparatus to promote post-swallow expiratory breathing and enable patients to quickly initiate a more effective ER response to expel residual material. In this study, the instrumentation used to measure respiration was not designed to quantify participants' lung volume during swallowing; therefore, definitive statements as to whether participants were in fact initiating swallow in the treatment and post-treatment conditions at lung volumes at or above functional residual capacity can not be made.

The second possible mechanism to explain how the barrel chest technique may have worked in the current study is based on research conducted by Gross, Atwood, Grayhack, and Shaiman (2003). The authors reported that swallows initiated with lung volumes at or below residual volume are executed significantly more slowly than swallows initiated at or above functional residual capacity. Hadjikoutis et al. (2000) hypothesized that under normal conditions, swallowing will inhibit respiration; however, if there is an urgent need for both swallowing and breathing, respiration will prevail and deglutition will be interrupted to allow airway needs to be met.

The maintenance of respiration is particularly important in conditions where the ability to breathe is limited as a result of repeated swallows and prolonged swallow apneas, which is often the case in patients with ALS. Therefore, initiating swallows with higher lung volumes may decrease the length of the swallow, thereby reducing the amount of time that respiration is inhibited, and leaving patients less starved for air at the end of their swallow. In the current study, swallowing speed was not measured so it was not possible to verify whether patients with ALS execute swallows faster when deglutition is initiated at higher lung volumes (i.e., at or above functional residual capacity).

The current findings extend the existing knowledge base regarding patterns of breathing during swallowing in neurological disorders. Most of what is known about breathing and swallowing coordination has been obtained from infants (Kelly, Huckabee, Jones, & Frampton, 2007), healthy adults (Martin-Harris et al., 2003; Martin-Harris et al., 2005; Shelley et al., 1989a; Wheeler-Hegland et al., 2009), and adults with various neurological impairments (Butler et al., 2007; Hadjikoutis et al., 2000; Selley et al., 1989b; Terzi et al., 2007). Data collected during this study, albeit with a small sample, add to the existing literature regarding respiratory-swallowing coordination in ALS specifically. Not only do the findings extend the knowledge base concerning the prevalence of post-apneic inspiration in this population, but this study also characterizes the prevalence of each of the four possible swallow-respiratory patterns (i.e., ex-ex, ex-in, in-ex, in-in) by examining the respiratory phase that precedes the apneic period.

Consistent with the findings of other studies involving patients with ALS, the participants in the current study had a fewer number of swallows followed by expiration than is typically observed in healthy adults. In the baseline condition, when participants were asked to swallow as they normally would, expiratory breathing following the apneic period occurred in only 66% of swallows, which is less than the frequency reported in studies with healthy adults. In comparison to a study in which similar procedures (i.e., a 20 ml water bolus self-administered from a cup) were used to those in the current study, Wheeler-Hegland et al. (2009) reported that 87% of total swallows by healthy adults were followed the expiratory phase of respiration. In other words, inspiratory breathing following the apneic period was only observed in 13% of total swallows in healthy adults, compared to 33% of total swallows in this study. Similarly, Martin-Harris et al. (2003) reported that post-apneic inspiration characterized 18-21% of total swallows in healthy adults. This result is consistent with the findings of other researchers who reported that patients with ALS were more likely to display an abnormal pattern of inhalation, rather than exhalation, after swallowing compared to normal subjects (Cleary et al., 2009; 2007; Hadjikoutis et al., 2000).

In terms of the prevalence of each of the four possible respiratory patterns surrounding swallowing (i.e., ex-ex, ex-in, in-ex, in-in), the respiratory pattern surrounding most swallows was the ex-ex pattern (i.e., 53% of total swallows), which is the most common respiratory pattern observed during swallowing in healthy adults. Although the present study did not include a healthy control group, several studies have reported that the ex-ex pattern of breathing is observed in 71% to 100% of healthy individuals (Martin-Harris et al., 2003; Martin-Harris et al., 2005; Shelley et al., 1989a; Wheeler-Hegland et al., 2009). The 53% observed in the current study indicates that, as a group, the participants with ALS had about 18-47% fewer swallows preceded and followed by expiration compared to healthy adults. Despite the overall lower proportion of ex-ex swallows, four participants displayed a pattern of breathing during swallowing that was consistent with the frequency observed in healthy adults (i.e., the ex-ex pattern within the 71-100% range).

The presence of normal swallow-respiratory patterns in four of the eight participants may be explained by factors that characterize ALS severity. For example, the participants with normal swallow-respiratory patterns had an ALSFRS-R bulbar score of 9 or 10 out of 12, which is slightly higher than some of the bulbar scores of participants who had abnormal patterns of respiration during swallowing. This finding is consistent with Hadjikoutis et al. (2000), who reported that patients with upper motor neuron bulbar signs were more likely to inspire, rather than expire, following the apneic period. However, while this finding may hold true for two participants, it does not explain the presence of abnormal swallow-respiratory patterns in two other participants who, despite ALSFRS-R bulbar scores of 9 and 10, had only four and three swallows, respectively, with the ex-ex pattern.

Given that this explanation does not account for all the variability in swallow-respiratory patterns observed in this population, other potential factors, such as ALS site of onset, may also be considered. In the current study, three of the four participants who displayed a proportion of ex-ex swallow comparable to that of healthy adults had limb, rather than bulbar, onset ALS. Of the four participants in the study who had limb onset ALS, only one (Participant #8) displayed abnormal patterns of respiration during swallowing. Specifically, only four of his ten swallows had the ex-ex pattern, whereas the other three participants with limb onset had the ex-ex pattern on 7/10, 7/10, and 9/10 swallows. The participant with limb onset ALS who was the exception presented with demographic (i.e., age and time post diagnosis) and disease severity (i.e., FVC, PCF, ALSFRS-R bulbar score) characteristics that were very comparable to other participants in the study, with one exception. Specifically, the participant had the lowest ALSFRS-R total score of all the participants in the study, which was 14/48. The next lowest ALSFRS-R total score was 33/48. Of the four participants in the study who had bulbar onset ALS, three displayed the ex-ex pattern less frequently than would be expected in healthy adults. Again, the participant who was the exception in this sub-group (Participant #2) had relatively comparable demographic and disease severity characteristics; however, of all eight participants in the study, she had the shortest time post diagnosis (i.e., one month). The significance, if any, of the individual differences with regard to ALSFRS-R total score and time post diagnosis in relation to swallow-respiratory coordination remains unclear, given the pilot nature of the study and the small sample size.

Despite some individual differences, group data suggest that the presence of abnormal patterns of respiration during swallow are unrelated to participants' age, gender, time post diagnosis, FVC, PCF, and/or ALSFRS-R total score. However, the presence of upper motor neuron bulbar impairment may be a potential factor in predicting the presence of abnormal swallow-respiratory patterns in patients with ALS.

This study also adds to existing literature regarding the effects of compensatory techniques on swallowing and respiratory coordination. Compensatory techniques are often utilized with patients who have dysphagia because they have been shown to be effective in facilitating airway protection. However, there is limited evidence to show whether compensatory swallowing techniques affect the normal respiratory–swallow pattern, which is an important factor to consider in airway protection. McFarland, Lund, and Gagner (1994) showed that postural alterations in adults can affect the expiratory phase of respiration during swallowing. Specifically, when adults swallowed while on their hands and knees (i.e., 'on all fours'), the swallow occurred earlier in the expiratory phase of the respiratory cycle. In contrast, when standing upright, swallows occurred later in the expiratory phase. More recently, a preliminary study conducted by Ayuse et al. (2006) also showed that some compensatory techniques can alter the relationship between swallowing and respiration. Researchers examined the effect of two common compensatory techniques (i.e., chin tuck and partial recline) on respiratory-swallow coordination in patients with dysphagia. Their findings showed that, when compared to the neutral, upright position, a 60^o recline from the vertical position with a 60^o chin tuck increased the duration of the apneic period and the total swallow duration, suggesting that compensatory swallowing manuevers affect aspects of swallow-respiratory coordination.

In the current study, researchers investigated alterations in respiratory-swallowing coordination brought about by a compensatory technique involving volitional modification of lung volume. The respiratory-swallow patterns observed in the post-treatment condition (i.e., with the use of the barrel chest breathing technique) differed from those in the baseline condition, suggesting that breathing patterns surrounding swallows can be altered by compensatory techniques. Although the ex-ex pattern was still the most frequently observed respiratory pattern during swallowing, it occurred more often in the post-treatment condition (i.e., 74%) compared to the baseline condition (i.e., 53%). The next most frequently observed pattern in the post-treatment condition was in-ex (i.e., 21%), followed by ex-in (i.e., 4%) and in-in (i.e., 1%) (see Figure 5).

These data support the use of volitional compensatory techniques to achieve swallow-respiratory patterns that resemble those of healthy adults. For example, in healthy adult populations, the two most common respiratory patterns during swallowing are the ex-ex pattern and the in-ex pattern, which occur in approximately 71-100% and 18-21% of swallows, respectively (Martin-Harris, 2003). In the current study, the frequency of the ex-ex pattern increased from 53% in the baseline condition to 74% in the post-treatment condition. Similarly, an increase in the frequency of the in-ex pattern was also observed in the post-treatment condition, making it the second most common pattern (i.e., 21%). Overall, 95% of swallows in the post-treatment condition were followed by expiratory breathing, which is in agreement with the findings of Wheeler-Hegland et al. (2009) who reported that 87% of healthy adults' swallows were followed by expiration. Although further research is needed, the preliminary findings of the study indicate that patients with ALS are able to achieve swallow-respiratory patterns similar to those of healthy adults after use of the barrel chest technique.

To date, this is the first study to assess the effects of breathing techniques on the coupling between respiration and swallowing in ALS. Although the sample size is too small to draw any firm conclusions about the effects of the barrel chest maneuver on swallow-respiratory coordination, the potential for increased post swallow expiration forms the foundation for future study of this and other techniques.

Secondary Outcomes

Participants' perceptions of their swallowing and the treatment. To assess participants' perceptions of swallowing before and after the introduction of the barrel chest breathing technique, participants were asked to complete the Mental Health subsection and a portion of the Symptom Frequency subsection of the SWAL-QOL. There were a total of nine Likert-type questions and higher scores indicated a better quality of life or more positive perspectives towards swallowing and eating (McHorney et al., 2006). The majority of participants' responses to questions from the Mental Health subsection of the SWAL-QOL were generally more positive in the post-treatment condition, resulting in slightly higher average scores compared to the baseline condition. In the N of 1 RCT pilot study upon which this thesis project was based, a subsection of the SWAL-QOL was administered to assess the participant's level of distress (Cleary et al., 2007). The study's findings were similar to the current study in that the authors reported that the participant rated his psychological distress as less frequent/ less severe (as indicated by higher scores) following the active treatment compared to the baseline and placebo. However, individual data analyses revealed that three participants' average score did not change between the two conditions and one participant's score was lower in the post-treatment condition compared to the baseline condition. Further, on the Symptom Frequency subsection of the SWAL-QOL, for seven participants, average scores on the Symptom Frequency subsection of the SWAL-QOL did not change between the baseline and post-treatment conditions. In both conditions, most participants responded that they never, or hardly ever, experienced any of the symptoms presented as response choices.

Findings from the administration of the SWAL-QOL should be interpreted with caution, primarily because the instrument may have lacked sensitivity to changes in perceptions of swallowing that might occur over the course of only one session. Specifically, participants may have found the questions from the Mental Health subsection difficult to answer given that they were required to base their responses on only their 10 most recent swallows of pudding. The questions are broad and responses are not meant to be based on a finite time period or a specific number of swallows. Indeed, participants qualified their answers by stating that although the items may have been true for them on a regular basis, they were not applicable during their most recent 10 swallows.

With regard to the Symptom Frequency subsection, participants' responses to the questions may also have been influenced by the bolus consistency, the nature of their dysphagia, and/or a combination of both. For example, if a patient did not present with dysphagia for thickened liquids, they may have been less likely than other participants to report symptoms such as coughing or choking during the pudding meal trial. Additionally, although participants were told to eat as naturally as possible (i.e., as they would any other meal), the presence of a researcher may have inadvertently influenced participants to eat slower and more carefully, which may have resulted in fewer incidences dysphagia-like symptoms. It is possible that the SWAL-QOL responses may have been different if the baseline and post-treatment measures had been collected over a longer period of time and with a range of bolus consistencies.

Participant perspectives regarding the barrel chest technique were also solicited through a semi-structured interview. The interview questions were designed to assess three general areas: characteristics of the treatment technique, participants' comfort and confidence in using the technique, and the quality and quantity of instruction provided. The perspectives of the eight participants were generally positive. Overall, participants' reported that they liked the breathing technique and several individuals elaborated by saying that they felt the technique improved the ease and safety of their swallow. Many of the participants were eager and willing to implement the breathing technique into their normal eating routines and stated that they felt confident using the technique. Only one participant reported that he did not feel he needed to use this technique as he did not have any difficulty swallowing (Participant #5). In terms of his respiratory-swallow patterns, 9/10 swallows in the baseline condition had the typical pattern (i.e., post-apneic expiration); therefore, the fact that he presented as asymptomatic for dysphagia, may explain why he was not as motivated as some of the other participants to use the breathing technique. Additionally, as a group, the participants were generally satisfied with the amount and type of instruction they received during the treatment phase, but two participants expressed that they needed more practice using the technique.

Overall, responses to the interview questions were brief but generally consistent between participants. The largely positive response from the participants reinforces that there is merit in continuing to explore this technique. The volitional breathing technique appears to be within the capabilities of the participants and no barriers were identified that would impede future research involving this volitional breathing technique.

Limitations of the Study

The primary limitation of the study was the research design, which lacked a control group and therefore does not allow firm conclusions about the effectiveness of the barrel chest maneuver in promoting normal swallow-respiratory patterns in patients with ALS. Although a within-subjects design was appropriate for a Phase I treatment outcomes study, One-Group Pretest-Posttest (O_1 X O_2) studies are considered weak experimental designs for studying treatment efficacy (Schiavetti & Metz, 2006). In this design, a single group is pretested, exposed to the experimental treatment, and posttested. It is considered a within-subjects design because all participants are tested under the pre- and post-conditions. Without a control group, it is difficult to assess the significance of an observed change in the group of participants. The change could be the result of historical changes unrelated to the treatment, the maturation of the subjects, an instrumentation error, an artifact of testing, or a consequence of statistical regression. These are factors that threaten the external and internal validity of a study with the One Group Pretest-Posttest design.

In the current study, history and/or maturation factors were unlikely to influence the treatment outcomes as there was very little time between the baseline and treatment conditions (i.e., less than 30 minutes) during which growth, development, and/or unanticipated events could have occurred to affect the post-treatment results. Similarly, statistical regression was likely not a threat to the internal validity of the study as none of the participants had extreme scores in terms of the number of abnormal swallows in the baseline condition. If no normal swallow patterns were observed in any of the participants in the baseline condition, statistical regression following post-treatment testing would be considerably more likely. Furthermore, the participants were not selected on the basis of displaying an extreme number of abnormal swallows, nor were they prescreened for presence of the aberrant swallow pattern. All selected participants were included in the study and received the treatment regardless of the frequency with which they displayed aberrant swallow-respiratory patterns.

Of the remaining threats common to One-Group Pretest-Posttest study designs, testing factors may have compromised the internal validity of the study.

Testing has the potential to threaten validity if the pretest somehow increases the participants' ability to perform well on the posttest. In the current study, it is not possible to rule out the influence of testing on the treatment outcomes observed. For example, prior to data collection in the post-treatment condition, participants had practiced approximately 15 swallowing maneuvers, 10 in the baseline condition and five during the treatment condition. Although it is more likely that fatigue would have negatively affected participants' performance in the post-treatment condition, it not possible to definitively determine whether the improvements noted in the post-treatment condition were due to the treatment, or to the practice effects resulting from the large number of swallows that occurred prior to posttest data collection.

Other limitations include procedural aspects of the study. First, although the equipment and instruments used for data collection were adequate for the purposes of this study, they were limited in their sophistication and ability to capture potentially relevant information. For example, during the treatment and post-treatment conditions, participants were told to initiate their swallows with higher lung volumes (i.e., "take a deep breath and hold it before swallowing"); however, the respiratory equipment used in this study did not permit accurate measurement of lung volume; therefore, it was not possible to verify whether participants' lung volumes were in fact higher when initiating swallows in the treatment and post-treatment conditions compared to the baseline condition.

This study could have been strengthened by using equipment designed to measure lung volumes, specifically total lung capacity and functional residual capacity, as well as the ability to provide visual feedback to participants to allow them to achieve the target lung volumes. Lung volume changes are reflected in the changes of the rib cage and abdomen volumes (Konno & Mead, 1967). Respiratory inductive plethysmography (e.g., The Respitrace system by Ambulatory Monitoring) is often used to transduce respiratory movements to determine volumes. For respiratory plethysmography, data are collected using two elastic cloth bands, which are placed around the rib cage and abdomen (Gross et al., 2003). The wire coils attached to the bands measure the changes in the cross-sectional area of the rib cage and abdomen. As respiratory inductive plesthymography does not involve any equipment that affects the face, it would be appropriate for measuring respiration during swallowing maneuvers. The signal generated by respiratory inductive plesthymography can also be used to provide visual feedback to participants to assist them in reaching their target lung volumes during treatment tasks. This feature would be ideal for future studies as it would also allow experimenters to ensure that participants are initiating their swallows with higher lung volumes when required to do so during the treatment and post-treatment phases of the study. Furthermore, it would permit researchers to determine precisely what lung volume (or percent of vital capacity) is required to execute a safe and successful swallow, without post-apneic inspiration.

The manner in which the accelerometer was utilized during data collection was also a limitation of the study. The pilot studies upon which this thesis is based, showed that the accelerometer was not sensitive enough to capture hyolaryngeal elevation when it was taped to the participant's neck (i.e., lateral to the thyroid cartilage). Despite hyolaryngeal elevation, the tape impeded the movement of the accelerometer such that it was not a reliable indicator of swallowing onset when it was taped to the neck. As a result, experimenters improvised and held the accelerometer during swallowing maneuvers. The placement of the accelerometer was largely dependent on whether the experiment was able to hold the accelerometer against the participant's thyroid cartilage without interfering with the swallowing maneuver. For some participants (e.g., those with long necks), the accelerometer could be gently held lateral to the thyroid cartilage and moved in an accentuated upward direction when the hyolaryngeal complex elevated. In other cases, it was not possible for the experimenter to hold the accelerometer against the participant's neck due to a limited range of motion with which to move the accelerometer. When this was the case, the experimenter held the accelerometer with one hand, and palpated the participant's thyroid notch with the other hand. When hyolaryngeal elevation was detected, the experimenter moved the accelerometer upwards once and then held it stable until the hyolaryngeal complex elevated for subsequent swallows. Although this reduced the validity of the study by introducing experimenter influence, the experimenter was an experienced speech-language pathologist who was also a Board Certified Swallowing Specialist and had extensive experience palpating patient's thyroid cartilages during swallowing maneuvers. Experienced speech-language pathologists have been shown to have high inter- and intra-rater reliability in judging hyolaryngeal elevation during thin liquid swallows (McCullough et al., 2000). In future studies, every effort should be made to try and find an alternative method of measuring swallowing onset time, so that the experimenter does not need to be directly involved. For example, potential options include increasing the sensitivity of the accelerometer if possible, and/or determining a more appropriate manner of attaching the accelerometer to the patient's neck such that its movement will not be impeded. Another option may be to use different instrumentation, such as a contact microphone, submental electromyography, or videofluoroscopy to mark the onset of swallowing activity.

With regard to methodology, a limitation of this study was that liquid bolus size was not tightly controlled. Although participants were given cups filled with 20 ml of water, some participants could not take all 20 ml in one swallow. When this occurred, it was not possible to determine precisely what volume of bolus the participant had consumed. Studies with healthy adults have shown that respiratory patterns during swallowing and the duration of the apneic interval are not influenced by various bolus volumes (i.e., 3, 10, and 20 ml) (Martin et al., 1994); however, it is unknown how bolus size affects the respiratory-swallow patterns in patients with ALS. Given the unknown interaction between bolus size and respiratory-swallowing coordination in this population, every effort should be made to ensure consistency in the administration of bolus volume within and between participants.

Future Research

The existing literature regarding swallowing and respiratory coordination is largely based on studies conducted with middle-aged healthy adults (Martin-Harris et al., 2003; Martin-Harris et al., 2005; Shelley et al., 1989a; Wheeler-Hegland et al., 2009). Only recently has new research begun to explore respiratory–swallow coordination throughout the human lifespan (Hirst, Ford, Gibson, & Wilson, 2002; Hiss et al., 2004; Kelly et al., 2007; Martin-Harris et al., 2005), and in the presence of neurological disease (Butler et al., 2007; Hadjikoutis et al., 2000; Selley et al., 1989b; Terzi et al., 2007), cancers of the head and neck (Charbonneau, Lund, & McFarland, 2005; Brodsky et al., 2010), and chronic obstructive pulmonary disease (Gross, Atwood, Ross, Olszewski, & Eichhorn, 2009). This study contributes to the growing literature of respiratory-swallowing coordination in impaired populations, specifically ALS. To date, this is the fourth known study that has involved examination of respiratory-swallow patterns in patients with ALS. The findings of this pilot project are consistent with those of the three other studies, which showed that abnormal patterns of respiration during swallowing (i.e., swallow apneas followed by inspiration) are observed in patients with ALS (Cleary et al., 2009; 2007; Hadjikoutis et al., 2000). These findings were contradicted by a study conducted by Shelley et al. (1989b), which reported that swallow-respiratory patterns were normal in patients with MND. However, the authors did not specify whether any of the participants with MND had diagnoses of ALS. Furthermore, the study was based on a small number of participants (i.e., five) and the procedures used to collect swallow-respiratory data were different than those used in the current study (i.e., participants swallowed 5 ml of juice from a teaspoon rather than 20 ml of water from a cup, as was the case in the current study). Further research is needed with a larger number of participants with varying degrees of severity and disease progression, to characterize the patterns of respiration during swallowing that are common to patients with ALS. Further understanding in this area would enable health practitioners to manage dysphagia by designing and implementing remedial techniques that target the nature of the dysfunction.

The existing literature is also limited by the fact that prior to this pilot project, no studies had examined and/or described the respiratory phase that precedes the apneic period during swallowing in patients with ALS. In studies with healthy adults, it is becoming common practice to report the prevalence of each of the four possible respiratory patterns surrounding swallowing (i.e., ex-ex, ex-in, in-ex, in-in). This study was the first to examine the frequency of each of these four patterns in patients with ALS (Figure 5). Further research is needed to reach a consensus about the respiratory patterns that precede *and* follow swallowing events in patients with ALS as this information may inform our knowledge about normal vs. aberrant patterns in affected individuals. Specifically, although we assume that post-apneic inspiration is maladaptive, additional factors may need to be considered in making this determination, such as examining where in the respiratory cycle the swallow occurs (B. Martin-Harris, personal communication, May 14, 2011). For example, a patient whose swallow occurs in the mid-expiratory phase of the respiratory cycle and is followed by inspiration may be considered more aberrant than a post-apneic inspiration that occurs following a swallow initiated at the bottom of the expiratory cycle. In the latter, it is possible that inspiration is necessitated by the fact that it is the next phase of respiration to occur in the patient's natural respiratory cycle, and this may in fact be considered normal. Further research is needed to explore this hypothesis and determine if the timing of the swallow within the respiratory cycle has a functional outcome on airway protection and swallowing safety.

In this study, the rationale for trying to help patients with ALS acquire normal respiratory-swallow patterns was based on the assumption that disruptions in swallowing and respiratory coordination have negative consequences on pulmonary health and swallowing safety. Specifically, patients with ALS who were lacking the post-apneic clearing mechanism (i.e., expiratory maneuver) during swallowing were assumed to be at increased risk for aspiration and aspiration pneumonia. However, the evidence in the literature for linking impaired swallow-respiratory coordination with the occurrence of aspiration and subsequent pulmonary compromise is not uniformly strong. Although some researchers have suggested that patients with post-apneic inspiration are more likely to aspirate food or liquid and develop pneumonia (Brodsky et al., 2010; Martin-Harris, 2008), other studies have failed to find such an association (Hirst et al., 2002; Hiss, Treole, & Stuart, 2001; Martin-Harris et al., 2005). Almost all studies that have failed to find a link have based their conclusions on outcomes observed with healthy adults. Few studies have addressed the clinical significance of impaired swallow-respiratory coordination in patients with ALS. Patients with ALS are unlike healthy adults in that they often have weak coughs, impaired mucociliary function, and compromised immune systems. As a result, they have a reduced ability to mechanically drive material out of their lungs, and their cellular defence mechanisms are ineffective at breaking down aspirated material. These factors have negative airway protective and bolus clearance implications for patients with ALS and may predispose them to aspiration and subsequent pulmonary compromise.

Currently, only one known study has attempted to clarify whether an abnormal pattern (i.e., swallow apnoeas followed by inspiration) is related to airway compromise in patients with ALS (Hadjikoutis et al., 2000). Although the study did not use videofluoroscopy to assess aspiration in patients, and the conclusions were based on a small number of participants (i.e., n = 32), authors concluded that abnormal breathing patterns were unrelated to chest infections, and episodes of coughing and choking. The authors suggested that the presence of post-swallow inspiration may best serve as an indicator of disordered swallowing, rather than as a cause of aspiration (Hadjikoutis & Wiles, 2001).

To date, the clinical significance of an abnormal pattern of respiration-swallowing integration remains uncertain, particularly when it occurs in the presence of neurological disease. Future studies are warranted to test the effects of swallow-respiratory instability on swallowing safety and health outcomes. Specifically, patients with ALS should be followed longitudinally to look for a possible causal relationship between impaired respiratory-swallowing coordination and incidents of aspiration and aspiration pneumonia. A better understanding of the nature and consequences of reversal or modification in swallow-respiratory patterns has the potential to inform clinical practice by evaluating whether therapeutic interventions and/ or behavioral remediation techniques are warranted in patients with aberrant patterns.

To our knowledge, this is the first study to attempt to promote normal respiratory-swallowing phase in patients with ALS. Although the preliminary findings were encouraging, additional clinical studies are needed to evaluate the impact of volitional modification of lung volume on swallowing and respiratory coordination in this population. Specifically, replication of this study with a larger sample size, a stronger research design, more sophisticated instrumentation, and a more controlled protocol would further the existing evidence regarding the viability of this technique in enabling patients with ALS to achieve more normal and safer respiratory patterns during swallowing. A Phase II study would be an appropriate next step in this line of research. In Phase II research studies, the goals are to establish and standardize treatment protocol and methods, as well as to formulate a potential explanation to account for the treatment effect. A small group experimental study with a treatment and control group would be appropriate for the next phase of research (Robey & Schulz, 1998). According to Portney and Watkins (2000), with an effect size of 0.50 based on a paired-samples t-test, a sample size of 28 participants is required to detect a significant difference between treatment conditions, if one exists.

With regard to refining the treatment protocol, future studies should include a review the features of the breathing technique to ensure they are optimal for maximizing mechanical function and airway protection. For example, one of the key features of this technique involves getting participants to increase their pre-swallow lung volume by taking a deep breath and holding it, similar to the supraglottic swallow maneuver. During breathing, the vocal folds are widely separated and in an abducted position to enable movement of air to and from the lungs. Maximum glottal size can be achieved during very deep inspirations (Sekizawa, Sasaki, & Takishima, 1985). In some patients, the instruction to take a deep breath and hold it doesn't result in closure of the vocal folds, as they will hold their breath by stopping chest wall movements (Martin, Logemann, Shaker, & Dodds, 1993). This results in an open airway. In the treatment protocol, participants were instructed to take a deep breath and hold it while they held the bolus in their oral cavity. For patients who display poor tongue control during bolus hold, premature spillage of the bolus into the pharyngeal cavity, or post-swallow residue, a strong inhalatory maneuver followed by a failure to close the vocal folds could leave them more vulnerable to airway compromise. Without closure of the laryngeal valve, food and liquid is more likely to enter the trachea. Perhaps a better administration of this technique would involve having patients take a deep breath, start to expire and then swallow to promote VF approximation prior to swallow initiation (Logemann, 1998).

Another aspect of the treatment protocol that should be evaluated in future studies is the effect of deep inspiration on hyolaryngeal positioning (B. Martin-Harris, personal communication, May 14, 2011). When patients are instructed to take a deep breath, their pulmonary apparatus expands and the diaphragm contracts downwards. The traction force of the diaphragm results in a downward pull on the larynx that tends to abduct the vocal folds (Hixon, Weismer, & Hoit, 2008); therefore, their ability to protect the airways is diminished. A second possible consequence is that the larynx may have greater difficulty moving up during a swallow because the downward contraction of the diaphragm tethers it from below. Not only does the larynx need to work against the downward pull of the diaphragm, but it also has further to elevate as it is starting its ascent from a lower position. Contrary to this assumption, Mitchinson and Yoffey (1947) x-rayed patients performing inspiratory and expiratory maneuvers and found that deep inspiration did not result in a downward 'tug' on the larynx as it was stabilized by contraction of supra- and infra-hyoid musculature. As hyolaryngeal elevation plays a critical role in swallowing function and safety, it is important to understand any mechanisms that may affect it and thus, further research is needed in this area.

In addition to refining the treatment protocol, another appropriate next step in phase II research involves further defining the target population (Robey & Schulz, 1998); therefore, in future research studies a goal could be to systematically evaluate the patient characteristics that influence responsiveness to treatment. Several factors could be explored, including gender, age, disease severity, respiratory health (e.g., measures of FVC, PCF, etc.), and the influence of bulbar vs. limb symptoms. Furthermore, as frontotemporal dementia (FTD) is not uncommon in patients with ALS (Woolley & Katz, 2008), future studies should address whether cognitive impairment, specifically deficits in executive functioning, influence the ability of patients to learn and successfully carry-out the treatment technique. Although this factor has been explored in two pilot studies (Cleary et al., 2009; 2007), additional research is needed in this area. Lastly, given the nature of the disease, future research should explore the impact of fatigue on treatment outcomes. For example, as this technique requires increased muscular effort, it may not be appropriate for patients in the moderate to severe stages of the disease who are experiencing muscle weakness and are easily fatigued.

Regardless of the purpose or scope of future research projects, it will be important to consider that many factors can influence the respiratory patterns surrounding swallowing. Emerging literature has shown that variability in respiratory-swallow patterns can occur as a result of manipulations in bolus size and consistency, posture, and swallowing task (i.e., sequential swallows vs. single swallows). For example, a recent study showed that post-apneic inspiration is more likely to occur following spontaneous, sequential swallows of large bolus volumes compared to smaller volume liquid swallows (Dozier, Brodsky, Michel, Walters, Martin-Harris, 2006). Bolus mastication has also been shown to influence respiratory-swallow patterns (McFarland & Lund, 1995). Further research is needed to specify the influence of these factors on swallowing and respiratory coordination as these are the factors that are often manipulated to facilitate airway protection and bolus flow in patients with dysphagia. If the influence of these factors on swallowing-respiratory coordination were better understood, clinicians could develop other compensatory techniques that promote normalization of aberrant respiratory–swallow patterns. In the meantime and until these factors can be systematically examined, future research should be interpreted with caution, especially when comparing findings between studies that involved different methodologies.

Conclusions

The findings of this study contribute to the growing literature regarding respiratory and swallowing coordination in patients with ALS. A comparison of the present results with those of prior investigations of healthy adults suggests that individuals with ALS display a greater number of swallows followed by inspiration. In additional, this study provides preliminary evidence to support the effectiveness of a respiratory-swallow intervention on promoting normal patterns of breathing during swallowing in patients with ALS. The statistically significant increase in the number of swallows that were followed by expiratory breathing in the post-treatment condition suggest that volitional modifications of lung volume may play a role in promoting normal patterns of breathing during swallowing. Given that the protection of the pulmonary airways during swallowing is dependent, in large part, on the coordination of respiration and swallowing, this breathing technique may reduce the risk of aspiration and aspiration pneumonia in patients with ALS. Until a cure is found, health practitioners will continue to play a crucial role in evaluating, educating, and treating patients with ALS. Through future research, it may be possible to develop a respiratory-swallowing phase training protocol that promotes optimal patterns of breathing during swallowing in patients with ALS. This may in turn improve long-term patient outcomes by significantly impacting health, survival, and quality of life in patients living with this disease.

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APPENDIX A: EXPERIMENTAL PROTOCOL

Data Collection Form   Starting time__________________

Participant Initials: Location: Date:

BASELINE CONDITION									
Forced Vital Capacity - ATS									
Use the microlab spirometer. Ensure that he/she is sitting erect with feet firmly on the floor. Attach nose clip, place mouthpiece in mouth and urge the patient to: seal his/her lips around "the mouthpiece close lips around the mouthpiece. Inhale completely and rapidly with a pause of < 1 s at TLC. “Breathe in fully -your lungs must be absolutely full”. Exhale maximally until no more air can be expelled while maintaining an upright posture.									
FVC #1:			**FVC #2:**			**FVC #3:**			
NOTES: Ideal PCF < 40L/m (.67L/sec) variability between highest & 2^{nd} highest result Ideal FVC < 0.2L variability for FEV1 & FVC between highest & 2^{nd} highest result									
Spontaneous Cough (PCF) - ATS									
Ensure that he/she is sitting erect with feet firmly on the floor. Apply a nose clip to the patient's nose; Urge the patient to: seal his/her lips around "the mouthpiece; “breathe in fully - your lungs must be absolutely full”; “then cough the air out - as hard as you can”									
PCF #1:			**PCF #2:**			**PCF #3:**			
NOTES: Ideal PCF < 40L/m (.67L/sec) variability between highest & 2^{nd} highest result									
Meal trial with pudding									
Provide patient with pudding. Minimum 8 bites; Maximum 10 bites or 5 minutes. Ensure the client is sitting. To allow the patient to get accustomed to task, they will complete 3 consecutive swallows without goniometer. Goniometer measurements will be taken on the 4^{th}, 6^{th} and 8^{th} swallow. Measurements will be taken in a subtle/discrete manner to prevent interfering with patient’s completion of task. Calculate average angle of flexion. No chest strap.									
☐ **S1**	☐ **S2**	☐ **S3**	☐ **S4**	☐ **S5**	☐ **S6**	☐ **S7**	☐ **S8**	☐ **S9**	☐ **S10**
Meal trial measures									
Upon completion of meal trial, patient will complete Mental Health and Symptom Frequency subsection of SWAL-QoL.									
☐ **SWAL-QoL Questions**									
Respiratory phase relationship									
Apply chest strap and accelerometer to patient. Patient will complete 30 seconds of normal tidal breathing. Save file. Patient will then swallow 10-20mL tepid water boluses.									
☐ **30 seconds of normal tidal breathing**									
☐ **S1**	☐ **S2**	☐ **S3**	☐ **S4**	☐ **S5**	☐ **S6**	☐ **S7**	☐ **S8**	☐ **S9**	☐ **S10**

<table>
<tr><th colspan="10">TREATMENT CONDITION ~10 minutes</th></tr>
<tr><td colspan="10">“Now I am going to teach you a new technique to use while you’re swallowing. While we are practicing these techniques, I’d like you to “THINK BIG”, which will involve:
1. Sitting in an upright position as tall as you can, with your shoulders touching the board behind you and your feet firmly on the floor.
2. Next, I’d like you to take a big, deep breath and hold it. When you do this, you should have a big, barrel chest.
3. Then, while holding your breath, I’d like you to swallow.
4. Before you try it, I’ll show you what I’d like you to do three times.
5. Now, I’d like you to try.
6. Remember to sit upright and as tall as you can, with your shoulders touching the board behind you and your feet firmly on the floor.
7. Now, take a big, deep breath and hold it. You should have a big, barrel chest. Then swallow when you’re ready.
8. Try it two more times.
9. Now we’re going to do the same thing three more times, but this time you’ll swallow water.”</td></tr>
<tr><td colspan="2">☐ Dry #1</td><td colspan="2">☐ Dry #2</td><td colspan="2">☐ Dry #3</td><td colspan="2">☐ Wet #1</td><td colspan="2">☐ Wet #2</td></tr>
<tr><th colspan="10">Client rests 5 minutes</th></tr>
<tr><th colspan="10">POST-TREATMENT CONDITION</th></tr>
<tr><th colspan="10">Respiratory phase relationship</th></tr>
<tr><td colspan="10">“Now we want to see if you can do this on your own.” Patient will swallow 10-20mL tepid water boluses. Ensure the patient is sitting in an upright position with their back/shoulders resting against the wood board. Provide feedback and verbal prompts as necessary to ensure proper use of technique.</td></tr>
<tr><td colspan="10">☐ 30 seconds of normal tidal breathing</td></tr>
<tr><td>☐ S1</td><td>☐ S2</td><td>☐ S3</td><td>☐ S4</td><td>☐ S5</td><td>☐ S6</td><td>☐ S7</td><td>☐ S8</td><td>☐ S9</td><td>☐ S10</td></tr>
<tr><th colspan="10">Meal trial with pudding</th></tr>
<tr><td colspan="10">Remove chest strap and accelerometer. Ensure patient is sitting in an upright position during the task and practicing the technique. Continue to provide verbal feedback and prompts, as necessary to ensure proper use of technique. “Now we want to see if you can use the technique while you’re eating a pudding snack.”</td></tr>
<tr><td>☐ S1</td><td>☐ S2</td><td>☐ S3</td><td>☐ S4</td><td>☐ S5</td><td>☐ S6</td><td>☐ S7</td><td>☐ S8</td><td>☐ S9</td><td>☐ S10</td></tr>
<tr><th colspan="10">Meal trial measures</th></tr>
<tr><td colspan="5">☐ SWAL-QoL</td><td colspan="5">☐ Qualitative Interview Questions</td></tr>
</table>

Note time at end of session: ___________

APPENDIX B: BASELINE AND POST-TREATMENT SURVERY AND INTERVIEW DATA COLLECTION FORMS

Pre-Survey

Below are some physical problems that people with swallowing problems sometimes experience. While you were eating the pudding snack, how often did you experience each problem as a result of your swallowing problem?

	Almost always	Often	Sometimes	Hardly ever	Never
Coughing	1	2	3	4	5
Choking when you eat	1	2	3	4	5
Having to clear your throat	1	2	3	4	5
Food sticking in your throat	1	2	3	4	5

While you were eating the pudding snack, how often were the following statements true for you because of your swallowing problem?

	Always true	Often true	Sometimes true	Hardly ever true	Never true
My swallowing problem depresses me.	1	2	3	4	5
Having to be so careful when I eat or drink annoys me.	1	2	3	4	5
I’ve been discouraged by my swallowing problem.	1	2	3	4	5
My swallowing problem frustrates me.	1	2	3	4	5
I get impatient dealing with my swallowing problem.	1	2	3	4	5

Post-Survey

Below are some physical problems that people with swallowing problems sometimes experience. While you were eating the pudding snack, how often did you experience each problem as a result of your swallowing problem?

	Almost always	Often	Sometimes	Hardly ever	Never
Coughing	1	2	3	4	5
Choking when you eat	1	2	3	4	5
Having to clear your throat	1	2	3	4	5
Food sticking in your throat	1	2	3	4	5

While you were eating the pudding snack, how often were the following statements true for you because of your swallowing problem?

	Always true	Often true	Sometimes true	Hardly ever true	Never true
My swallowing problem depresses me.	1	2	3	4	5
Having to be so careful when I eat or drink annoys me.	1	2	3	4	5
I've been discouraged by my swallowing problem.	1	2	3	4	5
My swallowing problem frustrates me.	1	2	3	4	5
I get impatient dealing with my swallowing problem.	1	2	3	4	5

How do you feel about using the technique you learned in this research study during your normal eating routines?

What did you like and dislike about the technique you learned during this research study?

How confident do you feel using this technique independently (without help or instruction from the researcher)?

Do you feel you need more training to use the technique you learned in this study? If yes, please explain.

Describe your level of anxiety when swallowing while using this technique.

APPENDIX C: PARTICIPANTS' BASELINE AND POST-TREATMENT RESPONSES TO QUESTIONS ON THE MENTAL HEALTH (MH) AND SYMPTOM FREQUENCY (SF) SUBSECTIONS OF THE SWAL-QOL

ID		MH #1	MH #2	MH #3	MH #4	MH #5	Average MH
1	Pre	5	4	4	4	4	4.20
	Post	5	5	5	5	5	5.00
2	Pre	5	4	5	5	5	4.80
	Post	5	4	5	4	4	4.40
3	Pre	4	4	5	5	5	4.60
	Post	4	5	5	5	5	4.80
4	Pre	4	4	4	4	4	4.00
	Post	5	5	5	5	5	5.00
5	Pre	5	5	5	5	5	5.00
	Post	5	5	5	5	5	5.00
6	Pre	5	4	4	5	5	4.60
	Post	5	5	5	5	5	5.00
7	Pre	5	5	5	5	5	5.00
	Post	5	5	5	5	5	5.00
8	Pre	3	3	3	3	3	3.00
	Post	3	3	3	3	3	3.00

ID		SF #1	SF #2	SF #3	SF #4	Average SF
1	Pre	5	5	5	5	5.00
	Post	5	5	5	5	5.00
2	Pre	5	5	4	5	4.75
	Post	5	5	4	5	4.75
3	Pre	5	5	5	5	5.00
	Post	5	5	5	5	5.00
4	Pre	5	5	5	5	5.00
	Post	5	5	5	5	5.00
5	Pre	5	5	4	5	4.75
	Post	5	5	4	5	4.75
6	Pre	3	5	5	5	4.50
	Post	5	5	3	5	4.50
7	Pre	5	5	5	5	5.00
	Post	5	5	5	5	5.00
8	Pre	4	4	4	3	3.75
	Post	4	4	4	4	4.00

APPENDIX D: PARTICIPANTS' RESPONSES TO QUALITATIVE INTERVIEW QUESTIONS IN THE POST-TREATMENT CONDITION

Question	Participant	Response
How do you feel about using the technique you learned in this research study during your normal eating routines?	1	"(I) would use (it)."
	2	"Good. Going to try (to use it)."
	3	"I'll try it."
	4	"Going to implement it right a way."
	5	"(I) don't have much trouble. Don't feel I need it at this point."
	6	"It went down smoother. If I thought about it, I'd use it."
	7	"It's going to help (me) to some extent."
	8	"Changed the rhythm of swallowing and had to get more involved."
What did you like and dislike about the technique you learned during this research study?	1	*Like:* "It forces you to breathe. Sometimes when you're not concentrating, you end up choking. Thanks for the pudding!" *Dislike:* "Nothing. I enjoyed it."
	2	*Like:* "If it will keep me from choking, I definitely like it. *Dislike:* "No."
	3	*Like:* "Hard to tell." *Dislike:* "No."
	4	*Like:* "It was easier to swallow with a full breath." *Dislike:* "No."
	5	*Like:* No response. *Dislike:* "No."
	6	*Like:* "It was okay." *Dislike:* "None."
	7	*Like:* "(I) learned a few things to help me." *Dislike:* "Nothing."
	8	*Like:* "It gives you more control because you are planning ahead. Have to think more about what you're doing when you swallow." *Dislike:* No response.
How confident do you feel using this technique independently (without help or instruction from the researcher)?	1	"Good. I'm good. I can do it."
	2	"I feel confident, but need practice."
	3	"I think I could do it."
	4	"Very confident."
	5	"I feel confident."
	6	"Wouldn't bother me a bit."
	7	"Fairly confident."
	8	"Yes."

Question	Participant	Response
Do you feel you need more training to use the technique you learned in this study? If yes, please explain.	1	"No, I think I'm okay. Now I just need to practice."
	2	"Not right now. I'll do what I can and see how it goes."
	3	"I would prefer more training."
	4	"No."
	5	"No."
	6	"Feel okay now."
	7	"No, I just have to remember to do it."
	8	"Keeps you busy mentally."
Describe your level of anxiety when swallowing while using this technique.	1	"I wasn't anxious. I felt comfortable. It was good."
	2	"I was somewhat anxious because I was trying something different. Getting air in isn't something I'm used to doing. But I think it will be better because you're automatically trying to push things out. I'm happy with it."
	3	"I'm not anxious but can't say I feel better."
	4	"None at all. Felt normal."
	5	"Anxious at the beginning but fine at the end."
	6	"No anxiety."
	7	"Not too anxious. Just being careful."
	8	"Too busy thinking to let anything else bother you."

APPENDIX E: SPSS OUTPUT FOR STATISTICAL ANALYSIS OF PRIMARY RESEARCH QUESTION

Descriptive Statistics

	N	Mean	Std. Deviation	Minimum	Maximum
Baseline xE	8	6.63	3.159	2	10
Post-treatment xE	8	9.50	1.414	6	10

Wilcoxon Signed Ranks Test

Ranks

		N	Mean Rank	Sum of Ranks
Post-treatment xE - Baseline xE	Negative Ranks	0[a]	.00	.00
	Positive Ranks	7[b]	4.00	28.00
	Ties	1[c]		
	Total	8		

a. Post-treatment xE < Baseline xE

b. Post-treatment xE > Baseline xE

c. Post-treatment xE = Baseline xE

Test Statistics[b]

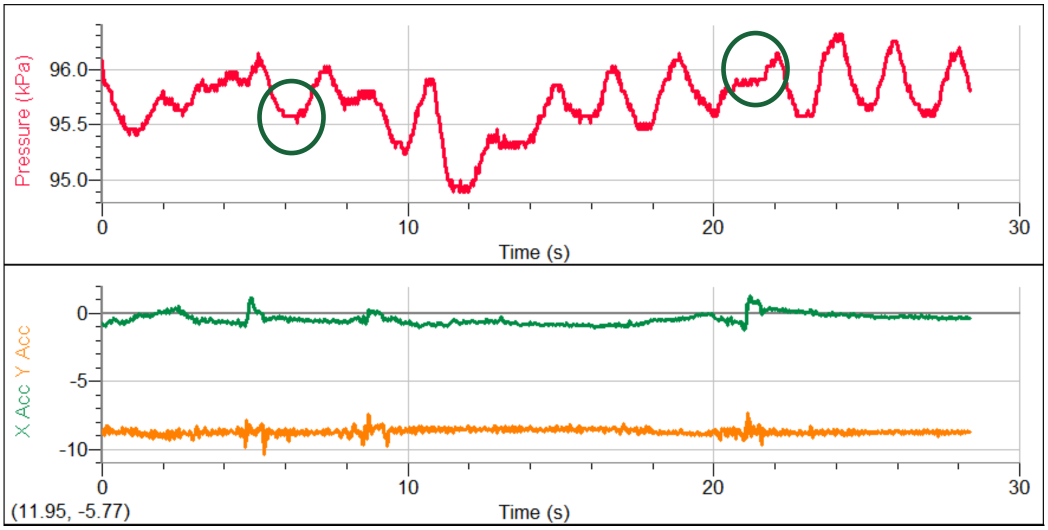
	Post-treatment xE - Baseline xE
Z	-2.388[a]
Asymp. Sig. (2-tailed)	.017

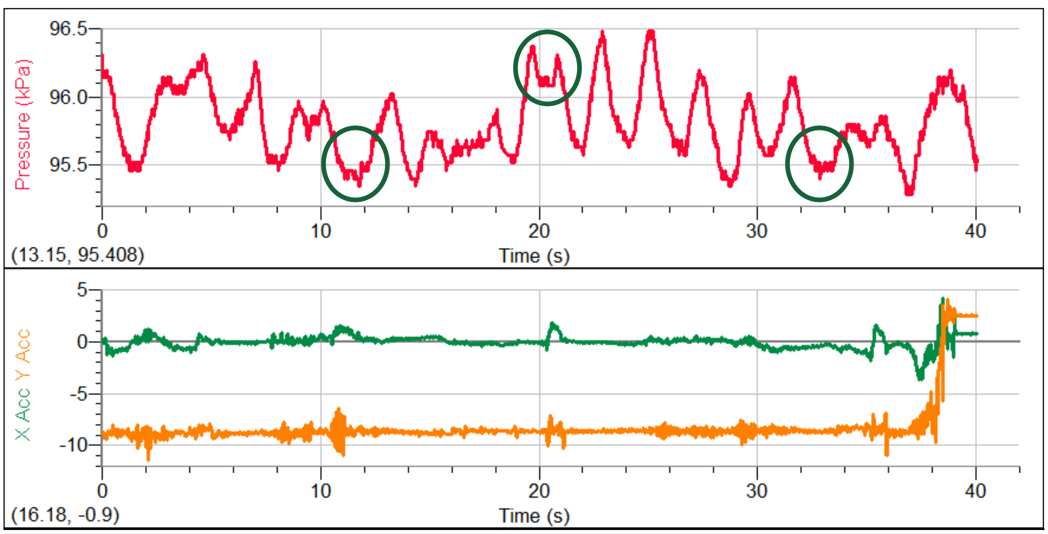
a. Based on negative ranks.

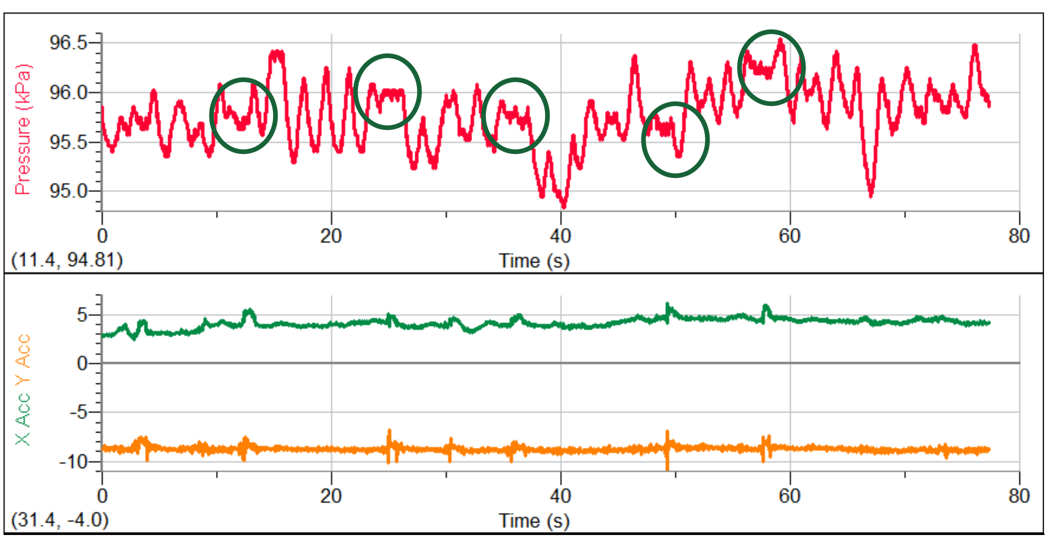
b. Wilcoxon Signed Ranks Test

APPENDIX F: PARTICIPANTS' BASELINE AND POST-TREATMENT RESPIRATORY PHASE DATA

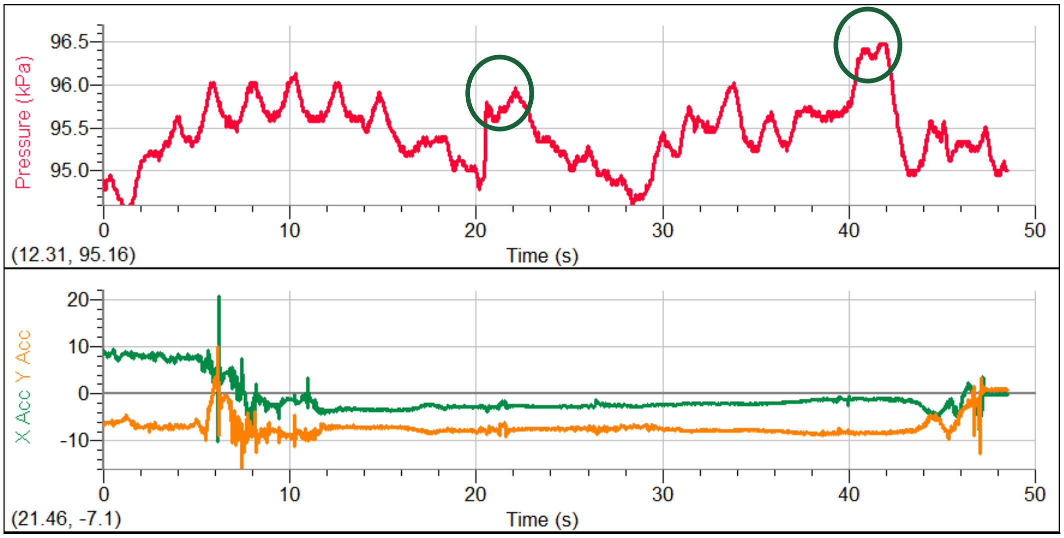
PARTICIPANT 1: BASELINE

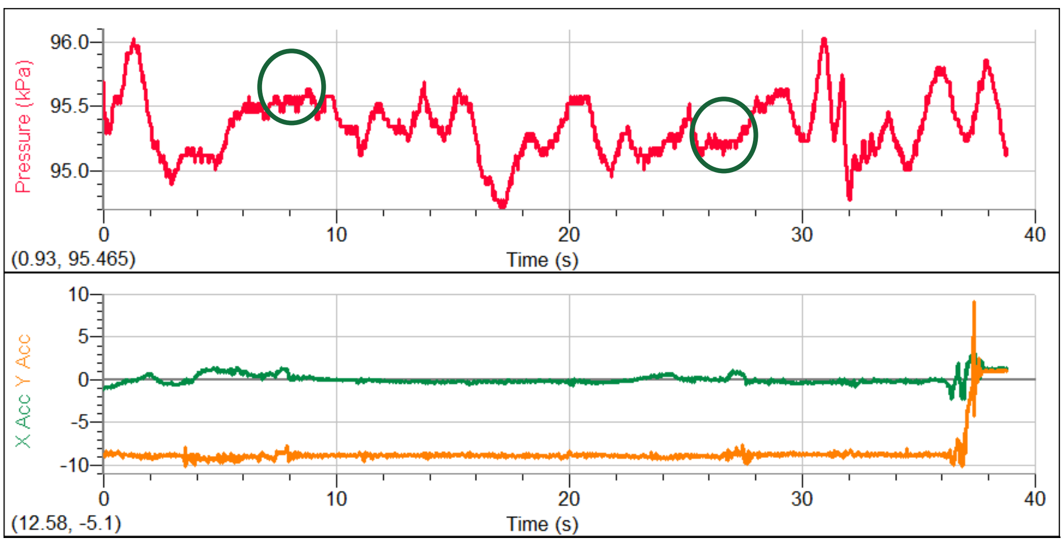


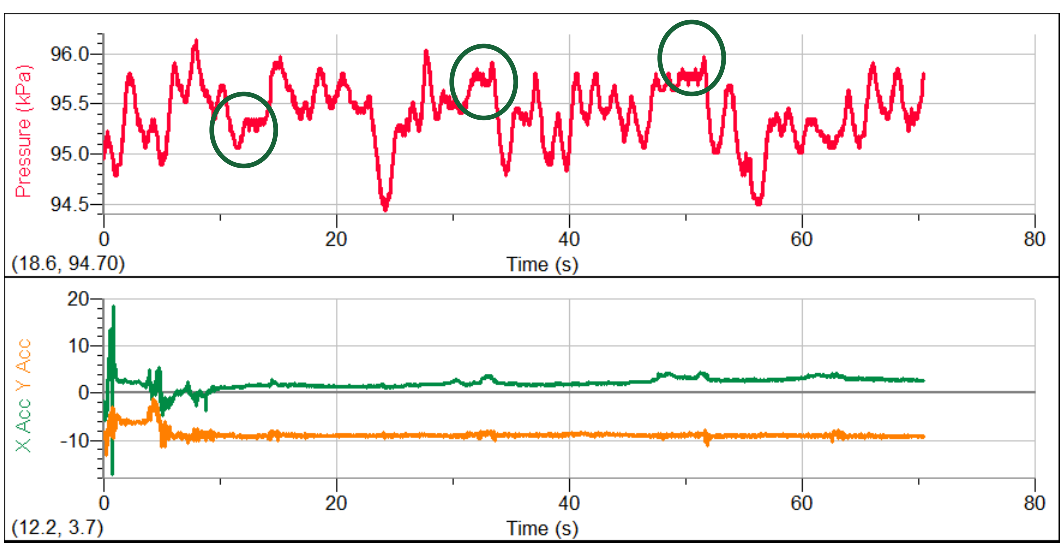




PARTICIPANT 1: POST-TREATMENT







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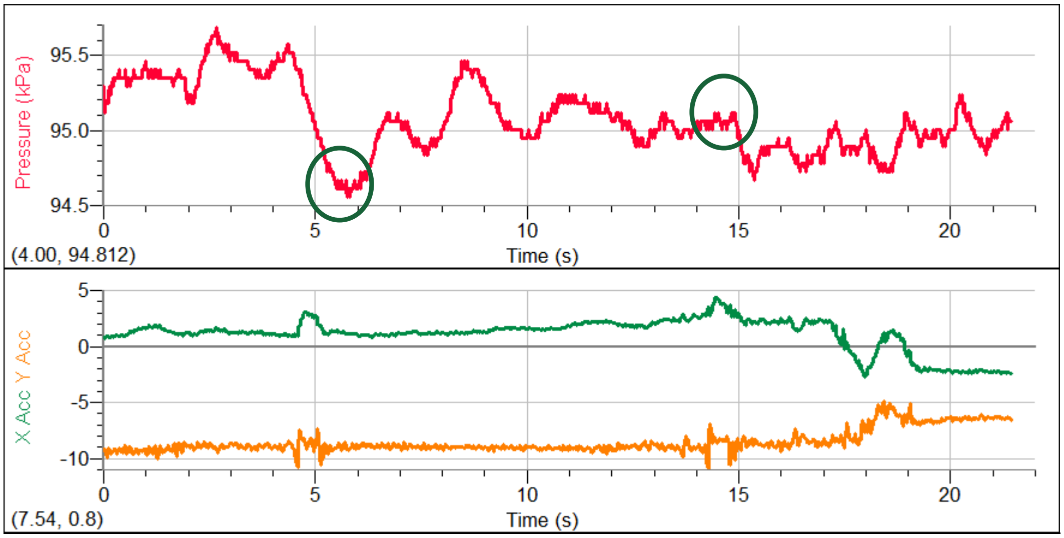
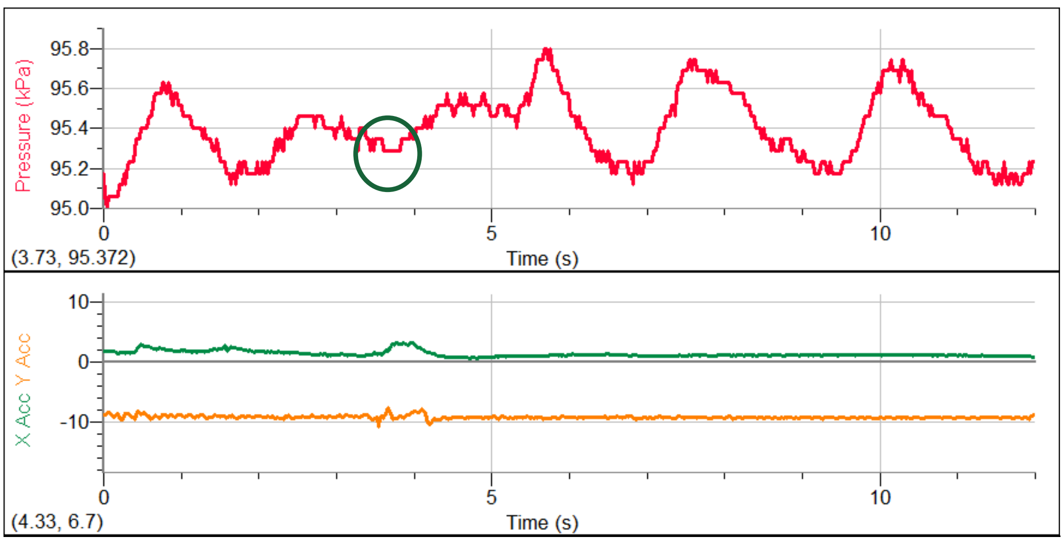
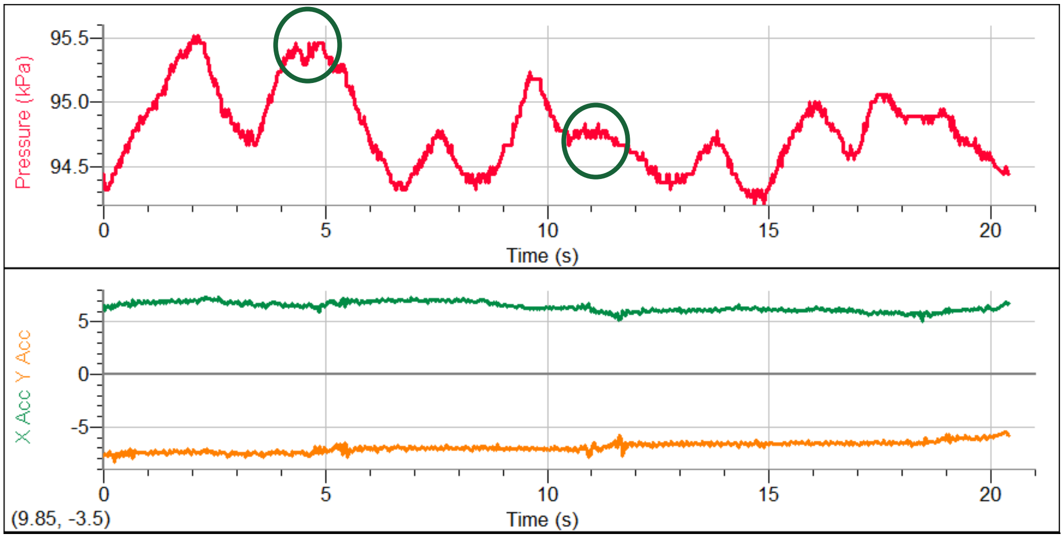
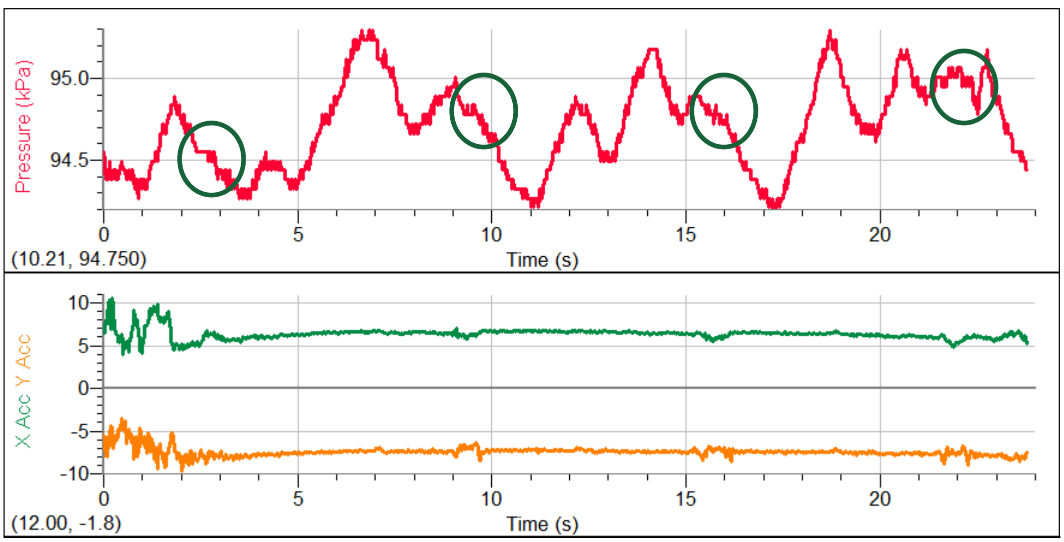
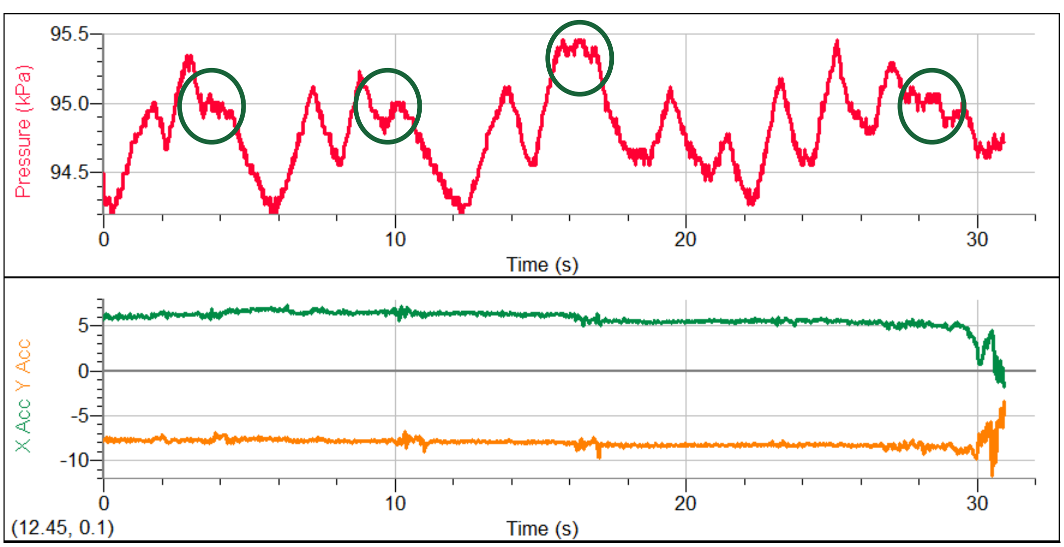




PARTICIPANT 2: BASELINE







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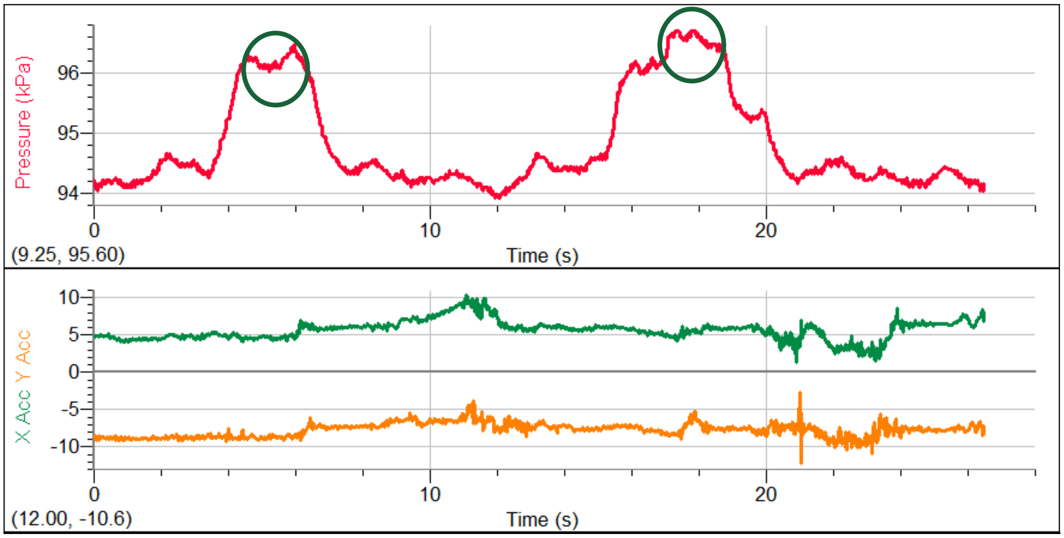

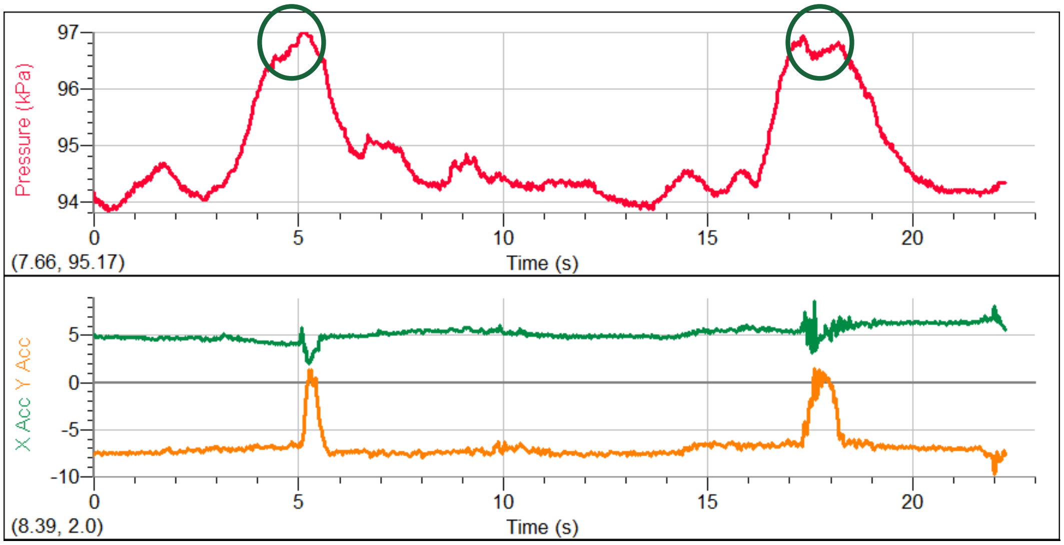

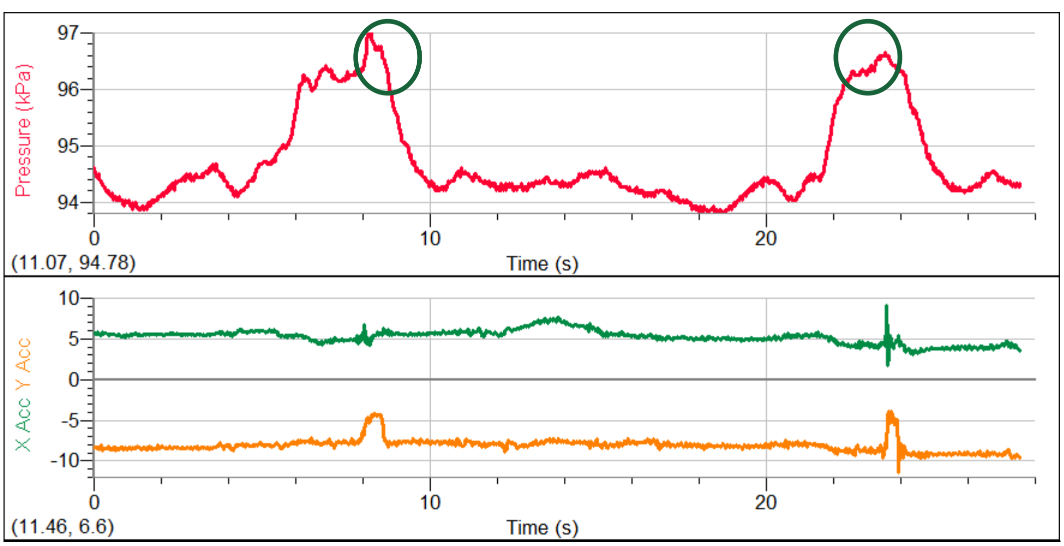

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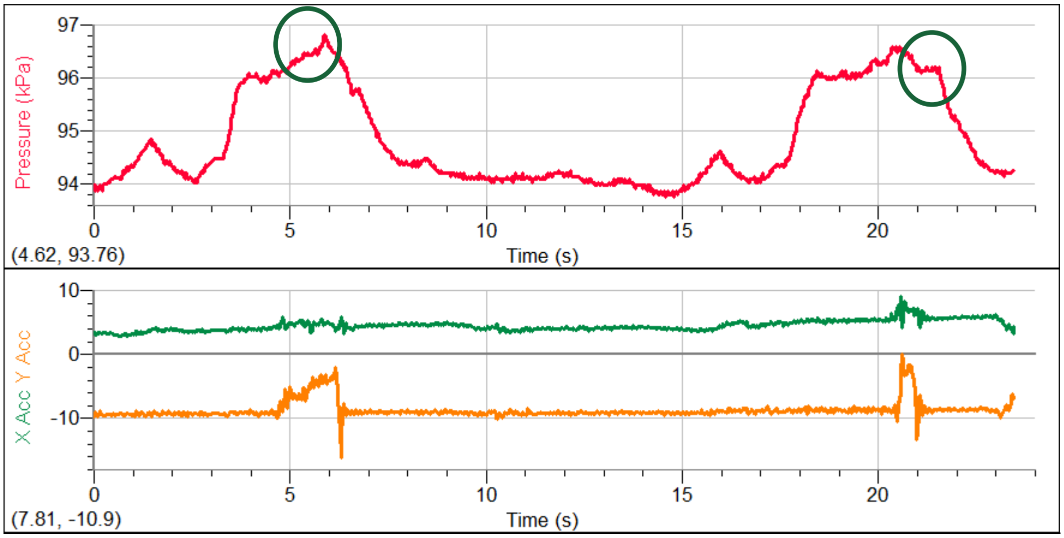

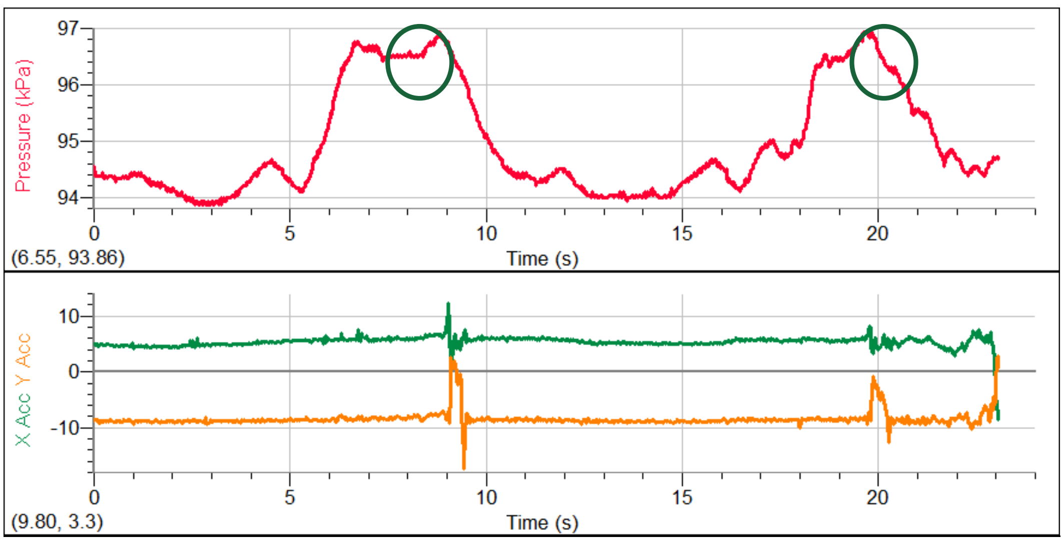

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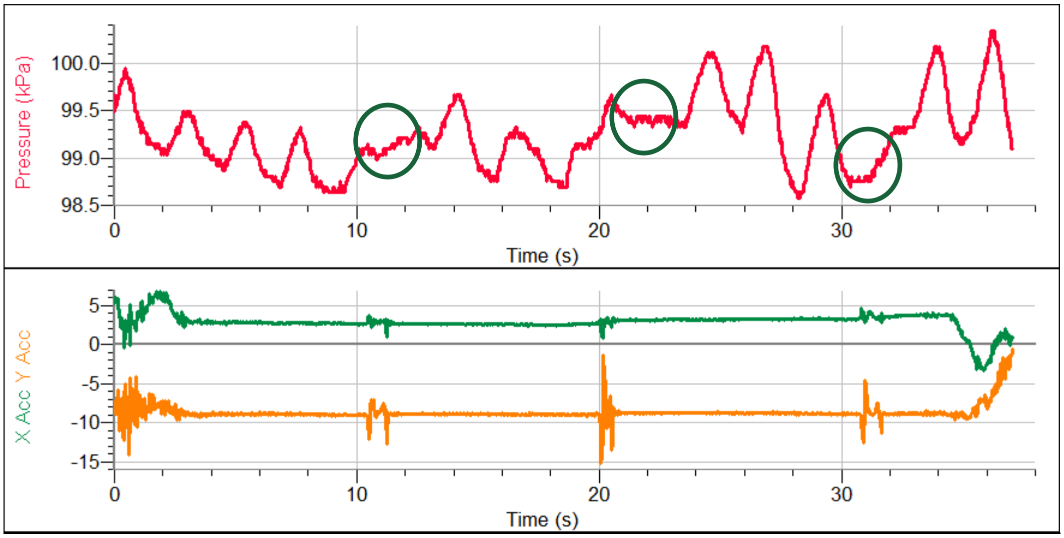

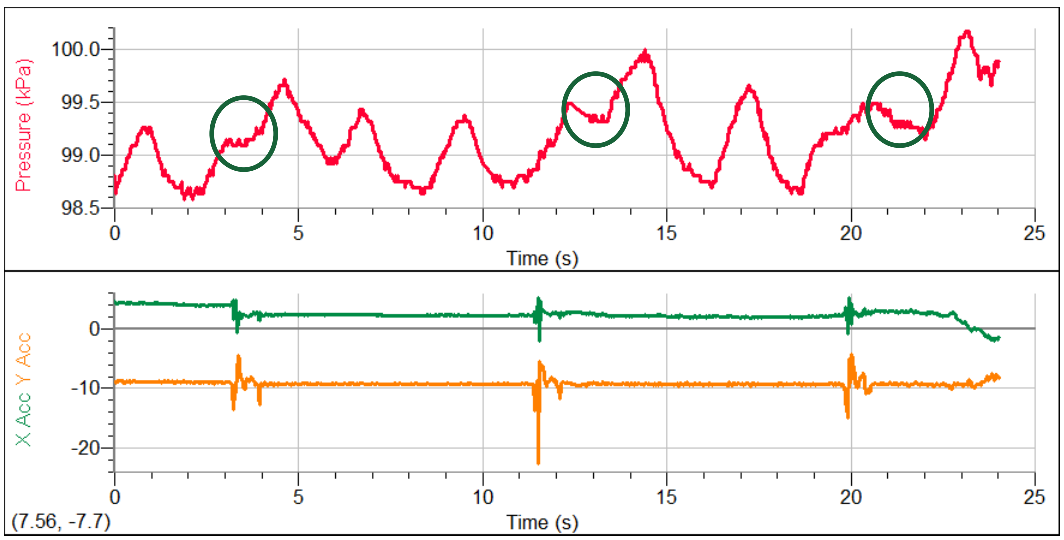

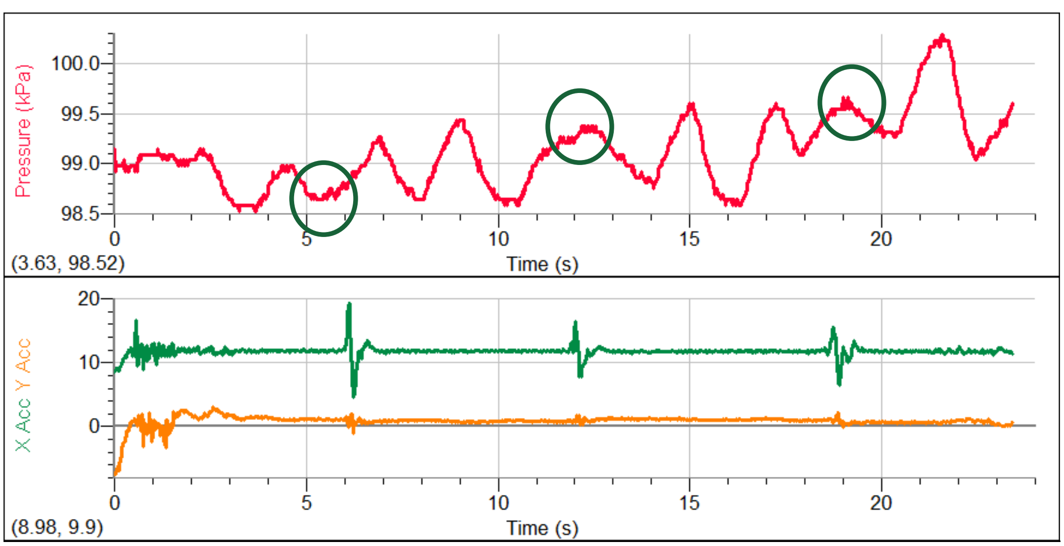

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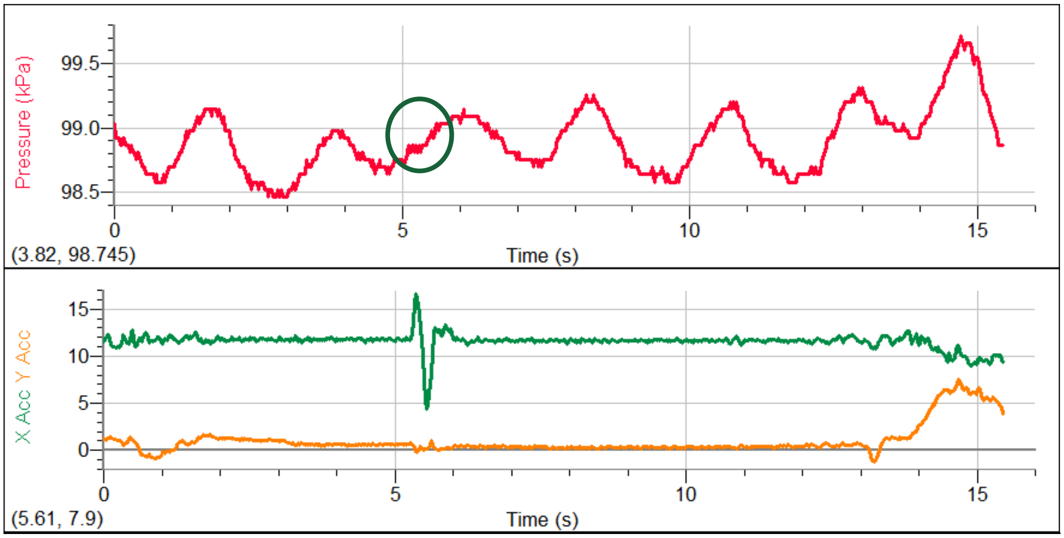

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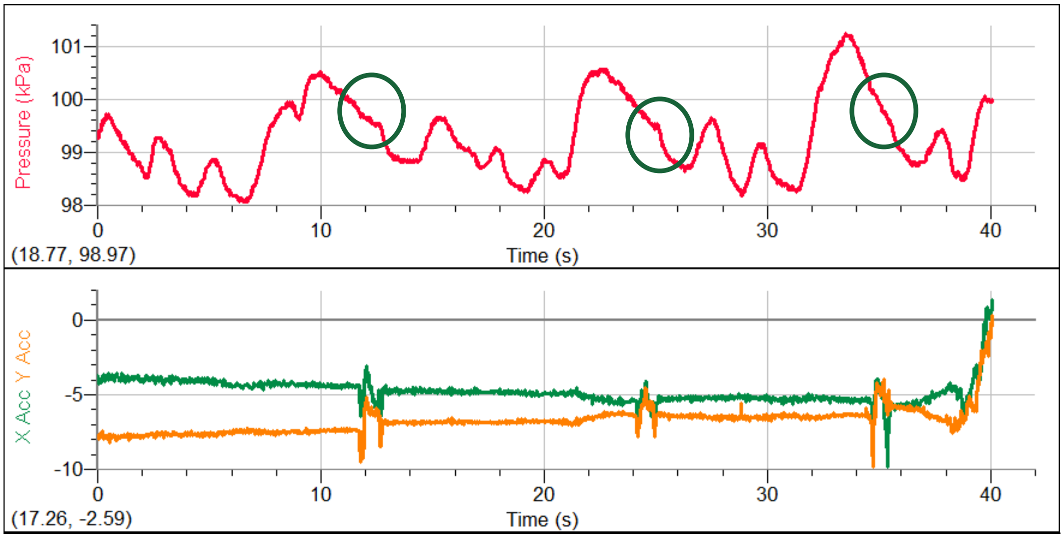

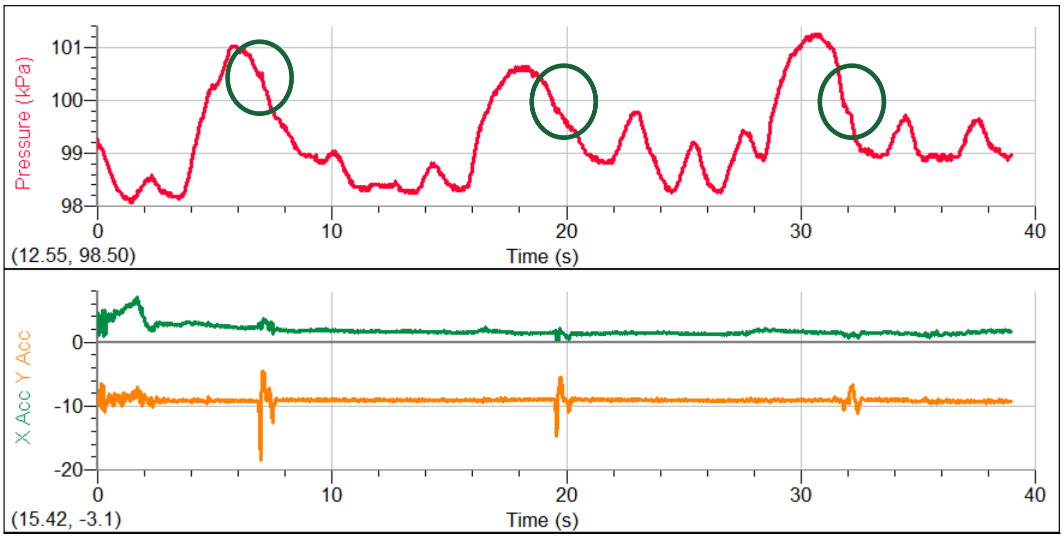

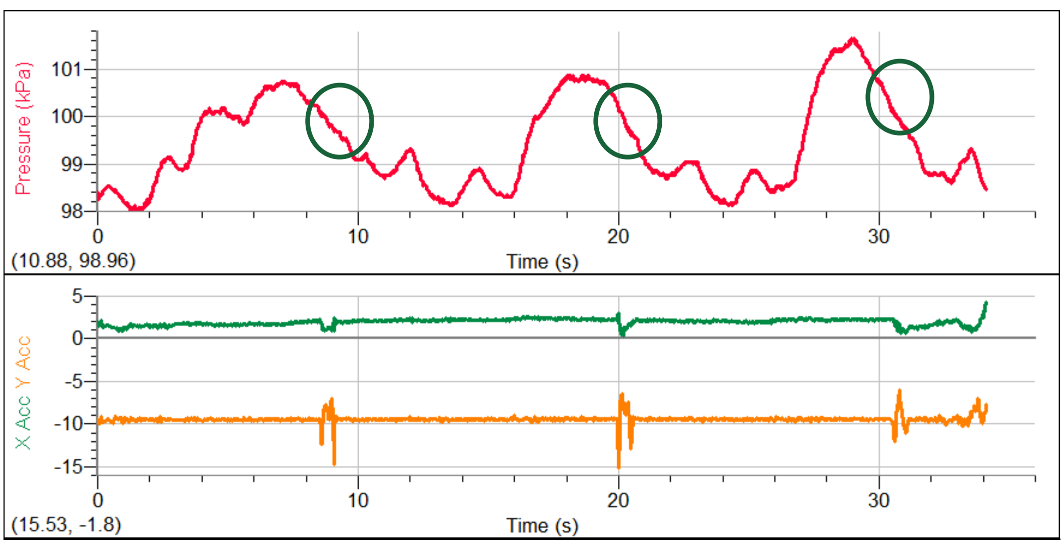

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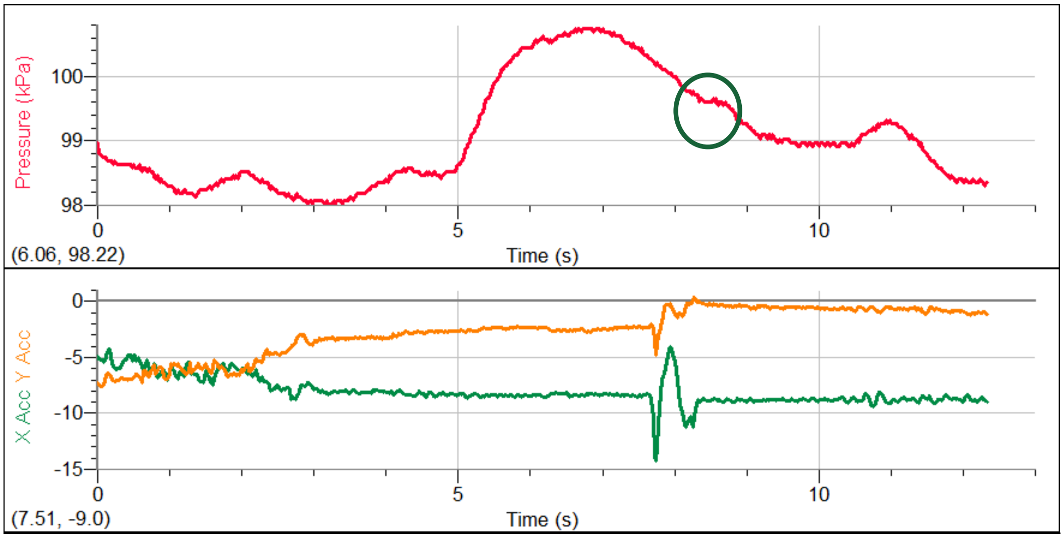

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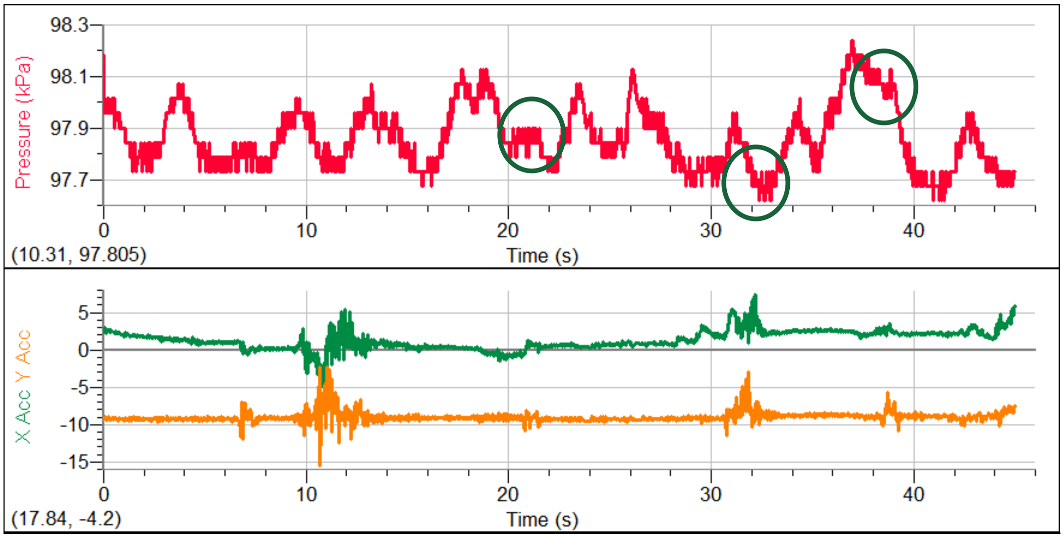

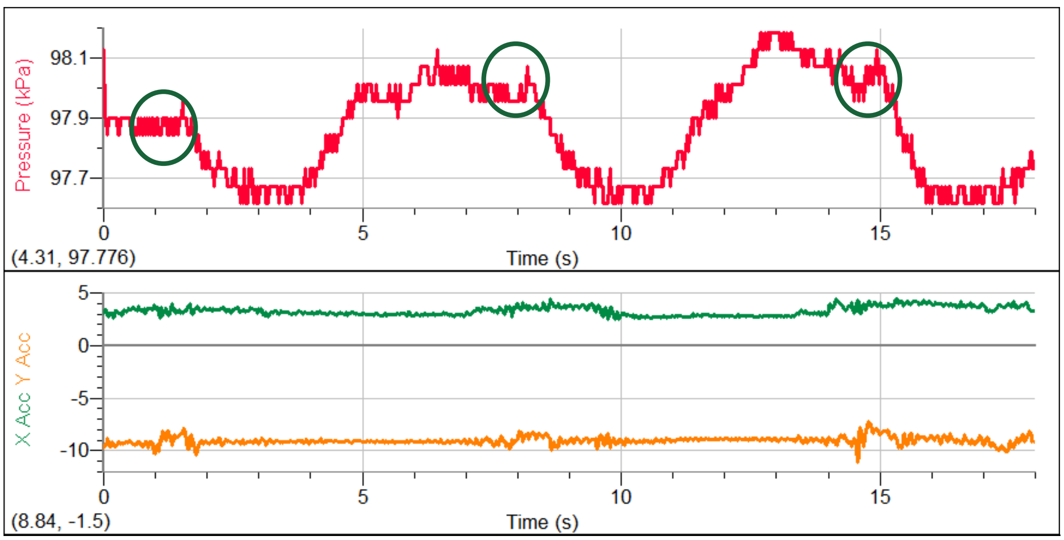

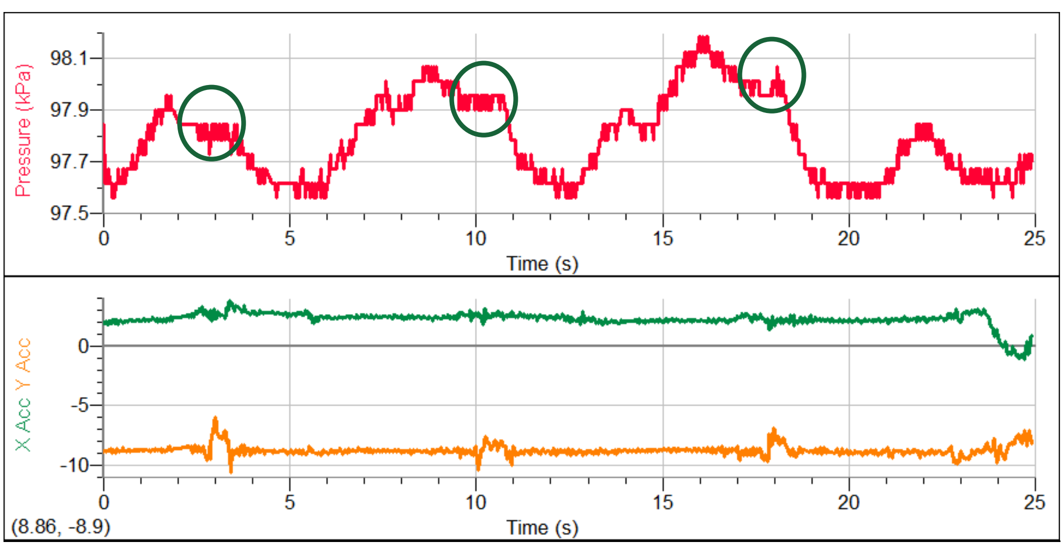

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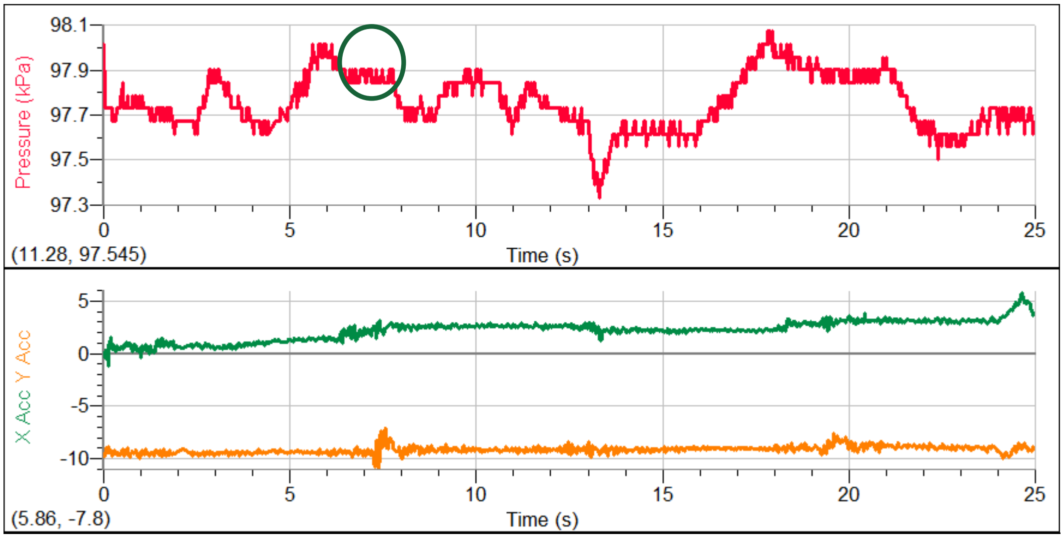

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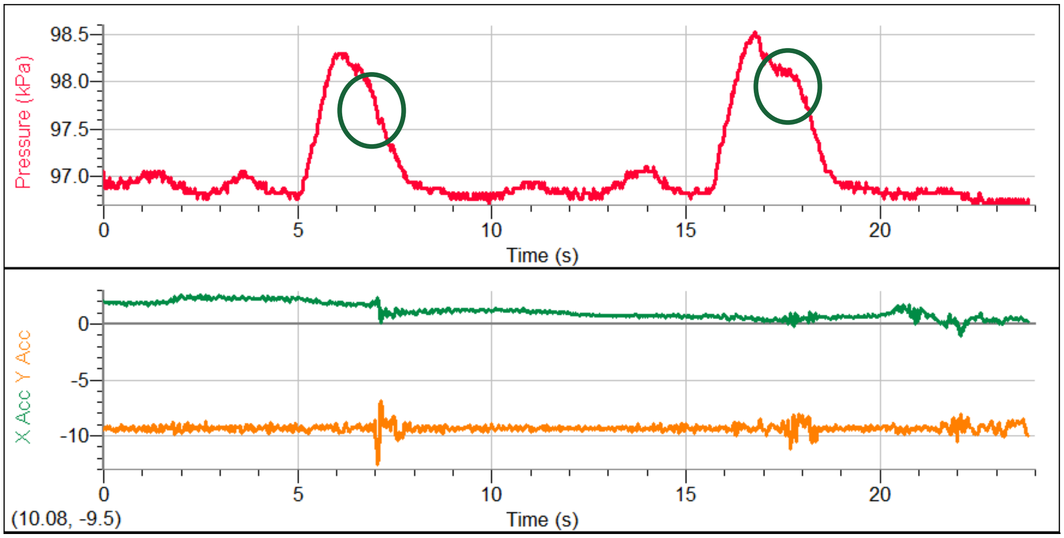

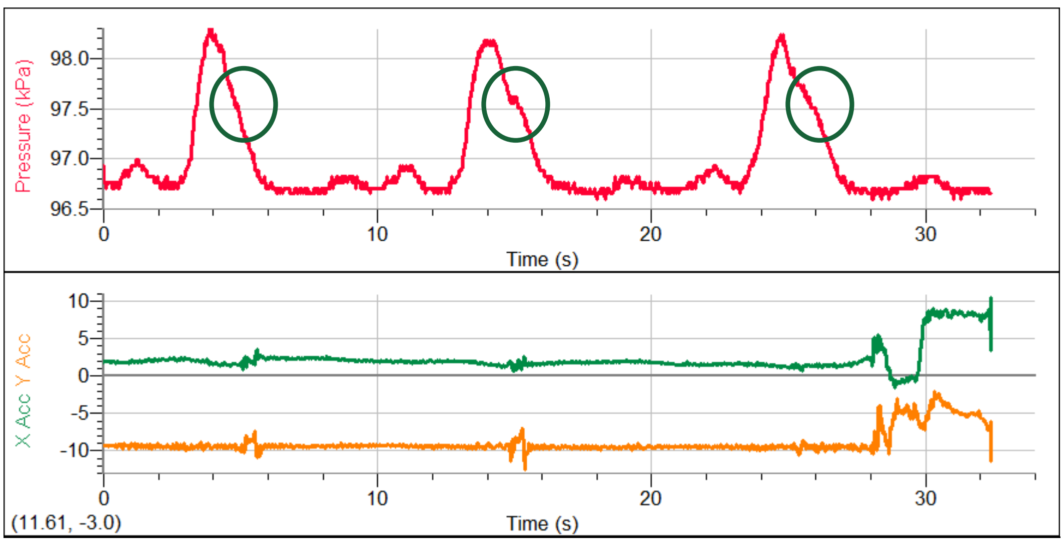

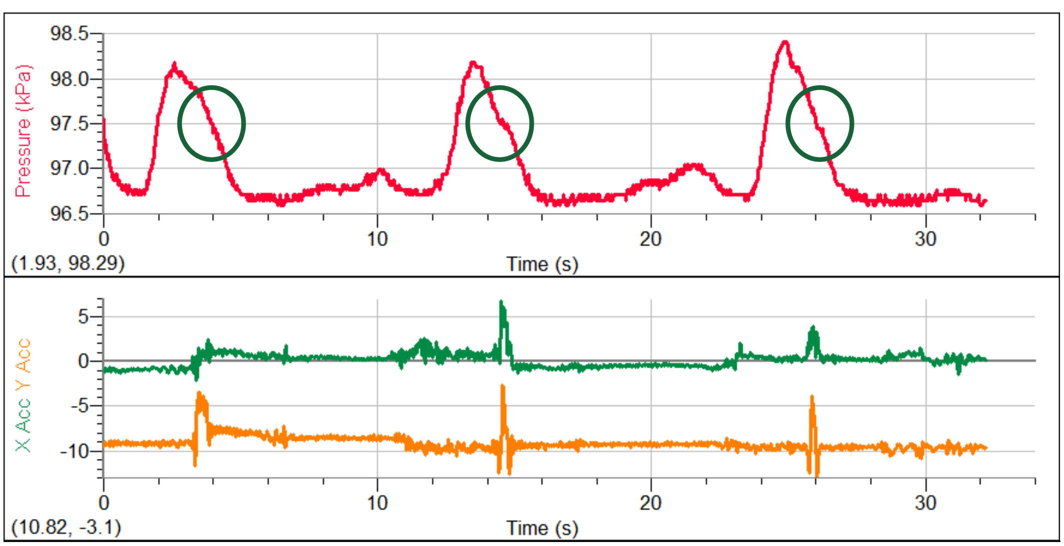

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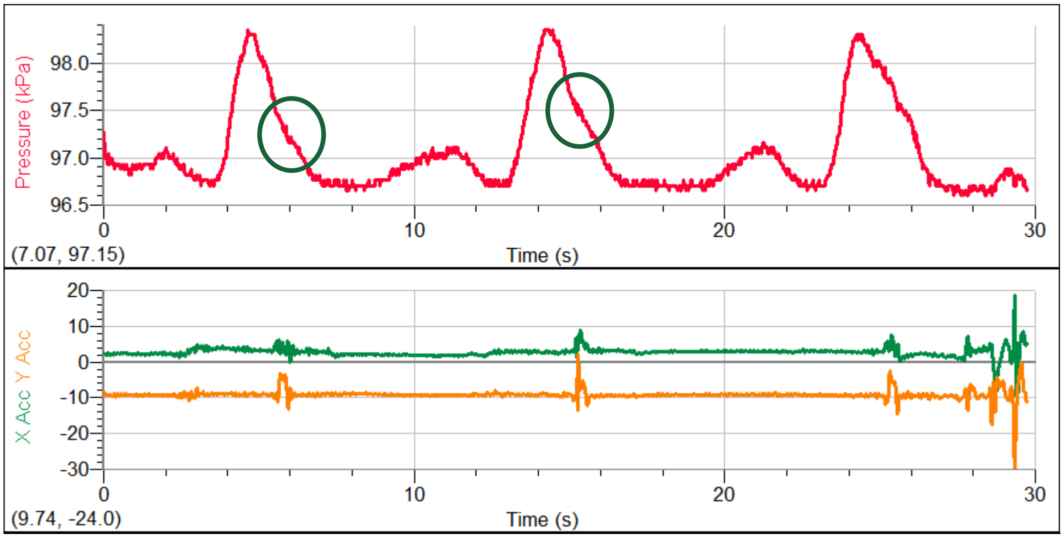

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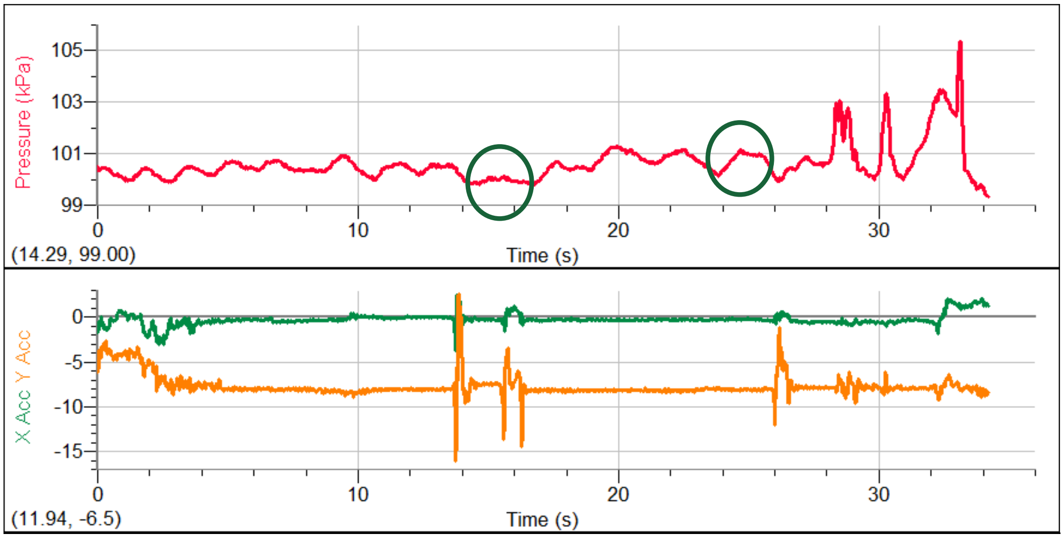

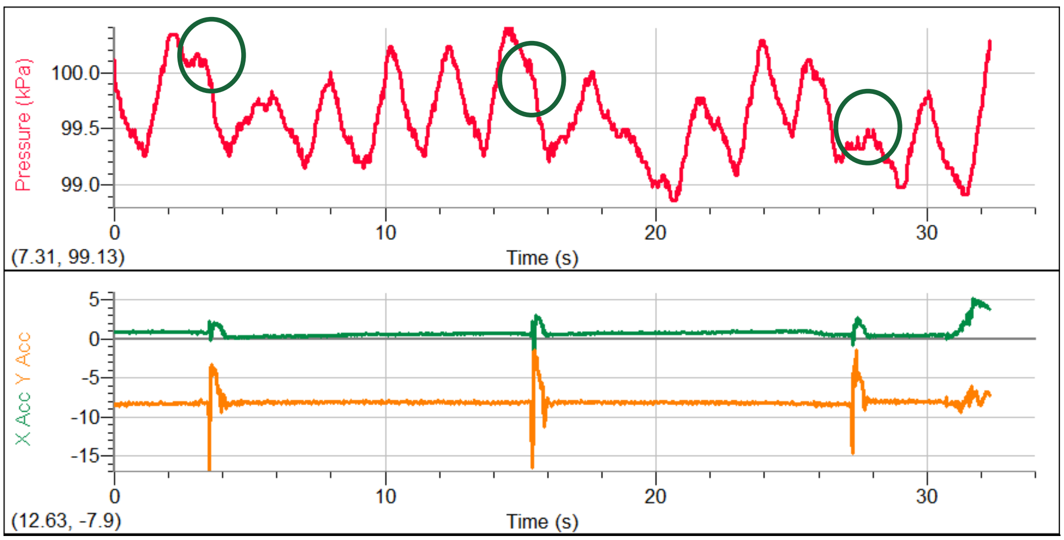

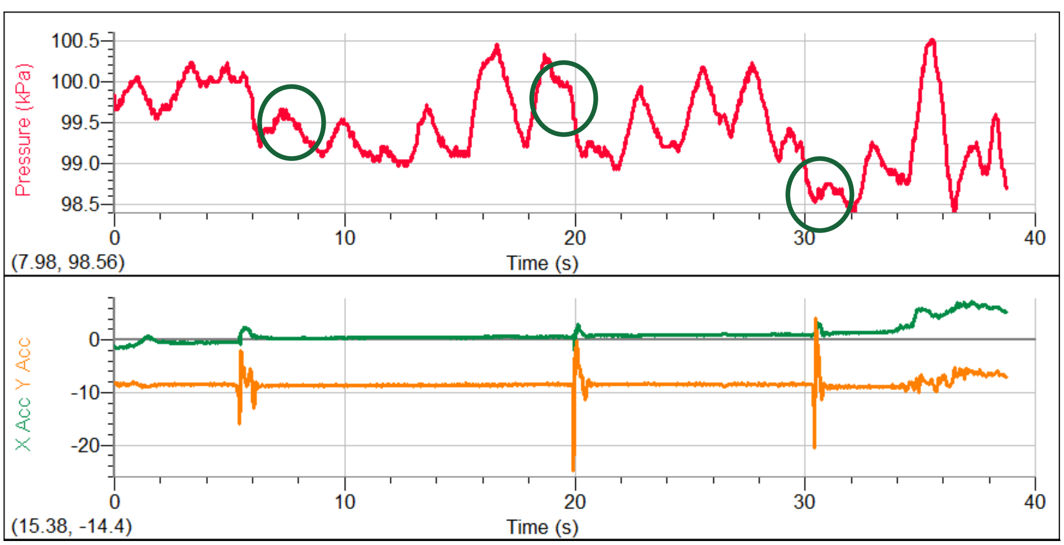

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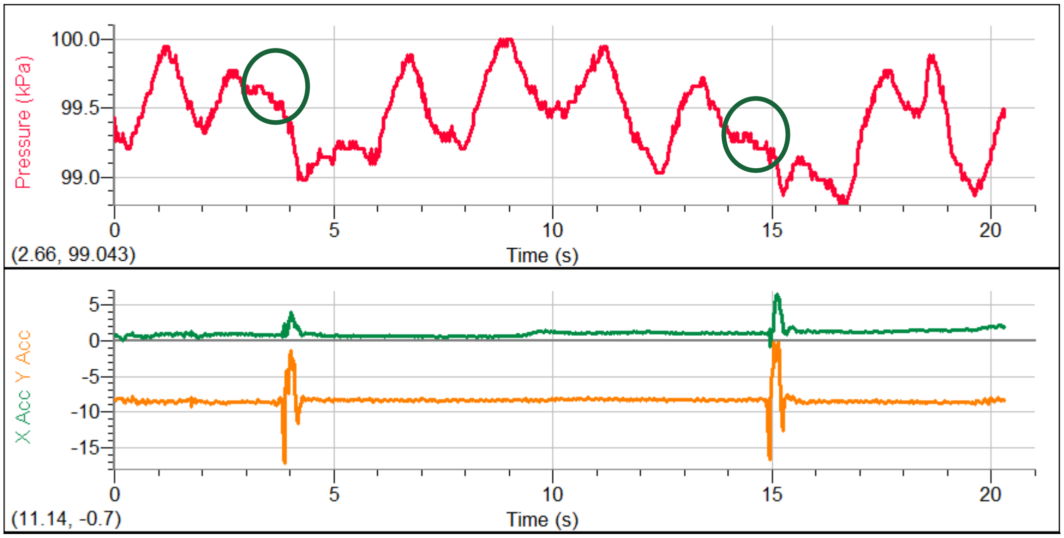
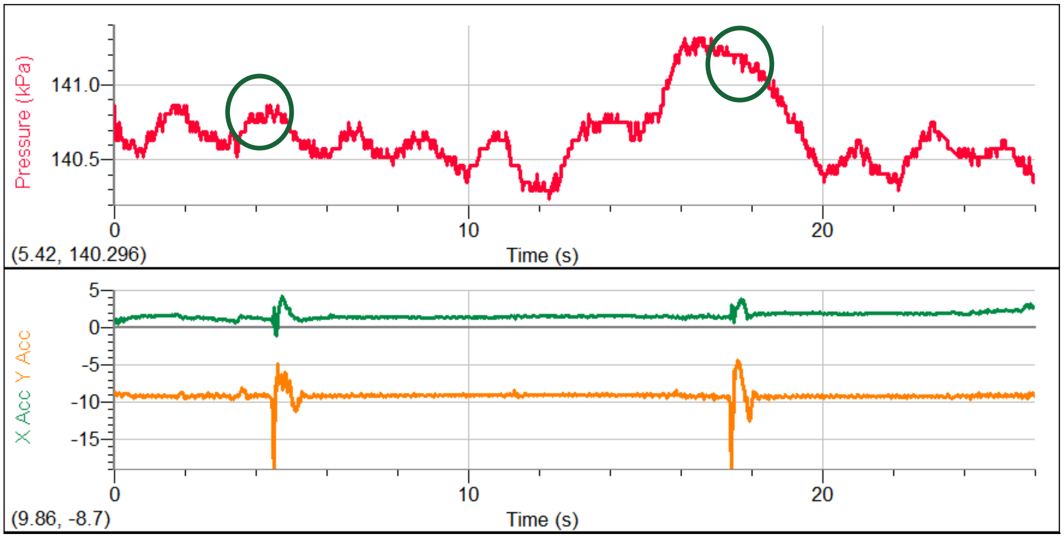
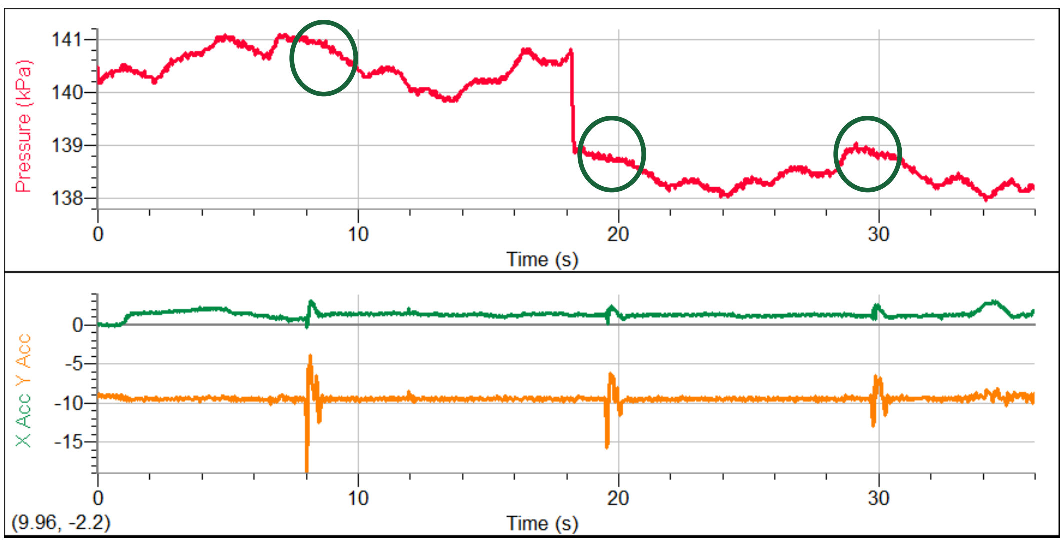
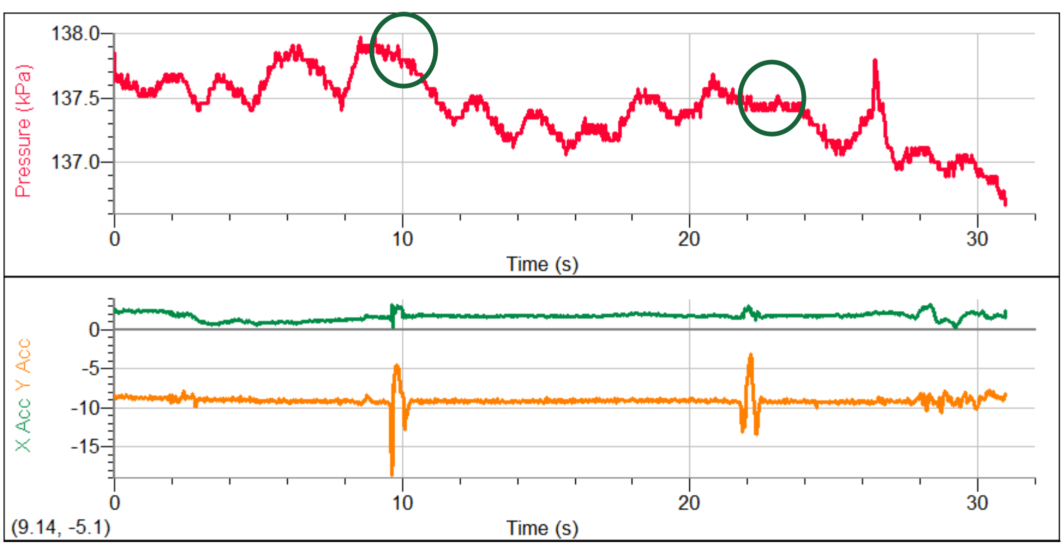
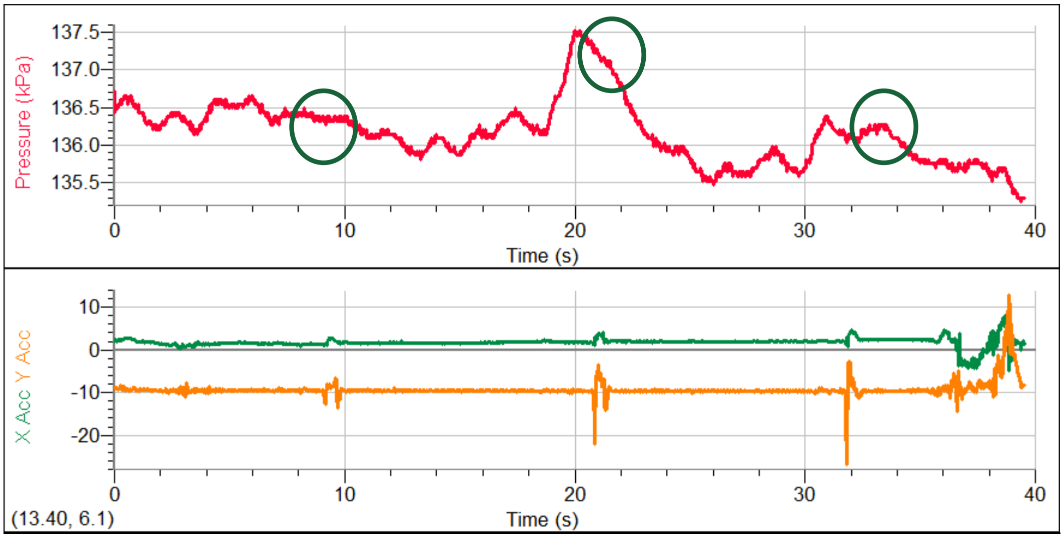


PARTICIPANT 5: POST-TREATMENT



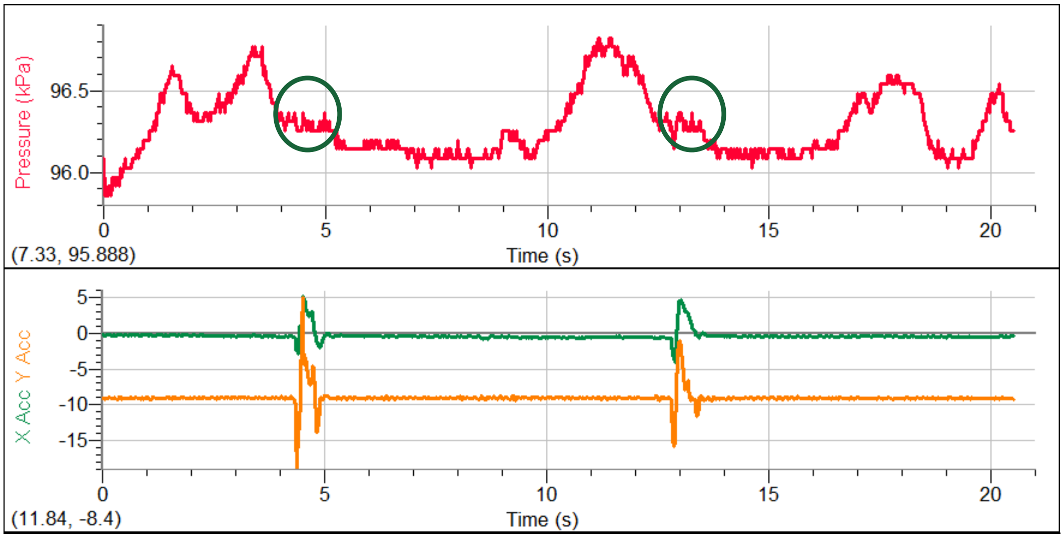


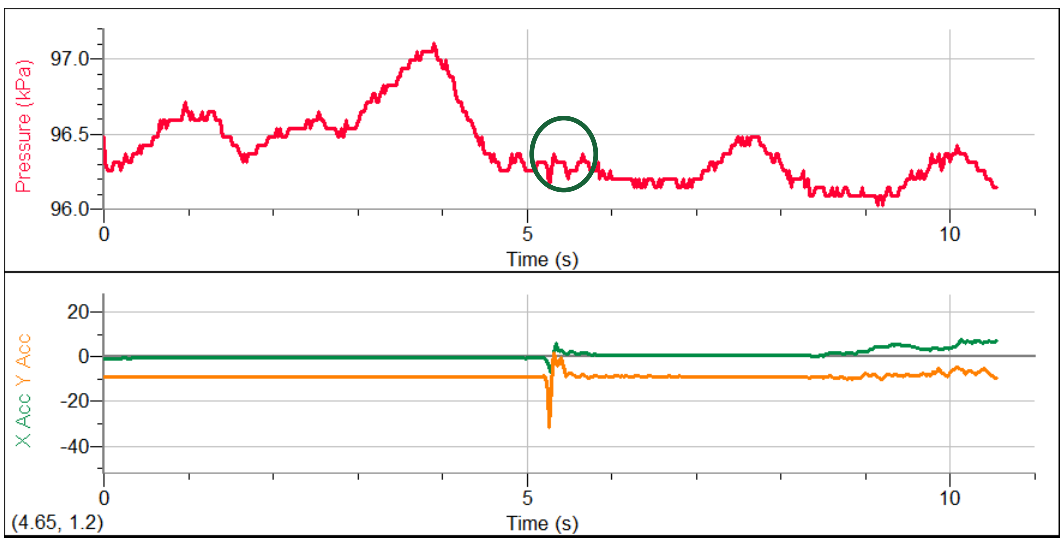


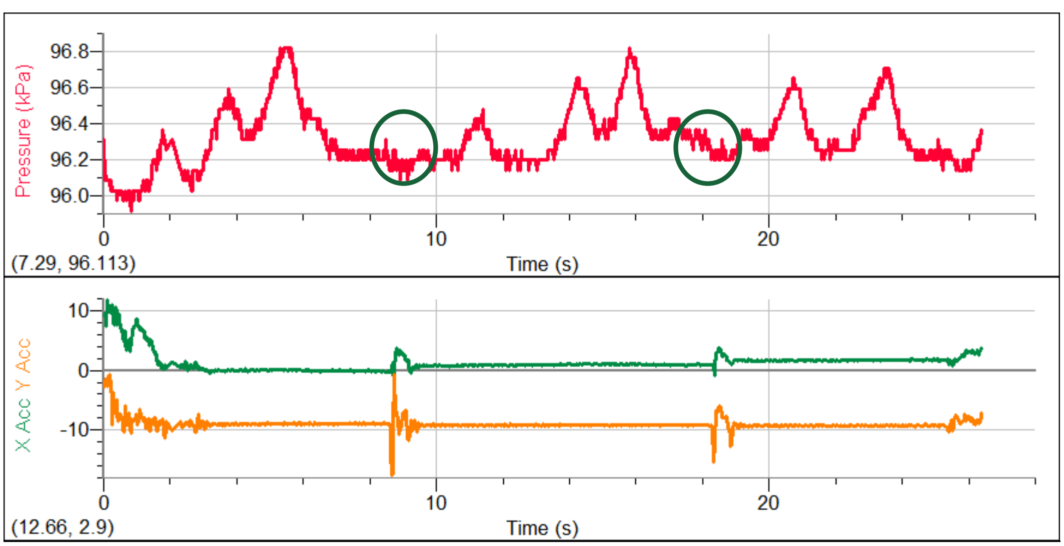


Pressure (kPa)
137.5
137.0
136.5
136.0
135.5
0
10
20
30
40
Time (s)
X Acc Y Acc
10
0
-10
-20
0
10
20
30
40
(13.40, 6.1)
Time (s)

PARTICIPANT 6: BASELINE







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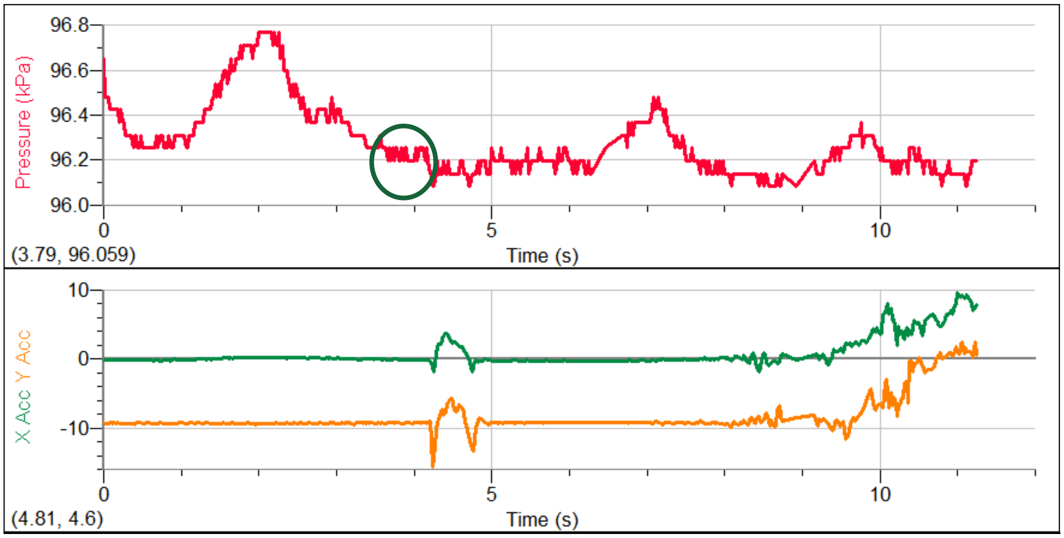
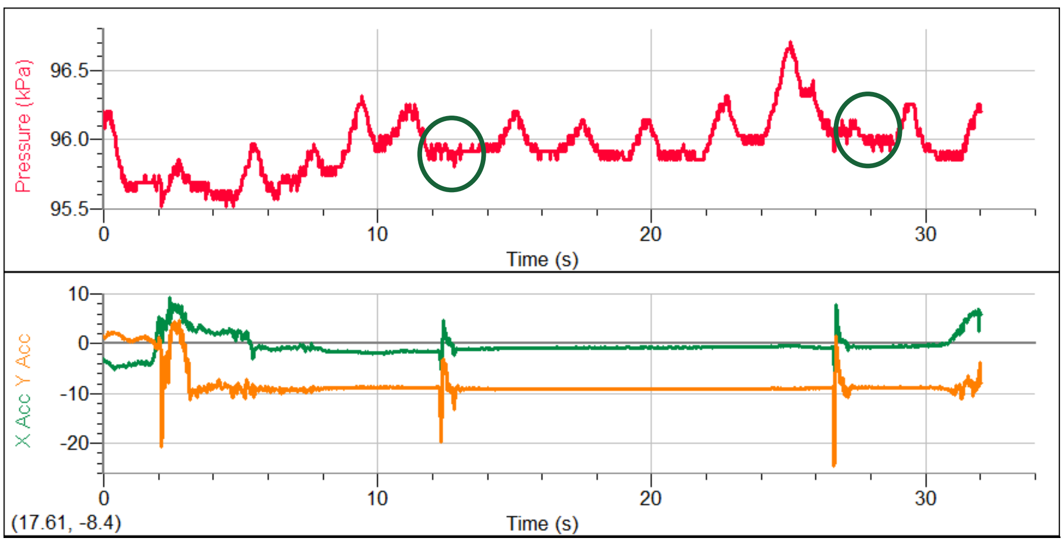
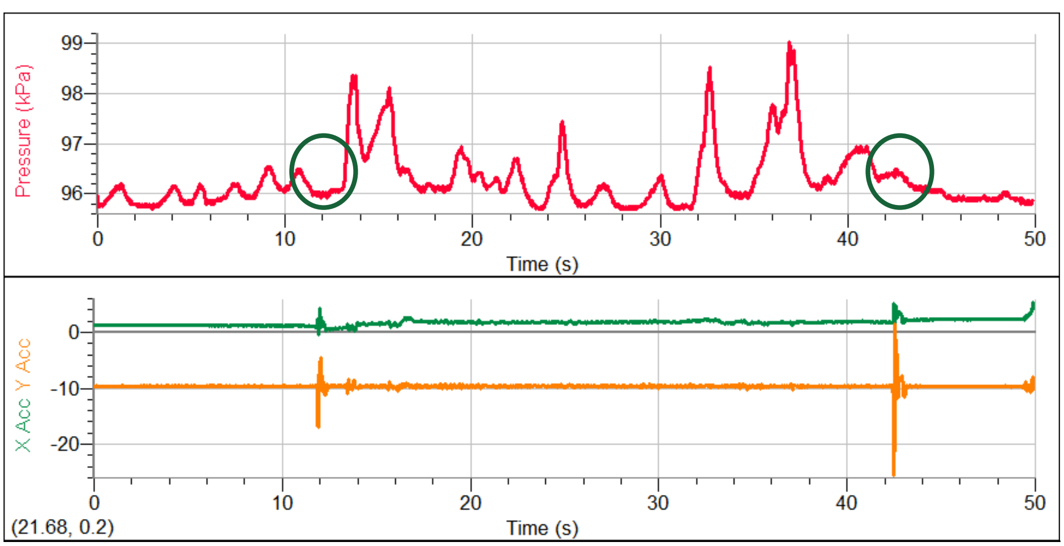
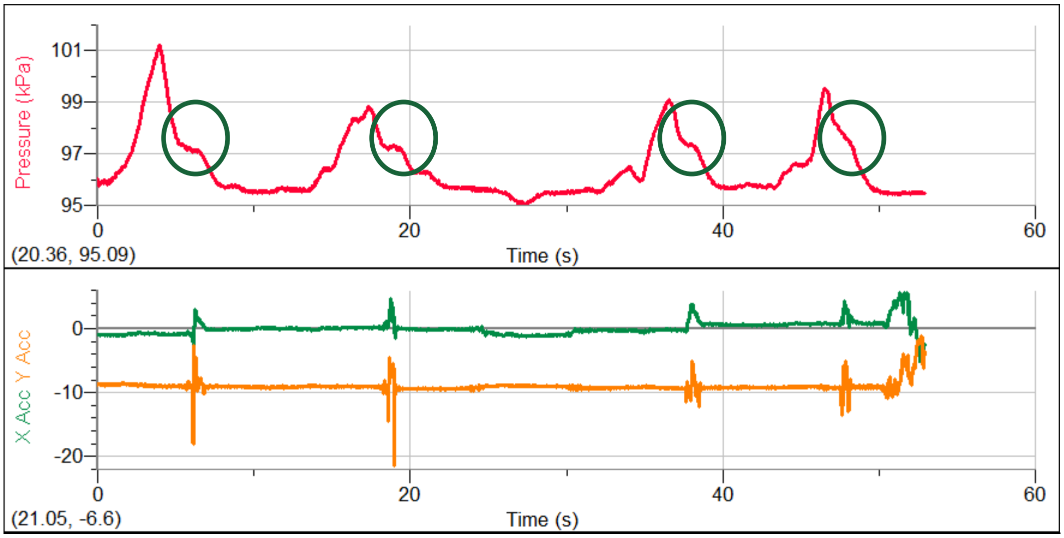
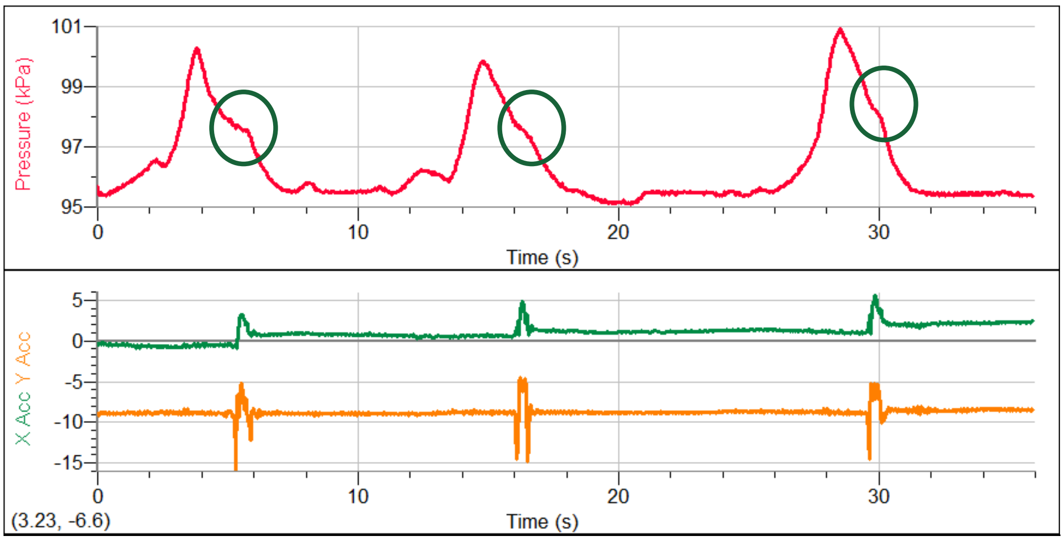


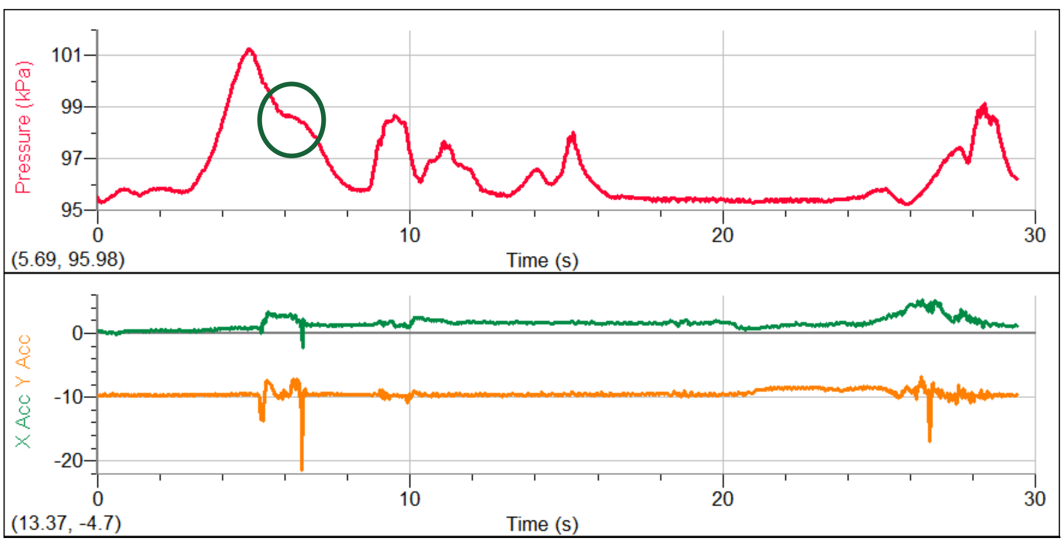




PARTICIPANT 6: POST-TREATMENT







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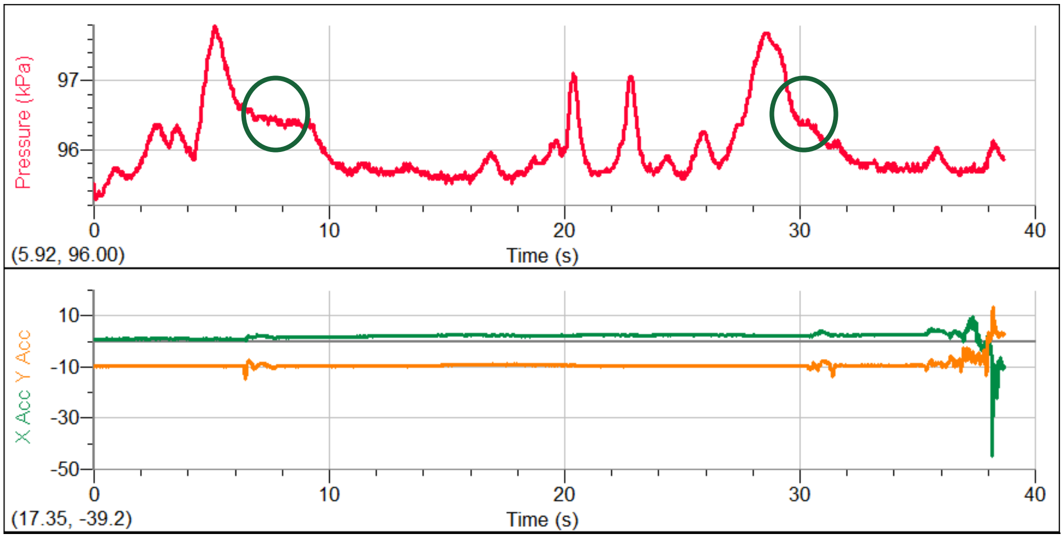
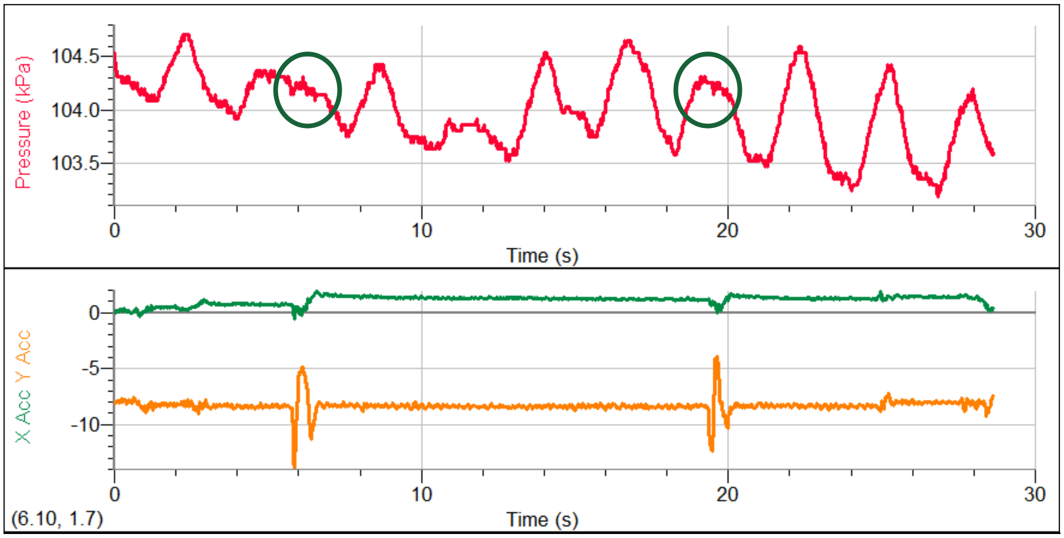
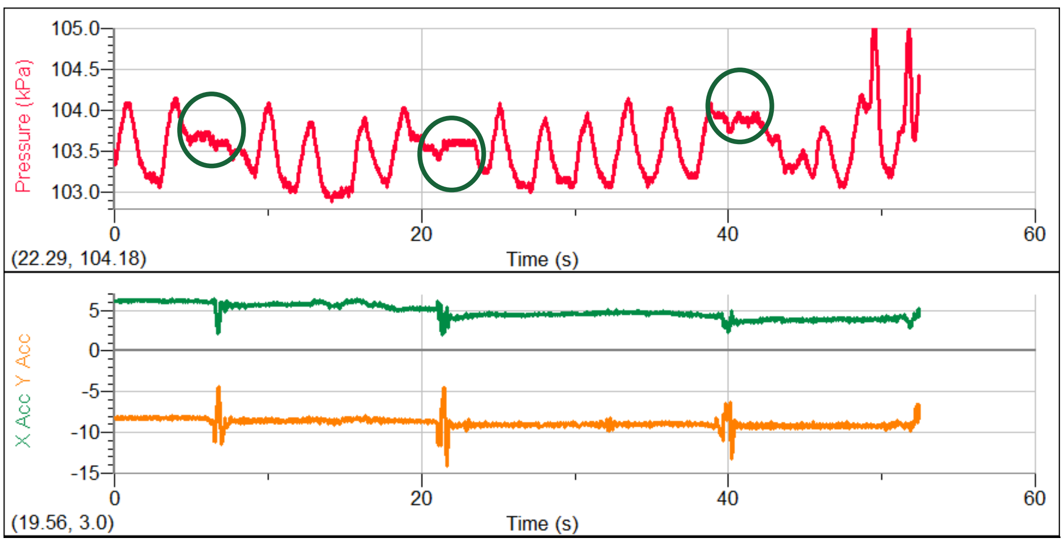
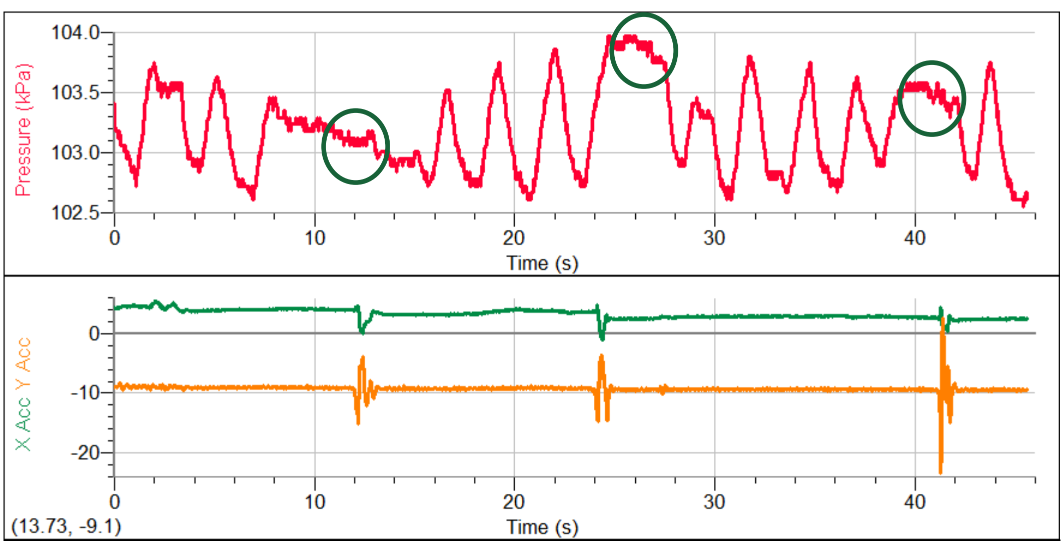
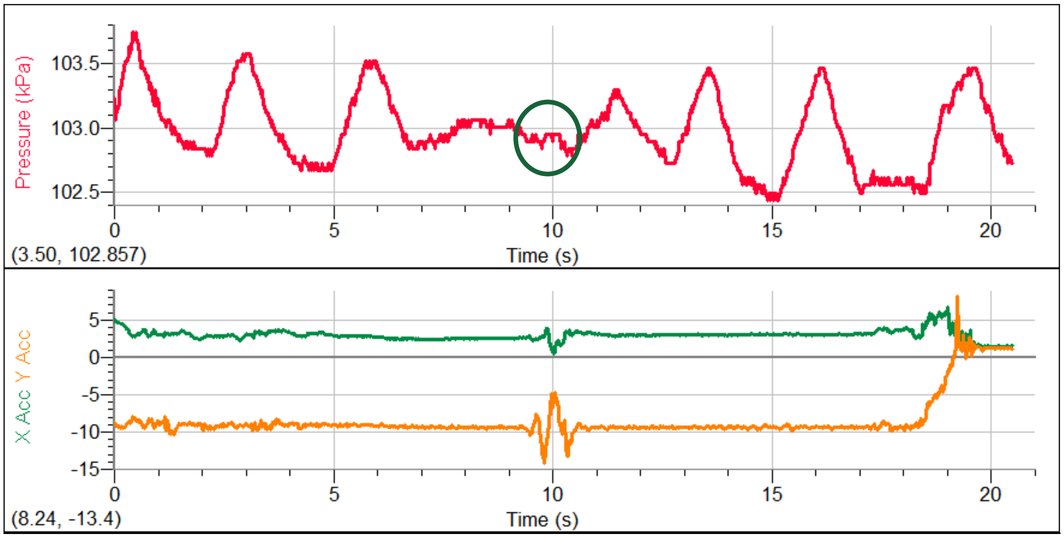


PARTICIPANT 7: BASELINE





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103.0
102.5
0
5
10
15
20
(3.50, 102.857)
Time (s)
X Acc Y Acc
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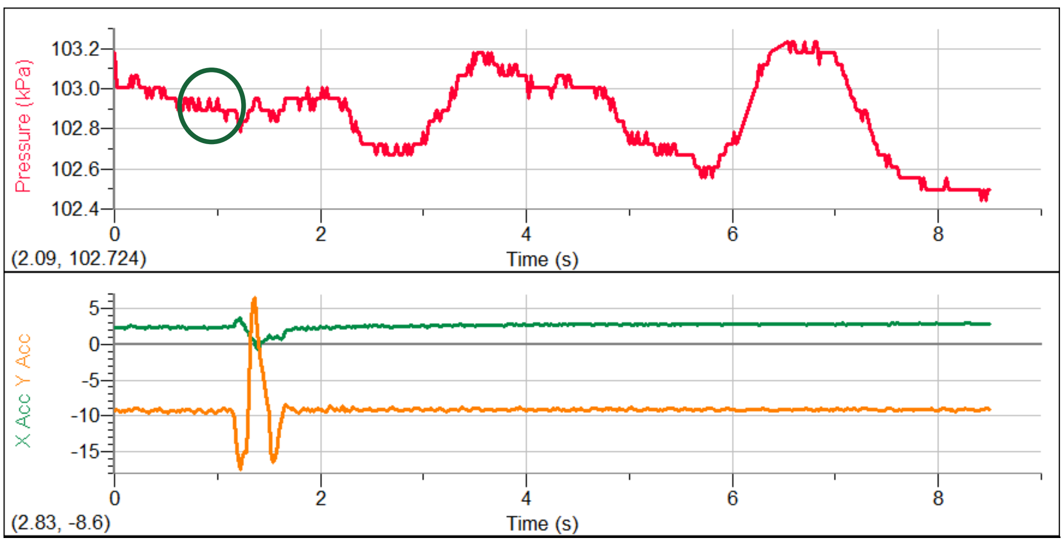
0
-5
-10
-15
0
5
10
15
20
(8.24, -13.4)
Time (s)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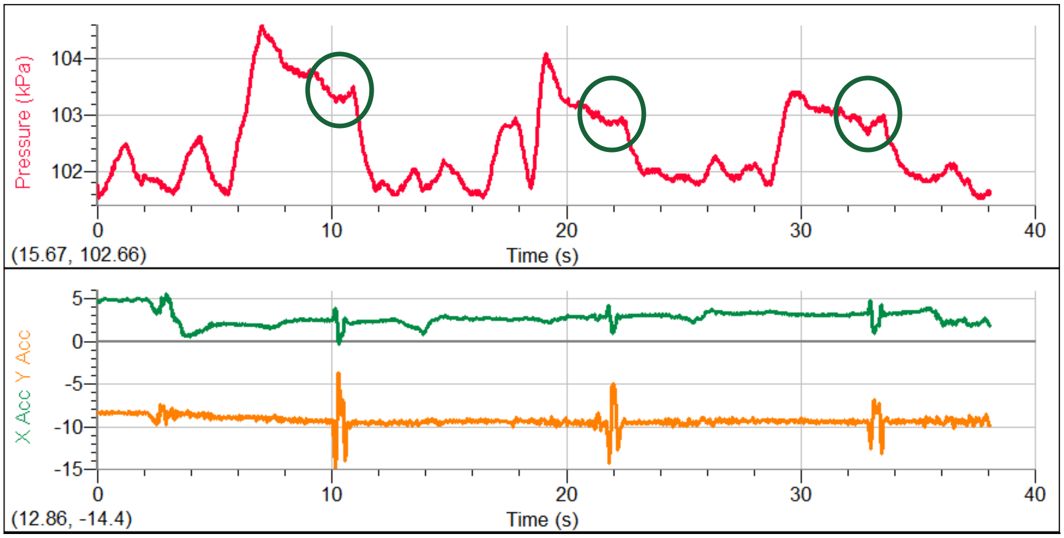
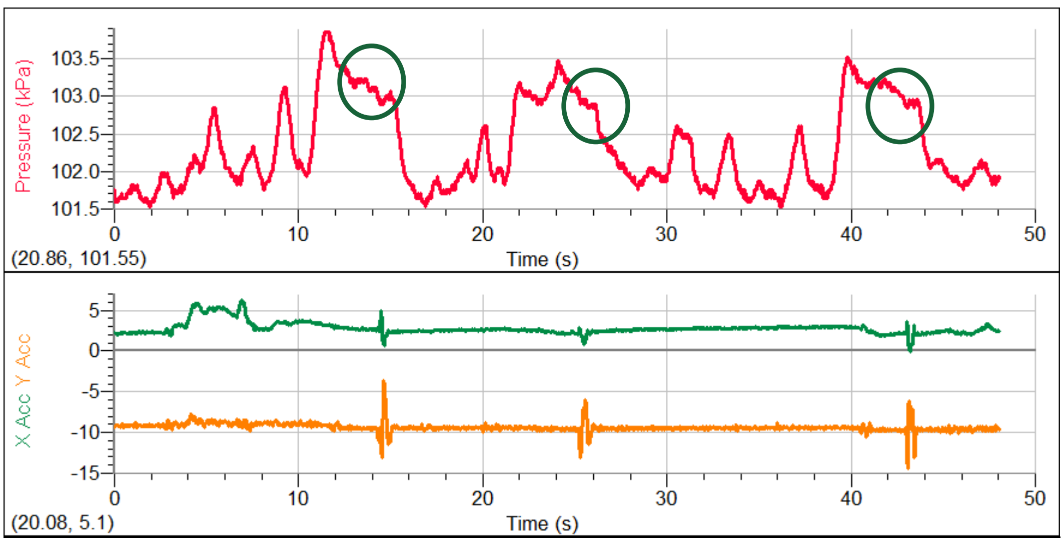
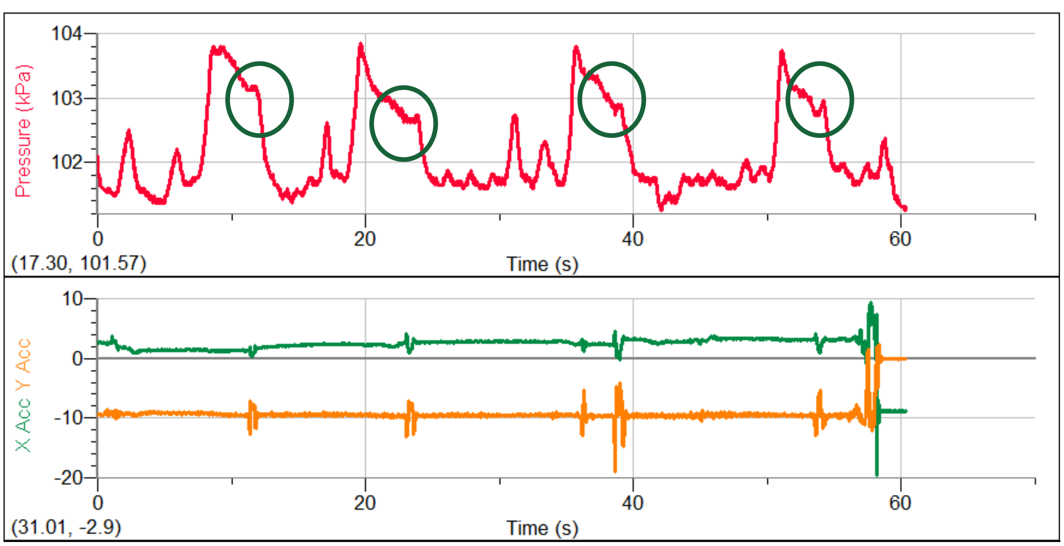
103.0
102.8
102.6
102.4
0
2
4
6
8
(2.09, 102.724)
Time (s)
X Acc Y Acc
5
0
-5
-10
-15
0
2
4
6
8
(2.83, -8.6)
Time (s)

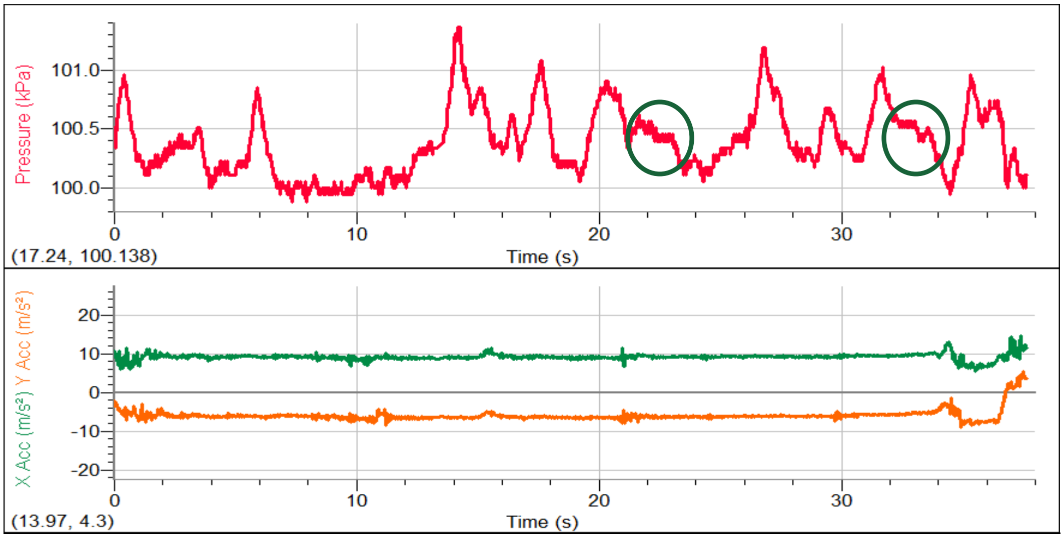
PARTICIPANT 7: POST-TREATMENT

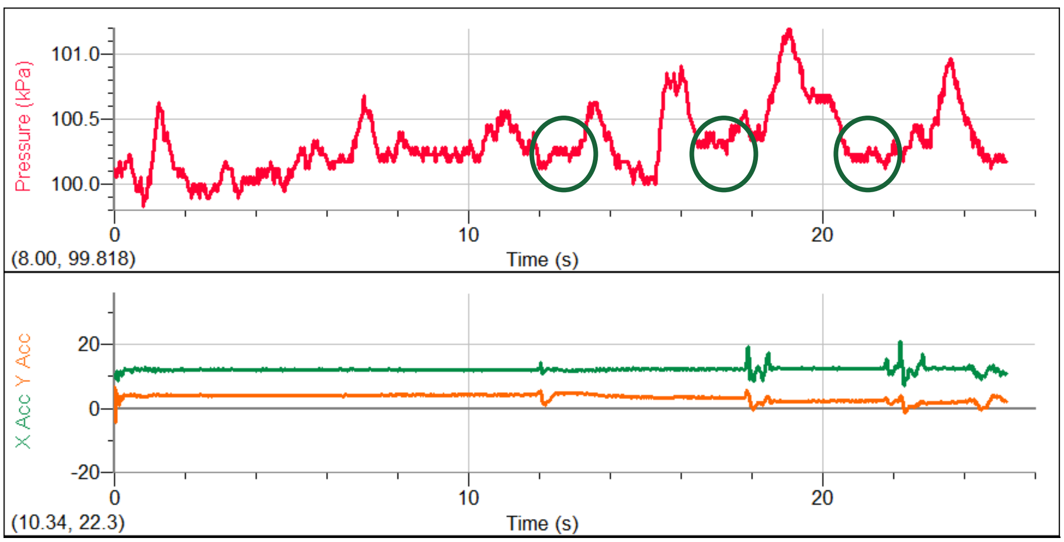


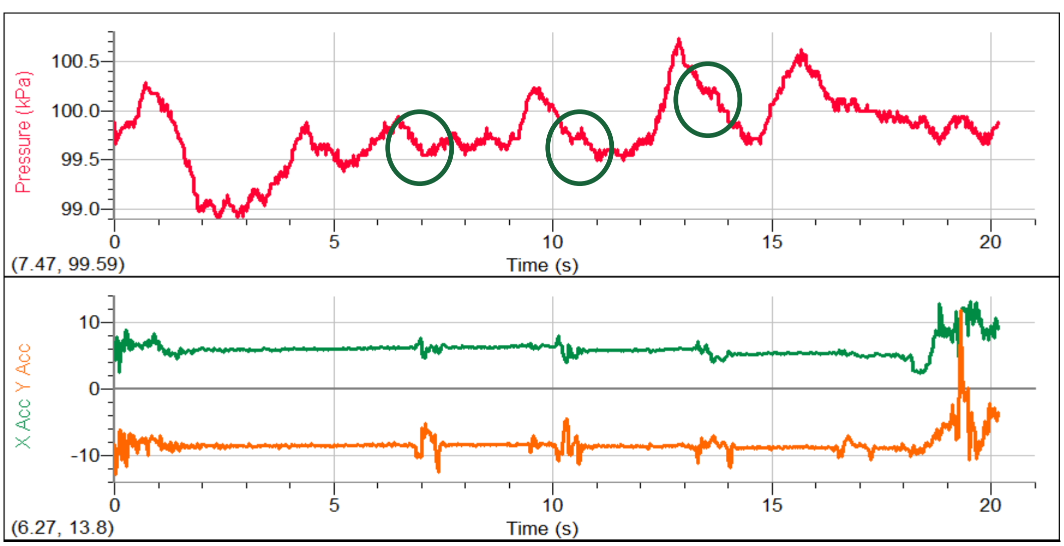




PARTICIPANT 8: BASELINE







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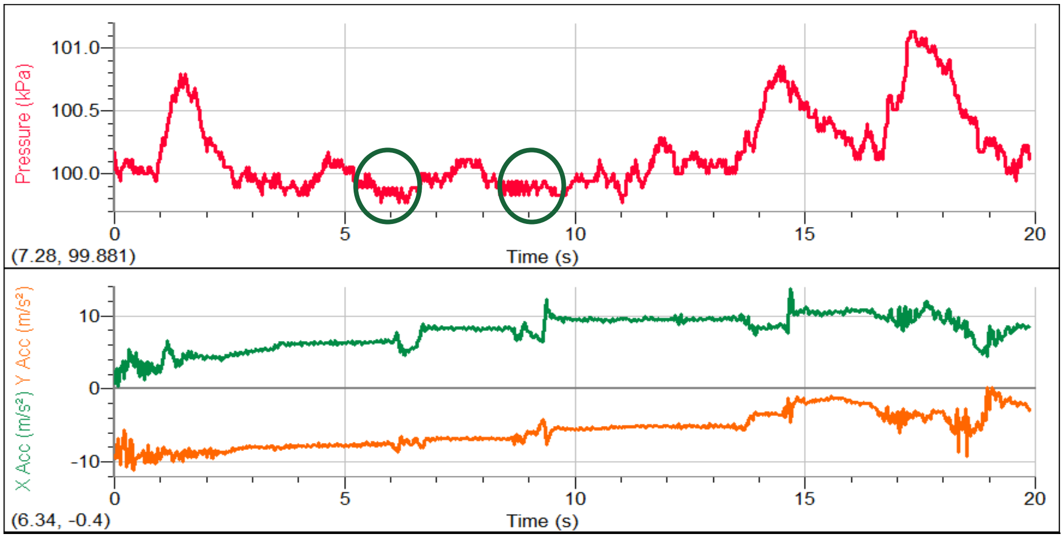
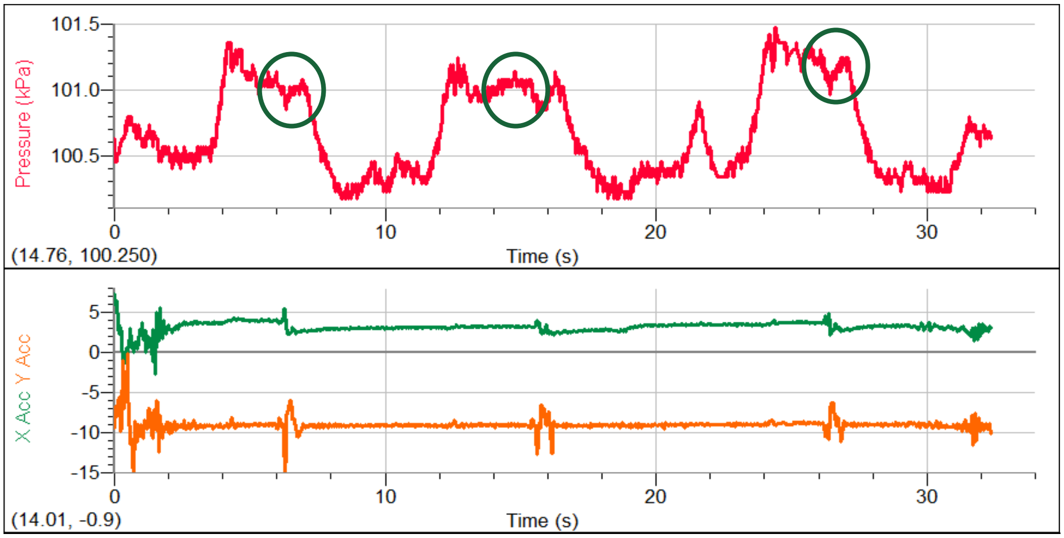
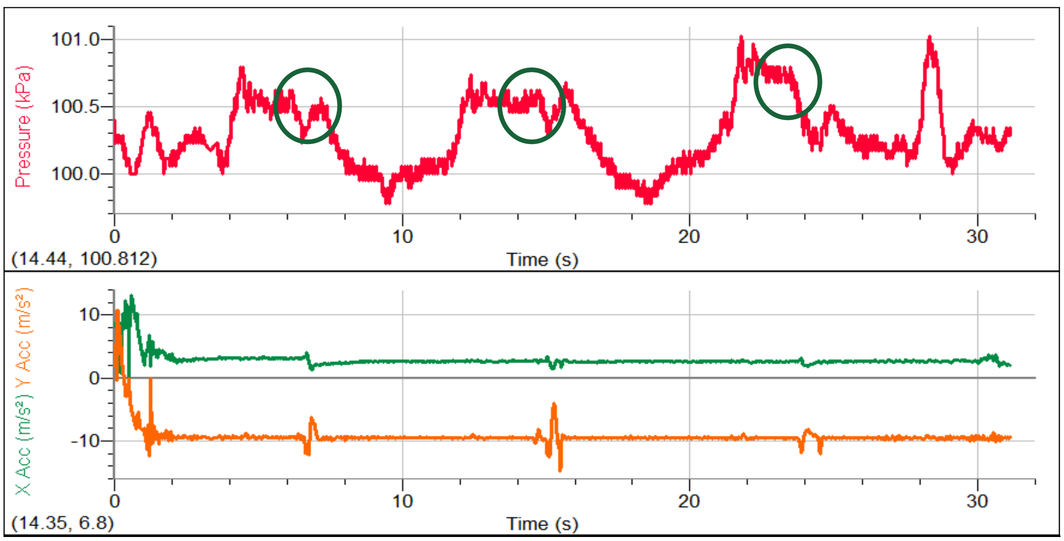
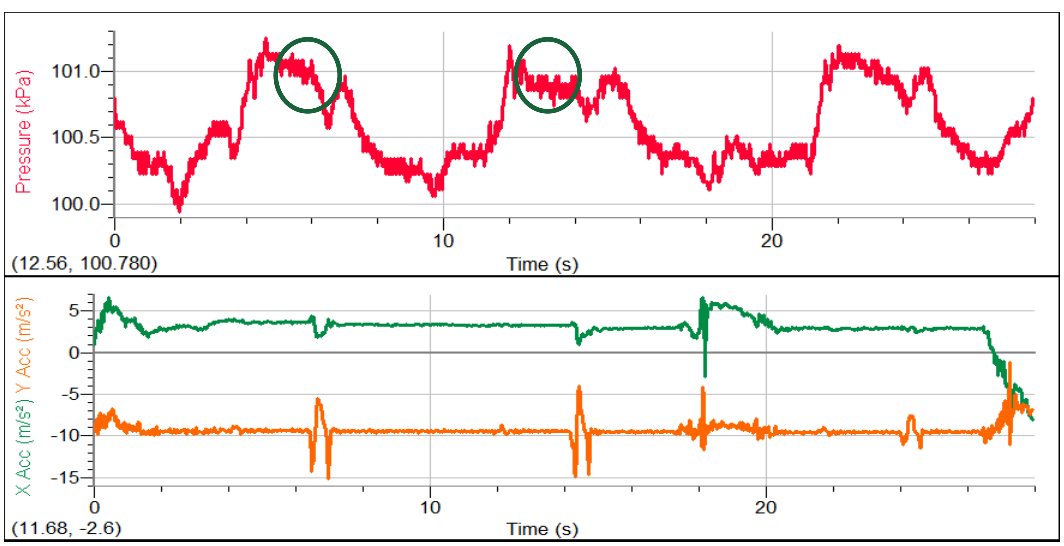


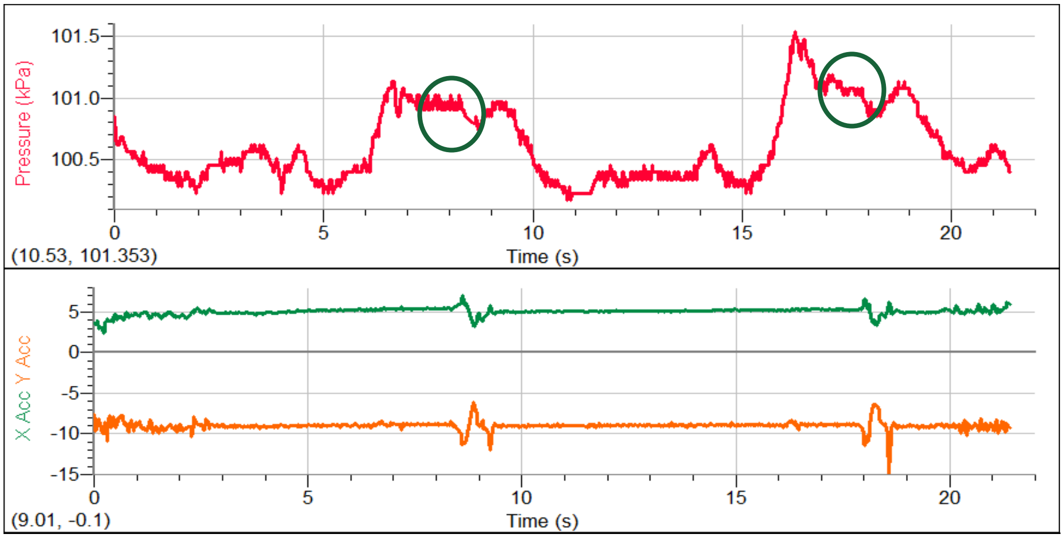
PARTICIPANT 8: POST-TREATMENT







… CONTINUED PARTICIPANT 8: POST-TREATMENT



APPENDIX G: INTER-RATER RELIABILITY PROCEDURAL PROTOCOL

INTER-RATER RELIABILITY PROCEDURAL PROTOCOL

1. **Random selection.** A total of 24 swallows from the baseline condition and 24 swallows from the post-treatment condition were randomly selected to be analyzed for the inter-rater reliability estimate. As there were 80 swallows collected in the baseline condition and 80 swallows collected in the post-treatment condition (i.e., eight participants completed ten swallows each), each swallow in both conditions was assigned a number from 1 to 80. A random number generator program was used to select 24 numbers between 1 and 80 from the baseline condition and 24 numbers from the post-treatment condition. The 48 random numbers were matched to the corresponding swallow and the Vernier file containing the selected swallow was copied into a new folder. When all the files had been copied into a separate folder, the files were sequentially arranged in ascending order according to the number (between 1 and 80) that was assigned for the randomization process. The files were then renamed from 1 to 48.

2. **Training.** A second person (who was not involved in data collection) was asked to analyze 48 individual swallows and judged whether the apneic period was followed by expiration (xE) or inspiration (xI). Respiratory phase data from a separate study were used for training purposes. An example of respiratory phase data was shown to the inter-rater judge and the following information was provided:
    - The respiratory data are displayed in the top graph. Time (in seconds) is located along the x-axis and pressure (kPa) is on measured on the y-axis. Expiratory and inspiratory respiratory movements are represented by the downward and upward direction of the signal, respectively. A flat area in the respiratory signal represents the swallow apnea.
    - Data collected from the accelerometer are displayed in the bottom graph. The peaks in the accelerometry signal indicate hyolaryngeal movement during swallowing. The green line represents anterior movement of the hyolaryngeal complex and the orange line represents superior movement. An increase in amplitude in the signal from the accelerometer indicates the onset of each swallow.

- Respiratory and accelerometry data are collected simultaneously and the two signals can be time locked along the x-axis to aid in analysis.

Following an explanation of the respiratory phase data, the inter-rater judge was given 10 sample graphs to analyze. The respiratory phase graphs were obtained from a previous research study. The first two graphs were analyzed with support from the researcher. The following instructions were given:

- Locate the flat area in the respiratory signal (swallow apnea) that occurs shortly before the onset of hyolaryngeal elevation (as evidenced by an increase in amplitude in the accelerometry signal). Note the subsequent upward or downward direction of the respiratory signal. If the respiratory signal moves upwards (i.e., an increase in pressure), write “xI” to indicate post-apneic inspiration. If the respiratory signal moves downwards (i.e., a decrease in pressure), write “xE” to indicate post-apneic expiration.

The inter-rater judge analyzed the remaining eight sample graphs independently. All eight graphs were analyzed appropriately. Following training, the inter-rater judge was given access to the Vernier files which contained the 48 swallows selected for the inter-rater reliability estimate. The inter-rater judge was instructed to record either xE or xI for each swallow.