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**Surface electromyography biofeedback to improve
swallowing function in persons with dysphagia and**

Parkinson Disease:

An intervention study

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A thesis submitted to the University of Dublin in partial fulfilment of
the requirements for the degree of

Doctor of Philosophy

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Supervisor

Dr. Margaret Walshe

Declaration

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Irene Battel

June, 28th 2020

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Summary

This thesis is concerned with a therapeutic approach aimed at improving swallow function in people with Idiopathic Parkinson's disease (IPD) and swallowing disorders. Despite the high prevalence of dysphagia and the severe clinical sequelae of dysphagia in IPD, there is not a recognised intervention to improve swallowing in individuals with IPD. The neural impairments in IPD manifest as reduced ability to plan motor acts based on internal cues. Providing external cues using sEMG can bypass the impaired neural mechanisms and improve swallowing function. This has already shown some effects in limb rehabilitation, suggesting that people with IPD benefit from feedback more than other groups of patients because it is hypothesised that cues help integrate different movement components. Little is known about the specific effects on biofeedback for swallowing treatment in IPD population.

Given the complexity of the swallowing intervention using biofeedback approach in people with IPD and dysphagia, this research study was informed by guidelines for developing complex intervention published by the UK Medical Research Council. This research comprises three key components: a systematic review of the literature, the development of a feasibility study and the cross cultural translation of a swallowing scale and finally the creation of an intervention protocol for a future study.

Firstly, a systematic review was completed to verify the evidence of the use of biofeedback as an augmentative tool for the improvement of swallowing function in people with IPD. The review found that biofeedback had positive effects on increasing swallowing function in people with IPD and dysphagia, although the quality of the evidence was graded as low. The included studies were heterogeneous in terms of type and frequency of biofeedback, study design and outcome measures. This made it difficult to draw firm conclusions on specific biofeedback interventions and it was difficult to formulate a robust theory on the exact mechanisms of biofeedback. Nevertheless, the narrative synthesis of the findings suggest that visual biofeedback as part of a swallow intervention programme for people with IPD and dysphagia was likely to benefits

swallowing function in particular increasing quality of life of people with IPD and dysphagia. Moreover, surface electromyography (sEMG) was the most common method to deliver swallowing biofeedback in people with IPD and dysphagia.

These findings combined with principles of neuroplasticity and motor learning led to a conceptual framework for a feasibility study on the implementation of sEMG biofeedback swallowing treatment in people with IPD and dysphagia.

Secondly, the context of this study is Venice (Italy) where the PhD student is a clinician working with people with IPD and dysphagia. This led to some challenges in using Italian validated scales for swallowing impairments. The second study therefore was a cross-cultural translation of the Functional Oral Intake Scale (FOIS) into Italian. This translated scale “the FOIS-IT” was used in the third study, the feasibility study.

The feasibility study was conducted at the Neurological Department of the Venice hospital (Italy). Twelve participants were recruited; two withdrew from the study at the beginning of the research, the remaining 10 participants completed the study. The intervention involved biofeedback with sEMG. Participants received this intervention for 1 hour per day, 5 days per week, for 4 weeks (20 hours). The intervention programme incorporated a progression of swallowing tasks using the sEMG biofeedback and the treatment approach was based on motor learning and neuroplasticity principles. The study included instrumental and clinical assessments which were carried out at four different times (2 time points pre-treatment and 2 post-treatment assessments).

The study also incorporated a longer-term post-treatment assessment in the study design in order to verify retention effects of the treatment. The study also included qualitative feedback from the participants to explore unexpected effects of this feasibility study. The participant point of view was fundamental for increasing understanding on the acceptability and adherence of the treatment for further studies.

Overall, the swallowing intervention programme, found positive results in people with dysphagia and IPD. There were statistically significant positive changes at the FOIS-It ($p < 0.05$) and in saliva and solid food pharyngeal residue ($p < 0.05$) assessed during

instrumental examination (FEES). The quality of life showed an improvement after treatment specifically the sub-part of food selection ($p < 0.05$).

These changes in oral intake and pharyngeal clearance for saliva and solid food were maintained three months post intervention suggesting an important effect of retention in IPD people. The intervention was well tolerated by the participants who reported additional benefits not only in swallowing-related functions such as saliva control and decreased duration of meal times but also in non-swallowing functions such as voice and cognitive attention skills. These results of the quality analysis were selected to be included in the protocol of future study.

The final chapter of the thesis presents a protocol for a further pilot study that is informed by the findings of this feasibility study. Directions for the next research phase are provided.

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List of abbreviation

α S	α -synuclein
BCR	Bolus Clearance Ratio
BiSSkiT	Biofeedback in Swallowing Skill Training
CBD	Cortico-Basal Degeneration
DBS	Deep Brain Stimulation
DOSS	Dysphagia Outcome and Severity Scale
EAT-10	Eating Assessment Tool
EMST	Expiratory Muscle Strength Training
ENT	Ear, nose, throat specialist
FEES	Fibreoptic endoscopic evaluation of swallowing
FOG	Freezing Of Gait
FOIS	Functional Oral Intake Scale
FOIS-It	Italian version of Functional Oral Intake Scale
H&Y	Hoehn & Yahr scale
IDDS	International Dysphagia Diet Standardisation Initiative
IPC	Inferior Parietal Cortex
IPD	Idiopathic Parkinson's disease
I-SWAL-QOL	Italian version of Swallowing Quality of Life Questionnaire
LBs	Lewy bodies
M1	primary motor cortex
MRC	Medical Research Council
MDS-UPDRS	Unified Parkinson's Disease Rating Scale
MMSE	Mini Mental State Examination
MoCA	Montreal Cognitive Assessment

MSA	Multiple System Atrophy
MUST	Malnutrition Universal Screening Tool
NRCTs	Non Randomised Control Studies
PAS	Penetration, Aspiration Scale
PCF	Peak of Cough Flow
POE	Pleasure of Eating
pre-SMA	pre-Supplementary Motor Area
PRISMA	Preferred reporting items for systematic reviews and meta-analyses
PSP	Progressive Supranuclear Palsy
QOL	Quality of Life
RCTs	Randomised Control Trial Studies
ROBINS-I	Risk Of Bias In Non-Randomized Studies - of Interventions
ROMP	Radboud Oral Motor Inventory for Parkinson's Disease
SD	Standard Deviation
SN	Substantia nigra
sP	Substance P
SPL	Superior Parietal Lobe
SENIAM	Surface Electromyography for the Non-invasive Assessment of Muscles
sEMG	Surface Electromyography
SWAL-QOL	Swallowing Quality of Life Questionnaire
TA	Thematic Analysis
TOMASS	Test of Mastication and Swallowing Solids
TWST	Timed Water Swallow Test
UES	Upper-Esophageal Sphincter
VaP	Vascular Parkinsonism
VFS	Videofluoroscopic swallowing study

WHO World Health Organisation
YPR-SRS Yale Pharyngeal Residue Severity Rating Scale

Publications to Date from this Thesis

Publications to Date from this Thesis

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2. Battel I, Calvo I, Walshe M, Biofeedback to improve safety and efficiency of swallow function in people with Parkinson's disease and dysphagia. European Society of Swallowing Disorders Congress, Dublin, Ireland, 25-29th2018. *Dysphagia* 2019; 34, (5):720 – 819

3. Battel I, Calvo I, Walshe M. Interventions involving Biofeedback to improve swallowing in people with Parkinson's Disease and dysphagia: A systematic review. *Archives of Physical Medicine and Rehabilitation*. 2020 Aug 27:S0003-9993(20)30526-8. doi: 10.1016/j.apmr.2020.06.033

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CONFERENCE PRESENTATION

Poster Conference Presentations

- 10th Congress of European Society for Swallowing Disorders (Online Congress, October 2020) “Biofeedback to improve swallowing function in persons with dysphagia and Parkinson Disease: An intervention study.”

- 8th Congress of European Society for Swallowing Disorders (Dublin, Sept 2018)
“Biofeedback to improve safety and efficiency of swallow function in people with Parkinson's disease and dysphagia”

- 7th Congress of European Society for Swallowing Disorders (Barcelona, Sept 2017) *“Cross-cultural validation of the Italian version of the functional oral intake scale*

Oral Conference Presentations

Italian Neurorehabilitation Society (Perugia, April 2019) *“Biofeedback to improve safety and efficiency of swallow function in people with Parkinson's disease and dysphagia”*

Chapter 1: Introduction

1.1 Overview of the Study

The current study explores the use of biofeedback for swallowing recovery in people with swallowing disorders (dysphagia) and idiopathic parkinson's disease (IPD). The main aim is to complete a feasibility study to examine the effectiveness of a specific intervention programme involving biofeedback and sEMG in people with IPD and dysphagia and to inform a larger clinical randomized controlled trial (RCT) study.

In order to explore biofeedback for swallowing recovery in IPD persons, the researcher adopted the guidelines of developing and evaluating complex interventions published by the Medical Research Council (MRC) (Craig et al., 2019). Firstly, the evidence the use of biofeedback for swallowing in IPD is identified through a systematic review. This facilitated the creation of a theory on how biofeedback for swallowing works in IPD population. Thereafter, specific research questions were formulated, and a feasibility study was conducted to address these questions. The results of this feasibility study builds the foundation for the development of a study protocol to verify the efficacy of this intervention in a larger RCT.

1.2 Key Concepts and Definitions

As an introduction to this thesis, this section describes the pathophysiology of IPD and swallowing disorders. The following sections form the basis of the study.

1.2.1 Pathophysiology of idiopathic parkinson's disease

IPD is classified under the group of movement disorder, which includes several neurological diseases resulting from the damage of the basal ganglia and its connections, recognised also as extrapyramidal diseases (Hawkes et al., 2009; Hunter et al., 1997). IPD is one of the most common progressive neurodegenerative disorders caused by a deficit of the basal ganglia and dopamine's circuitry. The cause of Parkinson's disease is

unknown and for this reason it is defined also as "idiopathic". Nevertheless, it seems to involve genetic components and environmental factors (Erro et al., 2018).

The diagnosis of IPD is based on medical history and a movement disorder (Postuma & Berg, 2017). The main motor signs are bradykinesia (slowness in initiating voluntary movements) muscular rigidity, tremor at rest, postural instability and gait impairments such as freezing of gait and festination (Hawkes et al., 2009) (Table 1.1). The disease involves also deficits in sensory system causing loss of proprioception, kinaesthesia, mechanosensation and olfaction (Cilia et al., 2015; Leow et al., 2012; Shah et al., 2009). The deterioration of basal ganglia structure and its connections worsen over the time affecting speech, swallowing and cognitive functions (Cilia et al. 2015; Dancis et al. 2015). The most typical speech disorder is hypokinetic dysarthria characterized by insufficient loudness, hypo-articulation and reduced prosodic features (Findley, 2007; Tjaden, 2008). Dysphagia is common, with about 80% of individuals with IPD developing swallowing impairment during the course of their disease and it is a clinically relevant symptom in IPD patients (Kalf, de Swart, et al., 2012; Pflug et al., 2018). In his first description of the disease, James Parkinson already recognized dysphagia as an essential symptom of IPD (Parkinson, 1817). This symptom is described further in the subsequent thesis chapters.

Table 1. 1 Major motor and non-motor symptoms in IPD

Motor symptom	Non-motor symptoms
Resting tremor	Impaired Olfaction
Rigidity	Dementia
Bradykinesia	Depression
Postural Instability	Fatigue
Festinating gait	Apathy
Micrographia	Sleep Disorders
Masked facies	Psychosis
Dysarthria	Vestibular Deficits
Dysphagia	Autonomic Dysfunction

1.2.2 Epidemiology of IPD

In industrialized countries, it is estimated that 0.3% of the general population is affected by IPD (Ball et al., 2019). Prevalence increases with age, being as high as 1.0% in the > 60 years age group, and 3.0% in those over 80 years (Balestrino & Schapira, 2020; Ball et al., 2019). IPD is approximately 1.4 times more frequent in men than women (Ascherio & Schwarzschild, 2016). A recent study published in the journal *Lancet* compared the epidemiological global analysis of IPD in 1990 and in 2016 (Dorsey et al., 2018). In 1990, 2.5 million of people had IPD and this number has almost tripled in 2016, with 6.1 million individuals worldwide. The authors suggest that this dramatic rise might be due to the increase in life expectancy but also by improvements in the accuracy of neurological diagnosis of IPD. In addition, a recent epidemiologic study by Wanneveich et al. (2018) confirmed that not only the population of IPD is increasing but also the disease duration with a rise of 3 years duration expected between the 2010 and 2030 expected. This is a consequence of population ageing and life expectancy improvement (Wanneveich et al., 2018). These factors have a direct effect on the increase in disability and demand for care. In Europe, estimated prevalence and incidence rates for IPD ranged between 65 and 257 per 100,000 and between 5 and 346 per 100,000 person-years respectively (Balestrino & Schapira, 2020; von Campenhausen et al., 2005). Twenty years ago, the incidence of IPD in the Italian population, where this research study is based, for people ≥ 65 years of age was 346/100,000 person years in 2000 (Baldereschi et al., 2000). A recent study on IPD epidemiology in the north-eastern regions of Italy, estimated that the regional incidence rate was 0.28 new cases/1000 person-years and the prevalence was 3.89/1000 inhabitants (Valent et al., 2018). In addition, the mortality associated with IPD has been rising in the Veneto Region, where this study is based, especially among males and mortality increases exponentially with age (Fedeli & Schievano, 2017). In the death certificates, mention of IPD was associated with dementia/Alzheimer disease and acute infectious diseases such as pneumonia/aspiration pneumonia, and sepsis (Fedeli & Schievano, 2017).

In this scenario, swallowing rehabilitation plays a key role on reducing the burden of disability, decreasing the risk of aspiration pneumonia and thus hospital admissions.

1.2.3 Prevalence of dysphagia in IPD

Worldwide, the prevalence of dysphagia in persons affected by IPD is the subject of ongoing debate. In the literature, it is widely accepted that major signs of dysphagia occur in the advanced phase of the disease (Fabbri et al., 2019; Müller et al., 2001). Coelho et al. found that severe dysphagia was reported in 68% patients in the late stage of the disease, about 10–11 years after the motor symptoms were reported (Coelho et al., 2010). However, mild oral-pharyngeal symptoms of dysphagia were also frequent in the early stages of IPD, and dysphagia might be the first sign of the disease.

Swallowing disorders are likely to be underestimated in IPD and instrumental assessments are required for accurate dysphagia diagnosis. In the IPD population, dysphagia can remain subclinical as individuals gradually adjust to the impairment with disease progression, hiding the initial signs of dysphagia (Miller, 2017; Suttrup & Warnecke, 2016). A meta-analysis conducted by Kalf and colleagues (2012) suggested that the prevalence of dysphagia in IPD is higher when documented by instrumental assessment (e.g. videofluoroscopy (VFS) and/or fibreoptic endoscopic evaluation of swallowing (FEES) (82%) compared to non-instrumental assessments (35%). Self-report of dysphagia tends to be poor in identifying dysphagia due to lack of awareness of swallowing difficulties associated with cognitive impairments. A recent study by Buhmann et al. confirmed these assumptions. In this study, dysphagia occurred in more than 95% (103/119) of people with IPD assessed using FEES and aspiration was seen in 25% (30/119) of them. However, while only 12%-27% of them reported swallowing problems in the specific patient reported questionnaires provided (Buhmann, Flugel, et al., 2019). For this reason, swallowing assessments in IPD people must incorporate instrumental evaluations.

1.3 Dysphagia in IPD

The occurrence of clinical signs of dysphagia differs enormously in the IPD population. The neuropathological mechanism of swallowing disorders in IPD is not already identified. In the literature it is accepted that swallowing disorders in IPD are characterised by a combination of impairments of both subcortical and cortical mechanisms and this could explain the unpredictability of dysphagic symptoms and the broad variety of swallowing impairments in IPD (Suttrup & Warnecke, 2016).

Swallowing difficulties have been reported for all phases of swallowing: preparatory, oral, pharyngeal and oesophageal phases of swallowing. In general, dysphagic symptoms are similar to those which affect the limb muscles including bradykinesia (slowness of movement), muscle rigidity, and prolonged initiation and reaction time (Miller, 2017). In the following paragraphs, the dysphagia symptoms are described according to the phases of swallowing.

1.3.1 Disorders of the oral preparatory phase

This is an initial phase of swallowing, in which the liquid or solid bolus is held in the anterior part of the mouth and saliva contributes to bolus formation. The oral cavity is sealed anteriorly by the lips and posteriorly by soft palate and the base of tongue, while the anterior part of the tongue manipulates the bolus against the hard palate and the upper dental arch (Shaw & Martino, 2013).

The oral preparatory phase is particularly compromised in IPD, including motor and sensory deficits (Table 1.2). Firstly, hypotonia of the facial muscles and especially of the buccinator result in a reduced lip seal (Miller, 2017; Simons, 2017). Inefficient lip seal is the major cause of leakage not only of saliva but also of liquid or solid bolus from the mouth. This is frequently associated with a proprioceptive deficit and so individuals tend not to be aware of food residue and or saliva in the lips (Miller et al., 2006). The oral preparatory phase in IPD is characterized also by tongue tremor, tongue pumping and

lingual festination movements, which are common impairments also in the limbs (Troche et al., 2008). Troche et al. (2008) analysed the swallowing of liquid and a thicker bolus using VFS. They found that the uncoordinated motion of tongue contributed significantly to increased oral transit time and decreased anterior-to-posterior bolus movement. Due to movement difficulties and incoordination of the tongue, persons with IPD can present with more difficulties in swallowing of solid rather than softer foods. In IPD, mastication is also compromised not only by slowed and limited mandibular excursion but also by hypotonia and festination of masseter and temporalis muscles, which cause deficit in the rotary moments for chewing (Wakasugi et al., 2017). Recently, Pflug et al. found that 50% of IPD persons in their study had deficits with bolus preparation while eating bread and biscuits revealing the importance of assessing solid food swallowing in this population (Buhmann, Flugel, et al., 2019; Pflug et al., 2018).

Table 1. 2 Summary of the oral preparatory and oral phase impairments in IPD

Oral preparatory phase and oral phase	
Motor- Sensory Disorders	Consequences for swallowing
Inefficient lip seal	Anterior loss of saliva, food and fluid; Difficulties with bolus formation
Loss of smell	Decreased appetite and taste associated with smell
Reduced oral sensation (hyposensitivity)	Biting tongue / cheeks Decrease saliva swallowing and drooling Reduced awareness of food residue in the mouth
Decreased buccal tone	Build-up of food in lateral sulci. Biting the inside of cheeks
Reduced jaw movement	Difficulty chewing. Impaired mastication and formation of bolus;
Abnormal tongue movements (Tremor, pumping and festination)	Impaired formation and containment of bolus; Reduced bolus pressure; Reduced clearing of oral residue; Difficulties initiating pharyngeal swallow Deficit in bolus transport;
Low tone in muscles of the tongue	Tongue lacks ability to flatten and cup around a food or liquid bolus leading to difficulty with organisation and control of bolus

1.3.2 Disorders of the oral phase

The oral phase begins with the posterior propulsion of the bolus by the tongue and ends with initiation of the pharyngeal swallow. Several studies of IPD confirm that lingual movements during the propulsion phase and palatal elevation are characterised by hypokinesia, bradykinesia and festination associated to rigidity (Leopold & Kagel, 1996; Miller, 2017) (Table 1.2). The hypokinesia and bradykinesia caused the reduced excursion of tongue movements and weak tongue pressure. As a result, the retropulsion of bolus is severely compromised and prolonged, requiring several attempts to transport the bolus posteriorly (Fukuoka et al., 2019). Not only hypotonia but also the 'rocking-rolling' festination movements of the tongue impede the propulsion of the bolus (Rosenbek & Jones, 2008). Furthermore, the abnormal tongue movements and the hypotonia of the oral pharyngeal muscles cause difficulties of the oral bolus control resulting in pre-swallow bolus loss, (Nienstedt et al., 2019; Warnecke et al., 2016). After solid, liquid bolus (IDDSI Level 0) represents, the most difficult consistency. The difficulties in oral control increases the risk of premature spillage and penetration and aspiration in the IPD population (Argolo & Nóbrega, 2013; Gaeckle et al., 2019).

1.3.3 Disorders of the pharyngeal phase

During the pharyngeal phase, the soft palate and pharyngeal wall seal off the nasopharynx. The vocal cords and arytenoids close off the laryngeal opening and the epiglottis covers the laryngeal vestibule. In the meantime, the hyoid bone and larynx move superiorly and anteriorly, the pharynx also widens and shortens and the upper oesophageal sphincter (UES) relaxes and opens in order to allow the bolus flow in the oesophagus (Kendall, 2002). IPD patients often present with difficulties from the initiation of the pharyngeal swallow, due to the motor and sensory deficits (Miller, 2017) (Table 1.3). Many studies using instrumental assessments confirmed the presence of increased residue in the valleculae rather than pyriform sinuses (Suttrup & Warnecke, 2016). Some report the presence not only of a food bolus but also of medication in the valleculae

(Buhmann, Bihler, et al., 2019; Miller et al., 2009; Warnecke et al., 2016). This is likely due to reduced tongue base movement, reduced pharyngeal sensation and pharyngeal wall approximation (Curtis et al., 2020a; Leopold & Kagel, 1996; Martinez-Ramirez et al., 2015). The anterior superior elevation movement of hyolaryngeal structure is particularly decreased in IPD people. In some cases, it is due by the hypotonia of the submental muscles, in other cases it is caused by rigidity, which affects IPD persons mostly during off phases (Miller et al., 2009; Warnecke et al., 2014). Interventions for improving on hyolaryngeal excursion during swallowing and non-swallowing task showed positive effects on swallowing in IPD (Athukorala et al., 2014; Troche et al., 2010). Recently, Gaeckle et al. found that the delay in reduced hyolaryngeal excursion and in the pharyngeal swallow were predictors of penetration-aspiration in IPD (Gaeckle et al., 2019). Nevertheless, the mechanism behind aspiration is more complex and it assumes also pharyngeal sensory and cough reflex impairments. In recent years, some researchers confirm the correlation of mechanism between the reduced protective cough mechanism and the aspiration in IPD (Pitts et al., 2010; Troche et al., 2010). There is an emerging evidence that reduced voluntary cough expiratory airflow and reflex cough parameters are significantly correlated with penetration and aspiration of bolus material during swallowing (Hegland et al., 2014; Troche, Brandimore, Okun, et al., 2014; Wheeler Hegland et al., 2014).

Table 1. 3 Summary of the pharyngeal phase impairments in IPD.

Pharyngeal phase	
Motor – Sensory Disorders	Consequences for swallowing
Weak tongue base	Poor bolus pressure, residue in valleculae
Pharyngeal delay in initiation of the pharyngeal swallow	Aspiration / laryngeal penetration before swallow
Reduced velopharyngeal closure	Nasal regurgitation
Weak Intra-bolus pressure and peristalsis in pharynx	Altered transit time, requiring repeated swallows, increase risk of penetration/aspiration of bolus, pooling of residue in the pharynx
Reduce laryngeal sensation	Reduced/absent cough reflex and risk of silent aspiration
Poor cough reflex	Poor airway protection
Reduced hyo-laryngeal excursion	Incomplete opening of UES
Reduced laryngeal closure	Increase the risk of penetration and aspiration
UES impairments in relaxation	Pooling of food in pyriform sinus, aspiration post-swallowing

1.3.4 Disorders of the oesophageal phase

Although it is well recognised that people with IPD present with disordered oesophageal motility, few studies have assessed oesophageal function in this population (Kim & Sung, 2015; Su et al., 2017). Suttrup et al. (2017) found abnormal oesophageal motility using high resolution manometry in 73% of IPD patients and 59% of them presented with a complete aperistalsis or multiple simultaneous contractions (Suttrup et al., 2017). In line with these results, Su et al. (2017) showed the oesophageal impairments using high resolution manometry and the Chicago classification. They found that 55% of the sample presented with ineffective oesophageal peristalsis, followed by ineffective oesophageal peristalsis and diffuse oesophageal spasm. In addition, 37% of people with IPD reported gastroesophageal reflux disease (Su et al., 2017).

Table 1. 4 Summary of the oesophageal phase impairments in IPD

Oesophageal impairments
Esophagogastric junction (EGJ) outflow obstruction
Diffuse esophageal spasm
Ineffective esophageal peristalsis
Gastroesophageal reflux disease: heartburn, regurgitation, chest pain

1.4 Consequences of Dysphagia

The main consequences are medical and psychological but there is also associated increased healthcare costs. These have consequences not just for person with IPD but also for family. This section focuses mainly on consequences of dysphagia on the individual with IPD.

1.4.1 Medical consequences

Medical consequences of dysphagia can be indirect. For example, they may be unable to benefit from the pharmacological treatments as people with IPD can have issues in taking oral medications. Studies have shown the presence of medications in the valleculae (Buhmann, Bihler, et al., 2019) suggesting that this medication can have limited impact on controlling symptoms. The major issues of swallowing disorders concern the aspiration of food and/or fluid into the airway. Although, inhalation of small amounts of oropharyngeal secretions is normal in healthy persons during sleep, aspiration in combination with poor oral hygiene is the major pathogenetic mechanism of most pneumonias (Mandell & Niederman, 2019). In IPD, large-volume aspiration (macro-aspiration) of colonized oral-pharyngeal residue associated with lack of weak cough response is directly associated with aspiration pneumonia (Donovan et al., 2013; Langmore et al., 1998). Aspiration pneumonia is considered one of the leading cause of death in people with IPD, showing to account for 70 % of the mortality (Akbar et al., 2015; Backstrom et al., 2018; Martinez-Ramirez et al., 2015). Weight loss, malnutrition and dehydration are associated with the difficulties of swallowing as well as the consequences of diet modification intervention (Namasivayam-Macdonald et al., 2019; Serra-Prat et al., 2017; Steele et al., 2015). These complications lead to a sequela of further neuro-motor problems and increase the need for hospitalization as well as for repeated hospital admissions (Martinez-Ramirez et al., 2015).

1.4.2 Psychosocial consequences of dysphagia in IPD

Swallowing difficulties have a direct impact on the social life, decrease mealtime enjoyment with a reduction in quality of life (Carneiro et al., 2014; Leow et al., 2010; Plowman-Prine et al., 2009). Many persons with IPD and dysphagia experience anxiety and fear of choking during meals, limiting not only the pleasure of eating and the social life but also impacting on family members (Carneiro et al., 2014). In line with this, some

studies confirmed a strong correlation between dysphagia and depression, although there are many different underlying factors which contribute to it (Miller, 2017; Miller et al., 2009). A further consequence of dysphagia in IPD is an increase in drooling.

1.4.3 Drooling in people with IPD

Drooling is one of the major problems affecting people with IPD. The prevalence varies from 10% to 74% due to the lack of a standard definition for drooling and/or the absence of objective assessments (Kalf, Bloem, et al., 2012; Nienstedt et al., 2018; Ou et al., 2015). Drooling seems to occur more in the advanced phase of the disease, impacting quality of life. Up to 52% people with IPD can report social and emotional negative consequences of drooling (Ou et al., 2015). The pathophysiology of drooling is not completely understood (Miller, 2017). It seems not to be directly associated with dysphagia, because severe drooling could occur also in patients with no clinically relevant dysphagia on FEES examination (Nienstedt et al., 2018; Srivanitchapoom et al., 2014). Srivanitchapoom et al. argue that drooling in IPD is multifactorial symptom including the sensory and motor components (Srivanitchapoom et al., 2014). Anterior drooling is associated with the lack of saliva proprioception, associated by mouth opening, flexed head posture (McNaney et al., 2019; Miller, 2017). Posterior drooling is associated with difficulty in initiating the swallow, retained saliva in the mouth and in hypotonia oral-pharyngeal structures.

Furthermore, recent studies shed light on the important role of cognition in saliva management (Reynolds et al., 2018; Troche, Okun, et al., 2014). Reynolds et al. found that people with IPD swallowed less frequently and had more drooling during cognitively distracting tasks (Reynolds et al., 2018). This suggests that saliva swallowing requires a level of attention in order to coordinate the process of monitoring, collection and clearance of saliva.

1.5 Assessment of Dysphagia in IPD

The methods of assessment of swallowing are grouped based on screening procedures, clinical assessments and instrumental assessments, which are summarized in Table 1.4.

1.5.1 Swallowing screening tests

In the IPD literature, there is a lack of systematic reviews on the diagnostic accuracy and effectiveness of screening tools. In 2012, Kalf et al. investigated the prevalence of swallowing disorders in IPD. Although, the aim of this review was epidemiologic, they found that questionnaires and patient reported outcome do not identify swallowing disorders (Kalf, de Swart, et al., 2012). The main reason is that IPD patients are not aware and underestimate the swallowing impairments. Hence, screening swallowing questionnaires, even if specifically designed for IPD such as Munich Dysphagia Test - Parkinson's Disease (MDT-PD)(Simons et al., 2014), are at high risk of false negative errors in IPD. Hence, self-reported assessments should not be adopted as the sole means of screening in this population. Screening tests such as the Timed Water Swallow (TWS) test have been used in people with IPD (Hughes et al., 1992). Hughes et al. 1992 showed that the TWS was a valid assessment to detect swallowing impairments in IPD people (Hughes et al., 1992). However, Pflug et al. have recently showed that TWS had a relatively high sensitivity (88%) but low specificity (19%) leading to high false-positive rate; in particular it does not identify the presence of silent aspiration (Pflug et al., 2019). In the last decades, several studies focused on assessment of the voluntary and reflex cough in order to detect silent aspiration in people with IPD and dysphagia (Brandimore et al., 2017; Pitts et al., 2010; Troche et al., 2016; Wheeler Hegland et al., 2014). To date, there is not a validated and reliable protocol to screen people with IPD at risk of silent aspirators.

1.5.2 Clinical swallowing examination and Instrumental assessments

Clinical assessments in IPD population incorporate mostly questionnaires.

Although the important value of the clinical assessments are recognised, most clinical guidelines in IPD suggest carrying out instrumental assessments to detect swallowing impairments (Kalf, de Swart, et al., 2012; Kwon & Lee, 2019; Miller, 2017). Both FEES and VFS are recognised reliable instrumental evaluations to assess oro-laryngo-pharyngeal structures and motility, to identify penetration or aspiration, and to observe physiological changes that are responsible for the symptoms (Kwon & Lee, 2019). A specific protocol for instrumental swallowing assessment in IPD people has not already been published. A German group validated a standardized protocol using FEES so called “FEES-Levodopa-test”, which assesses levodopa responsiveness in IPD patients with swallowing disorders (Warnecke et al., 2016) but it does not evaluate the pharyngeal residue associated with saliva and secretion.

Table 1. 4 Summary of the swallowing assessments for IPD.

Patient Reported Outcome Measures (PROM)	Clinical Assessments and screening	Instrumental Assessments
<ul style="list-style-type: none"> -Munich Dysphagia Test-Parkinson’s Disease (MDT-PD) - Swallowing disturbance questionnaire (SDQ) - Swallowing related questions of the Non-Motor Symptom Questionnaire (NMS-Ques) - Swallowing related questions of the MDS-Unified Parkinson’s Disease Rating Scale (MDS-UPDRS) 	<ul style="list-style-type: none"> - Timed Water Swallowing Test (TWS) -Tongue Pressure Measurement; - Swallowing Quality of Life questionnaire SWAL-QOL) 	<ul style="list-style-type: none"> - Fiberoptic evaluation of swallowing (FEES); - Videofluoroscopy (VFS) -High Resolution Manometry (HRM) -High-resolution impedance manometry (HRIM)

1.6 Dysphagia Management

There are a number of approaches to managing dysphagia. In this section, management is considered in the context of other approaches to managing swallowing in people with IPD, starting from the medical interventions, describing compensatory approaches and the rehabilitative and behavioural treatments. The sEMG biofeedback intervention which is the focus of this study, is part of the rehabilitative and behavioural interventions.

1.6.1 Pharmacological

In the literature it is well documented that pharmacological interventions alone are ineffective for the recovery of swallowing function and the evidence for the impact of levodopa on dysphagia in IPD is inconsistent. In 2009, Menezes and Melo conducted a systematic review with meta-analysis and found that levodopa was not related to an improvement of swallowing dysfunction in IPD (Menezes & Melo, 2009). Sutton et al. disagreed with the results of Menezes & Melo meta-analysis (Sutton, 2013), indicating that levodopa may improve certain components of the swallow process (e.g., tongue strength, oral transit time and pharyngeal pressure) (Sutton, 2013). Although the debate is still ongoing, it seems that specific medications contribute significantly to reducing the rigidity, festination or freezing episodes, which have positive effects on autonomy of feeding and manipulate food during meals with possible impact on mood and quality of life of people with IPD (Miller, 2017).

1.6.2 Deep brain stimulation (DBS)

Along with these medical interventions, it is important to mention deep brain stimulation (DBS), which is a recognised treatment for reducing motor symptoms in individuals with IPD. The evidence of this intervention for limb rehabilitation in the published literature is robust (Aum & Tierney, 2018; Kogan et al., 2019). However, the motor improvements following the application of DBS are not consistent with swallowing recovery, as it is documented by systematic review by Troche et al. (Troche, Brandimore, Foote, et al.,

2014). Research in this field is ongoing and it seems that the placement of the stimulation implant is important and can affect swallowing safety considerably in people with IPD (Troche, Brandimore, Foote, et al., 2014).

1.6.3 Compensatory approaches

Compensatory approaches are defined by Rosenbek and Jones (Rosenbek & Jones, 2008, p. 57) as: “*Compensatory refers to approaches that try to accommodate each person’s dysphagia rather than trying to improve it*”. These approaches include postural manoeuvres and changes to food consistency. Because of the presence of cognitive impairment also in people with IPD, compensatory strategies such as chin tuck, supraglottic swallow etc may be difficult for people to remember. In IPD populations, it has been recognised that the use of thickened fluids leads to positive immediate effects on elimination of aspiration in comparison with chin-posture. Nevertheless, there can be long-term side effects such as dehydration and urinary tract infections (Logemann et al., 2008; J. Robbins et al., 2008).

1.6.4 Rehabilitative strategies

Rehabilitation techniques aim to improve swallowing by altering physiology intended to achieve long-lasting improvement (Rosenbek & Jones, 2008). Despite the high prevalence of dysphagia and the severe clinical sequelae of this dysphagia in PD, few studies have documented the effects of swallowing rehabilitation in individuals with IPD. A systematic review by Van Hooren et al. confirmed the need to explore targeted training techniques for people with IPD and dysphagia using larger methodologically sound RCTs (van Hooren et al., 2014).

In general, swallowing rehabilitative exercises could be divided into two groups: (1) swallowing related exercises that do not involve swallowing and aim to change the underlying pathophysiology for example to augment strength; (2) exercises that involve swallowing in which the aim is to increase the skills of deglutition.

Most of studies in rehabilitation in IPD concern swallowing related exercises that do not involve swallowing. These include oral motor tasks (such as motion of the tongue, lips), respiratory exercises based on the assumption that swallowing difficulties are caused by muscles weakness. Argolo et al. examined the effect of oral motor exercise in 15 people with dysphagia and IPD. They found improvements in the timing of swallowing after treatment (Argolo & Nóbrega, 2013). However, they did not report retention effects at follow-up assessments. Other rehabilitation exercises focus on strength. Recent systematic reviews found that Expiratory Muscle Strength Training (EMST™) leads to beneficial changes in swallowing in individuals with IPD (Laciuga et al., 2014; Mancopes et al., 2020; van Hooren et al., 2014). EMST™ aims to increase the force generation capacity of the expiratory muscles which has an impact of voluntary cough and swallowing safety. Firstly, Pitts and Sapienza (2009) analysed the effects of EMST™ in order to enhance voluntary cough and swallowing in IPD (Pitts et al., 2009). They found a statistically significant increase in cough volume acceleration and a decrease of penetration and aspiration events immediately after treatment. Later, Troche et al. confirmed these results in a RCT, with a statistically positive effect of the treatment on hyolaryngeal elevation (Troche et al., 2010). This may explain the benefits of the exercise programme on swallowing safety in this study. Although positive effects were evident, these exercises did not directly address swallowing function. As Troche explained at the ESSD Congress in Dublin 2018, the recovery of swallowing function in IPD is complex and requires a specific treatment protocol, including strength exercises combined with specific swallowing tasks.

The exercises, which include the act of swallowing, incorporate the neuroplasticity principle of “Use or lose it” (Martin, 2009). According to this principle, the swallowing tasks, albeit only on “dry” or saliva swallows, may help preserve cortical and subcortical representations and it is hypothesised that they make the patient’s return to oral nutrition easier and faster. The effects of these exercises in IPD seems to be positive (Russell et al., 2010). The principles of neuroplasticity constituted the basis of these swallowing rehabilitation techniques and are described in more detail in Chapter 2.

1.6.5 Repetitive transcranial magnetic stimulation, direct current stimulation and neuromuscular electrical stimulation

In the recent years, there has been an increasing interest on the application of transcranial magnetic stimulation and direct current stimulation for swallowing rehabilitation, showing promising effects in patients with stroke and dysphagia (Langmore & Pisegna, 2015; Pisegna et al., 2016; Simons & Hamdy, 2017; Simons, 2017). Nevertheless, the effects of these techniques have not already been analysed in clinical trials and there are no large systematic review in IPD population. Thus-the evidence for these intervention is still weak.

Following this overview of dysphagia in IPD and its challenges in assessment and intervention, the next section introduces the context of the study.

1.7 Context of the Study

The researcher is an Italian speech and language therapist with twelve years' experience working in neuro-rehabilitation hospitals in Venice (Italy). Her clinical experience includes the treatment of people with stroke, traumatic brain injury, amyotrophic lateral sclerosis, multiple sclerosis as well as individuals with IPD. The work experience with IPD people led to recognition that this population is growing with little evidence for efficacy and effectiveness of swallowing treatment, albeit with severe medical, social and psychological consequences of dysphagia in this population. This motivated the student to carry out a PhD study in this field.

1.8 Research aims and objectives

The focus of this PhD study is improvement in swallow function and ultimately quality of life in people with IPD and dysphagia. The main aim is to complete a feasibility study to examine the effectiveness of a specific intervention programme involving biofeedback and sEMG in people with IPD and dysphagia and to inform for a larger clinical RCT study.

The objectives are to define how biofeedback improves swallowing in people with IPD and dysphagia; to assist in identifying potential problems with recruitment and the process of data collection so that these could be tackled prior to rolling out a larger RCT; to refine the therapy approach itself, as appropriate at the end of the study; to help determine the acceptability of the therapy regime to patients with IPD and to evaluate the suitability of key therapy outcomes and adapt them as required. The research questions are informed by the systematic review (Chapter 3) and the theoretical framework and the research questions are described in Chapter 4.

1.9 Outline of the chapters

This dissertation comprises nine chapters:

Chapter 1 This chapter has introduced the topic and describes the main symptoms and prevalence of the disease and management of dysphagia in people with IPD. It explains the context for the study as well as the motivation to conduct this research. The aims and objectives of the research are presented.

Chapter 2 This chapter provides the rationale for the use of biofeedback as an augmentative tool for rehabilitation in people with IPD.

Chapter 3 This chapter presents a systematic review on the use of biofeedback for swallowing recovery in people with IPD and dysphagia.

Chapter 4 This chapter discusses the theory underpinning the feasibility study and defines the research questions.

Chapter 5 This chapter describes the methodological approach of this feasibility study. It includes a detailed description of the design study, the research tools and ethical consideration. In addition, it provides information on the participants, method and analysis of data (quantitative and qualitative).

Chapter 6 This chapter describes the cross-cultural translation of the Functional Oral Intake Scale (FOIS) into Italian (FOIS-T).

Chapter 7 This chapter reports the results from the quantitative and qualitative findings.

Chapter 8 This chapter discusses the main findings of the feasibility study.

Chapter 9 This chapter concludes the thesis with a protocol for a future study.

Chapter 2: Biofeedback as a Complex Intervention for Dysphagia

2.1 Introduction

This thesis is concerned with biofeedback as a therapeutic intervention. This chapter discusses the use of biofeedback as an augmentative tool for rehabilitation in people with IPD. The intervention is recognized as a complex intervention and this framework influences the design and the implementation of the main study. The chapter is divided into three sections. Section 1 provides an overview of the process of developing a complex intervention. Section 2 describes the key concepts of neuro-rehabilitation associated with the use of biofeedback in IPD and the last section gives the rationale for biofeedback as an augmentative tool for swallowing rehabilitation.

2.2 Complex Interventions

Biofeedback as an augmentative tool for dysphagia treatment is considered a complex intervention. Complex interventions are defined as treatments with several interacting components in different dimensions from the theoretical to methodological and practical levels (Craig, 2019; O'Cathain et al., 2019). In rehabilitation, they constitute the majority of interventions as the type of population, the heterogeneity of the treatment approach and the range of possible outcomes increase the complexity of the intervention (Dowding et al., 2017).

In 2000, the Medical Research Council (MRC) published a *“Framework for the Development and Evaluation of RCTs for Complex Interventions to Improve Health”* to help researchers during the implementation of a complex intervention (Campbell et al., 2000). Recently, it has been updated and enhanced in order to guide researchers during all phases of implementation from the initial methodological choice to the design and

execution of the study (Craig, 2019). It is a clear and straightforward framework, which helps to examine the several variables during the development phase of the complex intervention (Baird et al., 2019).

The framework comprises four main stages from development through to implementation of a complex intervention (Figure 2.1): 1) Development; 2) Feasibility; 3) Evaluation and 4) Implementation. The stages interact with each other and do not follow a linear pathway in order to allow change and flexibility during the process.

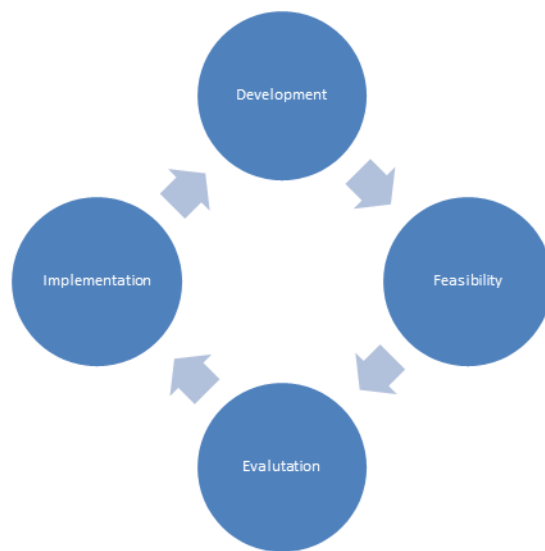


Figure 2. 1 Framework of the Development and Evaluation of Complex Interventions

2.2.1 Stage 1: Development

This stage consists of developing the knowledge base for an intervention in order to determine whether a specific approach, or undertaking a specific treatment, provide an effective benefit or not (O'Cathain et al., 2019). It involves the analysis of the evidence and the identification of an appropriate theory to guide the implementation. This evidence should be obtained throughout a systematic review. As complex interventions are difficult to compare and summarise, Higgins et al. suggests a descriptive approach to analyse the common patterns and characteristics within the included studies (Higgins et

al., 2019). This narrative analysis of the evidence leads to the creation of an appropriate theory. The theory incorporates the need for the new intervention, the rationale behind the study, the theoretical basis and what and how change is expected to achieve and measure (Baird et al., 2019). In this PhD study, the creation of a theory should give an explanation as to the use of biofeedback as a component of swallowing intervention and indicate the most appropriate swallowing outcome measures for people with IPD and dysphagia.

2.2.2 Stage 2: Feasibility

This phase is characterised by modelling the intervention in a small scale (Baird et al. 2019). It helps to create a deeper understanding of the contextual factors, which influence the treatment, variables within the research and the outcomes which need to be examined. It is recognised also as Phase 1 of the clinical research process (Claxton et al., 2002), as it allows one to estimate important features to help design the main intervention and also examines the safety of the intervention for clinical use. The aim of this stage is to verify if the intervention leads to the expected results and if the intervention is appropriate for the context and for the population in question. Several quantitative and qualitative assessments should be applied in this phase in order to provide a complete picture of the potential benefits and drawbacks of study. The guideline emphasises the importance of collecting data on unexpected changes, which should be documented accurately (Dowding et al., 2017). Interviews or open questions are suggested as useful tools with free form answers in order to understand positive or negative effects of the intervention (Campbell et al., 2000; O'Cathain et al., 2019). O'Cathain et al. suggest including also an analysis of the fidelity of the treatment, contextual factors and feedback from stakeholders and participants, which give a fundamental contribution for the development of further studies (O'Cathain et al., 2019).

2.2.3 Stage 3: Evaluating a complex intervention

This phase concerns the assessment of the effectiveness and efficacy of the complex

intervention in a larger RCT study. It involves the choice of research design; the appropriate outcome measures in order to verify if the intervention leads to positive changes in a larger scale (Craig, 2019). This PhD dissertation will end at this stage, proposing an intervention protocol for a RCT using swallowing biofeedback in people with IPD.

2.2.4 Stage 4: Implementation

The last phase of complex intervention evaluation and implementation involves disseminating the evidence and sharing the results in peer-reviewed journals and at national and international conferences (Dowding et al., 2017). Although, this is the last stage of complex interventions, this research student has already started disseminating the preliminary results of components of the thesis at European Society of Swallowing Disorders and has published two chapters of the PhD (Chapter 3 and Chapter 6). A key component of the theory on biofeedback in dysphagia intervention in people with PD and dysphagia is that it is linked with motor learning and neurorehabilitation. This is considered next.

2.3 The keys concepts of neurorehabilitation

As stated above in section 2.2.1, this first stage of designing and implementing a complex intervention involves the analysis of the evidence and the identification of an appropriate theory to guide the implementation. This section examines the key concepts of neurorehabilitation as the intervention proposed in this thesis involves these concepts.

The first main concept is **motor learning**. The aim of neurorehabilitation interventions is to reduce impairments and maximize functional ability through the process of motor learning. Motor learning has been defined as *“an internal processes related with practice or experience leading to the acquisition of a new motor skill”* (p. 302) (Schmidt & Wrisberg, 2008, p. 302). Some researchers have tried to develop a model to describe motor learning (Fig 2.2)(Roller, 2012). The process is characterised mainly by three stages.

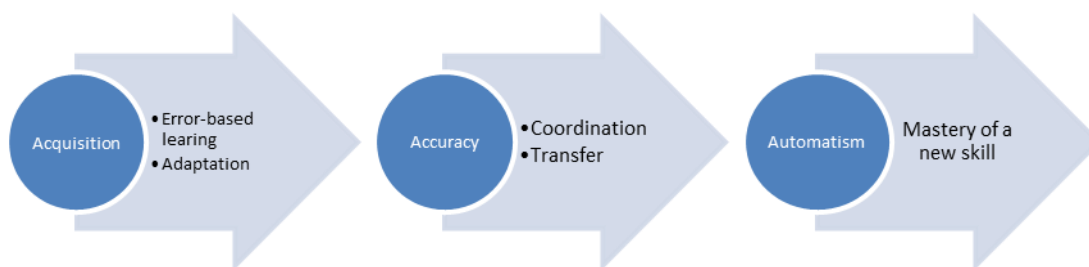


Figure 2. 2 The stages of motor learning (Roller, 2012).

The **acquisition** is accomplished by error-based and adaptation processes. In the error-based process, the learner acquires the new skills throughout the recognition of the mistakes during the execution of the skill (Krakauer et al., 2019). The adaptation process involves using the sensory information in order to predict performance and reduce errors (Krakauer et al., 2019; Shadmehr et al., 2010). The sensory system intervenes in two sensory loops. The first is the feed-back loop and the second is feed-forward (Schmidt & Wrisberg, 2008; Shadmehr et al., 2010). A **feed-back** loop provides the proprioception information, which allows one to monitor the execution of actions and provides a sensory response to the control command (Humbert & German, 2013; Petzinger et al., 2013). **Feed-forward** estimates if the motor command matches with the performance (Petzinger et al., 2013; Shadmehr et al., 2010). These loops allow the neuronal system to learn and to adjust the actions reducing the errors. The use of biofeedback during rehabilitation attempts to supplies the proprioceptive information when these loops are damaged.

The second phase regards **accuracy** of the movements, improving coordination and efficiency and transferring performance in different environmental contexts ((Krakauer et al., 2019). As it has been described in the following sub-sections (2.3.3), biofeedback intervenes in increasing precision and accuracy of the motor task. The last stage concerns the **automatism** and completes mastery of the new skills (Roller, 2012).

2.3.1 Neuroplasticity

Motor learning occurs when the brain adapts in response to practice or experience of a certain skill resulting in changes in the central nervous system that allow the performance of a new motor task (Krakauer et al., 2019; Schmidt & Wrisberg, 2008). Within this process, providing appropriate cues and external feedback is fundamental to foster motor learning and cerebral reorganization (Basmajian, 1981; Petzinger et al., 2013). Sensory information and training have the potential to boost the formation of synapses and remodelling dendritic connections in the cortex that create a long-term structural change in neuronal system (Khan et al., 2017).

This complex process is defined as **neuroplasticity**. Neuroplasticity is the last phase of motor learning and it occurs when the new skill has been integrated and creates a neuronal change (Nudo, 2006). The majority of the studies on motor learning and neuroplasticity has been conducted in stroke populations (Coleman et al., 2017; Dabrowski et al., 2019). They confirm that the brain could re-organise if the patient followed a specific treatment based on the neuroplasticity principles. In the IPD population, an emerging body of evidence indicates that intensive and specific rehabilitation interventions lead to the activation of several plasticity related events in the IPD brain (Hirsch et al., 2016).

2.3.2 The neuronal network of motor learning and implications for IPD

In the recent years, many studies have shown the involvement of cortical and subcortical structures during motor learning (Krakauer et al., 2019; Roller, 2012). The basal ganglia plays a fundamental role during the acquisition phase of a new skills (Section 2.3; Fig 2.2) (Petzinger et al., 2013). Recent studies have shown that during the error-based phase, there is an activation of ventral basal ganglia and the associative regions, involving dopamine and the dopaminergic D1 and D2 receptors (Petzinger et al., 2013). In the later automatism stages, there is major involvement of sensorimotor circuit including the dorsal basal ganglia and the sensorimotor cortex (Figure 2.3) (Ernster et al., 2018). These

circuits are severely compromised in IPD and this could explain the difficulties of motor learning in this population. The next section describes how biofeedback intervenes in order to compensate the damage of basal ganglia in IPD.

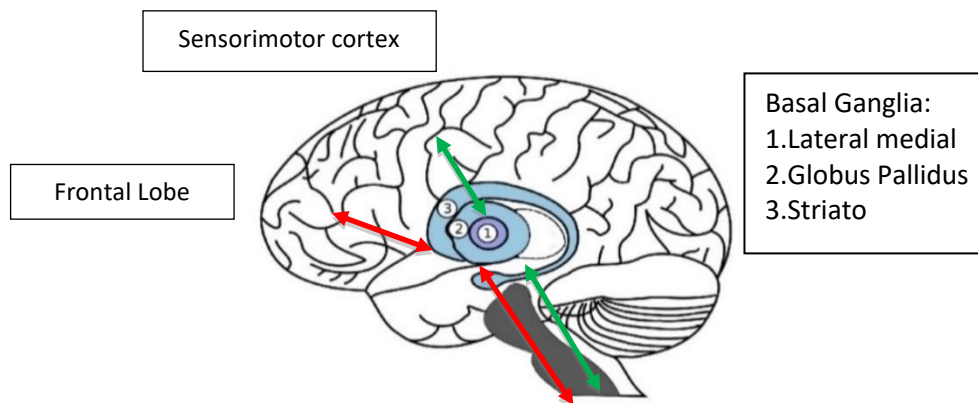


Figure 2. 3 Neurological involvement of motor learning process during the error-based and goal directed tasks (green lines) and during automatism tasks (red lines).

2.3.3 The neurological network of compensation mechanism of cueing in IPD

Several studies on neuroimaging found that the fronto-striatal pathways are particularly compromised in IPD patients (Storey et al., 1995; Tard et al., 2015). The role of feedback is to compensate the neurological impairments and activates the cortical involvement. That is, biofeedback activates directly the superior parietal lobe (SPL), inferior parietal cortex (IPC), pre-supplementary motor area (pre-SMA) and primary motor cortex (M1) in people with IPD, bypassing the basal ganglia (Fig. 2.4) (Petzinger et al., 2013; Tard et al., 2015). This explains the benefits of using cues and biofeedback in this population. Although the research is still ongoing and the underlying neural network of cueing is still the subject of ongoing debate, the neural network of cueing, which avoids the dopamine-deficient basal ganglia contribute to positive rehabilitation outcome in people with IPD.

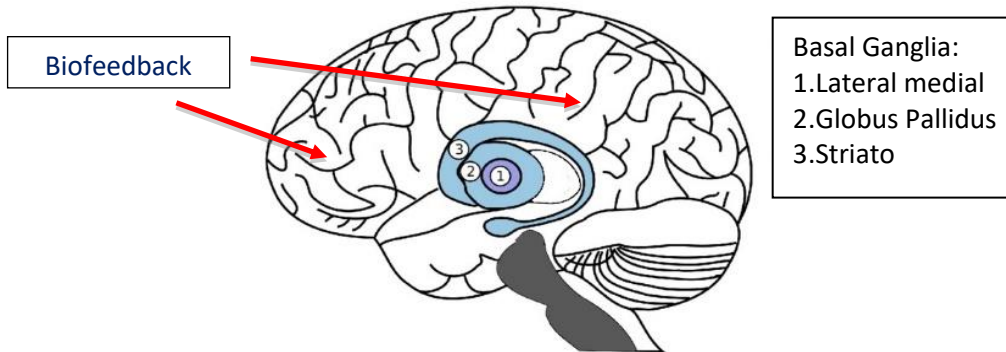


Figure 2. 4 The neuronal network during motor learning using biofeedback.

In conclusion, there is evidence that customised treatment based on goal-orientated motor learning exercises using feedback strategies increase proprioception and induce sustained benefits on motor outcomes especially in the IPD population. The following subsection concerns the importance of the use of biofeedback for motor recovery and for swallowing treatment in people with IPD and dysphagia.

2.3.4 The application of cues and biofeedback for motor recovery in IPD

It is widely accepted that the damage to basal ganglia and its cortical connections cause an impairment of motor learning in IPD at different levels (Bartels & Leenders, 2009). Nevertheless, the ability to learn is not completely lost in this population. Several studies, indeed, show that people with IPD acquire new skills easier even if the rate of learning is decreased when compared to healthy controls (Hayes et al., 2015). **Cues** are defined as discrete targets used to facilitate the execution of a movement (Ginis et al., 2017), while **feedback** are signals that allow the awareness of the ongoing movements in order to accomplish the correct performance (Basmajian, 1981).

The use of augmentative cues and feedback play a key role in helping the acquisition of new skills in IPD individuals, as they active a compensatory mechanism different from the dopaminergic circuits (Kearney et al., 2018; Kearney et al., 2019)

Biofeedback can involve the use of cues and/or sensors attached to your body to measure key body functions, facilitating awareness and/or control over the motor performance of the task (Basmajian, 1981). This information is recognised as augmented or extrinsic biofeedback, because it furnishes additional information such as movement-related information and it can be used also to supplement sensory proprioception. In rehabilitation, extrinsic biofeedback is extensively used to integrate weaknesses of intrinsic biofeedback mechanisms while increasing motor learning, generalization and retention (Basmajian, 1981). Biofeedback is argued to challenge patients beyond self-sensory abilities and facilitates awareness of movements that were previously automatic and unconscious (Ginis et al., 2017; Hirsch et al., 2016).

In the research literature, the evidence for the positive effects of cueing and biofeedback in IPD patients is well recognised. IPD patients seem to benefit more from augmentative cues than other parkinsonian syndromes and neurological disorders (Ginis et al., 2017; Kearney et al., 2018).

In the parkinsonian literature, two types of feedback are mainly adopted: visual and auditory Feedback (Deane et al., 2001; Tomlinson et al., 2014). Visual spatial feedback seems to increase motor functions such as walking and reduce freezing episodes (Rocha et al., 2014). An example of these are strips placed on the floor or coloured tiles, which facilitate the re-integration of sensory information particularly important for stepping movement. For this reason, they are specifically indicated for freezing of gait (Ginis et al., 2017). Ginis et al. systematically reviewed the literature on the use of cues in IPD individuals with freezing of gait syndrome and found that cues helped to focus the attention to gait reducing the interferences of other input (Ginis et al., 2017). Providing specific sensory cueing seems to facilitate cognitive and/or guide the motor execution, selecting the salient sensory information (Nonnekes et al., 2015).

The second type of feedback is auditory involving a metronome or music with positive effects on the timing of gait and limb coordination (Delval et al., 2014). A systematic review showed that auditory cues affected specifically the kinematic gait parameters,

increasing the speed of walking and so they are more effective in bradykinetic symptoms of IPD (Spaulding et al., 2013).

Nevertheless, the main limit in using these strategies concerns difficulties in retention and generalisation mostly in people with severe IPD. Some studies have confirmed that people with IPD are strictly dependent on the use of both cues and biofeedback and have difficulties with consolidation of strategies and also transference to new tasks (Ekker et al., 2016; Hayes et al., 2015; Heremans et al., 2016). This could be due to the cognitive impairment but also to modality of cue administration, which does not stimulate motor learning (Kelly et al., 2015; Zemankova et al., 2016). The delivery of constant and steady feedback indeed does not stimulate learning (Hula et al., 2008; Peterson et al., 2016). In this way, patients are likely to be accustomed to receiving an external cue and the motor learning skills are not challenged.

Although the evidence presented above concerns limb rehabilitation, it is argued that they could be applied to swallowing rehabilitation. This is considered in the next section.

2.4 Biofeedback in swallowing rehabilitation

Over the last two decades, biofeedback in swallowing rehabilitation has gained increasing interest and several studies have confirmed the benefits of feedback in the recovery of swallow function. Huckabee and Burnip summarised the conceptual basis for the use of swallowing biofeedback for motor learning during swallowing skill training. They proposed 5 stages from skill acquisition to generalisation (Huckabee & Burnip, 2018) (Fig. 2.5). In the first stage the use of biofeedback contributes to increasing the salience and specificity of the swallowing task. In combination with high intensive and repetitive practice they argue that the swallowing skills will be acquired.

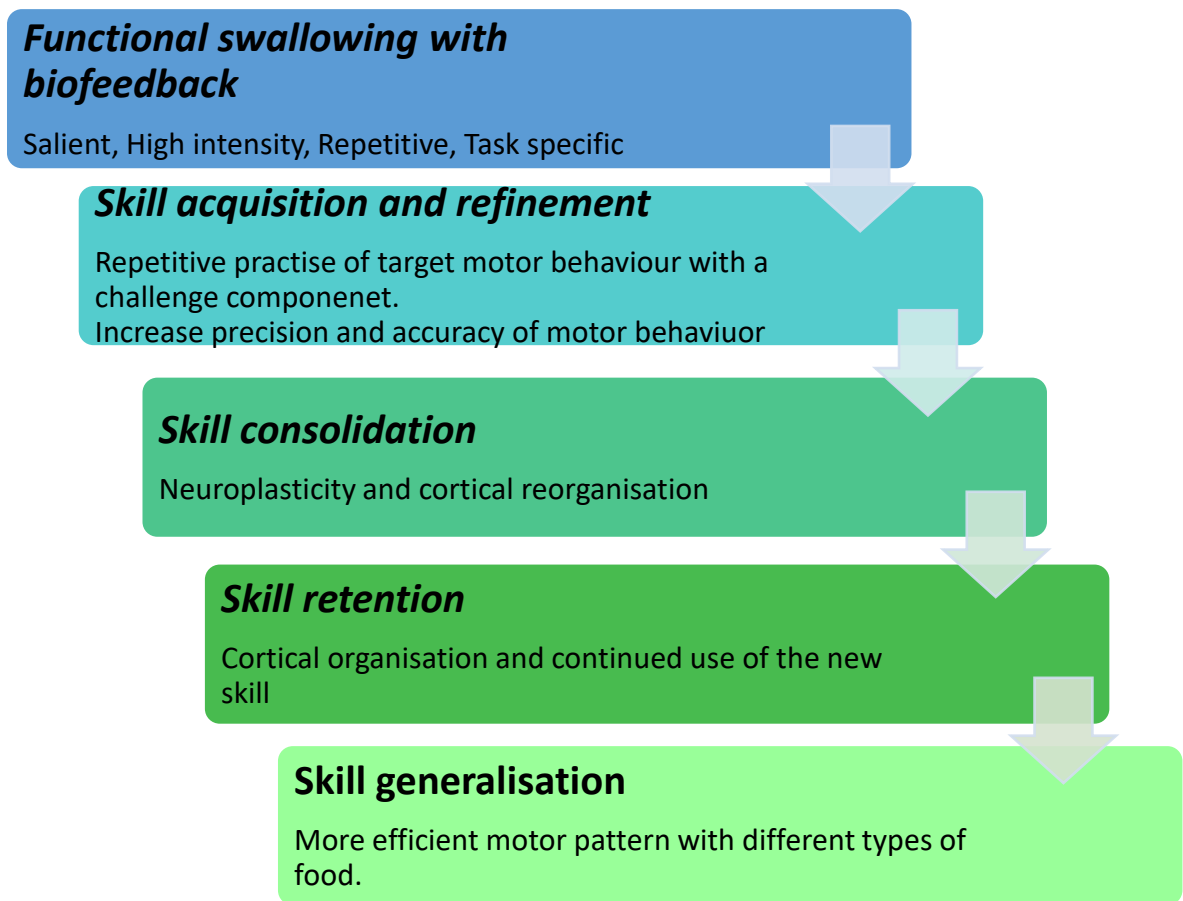


Figure 2.5 Theoretical stages of swallowing skill acquisition as defined by Huckabee & Burnip (2018)

A recent systematic review on the use of biofeedback for swallowing rehabilitation suggests that feedback is useful as an augmentative tool in interventions for dysphagia associated with a range of aetiologies with positive outcomes on maximum displacement of the hyoid bone during swallowing (Benfield et al., 2019). Nevertheless, the review showed considerable heterogeneity in terms of types of biofeedback devices employed, types of biofeedback provided, swallowing exercises used, and outcomes measured in studies using biofeedback for recovery of swallow function differ enormously. The main limit of this systematic review was the failure to analyse the effects of biofeedback based on the diagnosis or aetiology of dysphagia. It also does not indicate why biofeedback might work or how it works with specific populations. No systematic review specifically

on swallowing biofeedback in people, with IPD and dysphagia despite the recognised value of biofeedback in this population. The following sub section explores this but considers it also in terms of the types of cues applied to swallowing rehabilitation.

2.4.1 Types of cues and biofeedback applied to swallowing rehabilitation

In swallowing rehabilitation, the majority of cues are verbal (Macrae et al., 2014). Although, verbal cues have always been part of any swallowing treatment, few studies have analysed the accuracy and the method of delivery during swallowing rehabilitation. Macrae et al. found that verbal cues played a significant role on the acquisition of volitional laryngeal vestibule closure (vLVC) manoeuvre (Macrae et al., 2014). In dysphagia treatment, biofeedback has been used as an adjunctive treatment tool to increase awareness of the swallowing process and to increase the patient's control over performance, by offering concrete external monitoring that allows improvement of disordered swallowing (Humbert & German, 2013). The tools for providing biofeedback of swallowing can be summarized in main three groups:

1) FEES or VFS video. This feedback consists of video-recording the patient's swallowing performance during the instrumental examination using FEES or VFS. The videos are then used as feedback to increase the patient's awareness of the swallowing dynamic in order to enhance the recovery (Azola et al., 2015; Manor et al., 2013; Vose et al., 2019). This feedback is not typically simultaneous with the act of swallowing, and videos are watched after the examination. Nevertheless, they provide an image of internal pharyngeal movements and the actual swallowing function. The major limitation of this feedback is that both these instrumental procedures are recognised to be invasive and would not be used to provide biofeedback alone.

2) Manometry is a clinical procedure in which a thin, flexible catheter is placed in the pharynx and oesophagus for measurement of swallowing-related contact pressure against the catheter's closely-spaced pressure sensors (Jones & Ciucci, 2016). This tool has been recently used as biofeedback to improve the coordination of pharyngeal muscle

contraction (O'Rourke & Humphries, 2017). Huckabee et al. demonstrated the successful implementation of pharyngeal manometry in 16 patients who have mis-sequenced constriction of the pharynx during swallowing task (Huckabee et al., 2014). Like VFS and FEES however, this is also invasive and is difficult to justify using for biofeedback.

3) Surface electromyography (sEMG) is one of the most commonly used biofeedback tool for measuring submental muscle activity and its use in dysphagia rehabilitation is supported by several studies (Albuquerque et al., 2019; Crary et al., 2004; Huckabee & Cannito, 1999). This is readily available and not invasive. It is also less expensive than some of the techniques mentioned above. In this procedure the surface electrodes are attached to the submental muscles corresponding with mylohyoid, genioglossus and anterior belly of digastric muscles (Crary et al., 2004). This group of muscles responsible for hyoid movement associated with initiation of swallowing (Humbert et al. 2015).

The surface electrodes collect the electrical activity from the muscles to which they have been attached. For swallowing purposes the area of interest is the submental region, three electrodes are used: one is the positive; one is the negative and one acts as the ground electrode (Figure 2.6).

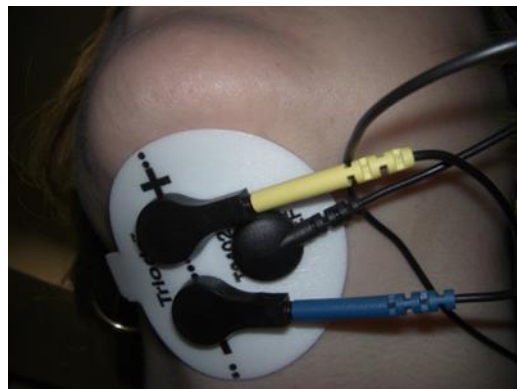


Figure 2.6 Example of surface electrodes attached to the submental region

The bioelectrical signals are amplified and filtered in order to be converted into a digital signals. On the screen, the signals are displayed in a form of a wave line shape in a graph

which provides information on the power of muscle contractions (ordinate axis) and the timing of contraction (abscissa axis). The amplitude of the wave line reflects the power of muscle activity in microvolt (μV). The power of a typical sEMG signal ranges from 0–450 μV (Stepp, 2012). The frequency of swallowing during sEMG biofeedback treatment is determined by the number of wave lines in the screen. The duration of the wave line during normal swallowing ranges from 1 to 3 seconds. Sometimes, the wave line is longer than 3 second, for example during Mendelsohn manoeuvres. During normal swallowing, the line can could present as two waveform peaks, which represent are the attempts to swallow (Figure 2.7).

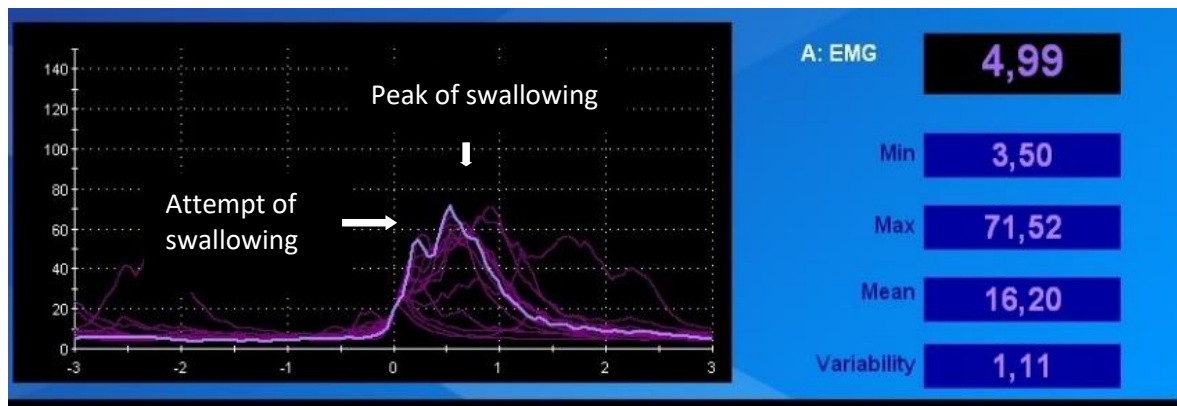


Figure 2.7: sEMG wave line during swallowing

One of the first case studies to investigate the use of sEMG as an adjunctive tool for swallowing rehabilitation was in a patient with swallowing disorder secondary to stroke conducted by Huckabee & Cannito (Huckabee & Cannito, 1999). They found positive outcomes for swallowing training using by sEMG biofeedback during effortful swallowing treatment (Huckabee & Cannito, 1999).

During effortful swallow task using sEMG biofeedback, an horizontal line is displayed and it plays a strength training target (Figure 2.8). The task is to exceed the target line with the peak of the swallowing amplitude.

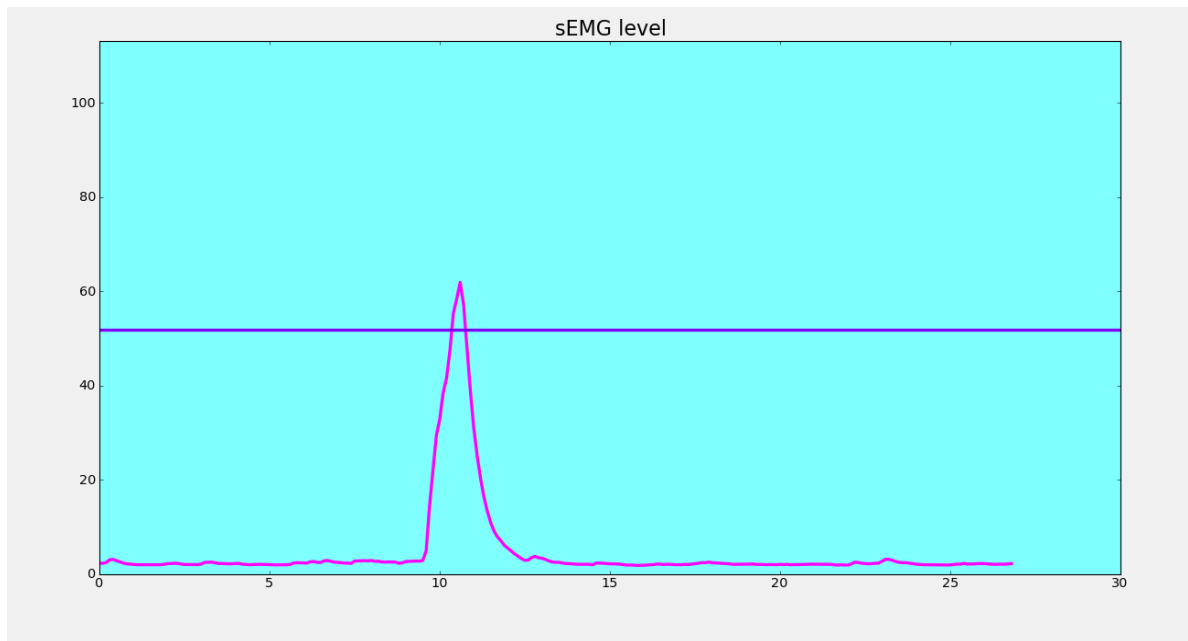


Figure 2.8: Example of effortful swallowing using sEMG biofeedback

The majority of first studies used sEMG mostly during swallowing strength treatment programme; for example, during effortful swallowing tasks or Mendelsohn manoeuvres (Crary et al., 2004; McCullough et al., 2012). The rationale for this was based on the notion of a positive correlation between the value of the maximum peak of the sEMG waveform and the degree of maximum hyoid excursion, as measured with VFSS (Wheeler-Hegland et al., 2008).

Recently, the swallowing laboratory coordinated by Dr Huckabee designed a software called Biofeedback in Strength and Skill Training Biofeedback in Strength and Skill Training (BiSSkiT), which was the first application of sEMG biofeedback during swallowing skill-based treatment approach (www.canterbury.ac.nz/rosecentre/products/bisskit-software). The task is to swallow with the peak of the sEMG waveform into the target box which require coordination of the timing and the strength of muscle contraction (Fig 2.9).

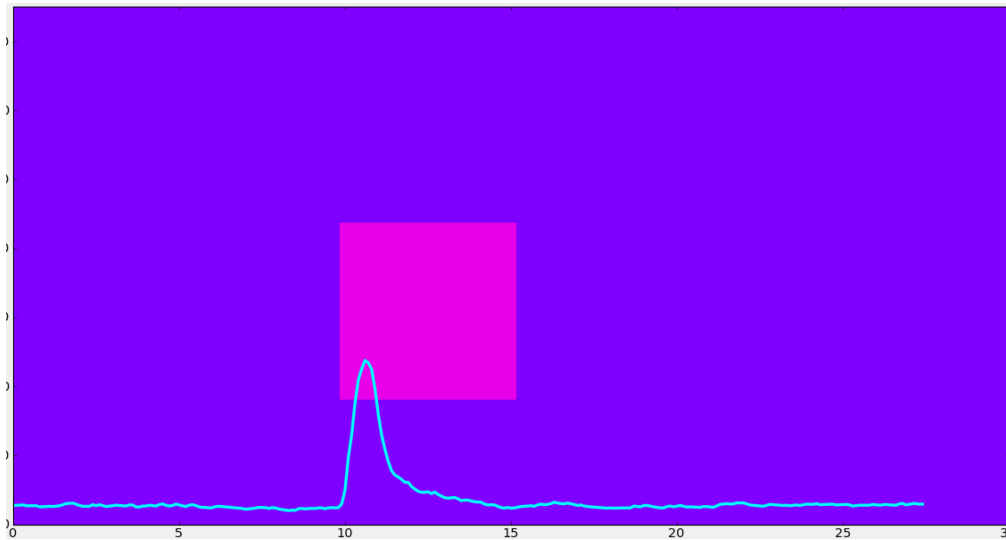


Figure 2.9: Example of swallowing skill task using BiSSkiT

In addition, a recent systematic review showed that the majority of the articles on sEMG swallowing biofeedback treatment included people with stroke and dysphagia, followed by IPD and individuals with dysphagia secondary to head and neck cancer (Albuquerque et al., 2019). The main outcome were increased hyolaryngeal excursion and maximal elevation of the hyoid bone (Albuquerque et al., 2019).

Biofeedback, therefore, seems to facilitate the recovery of swallowing function in IPD . One reason suggested earlier is that it embraces the principle of neuroplasticity. This is considered further below.

2.4.2 Swallowing biofeedback interventions and principles of neuroplasticity

The principles of motor learning and the importance of using cues for rehabilitation are fundamental in the recovery of dysphagia. Given the swallowing process is recognized to be highly dependent on sensory input, the external feedback increases the provision of sensory information and boosts the motor learning. The use of biofeedback, indeed, combined with the swallow tasks seems to embrace all the main principles of neuroplasticity (Martin, 2009). According to the ten principles of neuroplasticity, each principle is relevant to biofeedback swallowing treatment (Table 2.1) (Kleim & Jones,

2008; JoAnne Robbins et al., 2008).

The first principle is “**Use It or Lose It**” and it indicates that when a neural substrate is not stimulated, its function can degrade. There is evidence that “the loss of peripheral input to the sensory cortex produces a reduction of cortical somatosensory representation”(Kaas et al., 1983).

The second principle relates to the previous one and it is called “**Use It and Improve it**”. It is based on the notion that “using merely a function do not make changes in neural substrate, whereas using a function with increasing competence in terms of efficiency or accuracy could lead to neural modification and improvements” (JoAnne Robbins et al., 2008). Evidence of this application in swallowing biofeedback treatment was shown in Athukorala et al. and is described in the next chapter (Chapter 3) .

The third principle is “**Experience Specific**”, which assumes that “the exercises should involve swallowing activities and should be tailored for the patient”(JoAnne Robbins et al., 2008).

Table 2. 1 Summary of the 10 principles of neuroplasticity, including the description and the related swallowing intervention.

Principle	Description	Rehabilitation management
Use It or Lose It	It indicates that when a neural substrate is not stimulated, its function can degrade	Swallowing tasks
Use It and Improve it	Increasing competence in terms of efficiency or accuracy could lead to neural modification and improvements	Target swallowing exercises could lead to an improvement
Experience Specific	Exercises should be tailored in based of the specific deficit of participants	Customize the swallowing exercise based on deficit and interest of participants
Repetition Matters	Consistent practice is fundamental for learning and maintaining a function	Determine the number of swallowing exercises series
Intensity Matters	The concentration of exercise in a defined period	Quantify the number of swallowing exercises
Time Matters	The time of intervention and the duration of intervention influence the recovery	Early swallowing management reduces the risk of aspiration and pulmonary infections
Salience	Neural plasticity occurs when training is purposeful and related to the skill	Using food trials in the swallowing treatment
Age matter	Neural plasticity decreases over the life span	Younger people are more responsive to training than older ones
Transference	The ability of plasticity within one set of neural circuits to promote concurrent plasticity	Expiratory muscle strength training (EMST™) and Lee Silverman Voice Treatment (LSVT®)
Interference	The ability of plasticity within a given neural circuitry to impede the induction of new plasticity within that same circuitry	Inappropriate use of electrical stimulation in normal people, which inhibits swallows.

The fourth principle is “**Repetition Matters**”. It supports the notion that consistent practice is fundamental for learning and maintaining a function. In line with this, Maas et al., reviewed the literature in order to verify the amount of practice that results in greater retention in speech and language therapy (Maas et al., 2008) and found increased number of repetitions had positive effects in retention of improvements

The next principle is related to “Repetition” and it concerns the principle of “**Intensity Matters**”. Intensity could be referred to the concentration and the strength of exercise in a defined period (JoAnne Robbins et al., 2008)

The sixth principle is “**Time Matters**”. In the literature it is well recognized that immediate dysphagia management reduces the risk of aspiration and pulmonary infections. In the field of biofeedback, this principle embraces also the issue related to the timing of delivery of feedback (El Sharkawi et al., 2002; Hula et al., 2008) as it has been further described in section 2.4.

The seventh principle is “**Salience**”, which assumes that “neural plasticity occurs when training is purposeful and related to the skill”(JoAnne Robbins et al., 2008). This principle emphasizes the importance of using swallowing task specific training to generate neural adaptation and lead to neuroplasticity.

The eight principle is “**Age matters**”. It is broadly recognized that “neural plasticity decreases over the life span and young people are more responsive to training than older people”(JoAnne Robbins et al., 2008).

The next principle is “**Transference**”, which is one of the main principles for neuronal plasticity. It is defined as “the ability of plasticity within one set of neural circuits to promote concurrent or subsequent plasticity” (Kleim & Jones, 2008, p. S233). This principle presumes that the practice of specific movements could positively influence performance of other activities by improving somatosensory processing and optimizing neuromuscular firing pattern. This might explain why non-swallowing exercises enhance swallowing function, examples are the EMST™ (Pitts et al., 2009) and Lee Silverman Voice Training (LSVT LOUD®) (El Sharkawi et al., 2002)

The last principle is “**Interference**”. This is the “ability of plasticity within a given neural circuitry to impede the induction of new plasticity within that same circuitry”(JoAnne Robbins et al., 2008). The result is that learning or skill acquisition or reacquisition may be hampered.

2.5 Summary of the chapter

This chapter emphasised the importance of biofeedback for the limb rehabilitation in people with IPD. In addition, it has described the use of biofeedback as an augmentative tool for swallowing rehabilitation in people with dysphagia.

The next chapter is a continuation of the topic of this chapter, as it describes the results from a systematic review on the role of biofeedback for swallowing rehabilitation in people with IPD and dysphagia.

Chapter 3: A systematic review of interventions involving swallowing biofeedback in people with IPD

3.1 Introduction

This chapter describes the systematic review and provides an overview of the biofeedback application in people with IPD and dysphagia. Previous systematic reviews showed that swallowing biofeedback contribute in enhancing of the swallowing function in people with dysphagia (Albuquerque et al., 2019; Benfield et al., 2019) but is unclear if biofeedback is specifically effective in people with IPD.

Therefore, the aim of this systematic review was to find the evidence of this intervention in this population. The outcomes of interest were improvement in swallow function, changes in quality of life, nutritional status, and to confirm the safety of the intervention. Given that this project is the first step of a complex intervention study, the objective was to inform our understanding on how biofeedback works within intervention approaches for people IPD and dysphagia, identifying a theory of its implementation.

3.2 Methods

The protocol for this systematic review was registered on PROSPERO (CRD42017052477) and is available from

https://www.crd.york.ac.uk/PROSPERO/display_record.php?RecordID=52477.2.1

(Appendix A)

3.2.1 Selection criteria for studies

All published and unpublished randomized controlled trials (RCTs) and non-randomized controlled trials (NRCTs) were included. Relevant trials were those with at least one group with IPD receiving swallowing biofeedback treatment aimed at improving swallowing function and its impact. No language limits or date restrictions were applied to data base searches for studies.

3.2.1.1 Participants

Study participants were required to have a clinical diagnosis of IPD according to the United Kingdom Parkinson's Disease Society Brain Bank diagnostic criteria (Berg et al., 2015) or similar published clinical diagnostic criteria. Studies in which recruited people were at all stages of IPD, and all IPD disease severity levels presenting with all severities of dysphagia were included. Participants with parkinsonian syndromes and comorbidities associated with different neurologic disorders were excluded, as these conditions may impact swallowing pathophysiology differently.

3.2.1.2 Interventions

Interventions that used biofeedback as an adjunct or augmentation to swallowing therapy were selected. Biofeedback could involve any external visual, auditory, tactile cues or combination of cues using any type of device or equipment, which provided information to individuals with IPD on their swallow function.

3.2.1.3 Comparators

Comparison interventions comprised swallowing interventions, compensatory maneuvers or swallowing treatments without biofeedback or no intervention.

3.2.1.4 Outcomes

Outcomes selected were: (1) change in timing and efficiency of swallowing using valid outcome measures; (2) change in aspiration (food/fluid entering lungs) and safety of swallowing, as assessed by instrumental evaluation and validated rating scales; (3) change in residue as indicated on instrumental swallowing evaluation such as VFS and FEES or manometry and validated residue rating scales; (4) change in oral intake and/or in nutritional status documented on valid rating scales or as indicated through increased weight gain; (5) change in health-related quality of life as measured by quality of life measures or by participant and/or caregiver report. (6) Adverse events attributed to the intervention under investigation (e.g. increased dysphagia, fatigue, etc.). Studies that did not use instrumental assessments were included in the review but downgraded in terms

of methodological quality. Outcomes in three time frames were analysed: (1) immediate change in outcomes, (2) short term change (<12 weeks), and (3) long term change (> 12 weeks) in outcomes.

3.2.2 Search strategy

With the help of a university librarian, a systematic search strategy was designed and piloted to apply across all electronic databases identified as relevant to the research question (Table 3.1). This research student searched the following electronic bibliographic databases from inception to April 2019: EMBASE; PubMed; CINAHL; Web of Science; Elsevier Scopus; Science Direct; AMED; The Cochrane Database of Systematic Reviews; ProQuest Dissertations Theses A & I and Google Scholar. A follow up search was completed in January 2020. The research student and a colleague independently searched relevant conference proceedings from Dysphagia Research Society; European Society for Swallowing Disorders; International Congress of Parkinson's Disease and Movement Disorders and World Congress for Neurorehabilitation

Table 3. 1 Search Strategy

Database	Terms
Pubmed	(parkinson[Title/Abstract] OR parkinson's[Title/Abstract] OR parkinsons[Title/Abstract] OR "Parkinson Disease"[Mesh]) AND (dysphagia[Title/Abstract] OR dysphagic[Title/Abstract] OR deglutition[Title/Abstract] OR swallow[Title/Abstract] OR swallows[Title/Abstract] OR swallowing[Title/Abstract] OR swallowed[Title/Abstract] OR "Deglutition Disorders"[Mesh])
CINAHL Complete	(MH "Parkinson Disease" OR TI parkinson OR TI parkinson's OR TI parkinsons OR AB parkinson OR AB parkinson's OR AB parkinsons) AND (MH "Deglutition Disorders" OR MH "Swallowing Therapy" OR TI dysphagia OR TI dysphagic OR TI deglutition OR TI swallow OR TI swallows OR TI swallowing OR TI swallowed OR AB dysphagia OR AB dysphagic OR AB deglutition OR AB swallow OR AB swallows OR AB swallowing OR AB swallowed)
Web of Science Core (Thomson Scientific)	(parkinson OR parkinson's OR parkinsons) AND (dysphagia OR dysphagic OR deglutition OR swallow OR swallows OR swallowing OR swallowed)
EMBASE (Elsevier)	('parkinson disease'/exp OR parkinson:ab,ti OR parkinsons:ab,ti) AND ('dysphagia'/exp OR 'swallowing'/exp OR dysphagia:ab,ti OR dysphagic:ab,ti OR deglutition:ab,ti OR swallow:ab,ti OR swallows:ab,ti OR swallowing:ab,ti OR swallowed:ab,ti)
ScienceDirect (Elsevier)	(parkinson OR parkinson's OR parkinsons) AND (dysphagia OR dysphagic OR deglutition OR swallow OR swallows OR swallowing OR swallowed)
Database	Terms
Scopus (Elsevier)	(parkinson OR parkinson's OR parkinsons) AND (dysphagia OR dysphagic OR deglutition OR swallow OR swallows OR swallowing OR swallowed)
Cochrane Library	(parkinson OR parkinson's OR parkinsons) AND (dysphagia OR dysphagic OR deglutition OR swallow OR swallows OR swallowing OR swallowed)

ProQuest Dissertations and Theses	(parkinson OR parkinson's OR parkinsons) AND (dysphagia OR dysphagic OR deglutition OR swallow OR swallows OR swallowing OR swallowed)
AMED (EBSCO)	(parkinson OR parkinson's OR parkinsons) AND (dysphagia OR dysphagic OR deglutition OR swallow OR swallows OR swallowing OR swallowed)

3.3.3 Risk of bias and quality assessment

The methodological quality of the included studies was examined by the research student and colleague with the PhD supervisor using the Down's and Black checklist tool (Downs & Black, 1998) (Table 3.4).

3.3 Data collection and analysis

Reference manager software (Mendeley, www.mendeley.com) was used to manage references and remove duplicate publications. The research student and a colleague screened titles from the search using Covidence (www.covidence.org). Using this software platform, each person independently examined titles and abstracts for relevance to the review. Relevance of the search results was categorized as 'yes', 'no', or 'maybe'. If it was unclear from titles and abstracts whether a study should be included, full texts of these study reports were obtained for further examination. Letters to the editor were excluded. Any conflicts in inclusion were resolved through discussion with the PhD supervisor. The research student and a colleague extracted data and descriptive analysis was accomplished using Excel (Microsoft Corporation) spreadsheets.

3.4 Results

The electronic database search identified 10,785 records, of these 9,377 were duplicates, which were removed. The remaining 1,408 studies were screened against title and abstract. 1,384 of these studies did not meet the inclusion criteria. Full-text eligibility evaluation was carried out for 24 studies. Nineteen were excluded, as they were not the intervention of interest, and one was rejected, as it did not evaluate the outcomes selected for investigation. See PRISMA diagram (Figure 3.1). Four studies met the inclusion criteria for this review (Table 3.2). These were: Athukorala et al. (2014), Da Silva (2014),

Felix et al. (2008) and Manor et al. (2013).

There was considerable heterogeneity amongst included studies with regard to participant characteristics, study methodologies, swallowing outcome measures used, and specific swallowing tasks assessed. There was also incomplete data-reporting. Results are reported narratively.

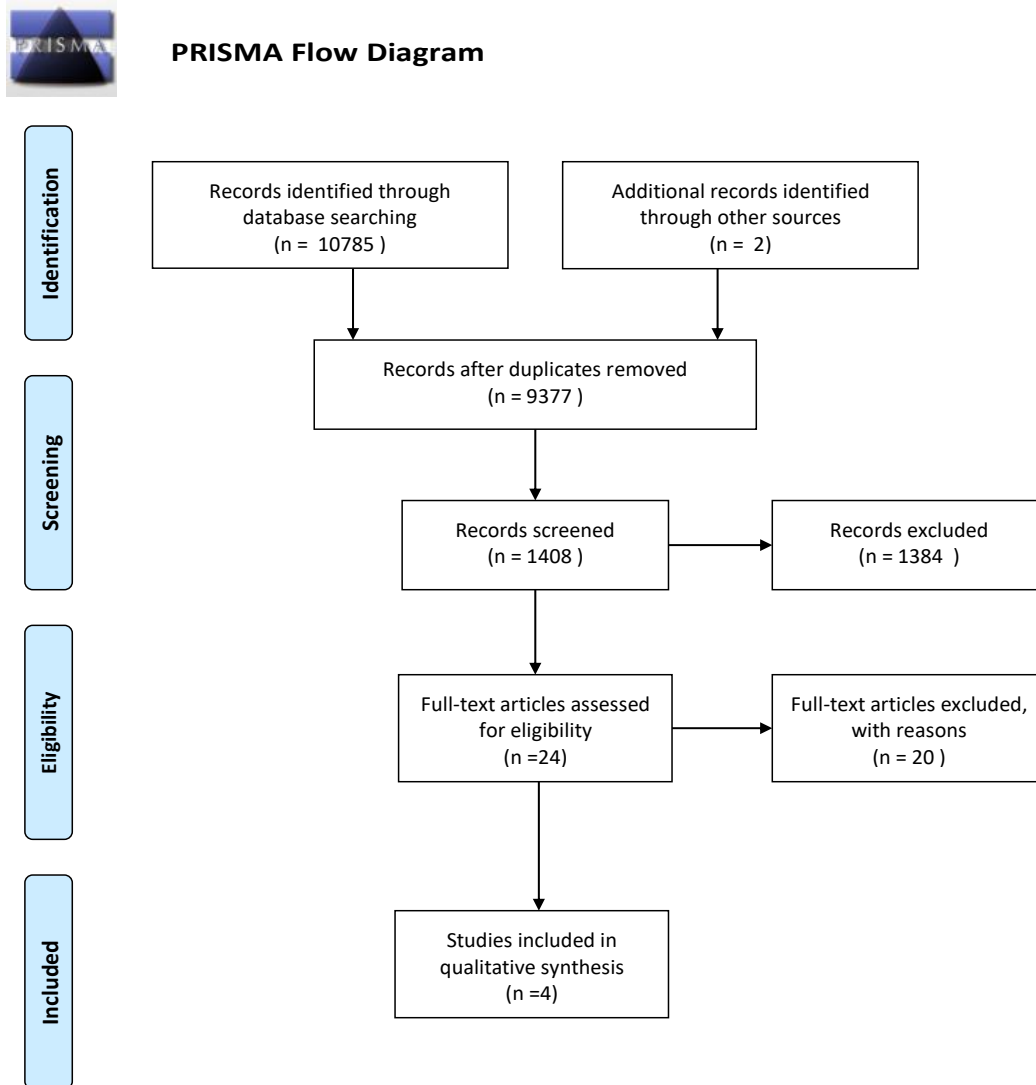


Figure 3. 1 Prisma Flow diagram.

Table 3. 2 Description of the included studies

Primary Author	Study Design	Participants	Disease Duration	Medication	Cognitive Status	PD Disease Severity	Outcome measures	Comparator Group	Intervention Group	Swallowing Tasks	Outcome	Follow-up
Athukorala et al. 2014	Within Subject Study	10 (3 female, 7 male; 67.4±8.6 yrs)	6.6±4	On Phase (Carbidopa/ levodopa)	-	H&Y 2.7±0.4	TWST, TOMASS and SWAL-QOL	Within study pre vs post treatment	Biofeedback Swallowing Skill Training (BISSKIT)	Saliva swallows (SKILL)	TWST (p=0.04)* TOMASS (p<0.001)* SWAL-QOL (p=0.18)*	2 weeks TWST (p=0.032)* TOMASS and SWAL- QOL (p>0.09)
Da Silva 2014	Case control study	6 (Males 73.1±6.2 yrs)	NA	NA	MMSE ±24	H&Y 2.3	FOIS DOSS (using VFSS) SWAL-QOL	The same treatment without SEMG biofeedback	Biofeedback Swallowing using SEMG	Oral facial, respiratory and voice exercises. Mendelsohn maneuver. Effortful swallow (STRENGTH)	FOIS (p=0.3) DOSS (p=0.6) SWAL- QOL (p=0.04)*	NA
Felix et al. 2008	Case series	4 (1 female, 3 male, 70.25 yrs)	9.25	On phase (Levodopa andBenserazide)	-	H&Y 3	Clinical swallowing assessment	Conventional Therapy	Biofeedback using an air balloon placed it in the anterior part of neck which was connected to a digital manometer	Effortful swallows (STRENGTH)	Clinical Assessment (p <0.001)*	NA
Manor et al. 2013	RCT	42 (18 female 24 male; 67.6±8.26 yrs)	7.43±4.66	NA	MMSE 28.14 ±1.59	H&Y 2.21 ± 0.79	FEES using non validated residue scale; SWAL-QOL; SWAL-CARE; POE	Conventional Therapy	Biofeedback Swallowing treatment using videos of FEES examination	Swallowing using compensatory manoeuvres (SKILL)	Reduced food residues at FEES (p<0.05)* SWAL-QOL (p<0.01)* POE (p<0.005)*	4 weeks and 6 months SWAL- QOL (p <0.5)*

MMSE: Mini-Mental State Examination ; H&Y: Hoehn and Yahr; TWST: Time Water Swallowing Test, TOMASS: Test of Mastication and Swallowing Solids (TOMASS); VFSS: Videofluoroscopy; FEES: Fiberoptic Examination of swallowing; DOSS: Dysphagia Outcome and Severity; SWAL-QOL: Swallowing Quality of Life; SWAL-CARE Swallowing Quality of Care (SWAL-CARE); POE: Pleasure of Eating
*statistically significant results

3.4.1 Study characteristics

Study design differed across the 4 studies (Table 3.2) with only one RCT (Manor et al., 2013). Manor et al. was the only RCT while Da Silva was a case control study, Athukorala et al. used a within subject pilot study design and Felix et al. (2008) was a case series study.

Studies were conducted between 2008 and 2014 and all were published in peer-reviewed journals with the exception of Da Silva. This was a degree thesis, with a poster presentation abstract from the European Society for Swallowing Disorders congress in 2015, published in the journal *Dysphagia*. This thesis was written in Portuguese and translated for the purposes of this review (Silva, 2014). All four studies were prospective studies; two were conducted in Brazil (Da Silva A., 2016; Felix et al., 2008), one in New Zealand (Athukorala et al., 2014) and one in Israel (Manor et al., 2013).

Recruitment methods was consecutive in three studies (Athukorala et al., 2014; Da Silva A., 2016; Manor et al., 2013) and unclear in Felix et al. 2008. Manor et al. and Felix et al.'s studies were carried out in a neurology department in Israel and Brazil respectively (Da Silva A., 2016; Manor et al., 2013). Da Silva's study was conducted in a neuro-rehabilitation hospital in Brazil (Da Silva A., 2016). Athukorala et al.'s study took place in an outpatient university clinic in New Zealand (Athukorala et al., 2014).

3.4.2 Population

Data on 62 patients with IPD were included across all 4 studies. Participants' age ranged from 62 to 83 years (mean 67.32 years). Severity of IPD was evaluated using H&Y scale (Hoehn & Yahr, 1967) in all included studies. Mean H&Y score provided by authors was 2.21 ± 0.79 ; range from Level 2 to Level 3, suggesting mild disease. Participants across the four studies represented a relatively homogeneous group in terms of age and disease (Table 3.2). All except Manor et al. indicated that participants received medication before the intervention and thus were "on phase" (Athukorala et al., 2014; Da Silva A., 2016; Felix et al., 2008). The medication status of participants is not mentioned by Manor et al. (Manor et al., 2013). Samples sizes were small in all 4

studies (Table 3.2).

Cognitive impairment was assessed using Mini Mental State Examination (MMSE) (Folstein et al., 1983) in Manor et al. and Da Silva's studies and in both studies MMSE scores were greater than 24 points indicating normal cognitive function for control and experimental groups (Da Silva A., 2016; Manor et al., 2013). No information of cognitive status was found in the other two studies.

3.4.3 Biofeedback Swallowing Intervention

3.4.3.1 Type of Biofeedback

Athukorala et al. and Da Silva used surface electromyography (sEMG) as biofeedback, which was simultaneous with swallowing tasks. The sEMG recording was visually displayed as a wave-line signal on a computer monitor or screen showing the timing and force of the muscle contraction involved during swallowing. In both studies, the sEMG electrodes were placed on the submental muscles to register the anterior movement of the hyolaryngeal complex during swallowing.

The goal of the intervention differed across both studies. The goal in the Athukorala et al. study was to improve the precision of muscle contraction to improve swallowing skills using immediate visual and auditory biofeedback. They used a specific biofeedback software programme (Biofeedback in Swallowing Skill Training) (BiSSkiT) (<https://www.canterbury.ac.nz/rosecentre/products/bisskit-software/>) that involved saliva swallowing tasks with increased the level of difficulty.

Unlike Athukorala et al., Da Silva focused on strength rather than skill of swallowing. They monitored the muscle contraction of participants' submental muscles using sEMG during different swallowing tasks such as effortful swallows, Mendelsohn manoeuvre, and McNeil training (Carnaby-Mann et al. 2010). It is unclear if the participants received auditory and visual or just visual cues. Like Da Silva, Felix et al. also focused on strength training and used visual and auditory cues. They designed a specific biofeedback device to provide information on increased strength of swallowing musculature during effortful swallows. They used an inflated air balloon and placed it

on the anterior part of the participant's neck. The balloon was connected to a digital manometer, which displayed pressures of the anterior part of the neck during effortful swallow exercise. Participants visually monitored the pressure measurements (mmHg) on the manometer during these effortful swallows and were verbally encouraged to increase the strength of swallowing.

In Manor et al., the focus was not skill or strength training but rather implementation of specific swallowing compensatory strategies (Manor et al., 2013). Participants watched videos of their own swallowing recorded earlier during FEES assessment while performing their individually tailored swallowing manoeuvre. They watched this video later using specified compensatory swallowing manoeuvres while eating and drinking and focusing on their new swallowing behaviour. This visual biofeedback was not done in real-time with swallowing events. The control group also received an individually prescribed swallowing manoeuvre but without video biofeedback. The focus of this study was not skill or strength training but rather implementation of specific swallowing compensatory strategies.

All four studies involved improving either skill or strength of swallowing or to change swallowing behaviour. It could be argued that motor learning was a core component of these interventions. The importance of motor learning was already emphasised in Chapter 2. To explore this further, analysis of biofeedback was carried out using motor learning principles framework described by Maas et al. (Table 3.3)(Maas et al., 2008).

All studies provided intervention that involved feedback with visual cues, although none of the studies described whether or not they used verbal cues and/or explanations during treatment (Table 3.3).

Table 3. 3 Description of biofeedback swallowing intervention based on the framework of Maas et al. 2008.

Primary Author	Practice Amount	Practice distribution	Practice variability	Feedback Modality	Type of feedback	Focus of feedback	Feedback schedule	Target complexity	Feedback frequency	Feedback timing
Athukorala et al. ¹⁶	10 therapy sessions. 1 session= 1 h 1 session = 100 trials (5 blocks of 20 swallows followed by break).	5 days per week for 2 weeks	Variable practice: increasing difficulties based on patient's performance during saliva swallowing	Visual feedback using sEMG	Feedback on the results	External focus on the target to reach (Hit the box)	Random real-time feedback: the target for the upcoming trial was not predictable to the patients	Complex Movements involving coordination and precision	After 3 consecutive swallows.	Immediate feedback
Da Silva ¹⁷	18 therapy sessions 1 session= 1h	15 sessions for 3 days per week. The last 3 once per week. 8 weeks in total	Variable practice: Effortful swallows, Mendelsohn Maneuver	Visual feedback using sEMG	Feedback on the performance	Internal focus on aspects of swallowing movements	Constant real-time visual feedback	Simple Movements	N/A	Immediate feedback
Felix et al. ¹⁸	10 therapy sessions 1 session:8 swallows trials	5 days per week for 2 weeks	Variable practice: 8 swallows with different food (starting with saliva and ending with biscuits)	Visual Feedback using manometer	Feedback on the results	External focus on the strength values of manometer	Constant Visual Real-Time feedback	Complex swallowing tasks	N/A	Immediate feedback
Manor et al. ¹⁹	6 therapy sessions 1 sessions: 30 minutes	5 therapy sessions during 2 weeks. The last 6 th session after 4weeks	Constant practice on compensatory swallowing maneuvers	Visual Feedback using FEES	Feedback on the performance	Internal focus on the FEES video	Off-line feedback	Simple task on compensatory swallowing technique	N/A	N/A

3.4.3.2 Timing

Information on feedback schedule, timing and frequency were not explicitly stated in any of the studies but it could be inferred that the feedback was immediate and constant in Felix et al. and Da Silva's studies, as participants watched the screen where visual cues were displayed continuously. Only Athukorala et al. reported that the biofeedback schedule was random and unable to be predicted by participants and frequency of feedback was after three consecutive swallows (Athukorala et al., 2014). Visual feedback was immediate and in real-time in three studies (Athukorala et al., 2014; Da Silva A., 2016; Felix et al., 2008) but was not simultaneous with swallowing in Manor et al. (Manor et al., 2013)

3.4.3.3 Treatment Dosage, Fidelity and Follow-up

Interventions in the included studies was delivered over two weeks in three studies (Athukorala et al., 2014; Da Silva A., 2016; Felix et al., 2008) with a longer duration (3 months) in the Da Silva study (Table 3.3). Each single session lasted one 1h, apart from Manor et al., where the session lasted 30 minutes. The amount of therapy was greatest in Athukorala et al. (100 swallowing trials per session). No information on the amount of practice were given in Da Silva and Manor et al. In Manor et al., intervention and control groups had 5 therapeutic sessions overall delivered over a 2-week period and a follow up (6th) session 4 weeks later with treatment distributed over a longer period of time than the other studies (Table 3.3). In Da Silva's study each participant received 18 therapy sessions. The entire intervention programme lasted 8 weeks. In contrast, intervention was concentrated over a shorter period of time in both Athukorala et al. and in Felix et al. studies. In Athukorala et al., each participant received 10 (1hour) skill training therapy sessions over a 2-week period and then follow up 2-week later.

None of the studies reported the assessment of fidelity of the treatment. Nevertheless, the use of sEMG in Athukorala et al. and Da Silva allowed to monitor the number of swallowing exercises and number of sessions. Hence, if these data are

saved in the system and computer, the fidelity of the treatment could be retrospectively verified.

Follow-up assessment was carried out only by Manor et al. 2 (6 months post-treatment) and by Athukorala et al. (two weeks post-treatment). At follow up, Manor et al. assessed only quality of life using the Swallowing Quality Of Life (SWAL-QOL)(McHorney et al., 2002) while Athukorala et al. completed all the outcome assessments again (Athukorala et al., 2014). No study examined outcomes greater than six months.

3.4.3.4 Swallowing Outcomes

Swallowing outcomes and outcome measures varied across all four studies. Manor et al. examined bolus flow time, bolus residue, penetration and aspiration of pureed, solid and liquid using FEES (Manor et al., 2013). Outcome measures for FEES were completed pre-treatment and post treatment using non validated clinical outcome scales. Residue outcome measures on FEES showed a statistically significant reduction in food residue in the intervention group (Table 3.3). Da Silva assessed the swallowing safety and pharyngeal residue through VFS using the Dysphagia Outcome and Severity Scale (DOSS) (O'Neil et al. 1999), Eisenhuber scale (Eisenhuber et al., 2002) and the FOIS (Craig et al. 2004). Results did not show a statistically significant difference between control and intervention groups on the DOSS and FOIS scales, but no statistical analysis was carried out for Eisenhuber scale. A statistically significant improvement was reported on the DOSS and FOIS scales for both control and experimental groups after treatment.

Athukorala et al. examined timing of swallowing on liquids and solids using timed water swallow test (TWST) (Hughes & Wiles, 1996) and Test of Mastication and Swallowing Solids (TOMASS) with sEMG (Huckabee et al., 2018). Athukorala et al. conducted two baseline assessment sessions and two post-treatment assessment sessions two weeks apart in order to evaluate stability of measurements, post-treatment effect and retention of effect (Athukorala et al., 2014). They found after intervention that swallowing rate for liquids using TWST decreased significantly

($P=.034$), sEMG durational parameters of premotor time also changed ($P=.003$), and pre-swallow time significantly improved ($P<.001$). A strong effect was seen from dry to water swallows ($P=.009$).

Felix et al. examined safety of swallowing, carrying out a subjective clinical evaluation of cough and wet voice quality during the swallowing of water, yogurt and biscuit. At the end of the intervention, they recorded a statistically significant reduction in the number of cough episodes and improvement in quality of voice. Quality of life was assessed in all the studies except for Felix et al. and the SWAL-QOL assessment was consistently used in the other three studies (Athukorala et al., 2014; Da Silva A., 2016; Manor et al., 2013). There was an increase in quality of life reported for participants in all the three studies (Table 3.2). Manor et al. , examined also swallowing quality of care (SWAL-CARE) (McHorney et al., 2002) and a non-validated “Pleasure of Eating” (POE) scale.

3.4.3.5 Adverse events

No included study examined or reported adverse events associated with the intervention. Three participants dropped out in Da Silva’s study; one patient had another associated disease and two did not conclude the programme. The reasons for dropout are not provided (Da Silva A., 2016).

3.4.3.6 Risk of bias in included studies

The methodological quality of the included studies was examined by the research student and colleague with the PhD supervisor using the Down’s and Black checklist tool (Downs & Black, 1998). The overall quality of all the included studies was considered “moderate quality”, with an average score of 10. (Table 3.4). Low ratings were generally associated with study design and the high risk of bias associated with design and implementation of the studies.

Table 3. 4 Down’s and Black checklist rating.

STUDY	RATING	DESCRIPTOR
Manor et al.	17	Good Quality
Da Silva et al.	12	Moderate Quality
Athukorala et al.	12	Good Quality
Felix et al.	4	Poor Quality

3.5 Discussion

This review aimed to evaluate the effectiveness and safety of biofeedback used as an augmentative intervention for the recovery of swallowing function in individuals with IPD and to develop a theoretical basis for its use within a swallowing treatment programme for people IPD and dysphagia. Most of the included studies were feasibility and pilot NRCT studies. This is appropriate as Phase 1 and 2 studies (Robey, 2004) exploring how a complex intervention such as biofeedback for people with IPD works. It was anticipated that an increased understanding on how this intervention might work to improve swallowing in people with IPD and dysphagia would be obtained. While the considerable heterogeneity within studies in terms of type of biofeedback, study design, outcome measures used make it difficult to draw firm conclusions and formulate a robust theory, it is argued that these review findings have some value. For example, there were changes in pharyngeal residue on instrumental assessment for two studies (Da Silva A., 2016; Felix et al., 2008). It could be hypothesised that also swallowing safety and efficiency improved in Athukorala et al. (2014) and Felix et al. (2008) but instrumental assessment is needed to confirm this (Felix et al., 2008). This would help inform the methods for the feasibility study (Chapter 5).

Change in quality of life across all studies is important given that it was the only consistent outcome measure across studies and quality of life did increase after biofeedback swallowing treatment. This might suggest that either changes in other aspects of swallow function were not detected with the outcome measures used or

that the intervention itself provided some psychological benefit to participants. All studies used the same assessment (the SWAL-QOL) and this assessment will also be considered in the feasibility study (Chapter 5).

The positive impact on swallowing in persons with IPD could be attributed to the fact that swallowing treatment with biofeedback embraces most of the principles of motor learning, but this requires systematic investigation and will be considered in the feasibility study (Chapter 5).

3.5.1 Biofeedback Intervention: skill versus strength

In this review two main categories of swallowing therapy programme were implemented. One focused on increased strength of swallowing musculature and the other aimed to improve the skill involved in swallowing (Table 3.2). Both approaches showed some improvement in swallowing.

Biofeedback during *skill swallowing tasks* showed an improvement in swallowing rate for liquids and solids in Athukorala et al. 2014 and in reducing pharyngeal residue in Manor et al. 2013 suggesting that visual swallowing biofeedback in IPD is beneficial for coordination and skill of swallowing (Athukorala et al., 2014; Manor et al., 2013). Although these assumptions are limited by the methodology of the studies, similar results are found in physiotherapy treatments for IPD, in which the main application of biofeedback focus on coordination during walking to increase balance and reduce freezing of gait. While *strengthening exercises* with biofeedback also resulted in improvement in swallow function in two included studies, it is argued elsewhere that strengthening exercises are not always beneficial to patients with dysphagia, particularly if they are not 'weak' (Huckabee & Steele, 2006). In the literature, it is recognized that strength and effortful exercise seem to be more appropriate in dysphagia associated with weakness or sarcopenia due to the hypotonia of oropharyngeal structures (Walshe, 2019).

3.6 Summary

This chapter provides the evidence of the use of sEMG for swallowing recovery in IPD population and it allows the creation of a theory underpinning the implementation of biofeedback swallowing treatment in IPD individuals with dysphagia. This is considered in Chapter 4.

Chapter 4. Theoretical Framework

4.1 Introduction

The previous chapters provide the background on the potential for biofeedback in IPD people and specifically its use as an augmentative tool for swallowing treatment in this population. This allows the creation of a theory defining the key elements in order to develop a feasibility study on the use of sEMG biofeedback swallowing in IPD people with dysphagia. In this chapter the theoretical framework is provided and the research questions are formulated at the end of this chapter.

4.2 Theoretical framework

The theoretical framework described here includes the fundamental factors underpinning the implementation of a feasibility study. This integrates the hypothesis regarding the need to develop a swallowing treatment in IPD people; the type of outcomes and assessments needed; including the assessment of acceptability and fidelity of the intervention, and the use of sEMG biofeedback treatment (Figure 4.1)

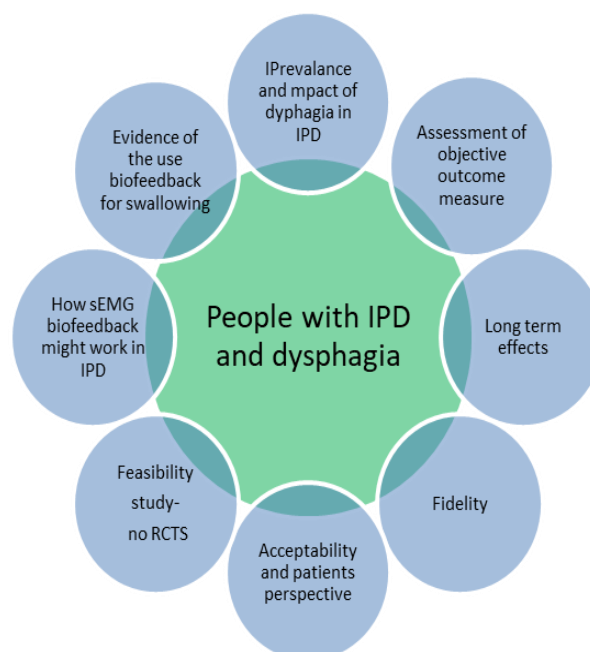


Figure 4. 1 Components to build theoretical framework

The impact of swallowing disorders in IPD was described in the first chapter. Despite the **high prevalence of dysphagia** and the severe clinical sequelae of dysphagia in individuals with IPD, few studies have documented the effects of swallowing interventions in individuals with IPD. Studies on the efficacy and effectiveness of swallowing rehabilitation in IPD are limited and lack methodological quality (Deane et al., 2001; van Hooren et al., 2014). The hypothesis of this researcher is that dysphagia has a significant impact on the person with IPD and new approaches to treatment must be rigorously tested to improve the impact on the person and their families

The evidence for the use of swallowing biofeedback in IPD has been revealed by previous literature reviews already discussed. These show that biofeedback during skill swallowing tasks had positive effects not only on the quality of life of the person with IPD and dysphagia but also on swallowing liquids and solids (Athukorala et al., 2014) and in reducing pharyngeal residue (Manor et al., 2013). In addition, evidence for using biofeedback for swallowing in patients with neurological disease had already been demonstrated, as described in Chapter 2 (Crary et al., 2004; Huckabee & Burnip, 2018; Huckabee & Macrae, 2014; Humbert & German, 2013). Furthermore, it is well accepted that IPD people benefit more from the use of biofeedback treatment than other neurological populations during motor learning tasks, as it has been explained in Chapter 2.

The argument for using sEMG biofeedback as a focus in this study was motivated by the findings of the systematic review (Chapter 3) in which the majority of studies used this modality for providing biofeedback in this population but studies were methodologically weak and no conclusive evidence on its usefulness is available. Nevertheless, the researcher hypothesises that this approach that augments intervention could be particularly appropriate for IPD people and it is worthy of further investigation. In contrast to other methods of biofeedback, sEMG allows to visualise the line of activation of submental muscles. Wheeler-Hegland et al. (2008) found a positive correlation between the value of the maximum peak of the sEMG waveform and the degree of maximum hyoid excursion as measured with VFS (Wheeler-Hegland et al., 2008). Recently, Albuquerque et al. (2019) confirmed that

sEMG applied to the submental muscles yielded positive effects for providing biofeedback for hyolaryngeal elevation (Albuquerque et al., 2019). Based on this research, it is hypothesised that biofeedback as part of an intervention programme facilitates awareness and control of swallowing function. sEMG biofeedback integrated into a carefully devised swallowing rehabilitation programme could be specifically effective for IPD and could provide an important change in swallowing function and quality of life in this population.

Nevertheless, the mechanism underpinning how sEMG biofeedback works for swallowing recovery in IPD is unclear (Fig 4.1). The systematic review in chapter 3 suggests that visual feedback is important and it is also this researcher's theory that a behavioural intervention such as this must include the principles of neural plasticity and motor programming.

The theoretical framework in this study further builds on the findings from the literature review (Chapter 3) that indicate a major shortcoming of previous studies which is the lack of instrumental assessment. Future studies in dysphagia rehabilitation in IPD must include **instrumental swallowing assessments with valid outcome measures**. These must include reported outcome measures and a validated quality of life scale, and some measure of the **acceptability of the treatment from the patient perspective**. A treatment may be highly effective, but if it is not acceptable to patients then it is of limited use. In literature, several studies confirmed that patient perspective is of fundamental importance during feasibility. It allows to verify whether an intervention is relevant and acceptable and could be implemented in real-world contexts (Turner et al., 2019).

In addition, as seen from the literature review, the long term effects of the intervention are important. If much time and effort has been spent on an intervention approach, it is important that return to pre-intervention baseline is not immediate. Therefore, it is part of the theoretical framework that **follow-up assessments** for a realistic time period (minimum 3 months) should be incorporated in research studies on interventions for dysphagia in IPD. Finally, few studies investigate adverse events

and it is hypothesised that these can exist but are not always reported. Adverse events include a deterioration in swallowing or a negative impact on quality of life and should be an integral part of any intervention on dysphagia in IPD. The absence of detail on timing of medication, reason for drop out in these studies included in the systematic review (Chapter 3), hampered the our ability to understand the mechanisms by which biofeedback worked. In addition, risk of bias was high across all studies and efforts to decrease the risk of bias with blinding of outcome assessors as a minimum should be implemented in the future feasibility study.

A further component of the theoretical framework that underpins this study is that fidelity to treatment and reporting of that fidelity is important. Cattaneo et al. found that the majority of interventions for dysphagia in IPD people did not report the **fidelity** of treatment (Cattaneo C, 2020). This threatens the validity and reliability of the findings if adherence to the study protocol, the intervention approach and methods of evaluation are not consistent. Fidelity and adherence of treatment enhances the validity and reliability of behavioural interventions (Krekeler et al., 2020). For this reason, the research assumes that assessment of fidelity plays fundamental role in the construction phase of feasibility studies.

Pring argues that much research is wasted in speech and language therapy because RCTs have been completed when the theory about how the intervention works, who it works best with, and why it works are not fully investigated (Pring, 2004). This is the case with sEMG and IPD and dysphagia. Therefore, a further hypothesis here is that a feasibility study that may ultimately lead to a pilot study and a RCT are required first. **Feasibility studies** provide a better understanding on the intervention itself and how the intervention has to be implemented. The results of this feasibility study provide the basis of a future study.

The aims of the study have been described earlier (Chapter 1). The focused research questions are now considered.

4.3 The research questions

This feasibility study aims to answer the following research questions:

(1) In people with dysphagia and IPD, does a swallowing intervention using sEMG as biofeedback change any of the following parameters when measured with instrumental evaluations and validated tools:

a) penetration and /or aspiration

b) pharyngeal residue

It is hypothesised that this intervention will result in an improvement with reduction in laryngeal penetration and/or aspiration events and decreased pharyngeal residue.

(2) Does a specific swallowing intervention using sEMG biofeedback improve the method of oral intake in people with IPD and dysphagia?

The hypothesis is that the intervention will result in changes to swallowing ability and functional oral intake.

(3) Does a specific sEMG biofeedback swallowing intervention reduce the self-perception of drooling in people with dysphagia associated with IPD?

It is hypothesised that this intervention will have an impact on the self-perception of drooling.

(4) What is the impact of this biofeedback swallowing intervention on a person with IPD's overall quality of life?

The hypothesis is that improvement in swallowing skills improves the quality of life of people with IPD.

(5) If swallow function changes after biofeedback swallowing intervention in people with IPD and dysphagia, is this change in swallow function maintained post treatment?

The hypothesis is that improvement in swallowing following this intervention in a person with IPD who is medically stable does not significantly decline at 3 months follow-up.

(6) What were the adverse events, if any, associated with the biofeedback sEMG swallowing intervention in people with dysphagia and IPD? This is exploratory and therefore no hypothesis is formulated.

(7) Is sEMG biofeedback treatment acceptable to people with IPD and dysphagia?

This is exploratory and no hypothesis is formulated

Chapter 5: Methodological Approach

5.1 Introduction

This chapter describes the research methods applied to this feasibility study with a rationale for this approach.

5.2 Feasibility study

As stated earlier, feasibility studies are fundamental to investigate acceptability of the intervention to participants, adherence to the treatment protocol, delivery of the intervention by researchers, recruitment of eligible participants and retention of positive outcomes for participants (Craig, 2019) (figure 5.1) .

Bleijenberg et al. describe the fundamental steps that should be considered when developing a feasibility study as a part of complex intervention study (Bleijenberg et al., 2018). These include: 1) determine the needs, 2) examine the context (Fig 5.1).

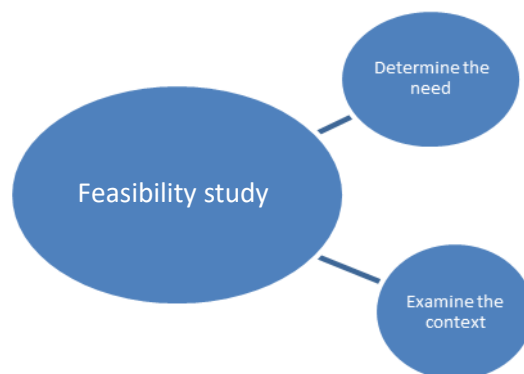


Figure 5. 1: The components for developing a feasibility study (Bleijenberg et al. 2018)

5.2.1. Determine the need

The concept of “Determine the need” involves the assessment of the demands, perceptions and capacities of the beneficiaries (participants) as well as the providers (researchers). Exploring the needs from different perspectives enhances the external validity, which is fundamental in clinical research with IPD population. In the literature, there is strong evidence on the impact of swallowing interventions in IPD people in the

reduction of malnutrition, dehydration and pulmonary infections (Ayres et al., 2016; Deane et al., 2001; van Hooren et al., 2014). On the other hand, it is established that people with IPD and dysphagia may not be aware of their swallowing difficulties and tend to underestimate their issues (Buhmann, Flugel, et al., 2019; Kalf, de Swart, et al., 2012). The researcher carried out some exploratory work with the Venetian section of Italian Parkinson Disease Association and explained the research concept to 50 people with IPD and their caregivers. There was interest and enthusiasm on the intervention concept and confirmed the importance of swallowing treatment for people with IPD supporting clinical experience and evidence from the literature.

5.2.2 Examination of the context

The analysis of the practice aims to investigate the context in which the intervention will be implemented (Bleijenberg et al., 2018). Feedback from people with IPD gives an important contribution to identifying barriers and facilitators in the implementation of the project (Turner et al. 2019). One of the main challenges was access of participants to the hospital for therapy. The context of the research is in the lagoon of Venice, in which there are two main hospitals. Both hospitals were potential sites for data collection. Based on the suggestions from the members of the Parkinson's Association, the Angel's Hospital was selected as the research site, as it is accessible by train, bus and private cars and is the main hospital for the Venetian province. The researcher is employed by Italian National Health System and could deliver the treatment at Angel's Hospital. Having decided that this would be a feasibility study, conducted in a large central hospital with easy access for participants, the study design was selected.

5.2.3 Study Design

Observational studies are an appropriate approach for feasibility studies as they provide a broad understanding of determining factors (Horn et al. 2012). Although observational studies are not considered robust methodologically, and they are susceptible to treatment selection bias and confounding variables, they are considered appropriate in preliminary rehabilitation research (Sedgwick, 2015).

Within-subject design is an observational study design in which individuals act as their own control. In rehabilitation interventions, this approach could be particularly indicated as it is well accepted that some participants recover better than others regardless of the same treatment. This method is recognised to show more power in detecting an effect of the independent variable than case-control studies (Horn et al., 2012).

As a result, the study is defined as a **within-subject prospective feasibility-pilot-study**. However, as the study also sets out to examine the acceptability of the intervention to participants, a qualitative approach is also required. Thus, this study is a mixed methods study with quantitative and qualitative components. The materials for conducting the study are considered next.

5.3 Materials

The research tools used in the assessment are described here.

5.3.1 Clinical non-instrumental swallowing assessments

In this study, the main clinical non-instrumental swallowing assessments were the assessment of quality of life; measurement of oral intake scale and measurement of drooling. The choice of tests and scales for clinical non-instrumental assessments in IPD is challenging. As explained in Chapter 1, people with IPD can underestimate dysphagia symptoms. Nevertheless, patient-reported outcome measures contribute significantly to swallowing-related symptoms such as drooling and psychological well-being. In addition, there is a paucity of translated and validated dysphagia assessment scales in Italian. Given that the research project was set in Italy and most dysphagia assessments are validated in English, the research student faced difficulties in finding reliable assessments for dysphagia translated and validated into Italian.

After an accurate analysis of available swallowing assessment scales, the research student selected the following outcome measures: SWAL-QOL (McHorney et al., 2002), FOIS (Crary et al., 2005) and ROMP-Saliva (Kalf et al., 2011) in order to assess the following outcomes.

5.3.1.1 Quality of Life

The SWAL-QOL questionnaire (McHorney et al., 2002) is a valid and reliable scale for measuring the satisfaction and well-being of people with IPD and swallowing impairments. The scale is validated into Italian. Ginocchio et al. 2016 completed a cross-cultural translation and validation (Ginocchio et al., 2016). I-SWAL-QOL is a 44 item tool that takes on average 15 min to complete and assesses 11 different sub-topics of quality of life; nine of them are dysphagia-related quality of life (food selection, burden, symptom frequency, mental health, social functioning, fear, eating duration, eating desire, communication) and two concern general quality of life concepts (sleep and fatigue). Each item is given a score from 0 to 4 (worst-best). Scoring in each domain is calculated by the sum of scores for each item expressed as a percentage of the maximum possible domain score. A total I-SWAL-QOL score is derived by summing each domain score and dividing by 10 giving a total I-SWAL-QOL score that ranges between 0 and 100 (worst–best).

5.3.1.2 Oral Intake

To measure change in the oral intake of following intervention the **FOIS** (Table 5.1) (Crary et al., 2005) was selected. The FOIS (Crary et al., 2005) is a valid reliable scale to record changes in oral intake (Table 5.1), which was initially validated on stroke population but it was adopted also in different population with swallowing disorders. As there was not an Italian version of the FOIS, the research student completed the cross-cultural translation and validation of the scale (Chapter 6) .

Table 5. 1 Functional Oral Intake Scale

Functional Oral Intake Scale (FOIS)	
TUBE DEPENDENT (levels 1-3)	
1	No oral intake
2	Tube dependent with minimal/inconsistent oral intake
3	Tube dependent with consistent oral intake of food or liquid.
TOTAL ORAL INTAKE (levels 4-7)	
4	Total oral intake of a single consistency
5	Total oral intake of multiple consistencies requiring special preparation
6	Total oral intake with no special preparation, but must avoid specific foods or liquid items
7	Total oral intake with no restrictions

5.3.1.3 Saliva Control

The **ROMT-saliva questionnaire** (Kalf et al., 2011) is part of an assessment of dysarthria and dysphagia symptoms in IPD. It was specifically designed to assess the subjective perception and impact of saliva in daily life activities in people with IPD. The questionnaire consists of 9 items; each is given a score from 1 to 5 (normal-deficit) and the score range is from 9 to 45 (Appendix B).

5.3.2 Clinical Instrumental Swallowing Assessments

The instrumental swallowing assessment chosen was Fiberoptic Endoscopic Evaluation of Swallowing (FEES). It is acknowledged that the VFS is more appropriate assessment than FEES for measuring the swallowing kinematics such as in order to explain improvements in swallowing safety and efficiency. Nevertheless, in Italy, few hospitals have dedicated VFS examinations and this was not available to the research student. However, FEES has established reliability and validity for detection of penetration, aspiration and residue which are main outcomes selected for this study. Several studies have confirmed its validity in assessing the presence of pharyngeal residue and swallowing safety in people with IPD (Leder & Murray, 2008; Pisegna et al., 2018; Pisegna et al., 2020; Warnecke et al., 2016). Two outcome measures were collected

during FEES: Penetration - Aspiration Scale (PAS) (Rosenbek et al., 1996) (Table 5.2) and the Yale Pharyngeal Residue Severity Rating Scale (YPR-SRS)(Neubauer et al., 2015) (Table 5.3). The PAS is recognised valid and reliable method to measure the severity of airway invasion during instrumental examination (Curtis et al., 2020a; Rosenbek et al., 1996; Steele & Grace-Martin, 2017). In this study an Olympus ENF-V2 Ultra slim rhinolaryngo videoscope was used with an Olympus OTV-SI compact video enabled digital processor and ADVAN 21” monitor (Olympus Corporation, Tokyo, Japan).

Table 5. 2 Penetration Aspiration Scale (PAS).

<i>Penetration Aspiration Scale (PAS)</i>
1. Material does not enter the airway
2. Material enters the airway, remains above the vocal folds, and is ejected from the airway
<u><i>Penetration</i></u>
3. Material enters the airway, remains above the vocal folds, and is not ejected from the airway
4. Material enters the airway, contacts the vocal folds, and is ejected from the airway
5. Material enters the airway, contacts the vocal folds, and is not ejected from the airway
<u><i>Aspiration</i></u>
6. Material enters the airway, passes below the vocal folds, and is ejected into the larynx or out of the airway
7. Material enters the airway, passes below the vocal folds, and is not ejected from the trachea despite effort
8. Material enters the airway, passes below the vocal folds, and no effort is made to eject

In order to measure the severity of pharyngeal residue, the Yale Pharyngeal Residue Severity Rating Scale (YPR-SRS) (Neubauer et al., 2015) was selected. The Italian version of this scale was validated by (Nordio et al., 2018).

Table 5. 3 Yale Pharyngeal Residue Severity Rating Scale on the vallecular and pyriform residue.

Valleculae			
I	None	0 %	No residue
II	Trace	1-5 %	Trace Coating of the mucosa
III	Mild	5-25 %	Up wall to quarter full
IV	Moderate	25-50 %	Up wall to half full
V	Severe	> 50 %	Filled to aryepiglottic fold
Pyriform			
I	None	0 %	No residue
II	Trace	1-5 %	Trace Coating of the mucosa
III	Mild	5-25 %	Epiglottic ligament visible
IV	Moderate	25-50 %	Epiglottic ligament covered
V	Severe	> 50 %	Filled to epiglottic rime

The FEES assessment compromised the administration food. The instructions were:- "Try to eat or drink as conformable as you can, without rush". Firstly it was administered two spoons of yogurt or applesauce (IDDSI 3), the participants was invited to take the food autonomously, if he needed he/she was helped during food administration. Secondly, the participant had to drink from the glass, it was helped if she/he could not hold the glass. In this case, it was not possible to measure the bolus volume as it depended on participant volume sip. Thirdly, it was administered a piece of cracker (IDDSI 7), the size of cracker were the same of the TOMASS (Huckabee et al., 2018).

5.3.3 Qualitative Feedback Instruments

This involved two open questions in a specifically devised questionnaire. There were two parts to this questionnaire. The first consisted of two open questions, posed by the research student at the end of each treatment session:

(1) *"How do you feel after this treatment session?"*

(2) *"What are your thoughts, feelings and feedback about this treatment?"*

The second part was qualitative methods was 4 point rating scale and an open comment section. A nurse gave the form at the follow-up assessment (T3) (Table 5.4).

Table 5. 4 Feedback form

1) How do you rate the sEMG procedure?	1	2	3	4
	_____	_____	_____	_____
	Very uncomfortable	Uncomfortable	Comfortable	Very comfortable
2) Would you like to recommend any changes?	_____			

5.4 sEMG Intervention tool

The sEMG is a non-invasive procedure with widespread potential application for the management of dysphagia (Albuquerque et al., 2019; Archer et al., 2020). It consists of an electromyogram, which records the electrical activity from a muscle or group of muscles through surface electrodes applied to the skin. The sEMG signal detects muscle activity under the electrode field.

For this project, three electrodes were used. Two active electrodes were placed on the submental muscles to detect the anterior movements of the hyo-laryngeal elevation (Stepp, 2012). The third electrode was the ground or reference electrode, allowing to reject the noise from the signals. The characterises of sEMG swallow pattern is a sharp upward slope from resting baseline, a recognizable peak, and a sharp downward slope. Several authors describe the swallow shape like a wave-line (Figure 5.2) (Crary et al., 2004; Huckabee & Steele, 2006; Stepp, 2012).

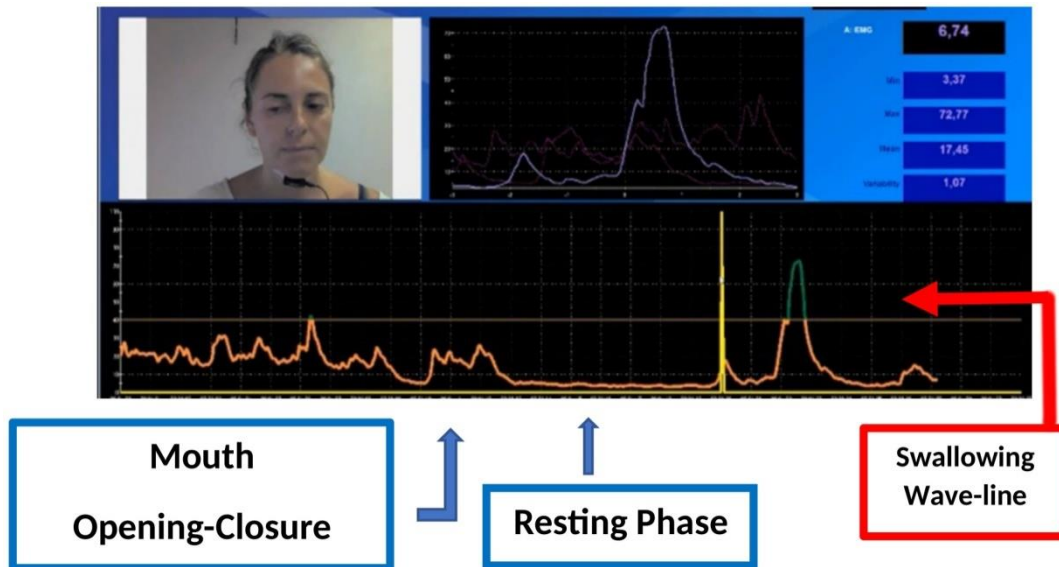


Figure 5. 2 The sEMG wave of muscles contraction during swallowing.

The main limitation of the application of sEMG in the submental muscles is that the activation signal could be triggered also during non-swallowing tasks such as mouth opening (Azola et al., 2015; Vose et al., 2019). This was controlled by the student which monitored participants during the intervention.

The choice of the type of sEMG equipment was determined by the following criteria:

- 1) The master-units must be compact and portable to facilitate treatment
- 2) The EMG signals should be clear and visible to participants and the format should be adjustable according to the need of participants.
- 3) Master-unit should include wifi or Bluetooth in order to facilitate the transmission of data from the intervention trials.
- 4) Electrode cables must be limited in order to allow ease of use
- 5) The software should include pre-set game programmes.
- 6) Surface electrodes must be small size and reusable if possible although with single patient use. If re-used the signal accuracy had to be checked each time. Equipment should be affordable to increase clinical utility if effective

The NeuroTrac® MyoPlusPro (Figure 5.3) was chosen as the sEMG equipment

The MyoPlus Pro device is small (length 18 cm; high 10.2cm and width 3.5 cm) and light weight (0.16 Kg) (Figure 5.3). It incorporates a small display which shows the programmes. It also includes Bluetooth which enables to transfer the data to the computer. The device is combined with the Neuro-Trac Software, which must be installed in the computer.



Figure 5. 3: The NeuroTrac® MyoPlus sEMG device.

Neuro-Trac Software was installed on a Dell, Latitude E6420 laptop computer. The software comprises several different game programmes as well as the sEMG raw data. For the purpose of this study, two programmes were selected : “open display” and “plane game”. The programme “open display” shows the wave line of activation. The bar on the left indicates the sEMG Range (from 0.2 to 2000 μV RMS continuous). The sEMG range could be calibrated base on participant performance (Fig 5.4).

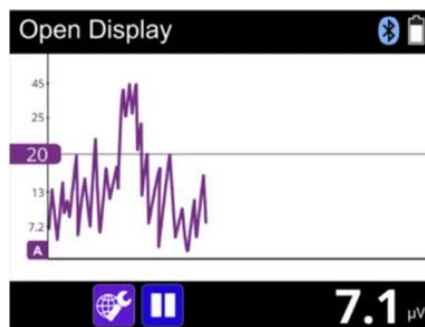


Figure 5. 4 The “Open display” sEMG modality.

The “plane game” (Fig 5.5) is work/rest type of sEMG training. The aim is to move up the plane to collect as many stars as possible by coordinating swallowing and increasing the activity of the submental muscles. Participants must contract the muscles for the aeroplane to go up the mountain and then relax below the lower

threshold for the aeroplane to descend. The choice of this exercise was motivated by three factors. Firstly, the motor learning principle of practice variability assumes that participants must be able to extract and manipulate movement patterns into different situations (Huckabee & Burnip, 2018). During this game, participants did not see the wave line but the plane. So, they were required to control the submental muscle contraction during swallowing in order to move the plane. Secondly, in this game participants had to swallow at a specific time, which was scheduled every 60 seconds. This was set in order to increase the frequency of swallowing. Finally, the game aims to motivate participants to adhere to the therapy.



Figure 5. 5 the “Plane Game” sEMG modality.

The surface electrodes were small (30mm diameter), allowing attachment them on the submental muscles with a distance of 1 cm between (5.6). The ground electrode was attached separately on the cheek. Two cables (red and black) connected the surface electrodes with the work-unit (5.3).

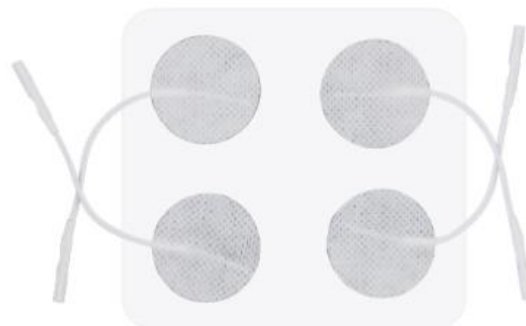


Figure 5. 6 Electrodes used for swallowing sEMG biofeedback treatment.

5.5 Ethical Considerations

This study received ethical approval from two committees. Firstly, the Joint Research Committee of the Faculty of Health Sciences Ethics Committee of Trinity College Dublin (Appendix D). This ethical approval was granted by the Joint Research Committee of the Faculty of Health Sciences Ethics Committee of Trinity College Dublin held on the 14th March 2018 (Ref: 180304) (Appendix D). Secondly, from the Italian Joint Research Committee of the Health Care System of Venice Province, Italy (Comitato Etico per la sperimentazione clinica della provincia di Venezia, AULSS 3, Serenissima) (Appendix E). The Italian Joint Research Committee of the Health Care System of Venice Province, Italy (Comitato Etico per la sperimentazione clinica della provincia di Venezia, AULSS 3, Serenissima) hold on the 13th September 2018 (Ref: 130918) approved the completion of the study (Appendix E).

5.6 Participants

Participants in this study were people with IPD and dysphagia. Inclusion criteria was as follows:

- a) The diagnosis of IPD, confirmed by neurologist following new International Parkinson Disease and Movement Disorder Society diagnostic criteria (Berg et al., 2015).
- b) Clinical stability as evaluated by a neurologist. Anti-parkinsonian medication therapy must be consistent throughout the duration of the study.
- c) Oropharyngeal dysphagia confirmed by instrumental examination (FEES). With scores higher than 2 on PAS (Rosenbek et al., 1996).
- d) An ability to provide autonomous consent to participate at this study.
- e) Absence of parkinsonism secondary to causes other than IPD, such as diagnosis of progressive supranuclear palsy (PSP), multiple system atrophy (MSA), cortico-basal degeneration (CBD), and vascular Parkinsonism (VaP).
- f) Absence of history of stroke or transient ischemic attack.
- g) Absence of severity dysphagic participants fed via percutaneous endoscopic gastrostomy (PEG) and with a FOIS-It (Battel et al., 2018) scores > 4 were

excluded. The programme of treatment involved the administration of food bolus which could compromise the safety of severity participants.

- h) Absence of dysphagia caused by pathologies other than IPD.
- i) Absence of cognitive impairments (MoCa score: 25 >) (Dalrymple-Alford et al., 2010).
- j) Absence of Deep Brain Stimulation implant.
- k) Absence of facial hair that will impede sEMG electrodes placement.

5.7 Sampling Method

Non probability sampling was employed.

The sampling pool was the Department of Neurology of Angel Hospital. The local clinical registry has estimated the presence of 2000 known cases of IPD in Venice Province. People with IPD are monitored and visited every 6 months by a team of neurologists who are specialist in PD.

5.7.1 Sample Size

Bell and Whitehead argue the importance of calculating the size of sample in feasibility studies because it increases the preliminary efficacy evidence (Bell et al., 2018). Nevertheless, in this feasibility study, it was not possible to calculate the sample size.

5.8 Recruitment

Participants with IPD were screened for eligibility by neurologists from September 2018 to February 2019. The team of neurologists at the research site oversaw the selection of potential candidates for the study based on the 2° sub part of the MD-UPDRS)(Goetz et al., 2007) as it has shown to be highly specific (94% specificity in screening swallowing disorders in people with IPD (Nienstedt et al., 2019) and there is a validated Italian version of the test. The criteria for selection was mild level (score 2 >) of sections 2.2 and 2.3 of the MD-UPDRS.

The neurologists provided the information leaflet (Appendix G, F). In addition, they organised the FEES appointment in collaboration with the ENT specialists in the otolaryngology department. This constituted usual procedure at the research site for

people at risk of dysphagia.

Participants interested in participating, contacted the student who arranged the first appointment in order to explain the research project, the protocol of assessment and treatment. During the first meeting, the student screened cognitive functions with MOCA test (Dalrymple-Alford et al., 2010). Participants with a score less than 26 were excluded from the study (Table 5.5).

Table 5. 5 Score level of MoCA and the cognitive severity level.

MOCA SCORES	SEVERITY LEVEL
26 >	Absence of cognitive impairments
18-26	Mild cognitive impairments
10-17	Moderate cognitive impairments
> 10	Severe cognitive impairments

The people with IPD who consented to participate, underwent a FEES assessment to determine the presence of swallowing impairments. Participants who presented with signs of dysphagia on FEES completed the whole assessment protocol. Finally, participants who met the inclusion criteria and gave written consent (Appendix H) were included in the study (Fig 5.7). The phases of implementation of the protocol of assessment run from January 2019 to October 2019.

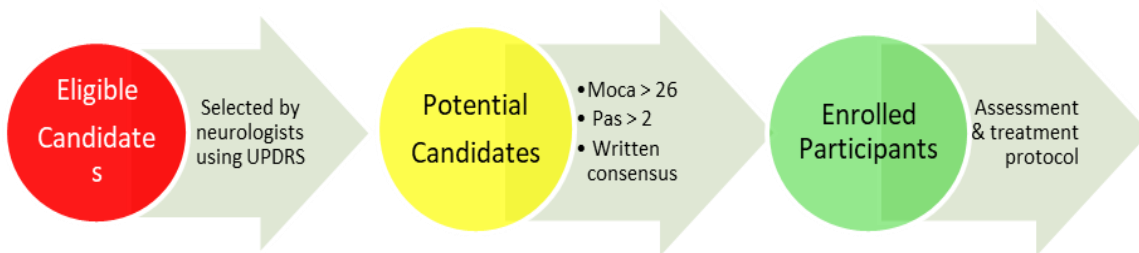


Figure 5. 7 Flow chart of recruitment procedure.

From October 2018 to February 2019, parkinsonian neurologist team screened 128 participants. Thirty-one people with IPD were selected as eligible participants. From

these only 12 met the inclusion criteria and they were enrolled (Fig 5.8) and two participants dropped out at the first week of treatment.

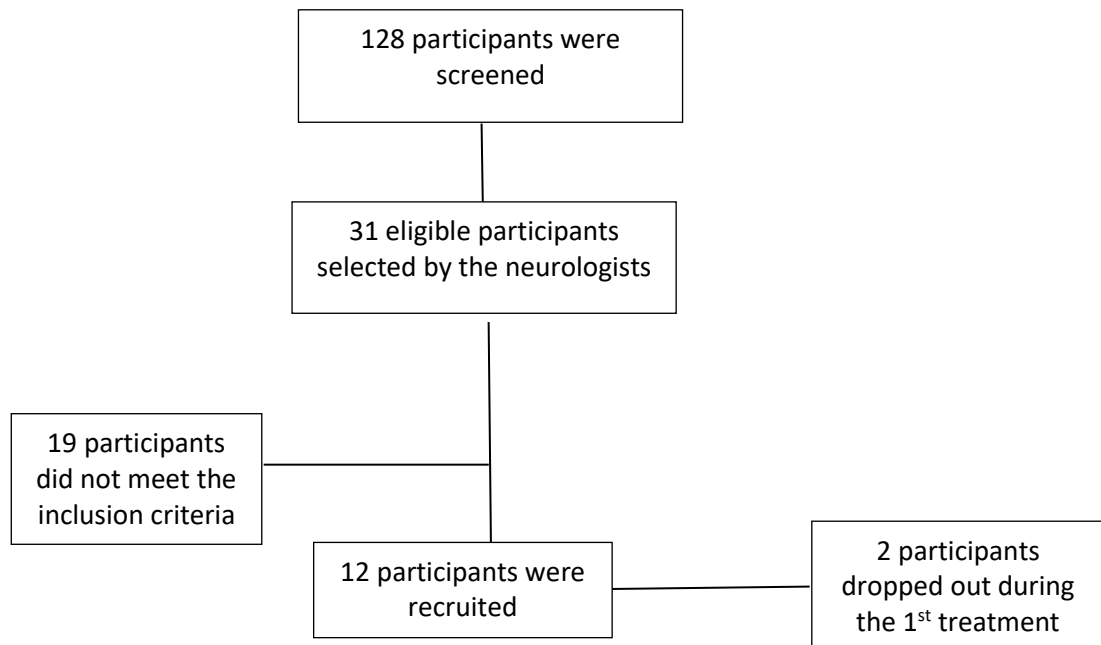


Figure 5. 8 Participant recruitment.

The reasons for non-recruitment are summarized in Table 5.6.

Table 5. 6 Descriptions for non-recruitment

Total number of participants	Description
6	Presence of cognitive impairments. Four participants had a mild impairment (MoCa scores ranging from 18-23) and two presented with severe cognitive deficit (MoCa score > than 10).
3	Presence of DBS
5	Co-morbidity. Three participants were recovered in Hospital, two for fractures and one for urinary tract infection. Two participants were trying new medication therapy as they suffered from rigidity and severe freezing episodes
3	Distance from the hospital (3 participants). Two participants lived on small islands than 2 h from the hospital by public transports. Unfortunately it was not feasible to carry out the treatment at participants' homes
1	Rejection of Fees examination (1 patient). One participant did not want to carry out the FEES examination
1	Rejection of treatment. One participant believed the treatment to be too intense and demanding

5.9 Descriptive characteristics of participants

The sample comprised 12 people with IPD and swallowing impairments. Two participants dropped out during the first week of treatment because the treatment was too intensive. The characteristic of these participants are described in the following sub-section.

5.9.1 Descriptive characteristic of participants who withdrew

The first participant was a male of 69 years old; the duration of the disease was 6 years. The severity of the disease was mild, the score of H&Y scale was 1.5 and UPDRS was 48. The swallowing assessment with FEES showed saliva traces coating in the valleculae (Yale Pharyngeal Residue Severity Rating Scale-Vallecular residue=2). No other impairments were noticed on instrumental examination. The participants complained of drooling and difficulties of saliva swallowing (ROMP-Saliva=35). He was on a full oral diet. Nevertheless he avoided crunchy foods such as crackers (FOIS=6). He dropped out at the first day of week 1, because the treatment was excessively demanding in terms of daily attendance and exercises workload.

The second participant who withdrew was 75 year old male with a 9 year history of IPD. The severity of the disease was mild, H&Y scale was 2 and UPDRS was 65. Swallowing assessment on FEES showed mild impairment with residue for both saliva and (Yale Pharyngeal Residue Severity Rating Scale-Vallecular residue: 3) and trace of residue in pyriform sinus (Yale Pharyngeal Residue Severity Rating Scale-Pyriform = 2). He presented with some episodes of drooling. However, these did not impact quality of life (ROMP-Saliva = 22). He was on a full oral diet (FOIS = 7). He dropped out at the third day of week 1 because he reported that his swallowing impairment was not severe to justify an intensive intervention.

5.9.1 Participants who completed the programme.

The remaining sample comprised 7 males and 3 females (Table 5.7). The overall mean age and SD were 69.1 ± 6.24 years. Males were slightly older than females. The mean and SD of IPD disease duration were 7.5 ± 2.6 years ranging from 4 to 11 years (Table 5.7). The overall severity of the disease was mild level in both assessments, the H&Y

scale and the MD-UPDRS scale (Table 5.7). The swallowing, speech and saliva subscales of the 2°part of the UPDRS scale indicated that participants perceived the deficits of speech more negatively than swallowing, although there was no statistically significant difference between both ($p > 0.05$). The mean quality of speech was the highest value (2.67 ± 1.03), followed by saliva and the drooling (2.50 ± 0.8) and by chewing and swallowing (2.08 ± 0.76) (Table 5.7). The scores of MoCA ranged from 25 to 30; mean and SD scores were 27.08 ± 1.32 (Table 5.7).

Table 5. 7 Descriptive information of participants

<i>ID</i>	<i>Gender</i>	<i>Age</i>	<i>Disease Years</i>	<i>MoCA</i>	<i>H&Y</i>	<i>UPDRS Tot</i>	<i>UPDRS 1°</i>	<i>UPDRS 2°</i>	<i>2.1</i>	<i>2.2</i>	<i>2.3</i>	<i>UPDRS 3°</i>	<i>UPDRS 4°</i>
<i>1P</i>	M	68	4	26	2.5	61	11	13	2	2	1	33	4
<i>2P</i>	F	73	5	27	3	79	17	17	1	2	2	37	8
<i>3P</i>	F	57	4	28	2	59	13	12	1	2	2	30	4
<i>4P</i>	M	72	9	26	3	64	15	12	3	2	2	32	5
<i>5P</i>	M	74	10	26	3	67	14	14	4	4	3	36	3
<i>6P</i>	F	69	6	29	2	48	9	11	2	2	1	25	3
<i>7P</i>	M	68	11	26	3	72	13	20	4	4	3	35	4
<i>9P</i>	M	58	8	27	2	52	12	10	3	3	3	28	2
<i>10P</i>	F	74	7	28	2.5	63	14	17	3	2	2	29	3
<i>12P</i>	M	77	8	25	3	78	14	20	4	3	3	40	4
<i>Mean</i>		69.1	7.25	27.08	3	65.92	13.33	15.83	2.67	2.50	2.08	32.92	3.83
<i>SD</i>		6.24	2.6	1.32	0.43	10.19	2.09	5.08	1.03	0.87	0.76	4.09	1.46

5.10 Data Collection

The assessment protocol was completed at 4 time points (1) 4 weeks before treatment (Time 0), immediately prior to treatment (Time 1), after 4 weeks of treatment (Time 2) and at 3 months follow-up (Time 3) (Fig 5.9). The location was the Neurology Department at Angel Hospital (Venice, Italy). The assessment was scheduled at the same time for each patient. The timing of anti-parkinsonian medications was scheduled in order to ensure they were “on-phase”.

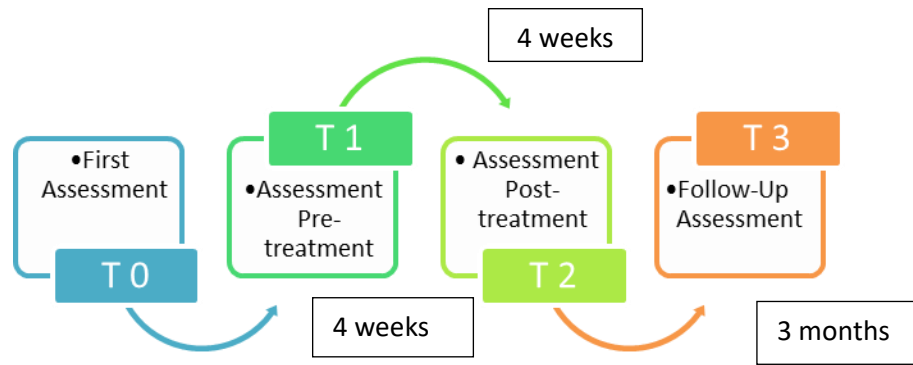


Figure 5. 9 Phase of the assessment protocol.

The assessment protocol consists of clinical non-instrumental evaluation and instrumental evaluation, which were previously described in the sub sections “Assessment Tools” (see Chapter 5.3) (Table 5.8). The protocol of assessment was carried out by two evaluators during the same day. Firstly, a SLP (not involved in the treatment) compiled the Italian version of the I-SWAL-QOL (Ginocchio et al., 2016), FOIS-it (Battel et al., 2018) and ROMP-saliva for each patient. Secondly, a senior ENT consultant carried out the FEES and completed the PAS (Rosenberg et al. 2016) and Yale Pharyngeal Residue Severity Rating Scale (Neubauer et al., 2015). The videos of FEES were saved and stored on a computer (Dell, Latitude E6420) using the participant reference ID code.

Table 5. 8 Summary of the outcome measures.

Outcome measures	Measures
Changes in the severity of airway invasion	PAS
Changes in presence of pharyngeal residue	YPR-SRS
Changes to method of nutritional intake	FOIS-It
Changes in dysphagia severity and swallowing related quality of life	I-SWAL-QOL
Changes in self-perception saliva	ROMP- Saliva

5.10.1.2 FEES Procedures

No topical anaesthetic was used in this procedure. At the beginning, the integrity of the nasal structures was assessed to verify where to insert the fibro-endoscope. The scope was gently inserted trans-nasally along the nasal floor below the middle

turbinate until it could be seen the nasopharyngeal vault. At this point the ENT consultant asked participants to breathe through the nose to allow the opening of the velopharynx. Then, the scope was inserted further into the oropharynx and placed above the epiglottis. This scope position allowed verification of the base of tongue, posterior pharyngeal wall, lateral pharyngeal walls, epiglottis, larynx and the presence of saliva in the valleculae and/or pyriform sinuses.

The swallowing assessments started with water trials (IDDS 0) (Cichero et al., 2017). In order to visualise liquids, 2 drops of blue dye for 150 ml of water were used, as per clinic protocol. Water trials were firstly administered using a 5 ml teaspoon. If no signs of penetration or aspiration occurred, participants were asked to drink the entire glass of water (150 ml). Thereafter, the examination involved yogurt trials (IDDSI Level 3). The type of yogurt (Yomo®) was provided by the hospital. Any allergies or food preferences were documented prior examination. In the presence of a dairy allergy, an apple sauce (IDDSI Level 3) was administered. Finally, small pieces of solid food (crackers) (IDDSI 7) (Cracker Gran Pavesi®) were given to participants.

The videos of all the examination were saved using the participant reference code by the student and they were used for inter-rater reliability

5.10.1.3 Blinding of assessors

The ENT consultant and the SLP acknowledged that the participants were recruited for this study, but none of them contributed in the protocol of treatment, ensuring some blinding of interventions.

5.10.1.4 Reliability of the outcome measure

Videos of FEES were used to evaluate the reliability of the scores among two different examiners. Inter-rater reliability was calculated in order to verify the level of homogeneity among assessors using the intra-class correlation coefficients (ICC); which is a recognised analysis used for continuous or ordinal data (Pisegna et al., 2018; Portney LG, 2015). The student randomly selected 20% sample of FEES video examination which were analysed by a second ENT. An ENT of the otolaryngology

department with a clinical working experience of 4 years, assessed 32 videos (20%) and completed the Yale Pharyngeal Residue Severity Rating Scale for each video. There was a strong level of agreement between the examinations (ICC=0.79)

In addition, most of the outcome measures obtained by clinical non-instrumental assessment involved participants self-administered questionnaires (SWAL-QOL and ROMP-Saliva). No reliability measurements were carried out for those assessments.

The inter-rater reliability of It-FOIS was carried for all the assessments. A SLP not involved in the study completed the forms based on the information given by the participants. The level of agreement between the form was excellent (ICC=0.97).

5.10.2 Treatment protocol

The intervention was carried after 1 hour from intake of medication. The treatment was delivered by the research student in the SLP office on the ground floor of the Angel Hospital. Participant was seated in a seat or in the wheelchair in front of the screen.

5.10.2.1 Intensity of the treatment

The treatment was scheduled for 1 hour a day, 5 days a week, for 4 weeks. This treatment regime was based on Lee Silverman Voice Treatment (LSVT)[®] which has demonstrated efficacy for IPD in the area of articulation and voice (McDonnell et al., 2018; Sapir et al., 2007). The treatment lasted about 1 hour. It was scheduled at same time and this was consistent for the following 4 weeks of treatment. If the participants had some problems, the time was changed according to participants needs. However, this modification could occur in a limited number of times.

The submental area was cleansed with an alcohol swab and the 2 electrode patches (round 30 mm each) were attached in midline underneath the chin, between the mental spine of mandible and superior palpable notch of thyroid cartilage. The electrodes were clipped into the MyoPlus Pro (Fig 5.3).

The sEMG signals from the device were transferred to portable computer using specialized NeuroTrac Software, as it has been described in sub-section 5.4. At the first meeting, the participants were registered in the programme. Then, it was selected

“EMG” programme from the home page. Two sEMG programmes were adopted in this study: “open display” and the “plane game” as previously mentioned (Section 5.3).

When participants required more information and time, this was given. If a participant had a dry mouth, it was used as an oral moisturiser. In addition, the programme involved yogurt swallowing tasks. The presence of any allergy and/or particular requests were collected prior to the treatment. As stated earlier, in case of an allergy, it was offered a food with similar rheology features of yogurt such as applesauce.

5.10.2.3 Verbal feedback

Verbal feedback was provided mostly on performances rather than results, especially when using the open display “software”. The rationale for this was that the treatment aimed to increase coordination of swallowing and not strength. For this reason, the instructions were: “Create a smooth line without interruptions”, which aimed to boost the coordination of swallowing skills. During the “plane game”, no verbal feedback was given to limit redundancy. In this programme, participants did monitor the plane instead of the wave-line of activation and were encouraged to increase the strength to reach the stairs. Hence, visual feedback was set on the results rather than on the performance.

Verbal feedback was delivered in order to increase internal error-detection and correction skills. These feedbacks were delayed 3-4 seconds after swallowing and scheduled randomly. In literature, it is recognised that frequent verbal feedback may limit self-assessment skills. Random and delayed feedback, instead, allow participants to independently evaluate the motor act and increase the awareness of motor control (Maas et al. 2008; Huckabee et al. 2018). Randomisation of feedback was scheduled using the software (www.randomizer.at).

5.10.2.4 Design of the treatment

The program involves a progression of specific motor tasks based on motor learning and neuroplasticity principles (Table 5.9). The type and frequency of verbal and visual feedback were strictly planned week by week.

Table 5. 9 sEMG swallowing tasks accordingly to neuroplasticity principles

Principle	Description of swallowing treatment tasks
Use It or Lose It	The intervention incorporated swallowing tasks
Use It and Improve it	The swallowing tasks were planned in an incremental order of difficulty week by week. sEMG and verbal feedback were set to increase of efficiency and accuracy of the swallowing function
Experience Specific	Exercises were tailored according to each participant skills.
Repetition Matters	Repetition and consistent practice of swallowing exercises
Intensity Matters	The treatment was intense, 1 hour a day, 5 days a week, for 4 weeks.
Salience	The treatment incorporated food trials and a game using sEMG

Description of the 1st week of treatment

The goal of this week was to familiarize participants with the equipment and the swallowing tasks. On the first day of treatment, the student demonstrated the swallowing task using sEMG biofeedback. Thereafter, participants tried the sEMG biofeedback, the student asked them to recognise the swallowing wave on the screen (Table 5.10). In addition, she invited participants to open the mouth or to contract submental muscles without swallowing in order to distinguish between the two sEMG signals (swallowing vs mouth opening) (Fig 5.3). The first swallowing tasks concerned saliva swallowing using “open display” (Fig 5.4). The last 6 swallows were executed using the plane game (Fig 5.5).

It is important to underlie that only during the first day of the treatment, verbal feedback were provided immediately and frequently at each swallow in order to allow the comprehension of the task.

Table 5. 10 Description of swallowing task during the first week of treatment.

1 st Week of treatment			
	1 st Day	2 nd to 4 th Day	5 th Day
Swallowing Tasks	15 saliva swallows, participants could have a break 4-5 minutes after every swallow.	- 3 consecutive saliva swallows followed by a break (for 3 times); - 3 consecutive saliva swallows followed by a break using the plane game (for 3 times).	3 saliva swallows.
Total number	15	18	3
Instructions	Try to swallow and create a smooth wave line on the monitor.	a) Swallow and create a smooth wave line, trying to produce a peak; b) Swallow and move up the plane, trying to reach the stars and then relax.	Try to swallow as best as you can.
Goals	Acquaintance and increase the understanding of the use of sEMG biofeedback.	The goal of this session was to enhance the skill of saliva swallowing, creating smooth wave line with high peak on command.	Assess the performance and mastery of the tasks.
Type of verbal Feedback	The feedback were on performance, provided immediately after swallowing and frequently at each swallow in order to allow the comprehension of the task. Verbal feedback was positive	Verbal feedback of the performance was delayed for 3-4 seconds after swallowing and provided randomly with a frequency of 50% of practice trials in order to increase the motor learning. The feedback was positive.	No feedback.

Description of the 2nd week of treatment

In this week, the treatment increased in difficulty, adding food trails during the treatments. This choice of using yogurt (IDDS 3) was motivated by several reasons. Firstly, the consistency and viscosity of yogurt are recognised to be not only safe but appropriate in IPD participants (Troche et al., 2013). Secondly, the inclusion of a food aimed to motivate participant during the tasks and to contribute to the neuroplasticity

principles of salience, transfer and generalisation.

In addition, the use of food could augment the saliva production which increased the difficulties of the task. In addition, the total number of swallows was increased up to 21, in order to augment the workload (Table 5.11).

Table 5. 11 Description of swallowing task during the second week of treatment.

2 nd Week of treatment		
	1 st to 4 th Day	5 th Day
Swallowing Tasks	a) 3 consecutive saliva swallows followed by a break (for 3 times). b) 3 consecutive saliva swallows followed by a break using the plane game (for 3 times). c) 3 consecutive yogurt swallows.	3 saliva swallows.
Total number	21	3
Instructions	a) Swallow and create a smooth wave line, trying to produce a peak; b) Swallow and move up the plane, trying to reach the stars and then relax.	Try to swallow as the best as you can.
Goals	The goal of this session is to enhance the skill of saliva ad yogurt swallowing, creating smooth wave line with a peak.	Assess the performance.
Type of verbal Feedback	Verbal feedback of the performance is delayed for 3-4 seconds after swallowing and provided randomly with a frequency of 50% of practice trials in order to increase the motor learning. The feedback was positive and encouraging the participants to enhance the performance.	No feedback.

Description of the 3rd week of treatment

The same protocol of treatment was maintained in the 3rd week, increasing slightly the number of swallowing. The goal of this week was to improve the retention of the skilled acquired in the previous week (Table 5.12).

Table 5. 12 Description of swallowing task during the third week of treatment.

3 rd Week of treatment		
	1 st to 4 th Day	5 th Day
Swallowing Tasks	a) 3 consecutive saliva swallows followed by a break (for 3 times). b) 3 consecutive saliva swallows followed by a break using the plane game (for 3 times). c) 3 consecutive yogurt swallows (for 3 times).	3 saliva swallows .
Total number	27	3
Instructions	a) Swallow and create a smooth wave line, trying to produce a peak; b) Swallow and move up the plane, trying to reach the stars and then relax.	Try to swallow as best as you can.
Goals	The goal of this session is to enhance the skill of saliva ad yogurt swallowing, creating smooth wave line with a peak.	Assess the performance.
Type of verbal Feedback	Verbal feedback of the performance is delayed for 3-4 seconds after swallowing and provided randomly with a frequency of 50% of practice trials in order to increase the motor learning. The feedback was positive and encouraging the participants to enhance the performance.	No feedback.

5.10.2.14 Description of the 4th week of treatment

In the last week of treatment swallowing tasks were delivered randomly. Thus, participants had to refine the motor control from one exercise to another. The randomisation of the exercises aimed to increase the transfer and generalisation principles (Table 5.13).

Table 5. 13 Description of swallowing task during the fourth week of treatment.

4 th Week of treatment		
	1 st to 4 th Day	5 th Day
Swallowing Tasks	a) 3 consecutive saliva swallows followed by a break (for 3 times). b) 3 consecutive saliva swallows followed by a break using the plane game (for 3 times). c) 3 consecutive yogurt swallows followed by a break (for 3 times).	3 saliva swallows.
Total number	27	3
Instructions	a) Swallow and create a smooth wave line, trying to produce a peak; b) Swallow and move up the plane, trying to reach the stars and then relax.	Try to swallow as best as you can.
Goals	The goal of this session is to enhance the skill of saliva and yogurt swallowing, creating smooth wave line with a peak.	Assess the performance.
Type of verbal Feedback	Verbal feedback of the performance is delayed for 3-4 seconds after swallowing and provided randomly with a frequency of 50% of practice trials in order to increase the motor learning. The feedback was positive and encouraging the participants to enhance the performance.	No feedback.

5.10.3 Fidelity of the intervention

Implementation fidelity has been defined as “*the methodological strategies used to monitor and enhance the reliability and validity of behavioural interventions*” (Bellg et al., 2004). Over the past year, several studies have confirmed the importance of addressing fidelity within research (Hayden et al., 2015; Hoffmann et al., 2014; Kaderavek & Justice, 2010; Slaughter et al., 2015). In this study, it has used the framework defined by Toomey et al. in order to ensure the fidelity to the intervention and adherence to the intervention protocol (Toomey et al., 2016). Each single sEMG session was saved in the computer allowing to verify the total number of swallows for each participant for each session. In addition, the verbal feedback of the SLP was recorded using the microphone on the computer (Dell Latitude E6420) where was installed the sEMG programme. This allowed the student to collect verbal feedback and the timing of delivering.

5.11 Data analysis

The data obtained in the quantitative and qualitative assessments were analysed using specific methods based on the nature of assessments.

5.11.1 Quantitative analysis

All the data were saved and entered into a spreadsheet in the Microsoft Excel. The student was responsible for verification of the accuracy of the inserted data.

Firstly, the data were analyzed descriptively indicating mean and SD for all the measures. The quantitative analysis was completed by the student with the help of a statistician using the software R (Development Core Team 2019). The test of normality of Shapiro-Wilk Test was assessed in order to verify how variables were distributed. It was used the Bartlett Test to assess homogeneity of variances.

The analysis of significance of the results in the four different assessments was calculated by analysis of variance (ANOVA). In case of non-parametric distribution, a Kruskal–Wallis test was used. The level of significance was set at a p-value < 0.05.

5.11.2 Qualitative analysis

The audio recordings were transcribed orthographically by the student. As recommended by the guidelines of Braun and Clarke (2006), she did not edit grammar errors or removing hesitations and pauses (Braun & Clarke, 2006). Thereafter, the videos were inserted in the NViVO software (Castleberry, 2014), which supports qualitative and mixed methods.

The transcriptions were analysed using Thematic Analysis (TA). TA is a method for systematically identifying, categorizing, and offering insight into patterns of meaning (themes) across a data set (Braun & Clarke, 2012). Braun and Clarke et al provided a framework constituted by the six phases of analysis (Braun & Clarke, 2012) which are summarised in Fig 5.10. In the first phase, the researcher read the transcription and noted trends in the . The second phase concerns the coding process, in which relevant features for research questions are identified and labelled. In the following phase, theme are assigned. A theme is a coherent and meaningful pattern in the data relevant to the research question. Searching for themes involves reviewing the coded data in order to identify areas of similarity or broad topics. In fifth phase, the themes are defined and named. The last phase is the writing up phase, in which the researcher must provide evidence of the data (Fig 5.10).

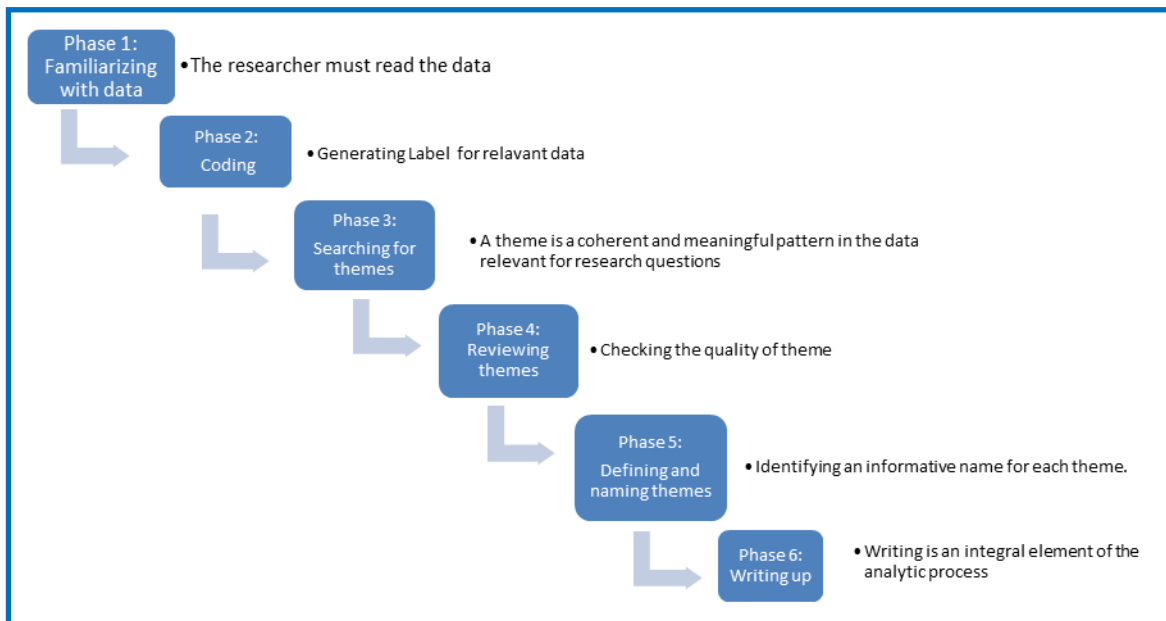


Figure 5. 10 Phases of Thematic Analysis (TA) by Braun and Clarke (2006).

5.12 Chapter summary

This chapter described the methods for this feasibility study including the process of recruitment and data collection. The assessment protocol and treatment was described,

Twelve participants were enrolled following the recruitment process and 10 completed the entire treatment programme.

The next chapter describes the process of cross-cultural translation of FOIS into Italian, which was an important outcome measure for data collection. The results of this feasibility study are described in Chapter 7.

Chapter 6: Cross-cultural translation of the Functional Oral Intake Scale

6.1 Introduction

This chapter describes the cross-cultural translation and validation of the Functional Oral Intake Scale (FOIS) (Crary et al., 2005) into Italian. Evidence based practice emphasises the importance of valid and reliable outcome measures in all fields of healthcare (Rodrigues, 2000). Currently, the majority of swallowing outcome scales are validated in English speaking populations, requiring a specific process for cross cultural translation for their use among non-English speaking populations. In clinical practice, an instrument is frequently directly translated from one language to another without a back-translation process, or with inappropriate translation techniques. This process does not guarantee a valid translated measure, which has content equivalent to the original version of the instrument (Cha et al., 2007). A scale should be appropriately translated and culturally adapted in order to be valid. The absence of assessments translated and adapted cross culturally for the Italian population is a significant drawback in providing high quality care to patients with dysphagia. Few scale has been validated into Italian.

The FOIS is a popular test that is routinely used internationally. FOIS has shown to be a valid and reliable tool to document change in functional oral intake of food and liquid in stroke patients (Crary et al., 2012; Kunieda et al., 2013). It has already been described in Chapter 5 Section 3.1.2 (see Table 5.1).

Albuquerque et al. suggest that the FOIS is one of the most used outcome measure to verify effectiveness of biofeedback swallowing treatment and it forms part of the protocol in this study (Albuquerque et al., 2019). While other outcome measures used in this study are translated and validated in Italian, the FOIS is not. Therefore, the aim of this study, which is part of the wider feasibility study, is to translate and validate the FOIS into Italian in order to facilitate its use in Italian clinical settings in this and future studies.

6.2 Materials and Methods

This prospective study was carried out according to the Declaration of Helsinki and received ethical approval from the Research Ethics Committee, School of Linguistic, Speech and Communication Sciences, Trinity College Dublin (Appendix I) . The study comprised two phases; the first was the process of translation and the second was the validation of the translated FOIS scale.

6.2.1 Translation Process

The process of translation followed the five stages described by Beaton et al. (2000) for cross-cultural adaptation of self-report measures (Figure 6.1). In addition, the COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) checklist was completed to verify the cross cultural validity (Mokkink et al., 2010). Permission of translation was obtained from the authors of the English version (Crary et al., 2005).

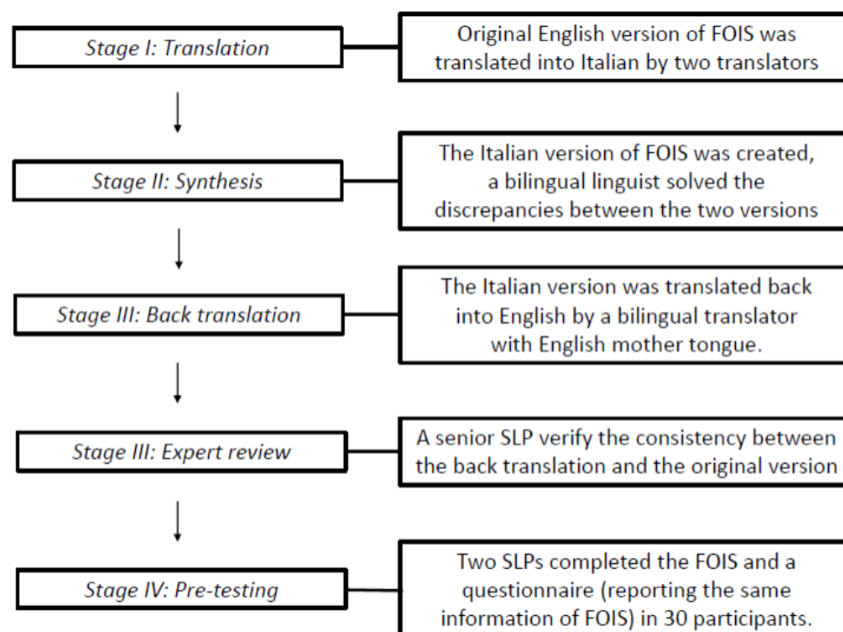


Figure 6. 1: The five stages of the cross-cultural adaptation described by Beaton et al.(2000).

6.2.1.1 Stage I: Initial Translation (English into Italian)

Two bilingual speech language pathologists (SLPs) independently translated the FOIS. Both had Italian as their mother tongue. Both SLPs had over ten years' experience in dysphagia rehabilitation, which was deemed important in selecting the appropriate clinical terms. Two independent translations were produced in order to facilitate the discussion on wording choices and discrepancies of the translation process.

6.2.1.2 Stage II: Synthesis of the Translations

At this point the translation process from Stage 1 was documented and any differences in translations and interpretation were addressed. Only one discrepancy existed, which concerned the term "Tube Dependent" whose direct Italian translation is "Tubo dipendente". However, this term is not used in Italian health care settings. Another co-author who is a bilingual (Italian-English) linguist was consulted in order to verify the pertinence of terms and also to analyse the rationale for translation choices. Following this team discussion, it was agreed to substitute the term "tube dependent" with "Enteral/Parental Nutrition". The Italian translation for this is "Nutrizione Enterale/Parenterale" (Appendix J).

6.2.1.3 Stage III: Back Translation (Italian into English)

This process ensures that the translated version of the scale presents the same item content as the original version of the scale. A bilingual psychologist whose mother tongue is English produced the back-translation from Italian to English. She was not informed of the concepts of the FOIS, as she did not have experience in dysphagia. This assisted in the preservation of information bias.

6.2.1.4 Stage IV: Expert Revision

The next phase involved agreement amongst the research team on the acceptability of differences between the back translation from Stage III and the original English version

of the FOIS. The final Italian version of the FOIS was consolidated (Appendix J) and consensus was reached amongst the translators and research team.

6.2.1.5 Stage V: Pre-testing

The final stage of adaptation involved the assessment of the validity and reproducibility of the new translated version of the scale. To test construct validity, a questionnaire (Appendix K) was developed containing the same information regarding tube feeding and modifications to oral intake as contained in the FOIS. To assess the feasibility and reproducibility of the new Italian FOIS version (FOIS-It), the questionnaire and the FOIS-It were administered separately by two SLPs, each in a different hospital site in Italy and the results compared. Each SLP first completed the questionnaire on a consecutive sample of 30 people with chronic dysphagia who were on their caseloads. Following completion, each individual questionnaire was coded and placed in a sealed envelope. Two days later the SLPs completed the FOIS-It on the same individuals. This short time interval allowed for some distance from recall of the contents the completed questionnaire but also attempted to avoid differing results as a consequence of change in swallowing or medical status.

The results of the questionnaire and the FOIS-It for each patient were collated and scores inputted onto an Excel spread sheet. The correlation between the completed questionnaires and the FOIS-It was excellent (Cronbach's alpha = 0.99).

The inter-rater reliability was calculated with other two SLPs, one with 3 years of dysphagia experience and one with 29 years dysphagia experience, completing the FOIS and questionnaire on the same sample of 30 patients within an interval of time of 24 hours. There were no lexicon concerns and the results reported an excellent inter-rater reliability (ICC showed 0.99). The final Italian version of the FOIS (FOIS-It) was now ready for validation.

6.2.2 Validation Process

The aim of this phase of the research was to validate the FOIS-It with a larger group of SLPs covering different clinical settings in Italy.

6.2.2.1 Participants

Ten Italian SLPs with a broad range clinical experience (range 3 to 29 years, mean 9.23 ± 7.9) were recruited from personal contacts (Table 6.1). These SLPs practiced at different hospital sites (neurorehabilitation hospital, acute teaching hospital, nursing home facility) in Italy. This ensured validation of FOIS-It in different clinical contexts.

Table 6. 1 Profile of SLP who completed the FOIS and questionnaire.

Participant SLP	Years' experience	Employment setting	Number of people with dysphagia assessed
1	3	Neuro-rehabilitation Hospital	21
2	3	Nursing Home	21
3	8	University Hospital (Department of Neurology)	21
4	6	Nursing Home	20
5	10	University Hospital (Department of Neurology)	34
6	29	Neuro-rehabilitation Hospital	29
7	10	Neuro-rehabilitation Hospital	21
8	9	Neuro-rehabilitation Hospital	21
9	4	Neuro-rehabilitation Hospital	20
10	3	Neuro-rehabilitation Hospital	19

6.2.2.2 Data Collection

All participating SLPs were asked to complete the FOIS-It and the questionnaire on individuals with different aetiologies of dysphagia, who had received a clinical swallow assessment and who were currently on their caseloads and medically stable. The same procedures used in Stage V (pre-testing) regarding data collection were used here.

Data collection was carried out over one month. No diagnosis and/or patient information were sought and the SLPs returned anonymized forms to the senior author.

6.3 Statistical Analysis

Data for completed sets of questionnaires and FOIS-It were inputted into an Excel database. Statistical tests were performed using the SPSS 20.0 statistical software (SPSS, Inc., Chicago, IL). Kolmogorov–Smirnov test was used to test the normality of the distribution. Internal consistency was assessed using Cronbach’s alpha coefficient.

6.4 Results

SLPs completed the FOIS-It and questionnaire on 227 people with dysphagia (Table 6.2). There were no reported issues in completing the FOIS-It. The distribution of FOIS was normal, with a mean score of 4.09 ± 2.09 and the distribution of questionnaire was normal with mean $4.09 \pm 2,10$. The internal consistency of scores was excellent ($\alpha = 0.99$).

Table 6. 2 Internal Consistency

	FOIS-It	Questionnaire
Scores	4.09 \pm 2.09	4.09 \pm 2,10
Internal Consistency	$\alpha = 0.99$	

6.5 Discussion

The cross-cultural translation of FOIS into Italian was carried out following a five-stage process as recommended by international guidelines (Beaton et al., 2000) and validated in a large sample of people with dysphagia by Italians SLPs. The process of translation required some adaptation of the FOIS to Italian health care setting specific terminology. One term was modified from the original version of the FOIS in order to have an accurate and pertinent tool in the Italian health care context.

One of the strengths of this study was the final validation process, which was completed in a relatively large sample of patients, recruited from different health care

settings on a heterogeneous population of people with swallowing disorders. The validation process using SLPs with a wide range of clinical experience suggest that use of the FOIS-It does not differ according to clinical experience of rates.

It is acknowledged that there are some shortcomings with the FOIS scale, for instance, it was originally validated on a stroke population, although it is widely used in other populations with dysphagia (Ciucci et al., 2016; Hutcheson & Lewin, 2013). The scale also refers to the individual's oral intake status but does not incorporate information on safety of swallowing at these levels. However, it remains a quick useful outcome measure of a person's oral feeding status at a specific point in time, facilitating the tracking of progress from non-oral to oral feeding and vice versa.

In this study, an analysis on the diagnosis and the severity of dysphagia was not carried out as the focus was only on the cross-cultural adaptation of the scale and the validity and reliability of the scale was already established in a previous study (Crary et al., 2005). Further studies might be verifying the reliability of the scale against instrumental examination in Italian people with swallowing disorders.

6.6 Summary

In conclusion, the Italian version of FOIS was translated following international guidelines and validated by Italian SLPs working with individuals with dysphagia. The FOIS-It has therefore some reliability to report swallowing outcomes in this study with an Italian population.

Chapter 7: Results of the Feasibility study

7.1 Introduction

In this chapter, the findings of this feasibility study are described according to the research questions. Firstly, the results of the quantitative analysis are presented in order to reveal if the specific biofeedback swallowing intervention using sEMG biofeedback improved swallowing function and if the changes were maintained in the follow-up assessment at 3 months. Secondly, the findings from the qualitative section of the study are provided in order to understand participant perspectives and to incorporate the suggestions from participants to inform a future study.

7.2 Impact of Intervention on Swallowing Parameters, Oral Intake and Quality of life

Overall, the results showed positive changes in the swallowing function measured using standardized assessments and scales. Some post-treatment assessments did not find statistically significant improvements, nevertheless none of the evaluations after treatment revealed a decrease in swallow function or a worsening of dysphagia signs or symptoms. The following sections describe the results according to the quantitative outcomes.

7.2.1 Laryngeal penetration and aspiration

Results from FEES assessment indicate that none of participants presented with aspiration events during the FEES examinations at any time periods (Table 7.1). The PAS scores ranged from 2 to 5 (Table 7.1), suggesting no aspiration. The participants with the most severe signs of dysphagia were participants 6P and 10P (Table 7.1) who presented with a PAS penetration score of 5 during solid food trials (cracker) (IDDSI 7) in both baseline pre-treatment assessments indicating that food contacted the vocal folds and was not ejected from the airway. The PAS scores improved slightly for both participants (respectively PAS=4; PAS= 3) after treatment. Changes in solid food residue are described in the following sub-section 7.2.2. Nevertheless, participant 6P

did not maintain these improvements at the follow-up assessment (PAS=5) (T3), whereas participant 11P showed a retention effect (PAS=3) (T3).

Overall, there were slightly more penetration events while swallowing water (IDDSI level 0) and solid food for 5 participants (3P; 6P; 9P; 10 P; 12P) in comparison with the other swallowing trials that involved saliva , yogurt and crackers (Table 7.1).

Table 7. 1 Penetration-aspiration scores using different food consistencies for all participants (N = 10).

	1° PENETRATION ASPIRATION SCALE				2° PENETRATION ASPIRATION SCALE				3° PENETRATION ASPIRATION SCALE				4° PENETRATION ASPIRATION SCALE			
	SALIVA	IDDSI 0	IDDSI 3	IDDSI 7	SALIVA	IDDSI 0	IDDS 3	IDDS 7	SALIVA	IDDSI 0	IDDSI 3	IDDSI 7	SALIVA	IDDSI 0	IDDS 3	IDDS 7
1P	4	4	3	3	4	4	4	4	4	4	3	3	4	4	3	3
2P	4	3	2	3	4	4	2	3	4	3	2	2	4	3	4	4
3P	2	3	2	4	3	3	2	5	2	3	2	3	2	3	2	3
4P	3	4	3	3	3	4	3	3	3	4	3	3	3	4	4	3
5P	3	4	2	4	3	4	2	4	3	4	2	4	4	4	2	4
6P	2	4	4	5	3	4	3	5	2	3	3	4	2	3	4	5
7P	3	3	1	3	3	2	3	3	3	3	2	3	3	3	2	3
9P	4	4	2	3	4	5	2	3	2	4	2	3	2	4	2	3
10P	3	5	3	5	3	5	3	5	3	4	3	3	4	5	3	3
12P	2	3	5	4	2	4	5	4	2	3	4	3	2	3	4	4
Mean	3	3,7	2,7	3,7	3,2	3,9	2,9	3,9	2,8	3,5	2,6	3,1	3	3,6	3	3,5
SD	0,8	0,6	1,1	0,8	0,6	0,8	0,9	0,8	0,7	0,5	0,7	0,5	0,9	0,7	0,9	0,7

Nevertheless, no statistically significant differences ($p > 0.05$) were found for laryngeal penetration across four FEES assessment time-points for any swallowing trial on any food or fluid consistency including saliva (Tables 7.1 and 7.2).

Table 7. 2 Statistical analysis results of PAS scores during different swallowing trials across the 4 assessment time points (T0; T1; T2; T3).

Scale	Swallowing Trials	SHAPIRO-WILK TEST		BARTLETT TEST	ANOVA
PAS	Saliva	T 0	0.6622	0.9953	0.9972
		T 1	0.3652		
		T 2	0.6622		
		T 3	0.6622		
	Water (IDDSI 0)	T 0	0.5571	0.9586	0.9865
		T 1	0.4563		
		T 2	0.5321		
		T 3	0.5574		
	Yogurt (IDDSI 3)	T 0	0.7742	0.9452	0.9752
		T 1	0.7654		
		T 2	0.6785		
		T 3	0.7895		
	Crackers (IDDSI 7)	T 0	0.6452	0.9658	0.9412
		T 1	0.6851		
		T 2	0.6923		
		T 3	0.6527		

7.2.2 Pharyngeal residue

Overall, on FEES, pharyngeal residue was observed more in the valleculae than pyriform sinus for all participants. In addition, three participants (5P; 7P and 9P) had medication residue at the base of tongue and valleculae during FEES examination (T0;T2). Participant 7P presented with a clear white layer between the base of tongue and the valleculae. Two participants (5P; 9P) had identifiable traces of medication in their valleculae. At the post treatment assessment (T2) no medication residue was found for any of these three participants. Nevertheless, at the follow-up FEES examination (T3), participant 7P again showed traces of medication between the base of tongue and valleculae, whereas no medication residue was noticed at T3 in the other two participants.

Yale Pharyngeal Residue Severity Rating Scale (YPR-SRS) (Neubauer et al., 2015) was used to score residue. Scores indicated more residue for saliva and cracker than water and yogurt swallowing trials for all participants. Analysis of scale scores revealed a

statistically significant reduction ($p < 0.05$), which are described in the following sections.

7.2.2.1 Saliva

At the first two assessments (T0 and T1), the scores on YPR-SRS (Neubauer et al., 2015) for saliva in the valleculae ranged from 2 to 5, indicating presence of traces of saliva in the vallecular mucosa (Score= 2-3) (Table 7.3). Overall, these scores were almost consistent during the two pre-treatment assessments (respectively the mean scores were 3.4 at T0 and 3.4 at T1) (Tables 7.3, and 7.4), confirming stability in function after 1 month. The residue of saliva in the valleculae decreased significantly ($p < 0.05$) after treatment from a mean score of 3.4 at T1 to a mean score of 2 at T2 and the improvement was maintained in the follow-up assessment (mean score 2.7 at T3) (Tables 7.3; 7.4 and 7.5). The YPR-SRS scores for saliva in pyriform sinus ranged from 1 to 4 (mean score: 2) at both T0 and T1 assessments. The saliva residue in the pyriform changed slightly after treatment from mean score of 2 (T1) to 1.3 (T2), although the difference was not statistically significant ($p > 0.05$) (Table 7.4). This reduction of saliva pooling in pyriform sinus was maintained also at the 3-month follow-up assessment (Tables 7.3; 7.4 and 7.5).

Table 7. 3 Yale Pharyngeal Residue Severity Rating Scale (YPR-SRS) scores during saliva swallowing trial for the participants. The scale ranges from 1 (None) to 5 (Severe), for all participants (N = 10)

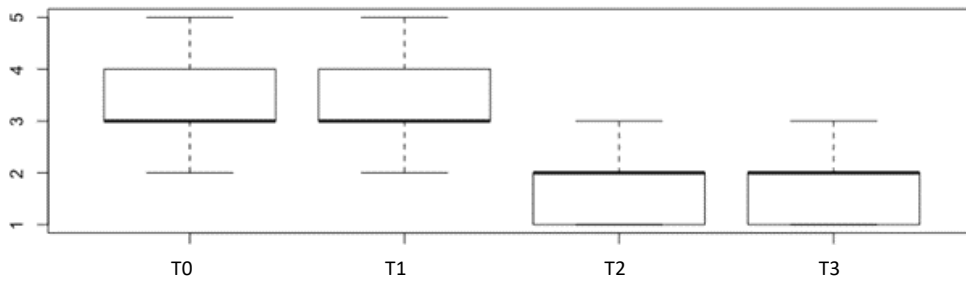
ID	SALIVA - Yale Pharyngeal Residue Severity Rating Scale							
	T0 ASSESSMENT		T1 ASSESSMENT		T2 ASSESSMENT		T3 ASSESSMENT	
	Valleculae	Pyriform	Valleculae	Pyriform	Valleculae	Pyriform	Valleculae	Pyriform
1P	3	2	3	2	1	1	2	1
2P	4	3	4	3	3	2	3	2
3P	2	1	2	1	1	1	2	1
4P	3	1	3	1	2	1	2	1
5P	3	2	3	2	2	2	2	1
6P	5	4	5	4	3	1	4	2
7P	3	1	3	1	3	1	3	1
9P	4	3	4	3	1	2	4	3
10P	3	2	3	2	2	1	3	2
12P	4	1	4	1	2	1	2	1
Mean SD	3,4±0.84	2±1.05	3,4±0.84	2±1.05	2±0.82	1,3±0.48	2,7±0.82	1,5±0.71

Table 7. 4 Saliva results of the Yale Pharyngeal Residue Severity Rating Scale (YPR-SRS)

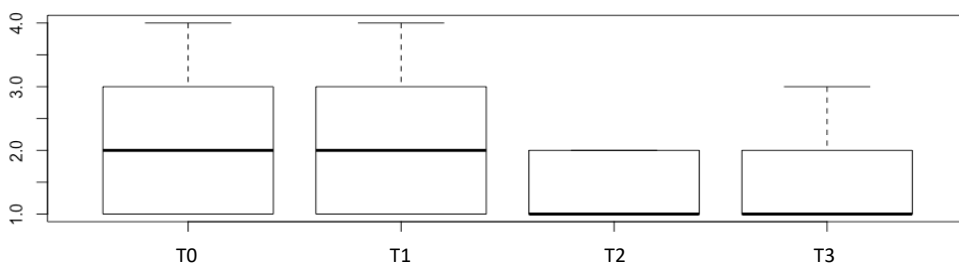
SALIVA SCORES		SHAPIRO-WILK TEST		BARTLETT TEST	KRUSKAL-WALLIS TEST
	VALLECULAE	T0	0.1716	0.9639	< 0.05*
		T1	0.1716		
		T2	< 0.05		
		T3	< 0.05		
	PYRIFORM	T0	0.07391	0.0998	0.2476
		T1	0.07391		
		T2	< 0.05		
		T3	< 0.05		

Table 7. 5 Boxplot of Saliva residue scores of the valleculae (A) and pyriform (B) collected in different times (T0; T1; T2; T3) using the YPR-SRS

A. Analysis of saliva scores in the valleculae



B. Analysis of saliva residue scores in the pyriform



7.2.2.2 Water (IDDSI Level 0)

The YPR-SRS (Neubauer et al., 2015) scores for water ranged from 1 to 5 at T0 (Table 7.6). Only participant 6P showed an increase in vallecular residue at T2 (Score = 5) (Table 7.6). The remaining scores ranged from 1 to 3 indicating traces coating the mucosa in both valleculae and pyriform across the all assessments (Table 7.6).

The vallecular residue during water swallowing trials did not show any statistically significant differences ($p > 0.05$) across four assessments (Tables 7.6 and 7.7). At the pre-treatment assessment, the mean overall score was 2 (T1) (Table 7.8). After treatment, the residue in the valleculae with water decreased but was not statistically significant; respectively from a mean score of 2 at T1 and to 1.7 at T2 ($p > 0.05$). The mean YPR-SRS score at the follow-up assessment was 1.7, showing a maintenance effect (Tables 7.7 and 7.8).

The presence of residue in pyriform sinus on water was small (mean score was 1.8 at T0) (Table 7.6). No statistically significant differences were found across the residue scores at all four assessment time points, the mean scores were T1=1.1; T2=1; T3= 1.1 (Tables 7.6).

Table 7. 6 Scores of Yale Pharyngeal Residue Severity Rating Scale during water swallowing for all participants (N = 10).

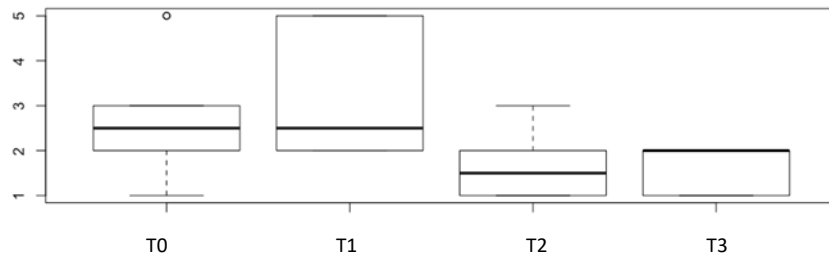
ID	WATER (IDDSI=0) Yale Pharyngeal Residue Severity Rating Scale							
	T0 ASSESSMENT		T1 ASSESSMENT		T2ASSESSMENT		T3 ASSESSMENT	
	Vallecula	Piriform	Vallecula	Piriform	Vallecula	Piriform	Vallecula	Piriform
1P	1	1	1	1	1	1	1	1
2P	2	1	2	1	2	1	2	1
3P	1	1	1	2	1	1	1	1
4P	2	1	2	1	1	1	2	1
5P	3	1	3	1	3	1	2	1
6P	3	2	5	2	3	1	3	2
7P	1	1	1	1	1	1	1	1
9P	2	1	2	1	2	1	2	1
10P	2	1	2	1	2	1	2	1
12P	1	1	1	1	1	1	1	1
Mean	1.8	1.1	2	1.2	1.7	1	1.7	1.1
SD	0.79	0.32	1.25	0.42	0.82	0	0.67	0.32

Table 7. 7 Yale Pharyngeal Residue Severity Rating Scale scores for residue on water

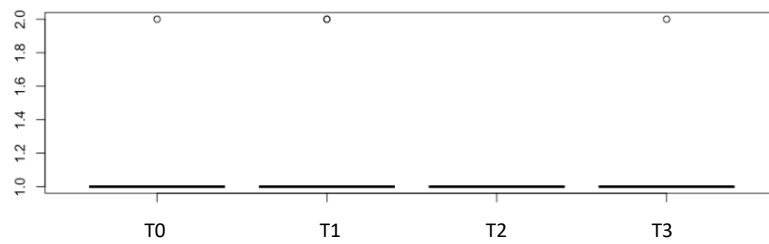
WATER SCORES		SHAPIRO-WILK TEST		BARTLETT TEST	KRUSKAL-WALLIS TEST	ANOVA
	VALLECULAE	T0	0.6833	0.9999		0.6116
		T1	0.6833			
		T2	0.8817			
		T3	0.8817			
	PYRIFORM	T0	0.399	0.9953		0.7305
		T1	0.3953			
		T2	0.3354			
		T3	0.5864			

Table 7. 8 Boxplot of water residue scores of the valleculae (A) pyriform (B) collected in different times (T0; T1; T2; T3) using the Yale Pharyngeal Residue Severity Rating Scale

A. Analysis of water scores in the valleculae



B. Analysis of water residue scores in the pyriform



7.2.2.3 Yogurt (IDDSI Level 3)

The YPR-SRS (Neubauer et al., 2015) scores for residue in the valleculae on yogurt ranged from 1 to 4, (mean score of 2) indicating traces on the mucosa. Participants 6P and 12P showed higher vallecular pooling (score= 4) than other participants during yogurt swallowing trials (Table 7.9) at the pre-treatment assessment. However, the residue decreased (score 3) at the post-treatment assessments for these two participants. Overall, the valleculae residue was reduced at the post treatment assessment and further diminished at follow up assessment. The mean YPR-SRS scores were respectively T1 = 2.1; T2=1.9 and T3=1.7 (Table 7.9). Nevertheless, these changes were not statistically significant ($p > 0.05$) (Tables 7.10 and 7.11).

The presence of yogurt residue on the pyriform sinus was small; the mean YPR-SRS score was 1.3 at T0 (Score 1= absence of residue) (Table 7.9) No statistically significant differences were found in pyriform residue ($p > 0.05$) on this consistency across the four assessments time points (Tables 7.10 and 7.11).

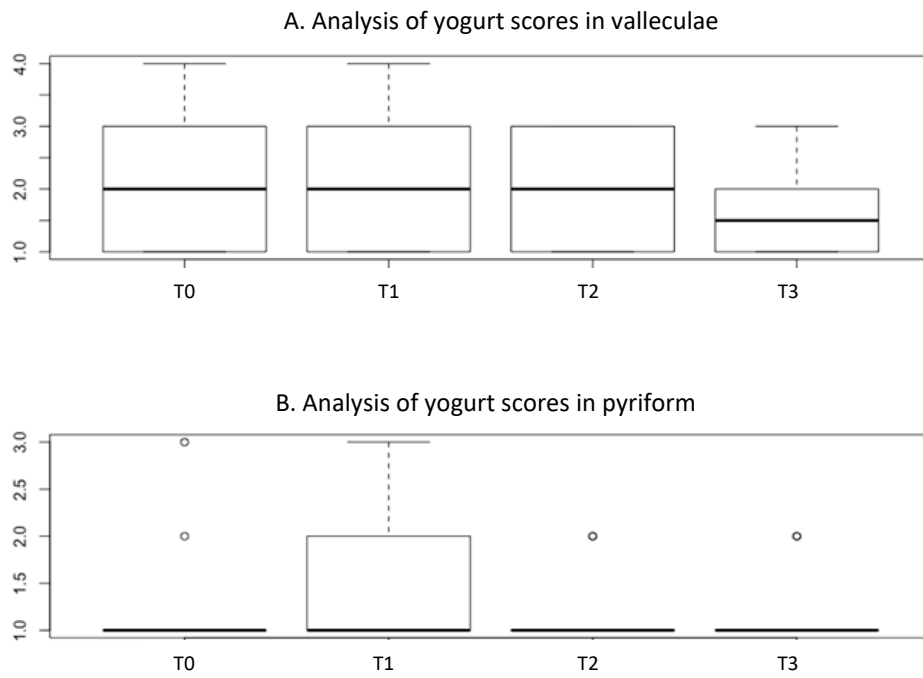
Table 7. 9 Yale Pharyngeal Residue Severity Rating Scale Scores during yogurt swallowing (IDDSI 3) for all participants (N = 10).

ID	YOGURT (IDDSI 3) Yale Pharyngeal Residue Severity Rating Scale							
	T0 ASSESSMENT		T1 ASSESSMENT		T2 ASSESSMENT		T3 ASSESSMENT	
	Valleculae	Pyriform	Valleculae	Pyriform	Valleculae	Pyriform	Valleculae	Pyriform
1P	1	1	1	1	1	1	1	1
2P	2	1	2	1	2	1	1	1
3P	1	1	1	1	1	1	1	1
4P	1	1	1	1	1	1	1	1
5P	2	1	2	1	2	1	2	1
6P	4	3	4	3	3	2	2	2
7P	1	1	1	2	1	1	1	1
9P	2	1	2	1	2	1	2	1
10P	3	2	3	2	3	2	3	2
12P	4	1	4	1	3	1	3	1
Mean	2,1	1,3	2,1	1,4	1,9	1,2	1,7	1,2
SD	1,20	0,67	1,20	0,70	0,88	0,42	0,82	0,42

Table 7. 10 Yogurt residue results of the Yale Pharyngeal Residue Severity Rating Scale.

YOGURT SCORES		SHAPIRO-WILK TEST		BARTLETT TEST	KRUSKAL-WALLIS TEST
	VALLECULAE	T0	< 0.05	0.5693	0.8799
		T1	< 0.05		
		T2	< 0.05		
		T3	< 0.05		
	PYRIFORM	T0	< 0.05	0.2656	0.9144
		T1	< 0.05		
		T2	< 0.05		
		T3	< 0.05		

Table 7. 11 Boxplot of Water residue scores of the valleculae (A) pyriform (B) collected in different times (T0; T1; T2; T3) using the Yale Pharyngeal Residue Severity Rating Scale



7.2.2.4 Solid Food (Cracker) (IDDSI Level 7)

Solid food (cracker) produced more pooling in the valleculae than pyriform sinus for all participants. It was more remarkable in participants 5P, 6P and 12P (Table 7.12). There was not a statistically significant difference ($p > 0.05$) between the two pre-treatment timepoints (T0; T1). The mean YPR-SRS scores were 3.4 at T0 and 3.5 at T1, revealing that the residue covered the vallecular mucosa, though the epiglottic ligament was visible (Table 7.12). Residue on the valleculae decreased significantly at the post-treatment assessment (mean score 2.3 T2) ($p < 0.05$) and the change was retained in the follow-up assessment (mean score 2 at T3) (Tables 7.12 and 7.13)

The pyriform residue on the cracker was consistent during T0 (mean score 2.1) and T1 (mean score 2.1) assessment time points. Only participants 6P and 2P showed higher YPR-SRS scores (respectively score 4 and score 3) at pre-treatment assessment than the rest of participants (Table 7.12). At post treatment assessment (T2), the residue on cracker was reduced from a mean score of 2.1 at T1 to a mean score 1.3 at T2, although the difference was not statistically significant ($p > 0.05$). The reduction in residue was not maintained at the follow-up assessment (mean YPR-SRS score 1.7 at T3) (Table 7.12; 7.13; 7.14).

Table 7. 12 Scores of Yale Pharyngeal Residue Severity Rating Scale during Solid Food (Cracker) (IDDSI 7) swallowing trial for all participants (N=10).

ID	SOLID FOOD (Cracker) (IDDSI=7) Yale Pharyngeal Residue Severity Rating Scale							
	T0 ASSESSMENT		T1 ASSESSMENT		T2 ASSESSMENT		T3 ASSESSMENT	
	Valleculae	Pyriform	Valleculae	Pyriform	Valleculae	Pyriform	Valleculae	Pyriform
1P	3	2	3	2	1	1	2	2
2P	3	3	4	3	1	1	1	2
3P	3	2	3	2	2	2	2	2
4P	3	2	3	2	1	1	1	1
5P	4	2	4	2	2	1	2	2
6P	5	5	5	5	3	2	3	2
7P	3	1	3	1	3	1	2	1
9P	3	2	3	2	3	2	2	2
10P	3	2	3	1	3	1	2	2
12P	4	1	4	1	4	1	3	1
Mean	3,4	2,2	3,5	2,1	2,3	1,3	2	1,7

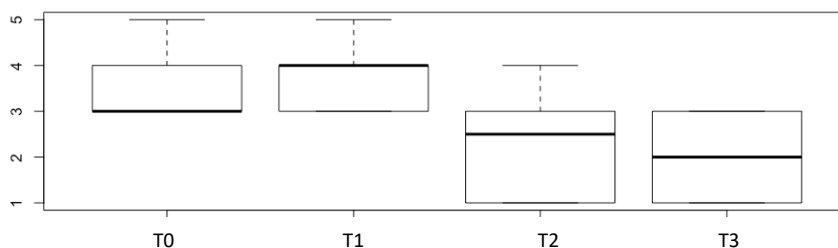
SD	0,70	1,14	0,71	1,20	1,06	0,48	0,67	0,48
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Table 7. 13: Solid Food (IDDSI Level 7) residue results of the Yale Pharyngeal Residue Severity Rating Scale

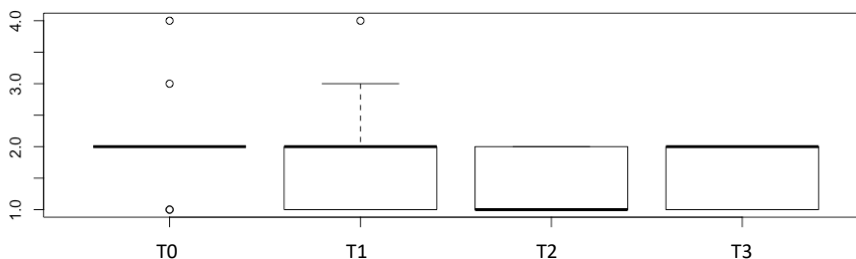
SOLID FOOD -CRACKER- (IDDSI Level 7) SCORES		SHAPIRO-WILK TEST		BARTLETT TEST	KRUSKAL-WALLIS TEST
		VALLECULAE	T0	< 0.05	0.4426
T1	< 0.05				
T2	0.1105				
T3	< 0.05				
PYRIFORM	T0	< 0.05	0.1515	0.1574	
T1	< 0.05				
T2	< 0.05				
T3	< 0.05				

Table 7. 14: Boxplot of Solid Food (IDDSI Level 7) residue scores of the valleculae (A) pyriform (B) collected in different times (T0; T1; T2; T3) using the Yale Pharyngeal Residue Severity Rating Scale.

A. Analysis of Solid Food scores on valleculae



B. Analysis of Solid Food scores on pyriform sinus



7.2.3 Methods of Oral intake

All participants were on an oral diet. At the pre-treatment assessments (T0;T1), five participants (4P; 6P; 7P; 9P; 12P) could eat only single consistency food (FOIS-It: 4). Four participants (2P,3P,5P,10P) consumed food that required special preparation (FOIS-It: 5) and 1 participant had to avoid specific food (FOIS-It: 6) (Table 7.15). The mean FOIS-It score at T0 was 4.8 and at T1 was 4.7, showing no important difference amongst the two assessments for the group. After treatment, the mean FOIS-It score increased from a score of 4.7 at T1 to 5.5 at T2 (Tables 7.15 and 7.16). This positive change in food intake was statistically significant ($p < 0.05$) and it was retained at the 3-month follow-up assessment (FOIS – It mean score 5.5 at T3) (Tables 7.15; 7.16 and 7.17).

Table 7. 15 Score of the FOIS-It at different assessment times for all participants

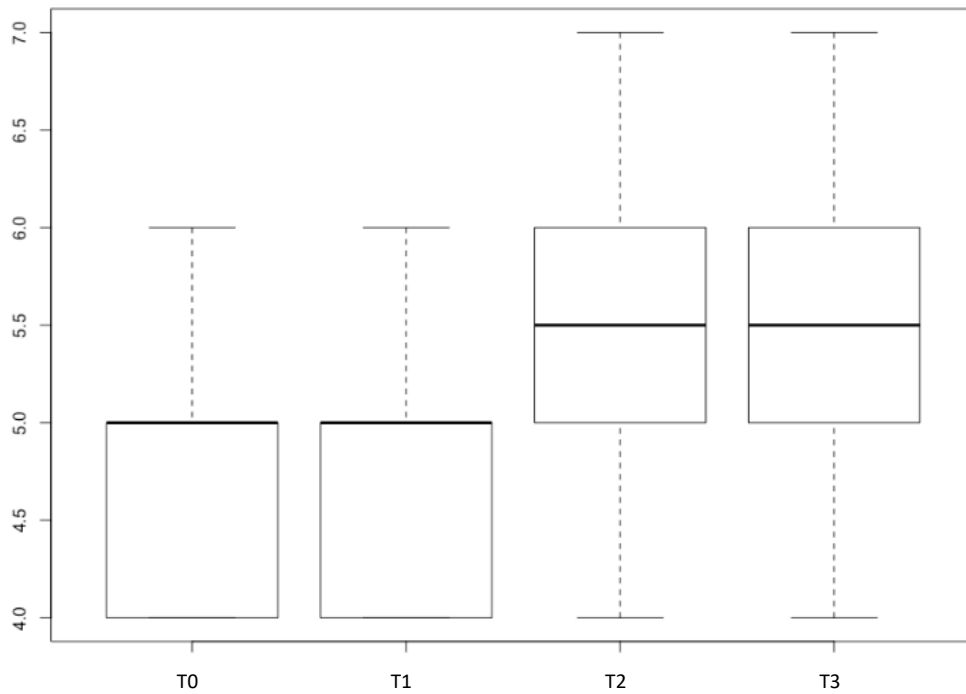
ID	FOIS-It at T0	FOIS-It at T1	FOIS-It at T2	FOIS-It at T3
1P	6	6	7	7
2P	5	5	6	6
3P	5	5	6	6
4P	5	4	6	6
5P	5	5	5	5
6P	4	4	5	5
7P	4	4	5	5
9P	5	4	4	4
10P	5	5	6	6
12P	4	4	5	5
Mean	4.8	4.7	5.5	5.5
SD	0.3	0.5	0.8	1.1

Table 7. 16 FOIS-It analysis.

FOIS-It	SHAPIRO-WILK TEST		BARTLETT TEST	KRUSKAL-WALLIS TEST
	T0	< 0.05	0.7523	< 0.05
	T1	< 0.05		
	T2	0.2576		
	T3	0.2576		

Table 7. 17 Boxplot of FOIS-It assessed in different period at T0; T1; T2; T3.

Analysis of FOIS-it



7.2.4 Self-rating of saliva

At T0, saliva control for all participants was assessed using the ROMP-Saliva scale (Kalf et al., 2011). Participants had a mean score of 22.8, suggesting the symptom required “special attention”. The ROMP-Saliva scale scores declined in a month at T1 before intervention, reaching a mean score of 26 (Table 7.18) suggesting a deterioration in drooling symptoms.

Table 7. 18 ROMP-Saliva mean and SD scores at different assessment timepoints for all participants (N = 10)

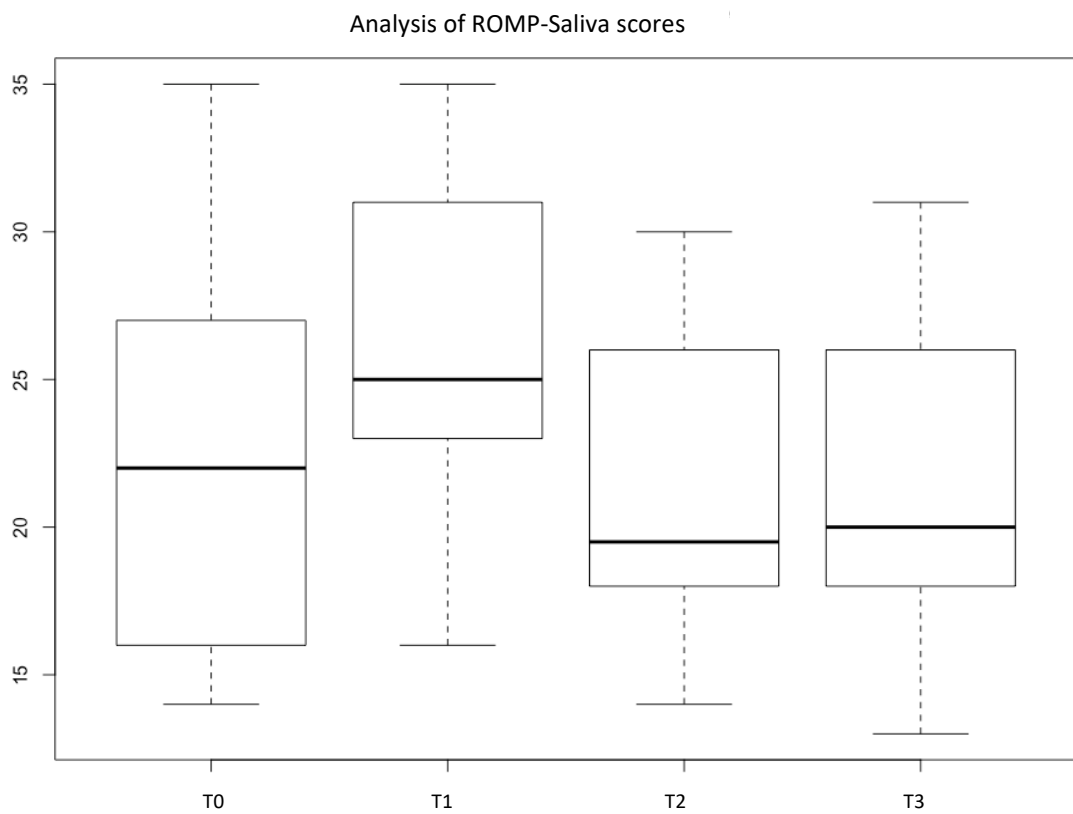
ID	ROMP-Saliva at T0	ROMP-Saliva at T1	ROMP-Saliva at T2	ROMP-Saliva at T3
1P	16	32	28	29
2P	29	30	22	20
3P	35	35	30	31
4P	22	30	23	24
5P	25	27	26	26
6P	32	23	19	19
7P	21	31	15	17
9P	15	16	14	17
10P	23	23	18	20
12P	32	32	20	28
Mean	22.8	26	21.2	21.7
SD	8.7	5.9	5.3	5.7

At the post treatment assessment (T2), the ROMP-Saliva scores showed a decrease from 26 (T1) to 21.2 (T2), although this reduction was not statistically significant ($p > 0.05$) (Table7.18). These changes were maintained at the follow-up assessment (T3) (Tables 7.18,7.19 and 7.20), indicating that the symptoms were mild in severity.

Table 7. 19 ROMP-Saliva statistical analysis.

	SHAPIRO-WILK		BARTLETT	KRUSKAL-WALLIS	ANOVA
ROMP-SALIVA	T0	0.6465	0.8579		0.3564
	T1	0.7872			
	T2	0.581			
	T3	0.7494			

Table 7. 20 Boxplot of ROMP-Saliva assessed in different periods at T0; T1; T2; T3



7.2.5 Quality of life

The first quality of life assessment at T0 using the I-SWAL-QOL (Ginocchio et al., 2016) revealed that the total mean score for all 10 participants was 135±27.4, indicating a slight negative effect of swallowing difficulties in the quality of life of all participants (Table 7.21). The overall score of the quality of life assessment did not change significantly across all the four assessments respectively T0=135; T1=133; T2=145 and T3=137 (Table 7.22). There was a small increase after treatment, respectively 145 at T2, but this was not retained at the follow-up assessment (137 at T3) (Table 7.21). Only the sub-part of quality of life associated with the food selection (I-SWAL-QOL 5) showed a statistically significant change after treatment ($p < 0.05$) and this was retained at the follow-up assessment (Tables 7.21;7.22; 7.23,7.24).

Table 7. 21 Mean and SD of I-SWAL-QOL at different times T0; T1; T2; T3.

Time	I-SWAL-QOL											
	Tot	Bur-den	Eating Duration	Eating Desi-re	Symp-toms	Food Selection	Commu-nication	Fear	Mental Health	Social Function-ing	Fatigue	Sleep
T0	135± 27.4	6±1.6	18±3.3	37±7.8	6±2.8	6±1.3	14±4	15±2.8	16±2.8	14±5.4	2±2.8	2±2.7
T1	133± 28.7	6±1.6	17±3.6	36±7.9	6±2.8	6±1.4	14±4.2	14±2.7	15±2.5	14±5.2	2±2.9	2±2.6
T2	145± 24.6	6±1.5	20±3.2	38±8.1	8±1.7	8±1.2	14±3.2	15±1.6	19±3.4	14±5.1	2±2.8	2±2.7
T3	137± 23.9	6±1.7	19±2.9	37±6.3	6±2.4	7±1.8	14±2.3	15±2.2	17±2.8	14±4.8	2±2.6	2±2.8

Table 7. 22 Scores of the I-SWAL-QOL at T0; T1; T2; T3 assessment time (N = 10) .

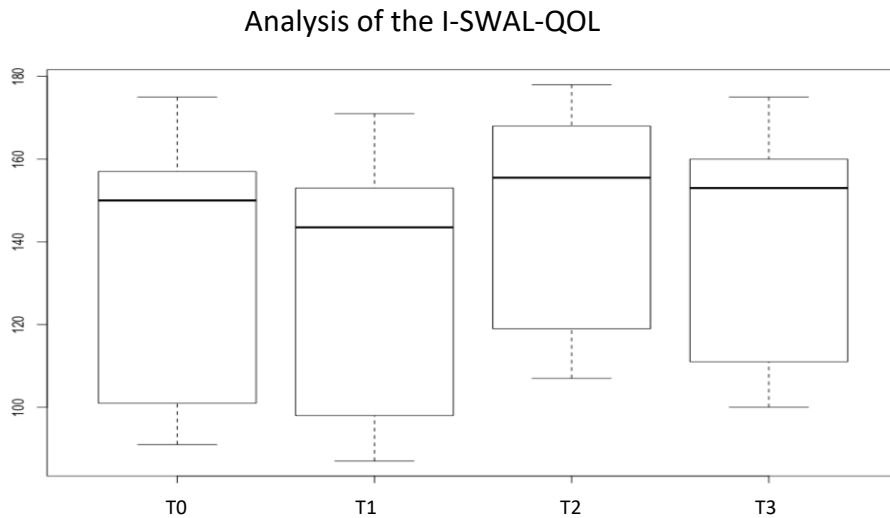
ID	I-SWAL-QOL T0											
PART	Burden	Eating Duration	Eating Desire	Symptoms	Food Selection	Communication	Fear	Mental Health	Social Functioning	Fatigue	Sleep	TOT
1P	9	23	46	8	7	19	20	23	15	2	3	172
2P	4	15	27	4	4	9	12	11	11	2	2	101
3P	6	19	34	10	6	20	15	18	17	2	2	149
4P	6	20	48	9	8	16	17	20	16	2	2	164
5P	5	18	44	8	6	15	16	21	16	2	2	153
6P	3	12	25	3	5	10	14	8	10	2	1	93
7P	7	20	42	9	6	17	19	19	15	2	1	157
9P	5	17	38	3	5	11	12	10	11	2	1	115
10P	6	21	40	5	6	18	14	21	16	2	2	151
12P	4	13	28	2	3	9	11	9	9	2	1	91
ID	I-SWAL-QOL T1											
1P	9	23	45	8	7	18	20	21	15	2	3	168
2P	4	15	25	4	4	9	12	11	10	2	2	98
3P	6	17	32	10	6	20	15	16	17	2	2	143
4P	6	18	47	9	8	16	17	17	16	2	2	158
5P	5	18	42	8	6	15	13	17	16	2	2	144
6P	3	12	24	3	5	10	13	8	10	2	1	91
7P	7	20	40	9	6	17	17	19	15	2	1	153
9P	5	16	35	3	5	10	12	10	11	2	1	110
10P	6	19	40	5	6	18	14	21	16	2	2	149
12P	4	10	27	2	3	9	11	9	9	2	1	87
ID	I-SWAL-QOL T2											
1P	9	24	46	8	9	19	20	23	15	2	3	168
2P	4	20	27	7	8	9	12	17	11	2	2	98
3P	7	21	34	10	6	20	15	22	17	2	2	143
4P	6	20	48	11	10	16	17	20	16	2	2	158
5P	5	18	44	8	8	15	16	21	16	2	2	144
6P	3	16	25	6	5	10	14	15	10	2	1	91
7P	7	21	42	9	12	17	19	22	17	2	1	153
9P	5	22	43	6	7	11	12	13	11	2	1	110
10P	6	21	40	7	8	18	14	22	16	2	2	149
12P	6	15	32	4	6	9	11	14	9	2	1	87

ID	I-SWAL-QOL T3											
	PART	Burden	Eating Duration	Eating Desire	Symptoms	Food Selection	Communication	Fear	Mental Health	Social Functioning	Fatigue	Sleep
1P	9	23	46	8	7	19	20	23	15	2	3	172
2P	6	18	28	6	6	9	12	11	11	2	2	111
3P	7	19	35	10	6	20	15	20	17	2	2	153
4P	6	20	48	9	9	16	17	20	16	2	2	165
5P	5	18	44	8	6	15	16	21	16	2	2	153
6P	3	14	26	3	5	10	14	12	10	2	1	100
7P	7	23	42	9	6	17	19	19	15	2	1	160
9P	5	18	38	3	5	11	12	13	11	2	1	119
10P	6	24	40	5	6	18	14	25	16	2	2	158
12P	4	17	30	2	5	9	11	11	9	2	1	101

Table 7. 23 I-SWA-QOL analysis.

	SHAPIRO-WILK	BARTLETT	KRUSKAL-WALLIS	ANOVA
I-SWAL - QOL Burden	T0	0.6833	0.9999	0.6116
	T1	0.6833		
	T2	0.8817		
	T3	0.8817		
I-SWAL - QOL Eating Duration	T0	0.743	0.7933	0.1083
	T1	0.9223		
	T2	0.4816		
	T3	0.4505		
I-SWAL - QOL Eating-Desire	T0	0.399	0.9953	0.7305
	T1	0.3953		
	T2	0.3354		
	T3	0.5864		
I-SWAL - QOL Symptoms	T0	0.1485	0.6826	0.5939
	T1	0.1485		
	T2	0.9706		
	T3	0.2475		
I-SWAL - QOL Food Selection	T0	0.7316	0.3921	< 0.05*
	T1	0.7316		
	T2	0.7376		
	T3	< 0.05		
I-SWAL- QOL Communication	T0	0.1671	1	0.973
	T1	0.08402		
	T2	0.1671		
	T3	0.1671		
I-SWAL- QOL Fear	T0	0.6622	0.9972	0.8847
	T1	0.3652		
	T2	0.6622		
	T3	0.6622		
I-SWAL- QOL Mental Health	T0	0.06422	0.6653	0.2386
	T1	0.1853		
	T2	0.07453		
	T3	0.1392		
I-SWAL -QOL Social Functioning	T0	0.05261	0.998	0.9696
	T1	< 0.05		
	T2	< 0.05		
	T3	0.05261		
I-SWAL-QOL Fatigue	T0	< 0.05	1	1
	T1	< 0.05		
	T2	< 0.05		
	T3	< 0.05		
I-SWAL- QOL TOT	T0	0.1012	0.945	0.4908
	T1	0.1533		
	T2	0.19		
	T3	0.07966		

Table 7. 24 Boxplot of I-SWAL-QOL at different times at T0; T1; T2; T3.



7.3 Impact of Intervention on swallowing outcomes over time

All the participants (N=10) underwent to the follow-up assessment at 3 months (T3).

The results are described according to the outcome measures.

7.3.1 Penetration and Aspiration

PAS scores did not change at the follow-up assessment for all 10 participants, the mean PAS score was 3 for all the swallowing trials in both T2 and T3 time points. This indicates that the swallowing function of participants did not deteriorate in the 3 months (Tables 7.1).

7.3.2 Pharyngeal residue

No difference in residue for saliva and solid food (IDDS 7) were detected in the valleculae between T2 and T3 assessments, confirming that the improvements were maintained over 3 months (Tables 7.3; 7.13).

The residue on saliva in the pyriform sinus slightly diminished after treatment (mean of 1.3 at T2), although the change was small, it was maintained also at the follow-up assessment (mean of 1.5 at T3) (Tables 7.3;7.4). Whereas, pooling of solid food (cracker) in pyriform sinus was slightly increased at the follow-up assessment, (score 1.4 at T2 to score 2 at T3) (Tables 7.12; 7.13), indicating that the improvements was

not maintained after treatment.

In addition, the small decrease of residue for water in the valleculae after treatment was retained at the follow-up assessment (Tables 7.6; 7.7).

Interestingly, the valleculae residue on yogurt decreased further after treatment. The mean scores during swallowing trials on yogurt diminished from 1.9 at the post treatment assessment (T2) to 1.7 at 3 months follow up assessment (T3), though this difference was not statistically significant (Tables 7.9,7.10).

7.3.3 Method of Oral intake

The positive changes of the modality of food intake (FOIS-It: 5.5 ± 0.8) at the assessment post-treatment (T2) were maintained at the follow-up assessment (T3) (FOIS-It: 5.5 ± 1.1). The mean score 5.5 at T3 confirmed that the participants were on food oral diet requiring only some special preparation (Tables 7.15; 7.16; 7.17).

7.3.4 Self-rating saliva

The self-rating drooling scores, which were slightly reduced after treatment (T2) (ROMP-Saliva: 21.2 ± 5.3), were maintained at T3 (ROMP-Saliva: 21.7 ± 5.8), indicating that the severity of perception of drooling was mild (Tables 7.18; 7.19; 7.20).

7.3.5 Quality of life

The total scores of I-SWAL-QOL showed a small decrease in the follow-up assessment from 145 ± 24 (T2) to 139 ± 23 (T3) (Table 7.21). Nevertheless, this reduction was not statistically significant ($p > 0.5$). The scores of sub-parts of the I-SWAL-QOL related to the food selection, which showed significant enhancement after treatment did not changed significantly at T3, indicating a retention of the quality of life related to the food selection (Tables 7.22; 7.23).

7.4 Adverse events

No adverse events occurred during the treatment protocol and the outcome measure did not show an increase or deterioration of swallowing symptoms. Three participants (3P; 4P; 10P) reported fatigue after treatment because it was considered an intense and demanding intervention. This feedback is described further in Section 7.6.

7.5 Adherence to treatment

Two participants decided to stop the treatment during the first treatment week because they thought that their swallowing disorders were not so severe to require an intensive intervention, as it was described in Chapter section . The remaining participants attended the clinic for treatment following a specific timetable. Two participants asked to change the time of treatment due to transport issues. No other issues were revealed during the 4 weeks of treatment. In addition, the participants who completed the treatment, underwent to the follow-up assessment (T3).

7.6 Qualitative analysis of the feedback from participants on intervention

This data was obtained from the interviews after each treatment sessions (Section 5.11.1). The data from the transcriptions (Appendix L) were grouped into thirteen basic themes on data analysis. These basic themes were grouped into five broad “Organizing Themes”(Braun & Clarke, 2012). These organizing themes were: ‘Benefits on swallowing’; ‘Benefits on swallowing related function’; ‘Feedback on intervention’ and ‘Adverse effects’ (Figure 7.1).

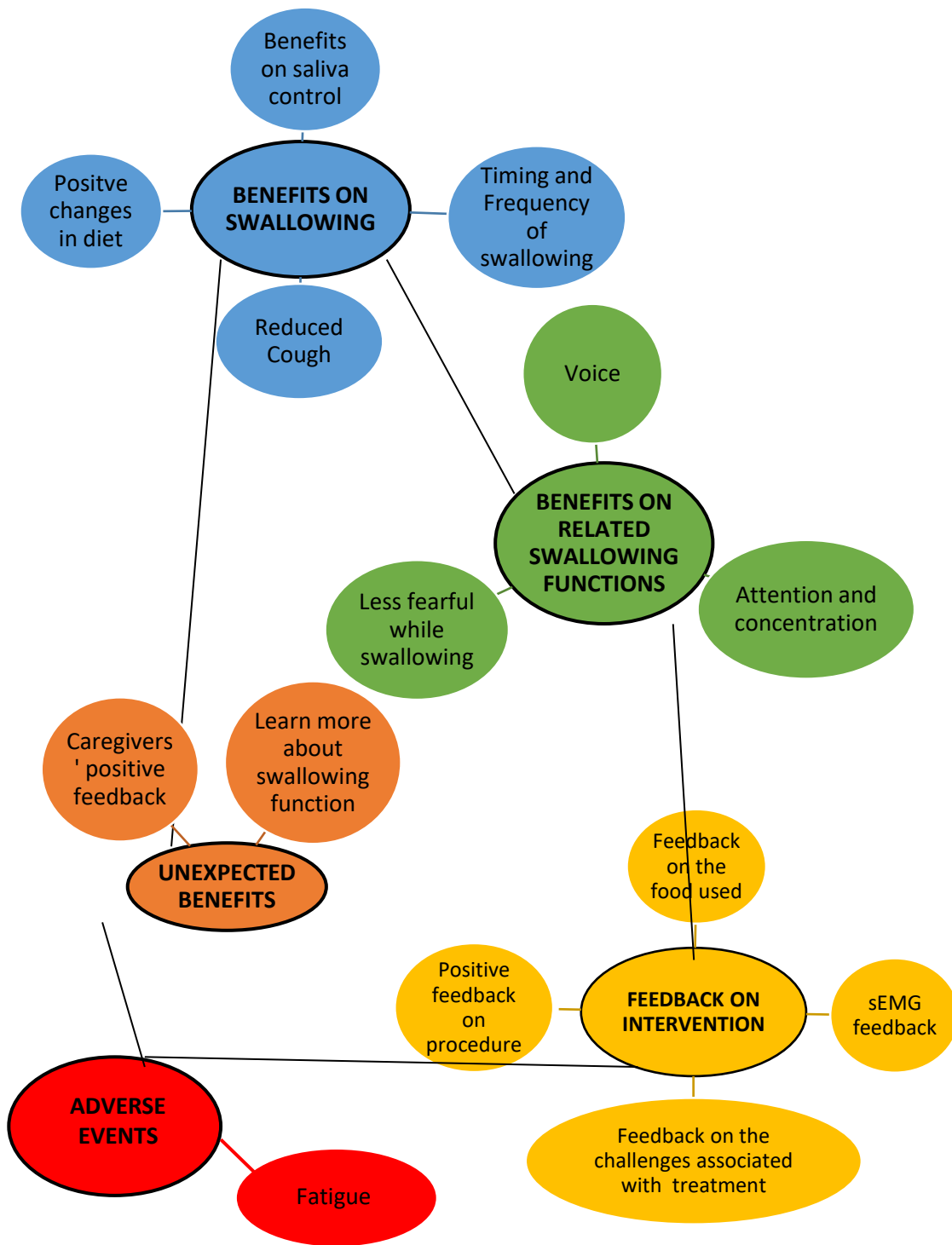


Figure 7. 1 The organizing themes and the basic themes

7.6.1 Benefits on swallowing

This organizing theme embraced all the benefits on swallowing that participants reported after treatment. It included improvements on the changes in diet, saliva control, the timing and frequency on swallowing, and the reduction of cough episodes during meals (Figure 7.2).

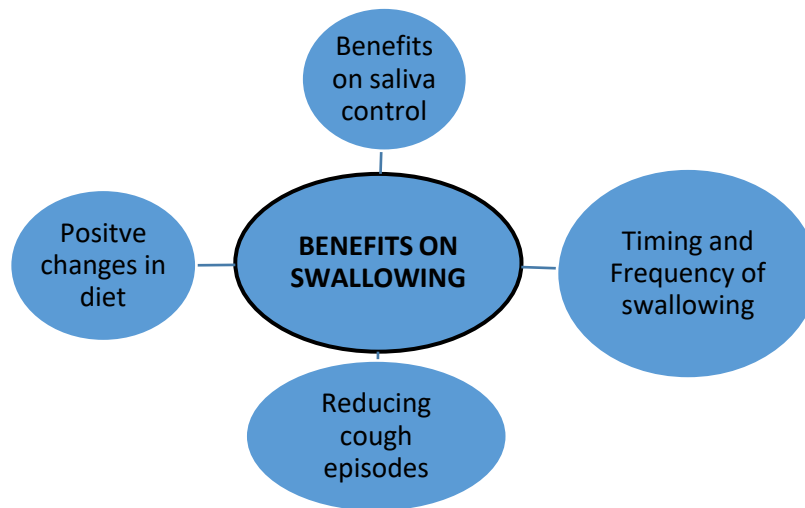


Figure 7. 2 The organising Theme: Benefits on Swallowing

➤ *Positive changes in diet*

This basic theme regarded the modifications of modality of food intake. Four participants (3P,4P,7P,11P) reported positive changes in diet modification. Two of them experienced improvements in solid food swallowing and two in liquid consistencies

Extract 7.1

"I had a piece of fried breast of chicken [...] it was almost 2 years since I had one" (6P).

"I returned to drinking espresso coffee" (7P).

➤ Benefits on saliva control

This basic theme concerned the improvements in saliva management. Five participants (1P,5P,6P,9P,10P) referred to an increased control of saliva swallowing also during non-swallowing related activities

Extract 7.2

"I am not losing so much saliva and I could read the newspaper without make it wet" (1P).

"If I feel saliva coming out, I swallow it several times." (5P).

➤ Timing and frequency related of swallowing

In this theme, two participants commented on improvements in the speed and rate of swallowing. One participant (5P) reported that she was not eating as slow as usual and the other (6P) said that he swallowed more frequently

Extract 7.3

"I've learnt that I have to swallow saliva often" (5P).

"I was eating not really slow at usual" (6P).

➤ Reducing cough episodes

After the intervention, three participants (4P,6P,7P) experienced a reduction in coughing episodes during meals

Extract 7.4

"The good things are that I am not coughing so often [...]" (4P).

"[...] no cough episodes" (6P).

7.6.2 Benefits related to swallowing function

In this organizing theme, improvements in function associated with swallowing were grouped together. This theme comprised the following three basic themes: voice changes, less fear while swallowing, and improved attention and concentration (Figure 7.3).

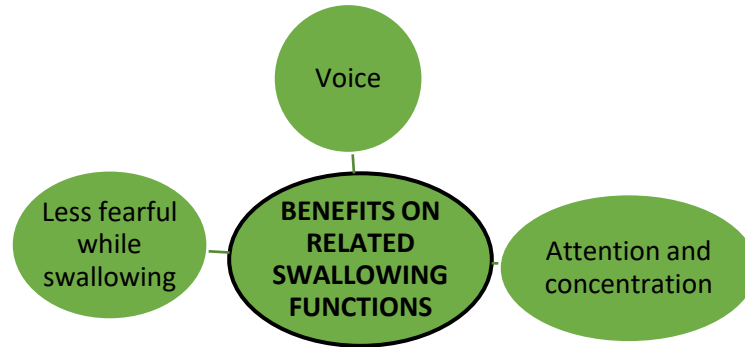


Figure 7. 3 The organising Theme: Benefits on related swallowing function

➤ Voice changes

Three participants (3P; 6P) reported benefits on voice production after this intervention.

Extract 7.5

"I am not sure if it is important for you but I also feel able to speak better and louder" (3P).

"I think I have a better voice [...] it is more clear I guess" (6P).

➤ Attention and Concentration

Five participants (1P,2P,5P,7P 10P) reported that the treatment increased their attention skills during eating.

Extract 7.6

"I think [...] I think I am more focused when I am swallowing" (1P).

"I have to say that I am pleased to see that I am swallowing with more attention [...]" (10P).

➤ Less fearful while swallowing

This basic theme comprises comments regarding the reduction of fear during meals. Two participants (3P,4P) said they felt less scared of eating after treatment

Extract 7.7

"I am not scared anymore while I am eating" (4P).

"But eventually I did not have any panic attacks" (3P).

7.6.3 Unexpected positive feedback

In this theme, participants made on unexpected benefits of the intervention by caregivers are included. These comments were unsolicited and given spontaneously from participants. Two basic themes form this group (Figure 7.4).

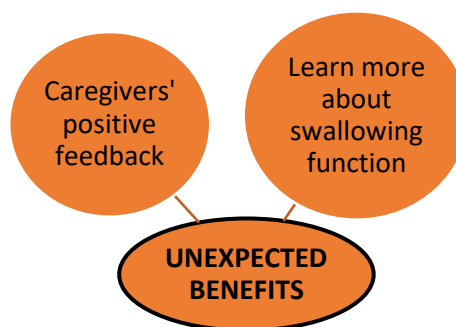


Figure 7. 4 Organising Theme: Unexpected Benefits

➤ Learning about swallowing function

Four participants (2P; 3P; 5P; 7P) mentioned that they acquired new swallowing skills

Extract 7.8

"I didn't know that I could swallow so many times" (7P).

"I think that my fellows with PD should know that it is important do exercises." (3P).

➤ Caregivers' feedback

The caregivers of two participants (6P; 9P) made comments that are reported by participants.

Extract 7.9

"My husband has noticed that I was eating not really as slow as usual and no cough episodes" (6P).

"My daughter said that I do not lose saliva so often and she thinks that I am speaking better" (9P).

7.6.4 Feedback on intervention

This organizing theme combined feedback related to delivery of the intervention and the sEMG equipment. Four basic themes emerged from the data: positive feedback on the food used; feedback on the treatment itself; positive feedback on the procedures and feedback on the sEMG delivery of intervention (Figure 7.5).

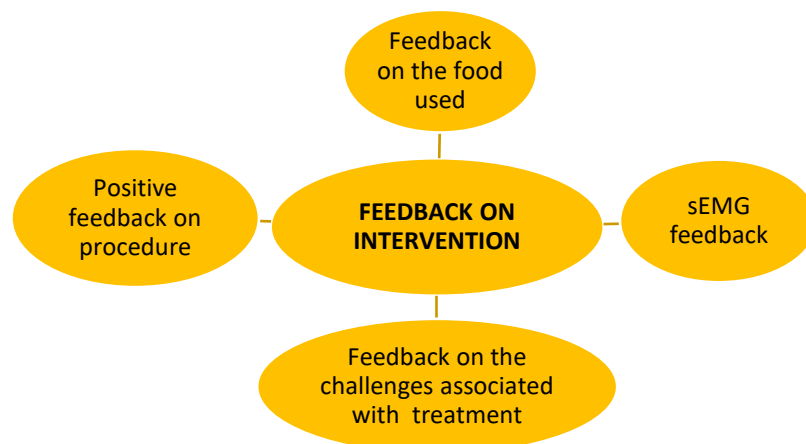


Figure 7. 5 Organising Theme: Feedback on intervention

➤ *Feedback on the food in the intervention*

Two participants (1P,3P) commented on the use of yogurt during treatment. Both comments were positive.

Extract 7.10

“It was better to swallow with yogurt instead of saliva” (1P).

“It is better to swallow with yogurt. I feel it going down (3P).

➤ *Feedback on the challenges associated with treatment*

Five participants (4P, 5P, 7P, 9P, 10P) commented on how the intervention was delivered. Five participants complained about the intensity of treatment schedule and difficulties encountered.

Extract 7.11

*"I haven't thought it was so intense [...] really intense" (7P).
"Well, it was not really easy to come everyday" (5P).*

➤ Positive feedback on procedures

This theme incorporated all the positive comments regarding the modality of treatment delivery. Seven participants (1P,2P,3P,4P,5P,9P,10P) described the positive aspects and their satisfactions with the treatment procedures.

Extract 7.12

*"It pushes to swallow and swallow and swallow" (10P).
"hmmm I enjoy coming here" (2P).*

➤ Feedback on sEMG

This theme encompasses all the comments made on sEMG equipment, including the swallowing signals, the device itself and software used. Among the six participants who reported comments on sEMG, 3 participants (1P,4P,9P) remarked on the entertainment features of the software and 3 participants (5P,6P,7P) described the issues with the signal responses and the electrodes used.

Extract 7.13

*"Sometimes I have lost the signals on the screen [...]" (9P).
"It likes a game [...] it is fun" (5P).*

7.6.5 Adverse Events

This organizing theme provided important information on the drawbacks of the treatment for people. With IPD and there was just one basic theme: Fatigue.

➤ Fatigue

Three participants (3P, 4P, 6P) reported that they were tired after treatment because the treatment was too intense

Extract 7.14

*"Well no doubts that it was intense and demanding treatment" (4P).
"After this treatment sometimes, I feel tired" (3P).*

7.7 Suggestions from participants

This next section provides the qualitative data collected at the 3-month follow-up assessment. Participants were requested to complete an anonymous feedback form, described previously in Chapter 5 (section 5.5.3).

Firstly, participants were asked to rate sEMG treatment procedure on a 4 point scale. The options were ‘very uncomfortable’, ‘uncomfortable’, ‘comfortable’, ‘very comfortable’). All ten participants selected the “Very Comfortable” box on the written form.

Secondly, to the open question: *“Would you like to recommend any changes?”* participants had to provide suggestions for enhancing the treatment. While the written data was at times illegible due to micrography and tremor, the legible answers were transcribed and analysed following the thematic analysis as described in Chapter 5 (section 5.11.1). Four main themes emerged: Treatment extension; Reduction in treatment sessions; telerehabilitation; inclusion of non-swallowing treatment. The comments below cannot be attributed to specific participants as the feedback was given anonymously.

7.7.1 Treatment extension

Four participants made suggestions that relate to their desire to continue treatment after 1 month.

Extract 7.15

*“It is a pity that it is finished, after 3 months I had forgotten everything”
“I would like to continue the treatment, because one month went by so quickly”.*

7.7.2 Reduction in treatment sessions per week

Three participants made recommendations on a reduction in the amount of treatment sessions. Two participants made this suggestion because it was difficult to come every day.

Extract 7.16

"I would recommend to schedule the treatment 3 times per week as it is difficult to come every day".

"I recommend to diminish the sessions per week for example 2 times per week".

"I would suggest reducing the sessions. For me it is a problem to come every day to the hospital".

7.7.3 Telerehabilitation

Two participants suggested the use of technology to deliver the rehabilitation programme at home.

Extract 7.17

"I recommend doing the treatment using Skype or internet, so I do not need to come every day".

"I suggest trying to do the treatment in telerehabilitation as the neurologist mentioned last week in a conference of our association".

7.7.4 Inclusion of non-swallowing treatments

This theme involved advice on augmentation of the treatment protocol to include non-swallowing related exercises. Two participants suggested the addition of a voice rehabilitation programme in order to communicate better. One participant recommended that the sessions be combined with physiotherapy treatment sessions.

Extract 7.18

"I want to suggest adding also the treatment for the voice in order to speak louder".

"I recommend combining swallowing exercise with the exercise for breathing and speaking".

7.8 Summary of the results

It was hypothesised that this intervention would result in an improvement in swallowing with a reduction in aspiration events and decreased residue. While the intervention did not produce a statistically significant change in the

penetration/aspiration measures using PAS in any of the FEES assessments, nevertheless, using YPR-SRS during FEES the results found statistically significant differences on saliva and solid food residue in the valleculae after treatment in this sample of people with IPD and dysphagia.

The second hypothesis was the intervention would increase swallowing ability and improve oral intake. This study confirmed this hypothesis. The results showed that this treatment had significant positive effects on improving oral food intake using FOIS-It in this sample of people with IPD and dysphagia. In addition, the improvement in dietary changes was confirmed by the qualitative reports during the post treatment interviews.

The third hypothesis was that this intervention would have some effects on the self-perception of drooling. The treatment reduced self-perception of saliva and some participants reported in the qualitative data that they had more control of saliva. Nevertheless, on the ROMP-Saliva questionnaire the decrease in drooling was not statistically significant at post treatment assessment.

The fourth hypothesis was that improvements in swallowing skills would positively affect the quality of life of this cohort of people with IPD. Overall quality of life increased slightly after treatment. Nevertheless, it was not statistically significant and any changes were not maintained at the follow-up assessment. The Food Selection sub-section of the I-SWAL-QOL was the only component which increased significantly after treatment.

The final hypothesis was that improvements in swallowing following this intervention in a person who was medically stable would not significantly decline at 3 months follow-up assessment. The statistically significant improvements in reduction of saliva and solid food residue in the valleculae and the positive diet changes were retained at the follow-up assessment. Nevertheless, the non-significant positive changes after treatment were not maintained at the follow-up assessments.

The sixth research question aimed to investigate any potential adverse events associated with the specific sEMG biofeedback intervention in people with dysphagia and IPD. No adverse events occurred during the treatment. However, some participants commented on feeling tired and experiencing fatigue after treatment.

The last research question concerned the acceptability of sEMG biofeedback treatment to people with IPD and dysphagia. Two participants dropped out at the beginning of the treatment, because the treatment was considered to be too intense, and they believed that their swallowing difficulties were not severe enough to justify the treatment. The participant interviews post-treatment revealed that the intervention was broadly acceptable to those who continued with the intervention. Most qualitative reports included benefits in the related and non-related swallowing function, positive feedback in the procedures and in the use of the sEMG software. Important information for study design and delivery was provided in this section of the study and is considered further in the final two chapters

Chapter 8: Discussion

8.1 Introduction

This chapter discusses the findings obtained from the feasibility study, highlighting the strengths and limitations, which will inform future study as well as clinical practice.

This chapter is divided into 8 sections. The first sections discuss the results from the quantitative component of the study and long-term effects of the intervention approach comparing the findings with the evidence from the literature. The remaining sections consider the qualitative findings and the final sections highlight the strengths and limitations of the study.

8.2 Changes in swallow function based on instrumental test findings

The results from FEES showed that the sEMG biofeedback swallowing intervention improved swallowing in this group of people with IPD and dysphagia, leading to substantial and consistent modification of the oral intake and reducing pharyngeal residue. Nevertheless, there were some outcomes that remained unchanged at the end of the intervention.

8.2.1 Laryngeal penetration and aspiration

All participants in this feasibility study did not aspirate at any of the four FEES assessments, suggesting that this cohort had mild dysphagia. It has been hypothesised that the absence of severe dysphagia with aspiration in this sample could be attributed to selection criteria of this study. The absence of cognitive impairments and the recruitment process based on neurological assessment might have contributed to selecting participants who had mild disease (2-3 level H&Y Scale). Further larger studies should aim to recruit a larger sample size that will allow for sub group analysis according to severity of dysphagia and IPD disease.

One could argue that the participants did not aspirate during assessment but may aspirate at other times but it is worth noting that the PAS scores did not reveal any clinical or statistical significant difference after intervention, reflecting perhaps the stability of this parameter.

Penetration slightly decreased after the intervention programme nevertheless no other changes were observed. No deterioration in aspiration or penetration suggests that the intervention is safe for IPD participants. In addition, the stability of the results at the four assessments showed that this assessment is consistent and reliable in the IPD sample.

8.2.2 Saliva management

It is well recognised that saliva secretions associated with anterior drooling and posterior pharyngeal secretions are common symptoms of IPD, affecting up to the 70% of IPD population (Kalf, de Swart, et al., 2012). As described in Chapter 1, anterior drooling in people with IPD is caused by facial hypotonia, poor head posture and inefficient lip seal associated with an infrequent swallowing (Miller, 2017; Miller et al., 2019). Posterior leakage of saliva is likely due by infrequent swallowing acts associated with oral-pharyngeal hypotonia and proprioception deficits (Miller et al., 2019; Srivanitchapoom et al., 2014).

Most of the assessments for drooling in this population are focused on anterior drooling using questionnaires (Miller et al., 2019). In this study, the scores of self-perceived drooling using the ROMP-Saliva scale, slightly decreased after treatment, although the findings were not statistically significant. This finding did not prove the initial hypothesis, as it was expected a significant improvement of drooling at the ROMP-Saliva scale but perhaps with a larger less homogenous population, different results may be seen. By contrast to the ROMP-Saliva scale findings, the qualitative assessment of the interviews showed that five participants referred to the fact that they noticed decreased drooling with better control of saliva, revealing that the treatment had an important effect on anterior saliva control.

The discrepancy between the ROMP-saliva results and the participants' feedback could be attributed by the choice of the saliva rating scale. Recently McNaney et al. 2019 have found the ROMP-Saliva did not detect statistically significant improvements, which were observed using visual analogue scales (VAS) in people with IPD. The authors suggest that ROMP-Saliva is not reliable or sensitive to capture the changes

over the intervention period in this population (McNaney et al., 2019). Given that this feasibility included only the ROMP-Saliva, it was not possible to draw the same assumptions. Nevertheless, it could be argued that more objective drooling assessments sensitive to change over time are fundamental in studies with people with IPD.

It is important to underline that this study was the first study, which documented positive effects of biofeedback swallowing treatment in reducing posterior drooling using assessment (FEES). Although the YPR-SRS was not specifically validated to assess secretions (Neubauer et al., 2015), it showed a statistically significant reduction of pooling of saliva in the vallecula after treatment and this positive finding was maintained also at the 3 months follow-up assessment.

The improvements in the anterior drooling and the reduction of saliva pharyngeal residue in this study were likely to be attributed to the increased frequency of saliva swallowing and the use of sEMG biofeedback. In the literature, several studies confirmed that the augmentation of saliva frequency produced a decrease of drooling (Carnaby et al., 2019; Miller et al., 2009; Miller et al., 2019). McNaney et al. trialled the use of a wrist-worn digital cueing device, which produces vibratory feedback in order to remind IPD participants to swallow often saliva (McNaney et al., 2019). Although they assessed only the severity of anterior drooling using a self-perceived questionnaire; they found positive results in the perceived severity and in the frequency of drooling. Based on these results, it is suggested that the increased number of swallows prompted by sEMG biofeedback in this feasibility study played a key role in the improvement of saliva management in this IPD sample. Furthermore, it is assumed that sEMG biofeedback was fundamental to direct the participants' attention to the saliva swallowing as well as to increase awareness of the swallowing control.

8.2.3 Pharyngeal residue

A positive reduction in pharyngeal residue after intervention was documented for all the food trials on FEES assessment. In the following sections, the results of pharyngeal

residue are discussed based on food trial: solid food (IDDSI 7); yogurt (IDDSI 3) and water (IDDSI 0).

8.2.3.1 Solid food (IDDSI 7)

The solid food residue were higher than water and yogurt residue in the study sample at the four assessments (T0;T1;T2;T3), indicating an increase of difficulty of swallowing this food consistency. The findings showed a statistically significant decrease of pooling of solid in both valleculae and piriform sinus after intervention, suggesting an effect of sEMG swallowing treatment in oral-pharyngeal clearance of solid food ingestion.

Solid injection difficulties included also swallowing of medication, which are critical to the management of IPD symptoms. In the present study, FEES assessments at T0 and T1 prior to intervention noticed traces of medications in the valleculae in 3 participants. This is not unusual. A recent study confirmed that pharyngeal residue with medication occurred in up to 28% of people with IPD, altering drug therapy effects (Buhmann, Bihler, et al., 2019). Of note, post-treatment FEES assessments in this study showed no medication residue for any participants, suggesting a possible direct effect of sEMG swallowing treatment in medication swallowing. Although this improvement may be not be retained for all participants at 3 months, as in this study, but it remains an important finding as it suggests a potential contribution of this intervention in the medication swallowing and pharyngeal clearance. All of these can contribute to positive effects on the health and well-being of the participants.

8.2.3.2 Yogurt (IDDSI 3)

The pharyngeal score residue during yogurt (IDDSI 3) swallowing trials showed the least amount of pooling among the food trials and did not differ greatly in the four FEES assessments. This could be due by the fact that yogurt was a safe food consistency for the IPD sample. These results are in line with Troche et al. and Newman et al., indicating that thicker consistencies such as yogurt were safer for people with IPD because of the viscosity, supporting the choice of using this food during the treatment protocol (Newman et al., 2016; Troche et al., 2008).

8.2.3.3 Liquids (IDDSI 0)

The results showed a slight improvement of pharyngeal residue during water swallowing after intervention, although these improvements were not statistically significant. Liquids are recognised to be the major threat for aspiration in IPD population (Gaeckle et al., 2019; Nienstedt et al., 2018; Warnecke et al., 2016). In this study, very little residue on liquids was observed. This finding is in contrast with previous studies which documented substantial liquid residue in people with IPD (Gaeckle et al., 2019; Nienstedt et al., 2019; Warnecke et al., 2016).

8.2.4 Positive changes to diet

Changes to oral intake in this present study might suggest that the swallowing sEMG biofeedback intervention has potential to make a substantial improvement to everyday life. All of the participants were on an oral diet, the majority of them ate one single consistency food, which was semisolid (IDDSI 3). After treatment almost all participants reported that they could take food of different consistencies requiring special preparation.

In this study FOIS-it showed some limitation, it is recognised to be a valid tool to detect the swallowing improvements associated with the use of sEMG biofeedback (Albuquerque et al., 2019; Carnaby-Mann & Crary, 2010; Crary et al., 2004). The levels of oral intake were too generic, and they were not sensitive to specific individual changes, which were reported in the interviews by participants.

In addition, it could be argued that FOIS scores could be due to placebo or education and not accomplished specifically by the biofeedback treatment. On the other hand, some participants reported that food changes were achieved by the treatment. Furthermore, some of the improvements impacted considerably the social life and were obtained in 3rd-4th weeks of treatment. If it was an effect of placebo they could occur also prior treatment or during the first week of treatment. For example, one participant described that he had better swallowing coordination which meant that he could drink an espresso, something which he could not drink for the past 3 years. Drinking espresso is an important social-cultural activity for Italian people who

typically have an espresso in a standing position at a coffee bar counter. The espresso is served in small cups, which increase the difficulties of drinking. People must tilt their head hold the coffee in their mouth and swallow it when the head returns to the normal position. This participant reported that he paid attention to the coordination of swallowing during coffee drinking, as he was doing during treatment with sEMG biofeedback. This achievement confirmed an important contribution of biofeedback in increasing the awareness of the swallowing act and in learning the swallowing motor sequence during coffee swallowing. In this case, the treatment led to the improvement of swallowing function and impacted enormously on the daily life of this participant.

In addition, other two patients reported benefits during the swallowing of solid food. One participant reported that he had attempted to eat pizza at home. Pizza belongs to a group of foods that are most difficult to swallow in IPD population, as the crunchy base associated with tomato sauce requires efficient mastication with lingual strength and coordinated actions of the oral-pharyngeal muscles. In these specific cases, the treatment may have improved the control and coordination of the swallowing pattern during solid food ingestion.

Of note, all these three participants mentioned that they paid attention to the way they were eating different food consistencies. It is suggested that sEMG biofeedback helped participants to direct their attention to swallowing tasks. This could potentially increase the awareness of participants on their swallowing skills and eventually make them confident to experiment with different foods.

8.2.5 Quality of life

The total scores of I-SWAL-QOL revealed that the intervention had no significant impact on the overall quality of life on the sample of participants in this study. This could be due by the mild severity of the sample, since Leow et al. found the IPD disease progression adversely affected QOL (Leow et al., 2010). Individuals with IPD in later and severe stages may experience greater reduction in the desire to eat, difficulties with food selection, and prolonged eating duration with overall lower SWAL-QOL scores than people at earlier stages of IPD in this study.

The overall scores of the QOL assessment using the I-SWAL-QOL changed marginally after treatment, but these changes were not statistically significantly different. Only the component that related to food selection (I-SWAL-QOL) showed a statistically significant improvement after treatment. The positive results on the Food Selection component were associated with the diet modifications and also with the increased choice in food to eat.

Two participants reported that they had increased the frequency and the speed of eating during meals. These findings are consistent with the results of Athukorala et al. They found the IPD participants after a skill swallowing intervention using sEMG reduced the time of swallowing during WST (Athukorala et al. 2016). Nevertheless, the current study did not measure swallow timing and further studies should consider this aspect as an outcome of intervention.

8.3 Maintenance of effects of sEMG biofeedback swallowing intervention in people with IPD and dysphagia

The reduction of saliva and pharyngeal residue on solids as well as the positive dietary changes were retained 3 months after treatment confirming that the swallowing skills learnt were consolidated and automatized into daily life in this time period.

It is important to highlight that the lasting effects of this study were found mainly for the outcome measures which showed statistically significant improvements immediately after treatment. For example, the reduction of saliva secretions and solid cracker residue in the vallecula were confirmed at the follow-up examination, whereas the minor changes on water residue were not found after 3 months. It could be suggested that the improvements of swallowing function, which contribute to substantial changes after treatment, were likely to be retained at the follow-up.

These important retention effects shed light on the possible contribution of the intervention in maintaining the swallowing function in this population, although the sample of this study was small. Several studies in physical rehabilitation showed that retention of motor improvements is significantly compromised in people with IPD (Abbruzzese et al., 2016; Heremans et al., 2016). Nevertheless, some recent research

showed that intensive treatment contributes to long term effects (Bouça-Machado et al., 2020; Ferrazzoli et al., 2018; Frazzitta et al., 2012; Ricciardi et al., 2016). Accordingly, there is a growing literature showing long lasting effects determined by intensive voice treatment in people with IPD (Körner Gustafsson et al., 2019; Ramig et al., 2018). In the field of dysphagia, few studies have investigated the long-term effects of an intensive behavioural treatment for improvement of swallowing function in IPD. Recently, Troche et al., studied the detraining effects of an intensive expiratory muscle strength training and found a significant retention effect on the safety of swallowing after 3 months post treatment in people with IPD and dysphagia (Troche, Rosenbek, et al., 2014). Based on these findings, the intensity of the current intervention may be an important contributing factor for the maintenance effects in people with IPD in this feasibility study and requires further investigation. The length of time for follow up also needs to be considered. This study examined retention of effects after 3 months but outcomes need to be measured also at 6, 12, 18 and 24 month periods. It is expected that training will not last long term given that these participants are also ageing more rapidly. Ramig et al found that some aspects of voice following LSVT® were retained 2 years after the intervention (Ramig, 2001). In future studies, it will be important to find the critical point at which detraining occurs and plan intervention programmes accordingly.

8.4 Adverse effects

This feasibility study did not show any adverse effects such as increased cough or choking episodes, presence of respiratory infection and/or increase in the progression of the disease.

Nevertheless, three participants reported that they were tired and fatigued because the treatment was intensive and demanding. Fatigue is common and disabling nonmotor symptom in IPD, which can manifest even during premotor stages of disease and limits participation in social activities, leading to impact the quality of life (Barone et al. 2009; Siciliano et al. 2018, Weintraub et al. 2011). In this study, the fatigue did not affect the completion of the intervention and it was only reported by 3 of the 10 participants. However, this aspect should be monitored in future studies, as it could

impact adherence to treatment. In addition, participants reported that they found it difficult to come daily to the hospital. This suggests that alternative means of delivering the intervention should be investigated in further studies. Two participants dropped out of the study at the beginning for this reason.

8.5 The positive effects on non-swallowing related functions in IPD participants

This feasibility study revealed unpredicted benefits of swallowing sEMG biofeedback intervention. Participants reported several positive effects also on non-related swallowing function indicating a transference effect of the intervention into other areas. The effects have been categorized in 3 areas: attention effects; effects on the fear of eating; effects on voice quality.

Half of participants reported an increase in attention and concentration during swallowing. Several authors found that IPD participants with mild cognitive and attention impairments presented worsening of swallowing safety during dual task conditions (Brody et al., 2012; Troche, Okun, et al., 2014). They hypothesised that a modified digit span task and swallowing tasks share the same neurological structures. This could partially explain benefits reported by the participants. Nevertheless, future studies should investigate this aspect in order to analyse the potential involvement of swallowing biofeedback treatment in enhancing overall swallowing attention .

Furthermore, the treatment using sEMG biofeedback seems to have an impact on reducing the fear of eating, as it was reported by two participants. Argolo et al. found that exercise reduced the fear of choking episodes during meals in IPD population. It is suggested that improvements in swallowing function could influence proprioception and self-confidence during meals.

Of note, three participants and one caregiver reported an improvement on the voice and speech quality of participants. Given that the treatment involved the orolaryngeal-pharyngeal structures, an increase in tone and mobility of these muscles might also have positive effects on voice quality and speech. This finding suggest that improvements in swallowing skills are transferred on other skills, presuming an effect

of the neuro-plasticity principle of transference. In line with this result, El Sharkawi et al. found that a similar but opposite result (El Sharkawi et al., 2002). An intensive treatment using feedback for increasing the speech volume (LVST) improved swallowing function. This explains a possible improvement of the voice quality after the swallowing treatment.

It is hard to define if these unexpected effects were exclusively achieved by this intervention, as some participants were doing physiotherapy and several social contextual components contributed to these improvements. Nevertheless, participants' feedback revealed improvements in fields, which should be investigated in future study using formal specific assessments.

8.6 Key contributors of motor Learning in this complex intervention

It is argued that designing the swallowing intervention following the motor learning principles was the main factor, which contributed to swallowing improvement in this feasibility study. Huckabee and Burnip summarized the motor learning approach applied for biofeedback dysphagia intervention, which were described in Chapter 2.4 (Huckabee & Burnip, 2018). In this section, the motor learning stages were applied to this feasibility study. The specific stages of implementation are described in Figure 8.1.

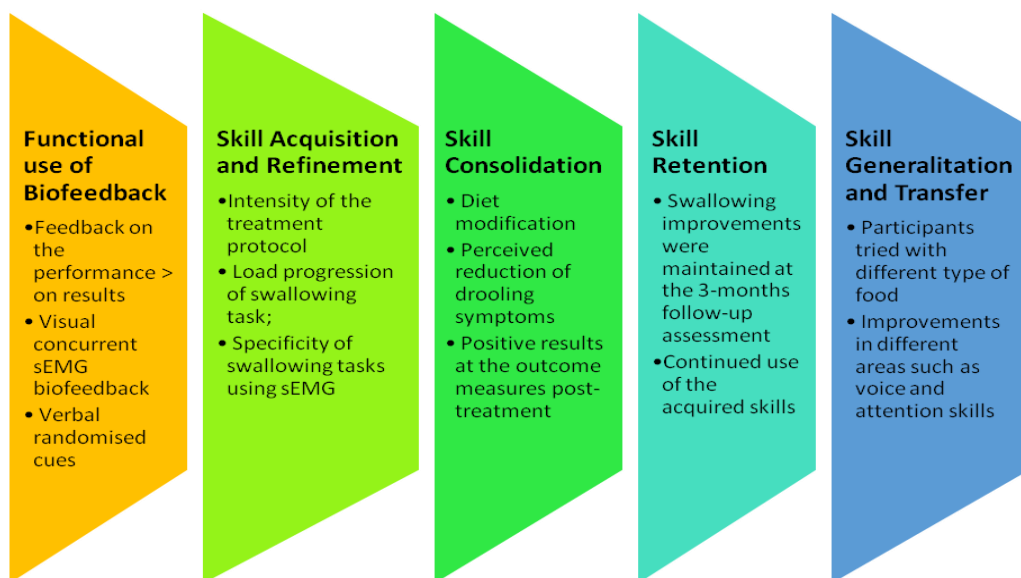


Figure 8. 1 Framework of the swallowing motor learning stages and contribution of this feasibility study

The first stage concerns the implementation of **functional swallowing tasks using biofeedback** (Fig 8.1). In the current study, the visual constant and simultaneous biofeedback of sEMG during swallowing compensates for the impairment of proprioception information, resulting in progressive improvement of pattern accuracy and stability during complex swallowing coordination pattern (Huckabee & Macrae, 2014; Maas et al., 2008; Shadmehr et al., 2010). The sEMG wave line allowed participant to focus on the submental muscles contraction and to modulate the strength and the timing of the contraction Furthermore, the use a game increased motivation and participants reported that it was an enjoyable task. The application of sEMG sensors revealed to be easy and fast procedure which does not require a high expertise to perform.

In addition, the verbal cues may have supported the improvements in this study. The important role of verbal cues is often underestimated in clinical and research field. Curtis et al. demonstrated that verbal cueing significantly affects respiratory-swallow patterning in people with IPD (Curtis & Troche, 2020). In the present study, the verbal feedback were delivered to increase the swallowing awareness and stimulate the coordination of muscles contraction during swallowing, as described in Chapter 5 (Section 5.10.2.8). In addition, the randomisation of delivering the verbal cues may increase the understanding of the swallowing motor pattern, consolidate the swallowing skills and boost the self-confidence during swallowing different foods.

The second stage of **skill acquisition and refinement** was accomplished by the intensity of the treatment protocol, load progression and specificity of swallowing tasks (Fig 8.1). As explained in Chapter 5, exercises must exceed the usual level of intensity in order to push the neuro-system beyond the accustomed level and to trigger the change (Burkhead et al., 2007). The effects of intensive treatment were well accepted for dysarthria recovery and in physiotherapy interventions (Bouça-Machado et al., 2020; El Sharkawi et al., 2002). Only recently, few studies found positive results in intensive swallowing treatment in people with IPD (Athukorala et al., 2014; Pitts et al., 2009; Wang et al., 2018). For this reason, it is suggested that the intensive schedule

of protocol treatment in the current study played a key role on the acquisition and the automatization of the swallowing pattern, although some participants dropped out from the study because of the intensity of the treatment protocol. This could be motivated also by the fact the swallowing impairments were not severe. It could be hypothesised that severe signs of dysphagia could increase the motivation to undertake an intensive dysphagia treatment.

Moreover, the use of sEMG biofeedback combined with saliva and yogurt swallowing tasks increased the specificity of the intervention and fostered the precision and accuracy of swallowing behaviour. The sEMG in this feasibility study was used to refine accuracy of muscles contraction without working on the strength of submental muscles. Participants were stimulated to recognize the appropriate swallowing movement pattern and adjust the contraction to meet the target. The positive results on reduction of pharyngeal residue suggest that the implementation of sEMG biofeedback in association with food trials contribute to increasing the swallowing coordination and pharyngeal clearance in the sample.

It is assumed that incorporating task specificity within a framework of adequate load, repetition and volume led to the **acquisition and consolidation** of the swallowing skills, which is the third stage (Fig 8.1). The positive effects on diet modification, pharyngeal residue and drooling suggest that the skilled were automatized and consolidated in daily activities.

The fourth stage concerns **retention of swallowing skills**. In this feasibility study, the majority of the results were consolidated and maintained at the 3 month follow-up assessments confirming that new swallowing skills were incorporated in daily activities.

The last stage is the “**generalisation and transfer**”, which is recognised as the last phase of motor learning and neuroplasticity principles, described in Chapter 2, section 2.3. It is assumed that this treatment lead to skill generalisation because participants experimented with other food such as espresso coffee and pizza. In addition, participants perceived improvements in functions not related to swallowing such as

voice. Their reports were suggestive of transfer effects, which should be investigated in a future study.

In conclusion, this feasibility study was able to apply a motor learning approach as described by Huckabee and Burnip (2018) to all the stages of the swallowing.

8.8 Strengths, challenges and limitations of the study

The main strengths of this feasibility study were the design of the study, the interventions using sEMG biofeedback, the motivation and adherence of participants to the intervention and the positive outcomes. The within-subject design allowed to find differences across levels of independent variables, although the sample of the study was small. A further strength of this study was the ease of use and the acceptability of sEMG biofeedback. Seven participants described benefits of the treatment procedures, and three participants enjoyed the sEMG biofeedback especially during the use of game, indicating an important contributor of this tool in increasing compliance and acceptability of this treatment. In addition, it was relatively inexpensive and easy to administer and no adverse events were detected. The main strengths of this feasibility study were the positive outcomes of the interventions. Although some evaluations were not included, the qualitative analysis of patient perspective was fundamental in order to detect the unexpected improvements such as attention and voice quality, which should be assessed objectively in future studies. The findings are important in shaping a protocol for a further study (Chapter 9)

There are, however, a number of limitations

1. Despite the fact that this is predominantly a feasibility study, the small sample size was a barrier to assess fully the efficacy and effectiveness of the treatment. This sample size lends to possibility of a Type 1 or Type 2 error so the statistical significance must be interpreted with a degree of caution.
2. The recruitment phase showed that more than half of eligible candidates were not enrolled in the study. This could be caused by the restricted inclusion criteria . Among the criteria, the presence of cognitive impairment was one of the most common for exclusion of the study (Chaper 5.6), which was needed in order to be

able to follow this intervention but should be reviewed in the future study.

3. There were barriers related to the site of the hospital and the lagoon of Venice. Participants reported that it was demanding to come every day to the hospital. Difficulties of mobility and ambulation could affect the fulfilment of the treatment. So, future studies should investigate new ways of treatment delivery including tele-rehabilitation.

4. Some assessments were not appropriate to detect some changes such as the changes in the frequency and severity of drooling, which should be documented by objective scales.

5. The follow-up assessment at 3 months post-treatment does not detect the long term effects. Future studies should include long-term retention of skills after treatment.

8.9 Summary

In conclusion, this study revealed that the study protocol is appropriate and given that the sEMG biofeedback intervention improved components of swallowing function of sample., it is worth investigating further

This study signals the end of the developmental phase of the complex intervention, and provides the basis for the implementation of a future trial. This is outlined in the last chapter of this thesis.

Chapter 9: Protocol for a future study

9.1 Introduction

As part of the design of complex intervention studies, the final phase concerns the development of a new trial based on the findings obtained by this feasibility study (Craig, 2019). The positive findings here suggest that sEMG biofeedback swallowing intervention has improved swallow function for the study participants. Nevertheless, some modifications are required. This chapter describes a protocol for a clinical trial based on the evidence from this study.

The design of the protocol is informed by the reporting guidelines of Standard Protocol Items Recommendations for Interventional Trials (SPIRIT) (Chan et al., 2013), recently updated by Calvert et al. to include content relating to patient reported outcomes (PROs), such as health-related quality of life and patient-feedback (Calvert et al., 2018). Hence, the new treatment protocol will be designed following the SPIRIT-PROs guidelines (Calvert et al., 2018). The following subsections describe the rationale for the therapy, its delivery, type of assessments used and study design according to SPIRIT-PROs. However, the lessons learnt from this study are considered first.

9.2 Lessons learnt from this feasibility study

The study findings supported the hypothesis that people with IPD benefit from the use of sEMG swallowing biofeedback intervention.

The intervention, guided by neuroplasticity and motor learning principles, was effective in improving the swallowing function and in maintaining improvement at 3 months post treatment. It is believed that the type swallowing exercises, dosage, modality of feedback and intensity of treatment played a key role in determining the positive effects. The sEMG biofeedback showed itself to be a versatile tool, acceptable to participants and could be easily customised based on the participant's swallowing skills. For this reason this sEMG biofeedback swallowing treatment should be maintained in the future study.

However, some participants reported fatigue mainly related to daily sessions at the hospital. They suggested to reduce the number of treatment sessions per week and other participants proposed the use of technology to deliver the treatment at home. This was not unexpected. In people with IPD, the major barriers to rehabilitation service include geographical location, the motor and psychological symptoms of IPD, and cost (Campbell et al., 2012). The level of effort required, particularly for intensive treatment protocols, in combination with the timing of medications and motor issues, are recognised to have a negative impact on the compliance with therapeutic intervention (Spurgeon et al., 2015). Telerehabilitation or telehealth has the potential to provide early and intensive treatment specially in IPD throughout the course of the disease (Theodoros et al., 2019). A technology-based approach can facilitate optimal timing, intensity and sequencing of intervention and the delivery of services in the home (Winters & Winters, 2004). Recently, Theodoros et al. completed a scoping review on the technology-enabled management of communication and swallowing disorders in people with IPD (Theodoros et al., 2019). They found that a variety of technologies used to deliver therapy at participants' home and participant perspectives on the implementation of telerehabilitation intervention were very positive (Theodoros et al., 2019). Based on these findings, it is indicated that the new protocol should verify the effectiveness of the sEMG biofeedback using a telerehabilitation approach. It has been assumed that this approach will ensure the intensity of treatment and compliance of the intervention in people with IPD and swallowing impairments. However, the ability to use the sEMG device with correct adherence of electrodes to the submental region needs to be considered and there will be increased cost associated with supplying additional devices to participants. An alternative way of delivering biofeedback will need to be explored if telerehabilitation is used and the biofeedback approaches covered in the systematic review (Chapter 3) may help in constructing this approach.

Other lessons learnt included the use of more sensitive outcome measures, an additional instrumental assessment (VFS) and the addition of a qualitative arm to the study. These are considered further in section 8.2.3.

9.3 The aim and research questions

The aim of the next study will be to establish the efficacy and safety of swallowing treatment using sEMG biofeedback but to extend the study to compare outcomes between interventions delivered at home using a telerehabilitation approach versus in clinic in people with IPD and dysphagia. It has been hypothesized that this treatment will have positive effects on increasing swallowing function, which could allow participants to change methods of oral intake and recover from functional swallowing difficulties.

This feasibility study aims to answer the following research questions:

(1) In people with dysphagia and IPD, does a swallowing intervention using sEMG as biofeedback change any of the following parameters when measured with objective and validated tools:

- a) secretion and saliva residue using FEES and a validated scoring scale
- b) pharyngeal food residue using FEES and a validated scoring scale
- c) medication residue using FEES and validated scoring scale
- d) swallowing kinematics and laryngeal penetration and /or aspiration assessed using VFS and validated scoring scales

It is hypothesised that this intervention will result in an improvement with reduction in secretions, medical and food pharyngeal residues, as well as swallowing kinematics and laryngeal penetration and/or aspiration events

(2) Does a specific sEMG biofeedback swallowing intervention reduce the self-perception of drooling in people with dysphagia associated with IPD using validated scale?

It is hypothesised that this intervention will have an impact on the self-perception of drooling.

(3) Does a specific swallowing intervention using sEMG biofeedback improve the method of oral intake in people with IPD and dysphagia using validated scale?

The hypothesis is that the intervention will result in changes to swallowing function and thus improve functional oral intake

(4) What is the impact of this biofeedback swallowing intervention on a person with IPD's overall quality of life using validated scale?

The hypothesis is that improvement in swallowing skills improves the quality of life of the study sample

(5) If swallow function changes after biofeedback swallowing intervention in people with IPD and dysphagia, is this change in swallow function maintained at medium > 3 months and follow-up (6 months)?

The hypothesis is that improvement in swallowing following this intervention in a person with IPD who is medically stable will be retained at or for longer than 6 months follow-up.

The secondary aims involved the following research questions:

(5) Does a specific biofeedback swallowing intervention modify the time for swallowing which will be assessed using the validated scale?

The hypothesis is that the intervention will increase the speed of swallowing of the study sample.

(6) Does a specific biofeedback intervention increase cognitive and attentional function as assessed using validated scale?

It is hypothesised that this intervention will have positive effects on attentions skills on the study sample.

(7) Does a specific biofeedback swallowing intervention change the voice quality and intensity of the voice using objective measures.

It is hypothesised that this intervention will increase the voice quality and the vocal intensity of the people with IPD.

(8) Does a specific biofeedback swallowing intervention change have an adverse event (eg effect on the general fatigue) on people with IPD.

It is hypothesised that this intervention may have an effect on the fatigue levels of people with IPD.

(9) Does a specific biofeedback swallowing intervention delivered using telerehabilitation approach increase adherence and acceptability to the swallowing intervention approach?

It is hypothesised that the intervention delivered via tele-medicine will be more acceptable to participants and increase the level of adherence to the programmes than that delivered in the clinical setting .

9.4 Methods

9.4.1 Study Design

The design of this study will be within-subject crossover feasibility-pilot-study. (Figure 9.1). Feasibility studies are fundamental to investigate the effects of specific intervention as well as acceptability of the intervention to participants, adherence to the treatment protocol as it was described in chapter 5. In addition, within-subject allows to find differences across different level of independent variables.

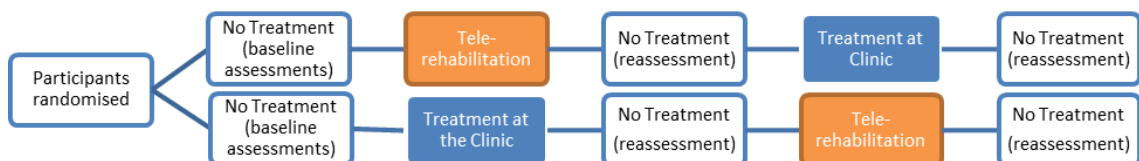


Figure 9. 1 Proposed trial design

9.4.2 Participants

People with IPD who meet the inclusion/exclusion criteria with confirmed dysphagia on instrumental assessment will be recruited (Table 9.1).

9.4.2.1 Inclusion/Exclusion criteria

The inclusion and exclusion criteria are described in table 9.1. The differences from the criteria from the previous feasibility study are :

- 1) The inclusion of people with mild-severe dysphagia, severity confirmed using VFS and a PAS score < 6. People, who were fed only by PEG, will be excluded (FOIS >1).
- 2) The inclusion of people with mild cognitive impairments. People with severe cognitive impairment (MoCA >18) will be excluded.

Table 9. 1 Inclusion and Exclusion Criteria

	<i>Inclusion criteria</i>	<i>Exclusion criteria</i>
a)	The diagnosis of IPD should be confirmed by neurologist following new International Parkinson Disease and Movement Disorder Society diagnostic criteria.	The presence of parkinsonism secondary to causes other than IPD
b)	Clinical stability has to be evaluated by the neurologist. The anti-parkinsonian medication therapy must be consistent throughout the duration of the study.	History of stroke or transient ischemic attack.
c)	The presence of oropharyngeal dysphagia must be confirmed by instrumental examination (VFS) and PAS > 6.	Severe dysphagic participants fed via PEG and FOIS > 1
d)	The ability to provide autonomous written and verbal consent	The presence of severe cognitive impairments (MoCa score > 18)

9.4.3 Intervention

The treatment protocol is the same as the previous feasibility study (Chapter 5. section 5.10.2). It is proposed to maintain this in the new study protocol. The only discrepancy between the two group interventions will be the method of delivery: at the clinic versus at participant's home using telerehabilitation. The treatment at the clinic will be as described in Chapter 5. The telerehabilitation intervention group will be informed

by an advisory group that involves people with IPD, carers and specialist clinicians. Specific tools will be selected in order to facilitate the delivery at home such as the use of flexible sub mental-sensor patch for surface electrode placement.

In addition, adherence to intervention protocols will be routinely monitored throughout use of sEMG software and a model of adherence described by Krekeler et al. will be used as a framework to organize and categorize the factors that may be affecting adherence to the intervention (Krekeler et al., 2020).

9.4.4 Outcomes

The feasibility study revealed that some of the outcome measure were valid tools; some of them were not appropriate to detect the changes during swallowing sEMG biofeedback treatment in IPD people and some additional outcome measure should be included.

The instrumental examination using the FEES showed positive and negative implications. On the one hand, it allowed assessment for the presence of secretions, bolus pooling, laryngeal penetration and aspiration. In this study, it was of fundamental importance for detecting the presence of medications in three participants (Chapter 7, section 7.2.2). On the other side, it did not provide important information on the swallowing kinematics, which are important for the understanding the coordination of oral-pharyngeal contraction in people with IPD. Recently, Curtis et al. showed that VFS assessment was specifically important in the swallowing assessment in people with IPD (Curtis et al., 2020a; Curtis et al., 2020b).

Given that FEES examination was fundamental to document the presence of secretion and medication residue and VFS could contribute in the understanding the swallowing kinematics and pharyngeal residues, the new protocol of assessment should include both instrumental examinations, adopting specific scales for scoring of parameters.

9.4.4.1 Primary outcome measures

(1) Changes in penetration and aspiration will be measured using FEES and VFS incorporating the PAS (Table 5.2) (Rosenbek et al., 1996).

(2) Changes in pharyngeal residue, swallowing efficiency and kinematics will be measured using FEES and VFS. The Yale Pharyngeal Residue Severity Rating Scale (Table 5.3) (Neubauer et al., 2015) with FEES will be used, whereas bolus clearance ratio (BCR) (Leonard, 2017) and the ten swallowing kinematic measures (Curtis et al., 2020b) will be used with VFS (Table 9.2).

Table 9. 2 Spatial and temporal swallowing kinematic measures (Curtis et al. 2019)

Spatial and temporal swallowing kinematics
Peak hyoid position
Peak laryngeal position
Maximal laryngeal constriction area normalized
Maximal pharyngeal constriction area normalized
Maximal PES displacement
Onset of hyoid displacement
Onset of laryngeal vestibule closure
Duration of hyoid movement
Duration of laryngeal vestibule closure
Duration of upper oesophageal sphincter opening

4) Changes in medication residue will be assessed using the rating scale described by Buhmann et al. (Buhmann, Bihler, et al., 2019) (Table 9.3).

Table 9. 3 Rating scale of medication swallowing

Rating	Description
No	No problems swallowing oral medication
Mild	Oral medication remains initially in the oral cavity or pharynx but is felt by the patient and cleared spontaneously or by a swallow of water
Moderate	Oral medication remains in the oral cavity or pharynx and is either not recognized or cleaning is ineffective
Severe	Direct or indirect (coughing during or after swallow) signs of aspiration. Oral medication can only be administered with puree or has to be crushed

5) Changes in posterior drooling and pharyngeal secretions will be assessed using New Zealand Secretion Scale (Miles et al., 2018) (Table 9.4)

Table 9. 4 New Zealand Score

Category	Symptom	Score
Location	Nil significant pooled secretions in pyriform fossae or laryngeal vestibule	0
	Secretions in pyriform fossae (above 20%)	1
	Secretions in laryngeal vestibule (beyond healthy lubrication of mucosa)	2
Amount in pyriform	Nil significant pooled secretions in pyriform fossae (0–20%)	0
	Secretions in pyriform fossae, not yet full (20–80%)	1
	Secretions filling (80–100%) or over spilling pyriform fossae/interarytenoid	2
Response (do not score if no significant pooling of secretions)	Normal airway responses in the pharynx or laryngeal vestibule may include spontaneous coughing, throat clearing, and/or swallowing. Secretions in pyriform fossae or laryngeal vestibule effectively cleared	0
	Ineffective attempts to clear or no response to secretions in pyriform	1
	Ineffective attempts to clear secretions from laryngeal vestibule	2
	No response to secretions in laryngeal vestibule	3

6) Changes in self-perceived anterior drooling will be assessed using 100mm visual analogue scale (VAS) where participants have to place a cross (X) on a 100-mm line (with 0mm being “no problem” and 100mm being “as bad as can be”) to indicate the number of separate incidents they feel that drooling occurred (frequency), how long in minutes they feel drooling occurred (duration), and how severe they feel drooling was (severity) (Hauser et al., 2004; McNaney et al., 2019). This scale may be more sensitive to change in frequency and severity.

7) Changes in method of nutritional intake. This will be graded using the FOIS (Crary et al., 2005).

8) Change in swallowing related quality of life through SWAL-QOL (McHorney et al., 2002).

9.4.4.2 Secondary outcome measures

The secondary outcomes incorporate the assessments, which were suggested by participants in this earlier feasibility previous study. Some of them relate to swallowing function.

a) Changes in the speed of swallowing will be assessed using the validated scale of the WST (Hughes & Wiles, 1996) and solid bolus using TOMASS (Huckabee et al., 2018) although speed may not always be a positive functional outcome in IPD, it will help explore the participants' reports of feeling faster at swallowing.

c) Cognitive and attentional function will be assessed using the MoCA (Dalrymple-Alford et al., 2010).

d) Changes of voice will be assessed using objective measures such as the sound pressure level meter, software such as PRAAT (<https://www.fon.hum.uva.nl/praat/>) and clinical dysarthria assessments using Frenchay Dysarthria Assessment – 2 (Enderby and Palmer 2008)

e) Assessment of fatigue using specific IPD scales that have yet to be explored. Fatigue in people with IPD is characterized by several different factors. Kluger et al. categorized the fatigue measures into three general domains: (1) measures of perceptions of fatigue and subjective fatigue complaints; (2) measures of performance fatigability; and (3) physiologic factors associated with fatigue or fatigability (Kluger, 2017). In this study fatigue will be assessed using the "The Parkinson Fatigue Scale (Brown et al., 2005)

f) Assessment of adherence to treatment counting the number of sEMG lines of activation during swallowing tasks, the number of people who drop out of the study, and the number of people who complete the weekly tasks.

g) Analysis of cost-effectiveness of the intervention based on the health economic evaluation which include human resource costs, equipment costs, (Burns et al., 2020).

9.4.4.3 Qualitative Feedback

One of the most valued part of this feasibility study was the qualitative feedback provided by the participants in the interviews. It provided the most accurate and authentic findings on impact of the treatment on the life of IPD participants. This methodology will be expanded in the new proposed study intervention. The questions will be addressed to participants and care-givers. The key selected questions for this component are:

- *What do you believe are the benefits of this treatment?*
- *What do you believe are the drawbacks of this treatment?*
- *Which modality of delivery the treatment do you prefer (home vs clinic)?*
- *If you had to change the intervention, what would you change?*

9.3.5 Enrolment

The time schedule for enrolment is summarised in figure 9.2.

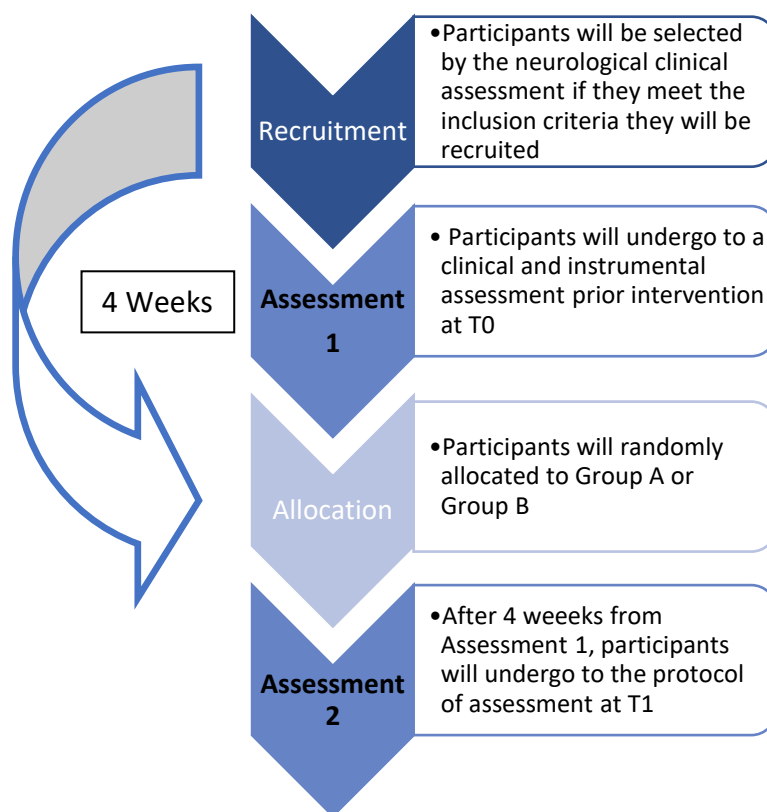


Figure 9.2 Recruitment Phase

The process of the treatment enrolment is summarised in the figure 9.3.

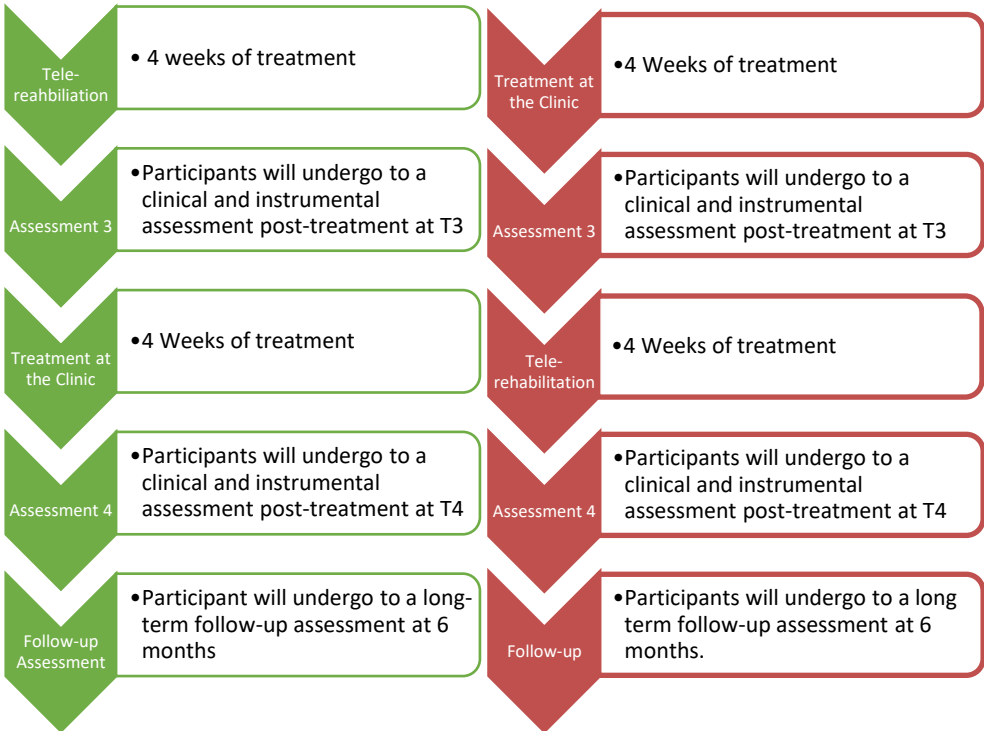


Figure 9. 2: Phases of treatment enrolment

9.5 Conclusion

This PhD study allowed the researcher to complete the first phases of the development of complex intervention on the use of sEMG biofeedback for people with dysphagia and IPD. The student believes that this intervention has clinical value and this assumption is supported by the findings of the feasibility study.

The current study was unfunded, which limited the resources available. On the other hand it reflects the difficulties of clinical practice, which has to operate with small budget, showing the values of practicability and affordability of this intervention. This protocol now prepares for the next stage and for the application for funding.

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APPENDIX A

PROSPERO International prospective register of systematic reviews

Biofeedback to improve the safety and efficiency of the swallow function in people with Parkinson's disease and dysphagia

Irene Battel, Irene Calvo, Isolde Harpur, Margaret Lawler, Margaret Walshe

Citation

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Review question(s)

What is the effectiveness of interventions using biofeedback to improve swallow function in adults with Parkinson's disease?

What is the safety of interventions using biofeedback to improve swallow function in adults with Parkinson's disease?

Searches

We will conduct the following searches:

Electronic bibliographic databases:

1. EMBASE (inception – January 2017),
2. PubMed (inception - January 2017),
3. CINAHL (inception - January 2017)
4. Web of Science (inception - January 2017),
5. Elsevier Scopus (inception - January 2017),
6. ScienceDirect (inception - January 2017),
7. AMED (inception - January 2017),
8. The Cochrane Database of Systematic Reviews (inception - January 2017),
9. ProQuest Dissertations and Theses A & I (inception - January 2017), and
10. Google Scholar (inception - January 2017).

Relevant conference proceedings: Conference proceedings of the annual congresses of Dysphagia Research Society, European Society Swallowing Disorders, International congress of Parkinson's disease and Movement Disorders, World Congresses for Neurorehabilitation will be hand-searched by Irene Battel (IB) and Irene Calvo (IC).

Reference lists: IB will search the reference lists of included studies to screen for the presence of any novel citations, which were not identified during the initial systematic search.

Authors will not apply any language or date restrictions within searches.

Types of study to be included

We will include randomised control studies (RCTs) and non-randomised studies. We define 'non-randomised studies' as any studies estimating the effectiveness of a recruitment intervention that does not use randomisation in order to allocate participants to the intervention or comparison groups. These types of studies are referred to by multiple names in the literature including, but not limited to: observational studies, cohort studies, and case-control studies. We will also include case series with either post-test or pre-test/post-test concrete outcomes. There are no restrictions on the language of publication. The methodological quality of included studies will be graded using "Checklists of methodological issues for review authors to consider when including non-randomized studies in systematic review" (Wells et al. 2013).

Condition or domain being studied

Swallowing disorders (dysphagia) occur frequently in Parkinson's disease (PD) patients. Several studies confirm that the vast majority of people with PD will develop dysphagia during the course of the disease (Miller et al. 2009). A meta-analysis based on studies using objective instrumental evaluation suggests that the prevalence of dysphagia in PD is as high as 87% (Kalf et al. 2012).

The nature of swallowing impairment is characterized by drooling, poor bolus formation, a deficiency of tongue movements and prolonged oral transit which results in a delay in the triggering of the pharyngeal swallow reflex (Miller et al. 2009), vallecula and pyriform sinus pooling, somatosensory deficiencies and a high risk of penetration and/or aspiration (Troche et al. 2014). In PD patients, dysphagia is characterized not only by weakness and tremor, but also by a deficit in motor planning and coordination. Many studies have suggested that the neural impairments in PD are manifested as a reduced ability to plan motor acts based on internal cues, and that these could be the causes of the freezing and coordination deficit during swallowing (Suttrup et al. 2016, Athukorala et al. 2014).

Participants/ population

Participants are male and female adults over 18 years of age with a clinical diagnosis of PD according to the United Kingdom Parkinson's Disease Society (UPDRS) Brain Bank diagnostic criteria (Hughes 1992), or other similar published clinical diagnostic criteria. We will include studies of people at all stages of the disease, at all disease severity levels presenting with all severities of dysphagia.

We will exclude participants with parkinsonian syndromes (multiple system atrophy, progressive supranuclear palsy, and dementia with Lewy bodies).

Intervention(s), exposure(s)

The intervention of interest is biofeedback. We will include studies that provide swallowing treatment using biofeedback. The biofeedback will involve any external visual, auditory, or tactile cue or combination of cues delivered using any type of device or equipment, which gives information to the individual with PD on their swallow function.

Comparator(s)/ control

The comparator interventions will be:

- (1) No intervention;
- (2) Usual care that does not involve biofeedback (e.g diet modification, postural changes, safe swallowing strategies);
- (3) Sham interventions;
- (4) Other swallowing interventions that do not include biofeedback.

Outcome(s)

Primary outcomes

- (1) Change in timing and efficiency of swallowing assessed by instrumental evaluation (videofluoroscopy or FEES or manometry) using valid reliable outcome measurement scales (e.g penetration-aspiration scale; residue scales etc.).
- (2) Change in frequency and amount of aspiration (food/fluid entering lungs) and safety of swallowing as assessed by instrumental evaluation.

Secondary outcomes

- (1) Change in oral intake status as measured by validated scales (e.g. functional oral intake scale, Crary et al. 2005);
- (2) Change in nutritional status as measured using a validated scale (e.g. malnutrition universal screening tool, Stratton et al. 2004);
- (3) Change in health-related quality of life as measured by quality of life measures and swallowing quality of life measures by participant and/or caregiver report;
- (4) Compliance with intervention;
- (5) Adverse events attributable to the intervention (e.g increased dysphagia, fatigue etc.).

Three timeframes will be examined (1) immediate change in outcome measures; (2) short-term change in outcome measures (1-12 weeks); and long-term change (12 weeks +).

Data extraction, (selection and coding)

Studies will be selected following a database search. Isolde Harpur (IH) will design and pilot a systematic search strategy to apply across all electronic databases identified in the protocol as relevant to the subject. Searches of electronic databases using this search strategy will subsequently be conducted by two other authors (IB and IC). These authors will also hand-search the annual scientific meetings identified as relevant in this protocol. A reference manager software (Zetero www.zetero.org) will be used to manage references.

Screening of titles will be conducted using Covidence by three authors (Irene Battel-IB; Irene Calvo-IC and Margaret Walshe-MW). Following duplicate deletion, double screening of titles and abstracts will be conducted by IB and IC, to exclude obviously ineligible results. Following this, articles identified as potentially relevant will be subsequently screened for eligibility by IB and IC. IB will scan the reference lists of included studies to identify further relevant articles. A third author (MW) will act as an independent reviewer and any discrepancies will be resolved through discussion.

The following data will be extracted:

Study design, setting, type of intervention, comparison intervention if relevant, population characteristics (severity of PD; severity of dysphagia; gender, number of participants, outcome, differences in outcome measures pre- and post-intervention, and compliance with the intervention), adverse effects, and the methodological quality of the studies.

This will be done using a specifically devised data extraction form.

Data will be extracted regarding: study design, setting, participant demographics, outcome measurement, prevalence figures and statistical robustness.

Descriptive analysis will initially be accomplished using Excel (Microsoft Corporation) spreadsheets, with subsequent exportation to statistical software, as appropriate. Statistical advice will be given by Margaret Lawler (ML).

IB will address missing/unclear data by contacting authors for studies published within the last ten years. Studies will be excluded if no response is received following two contact attempts.

Risk of bias (quality) assessment

Two review authors (IB, IC) will independently assess risk of bias in all included studies. The Cochrane Collaboration's Risk of Bias tool for assessing risk of bias will be used for randomised controlled trials, and will address such issues as sequence generation, allocation sequence concealment, blinding of participants and personnel, blinding of outcome assessment, incomplete outcome data, selective outcome reporting, other potential threats to validity, and other biases such as differences in baseline characteristics between the experimental groups, sources of funding, e.g. the pharmaceutical industry, and conflicts of interest (Higgins 2011).

The Risk Of Bias In Non-randomized Studies - of Interventions (ROBINS-I; <https://sites.google.com/site/riskofbiastool/>) tool will be used to assess the risk of bias due to confounding, and will consider aspects of methodological quality such as participant selection, measurements of interventions, departures from intended interventions, missing data, measurements of outcomes and selection of the reported results. Each study will be rated as having a critical, serious, moderate or low risk of bias based on a judgement on the gathered information. Reporting of information on the flow of participants through the trial (e.g. from a CONSORT diagram) will also be recorded.

We will resolve any disagreements by discussion and consensus agreement within the review team.

Strategy for data synthesis

We anticipate there will be limited scope for meta-analysis due to the range of outcomes and the small number of RCTs that exist in the area. Trials will be analysed according to the type of intervention used in the study. Interventions will be grouped when their form or content is deemed to be sufficiently similar.

The analysis will be narrative description of the data, and we intend to present the data in tables, grouped by intervention type. Where population, intervention and outcome are sufficiently similar to allow pooling of data in a meta-analysis, we will look for both visual evidence of heterogeneity in forest plots and statistical evidence of heterogeneity using the chi-square test for heterogeneity (Gardner et al. 2016).

Analysis of subgroups or subsets

If sufficient data are available, we will perform a subgroup analysis, in which the following intervention characteristics will be considered:

- (1) Type of biofeedback (e.g. visual /auditory /verbal);
- (2) Intensity of swallowing interventions;
- (3) Stage of the disease and severity of dysphagia.

Dissemination plans

The authors intend to submit this study in its completed form for publication in an appropriate peer-reviewed international journal. Also, the primary author intends to present the results at the annual congress of the European Society for Swallowing Disorders.

Contact details for further information

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Organisational affiliation of the review

Trinity College Dublin

Review team

Miss Irene Battel, Trinity College Dublin
Miss Irene Calvo, Casa di Cura Privata Policlinico (Milano)
Miss Isolde Harpur, Trinity College Dublin
Dr Margaret Lawler, Trinity College Dublin
Dr Margaret Walshe, Trinity College Dublin

Anticipated or actual start date

05 December 2016

Anticipated completion date

01 June 2017

Funding sources/sponsors

This project is unfunded

Conflicts of interest

None known

Language

English

Country

Ireland, Italy

Subject index terms status

Subject indexing assigned by CRD

Subject index terms

Deglutition; Deglutition Disorders; Humans; Neurofeedback; Neurological Rehabilitation; Parkinson Disease; Patient Safety; Safety; Treatment Outcome

Stage of review

Ongoing

Date of registration in PROSPERO

24 January 2017

Date of publication of this revision

24 January 2017

Stage of review at time of this submission	Started	Completed
Preliminary searches	Yes	Yes
Piloting of the study selection process	No	No
Formal screening of search results against eligibility criteria	No	No
Data extraction	No	No
Risk of bias (quality) assessment	No	No
Data analysis	No	No

PROSPERO

International prospective register of systematic reviews

The information in this record has been provided by the named contact for this review. CRD has accepted this information in good faith and registered the review in PROSPERO. CRD bears no responsibility or liability for the content of this registration record, any associated files or external websites.

APPENDIX B

ROMPT-SALIVA

(Kalf et al. 2011)

I. Do you experience loss of saliva during the day? 1. I do not lose saliva during the day and do not feel accumulation of saliva in my mouth.

2. I do not lose saliva, but I feel accumulation of saliva in my mouth.
3. I lose some saliva in the corners of my mouth or on my chin.
4. I lose saliva on my clothes. 5. I lose saliva on my clothes, but also on books or on the floor.

II. How often do you experience increased amounts or loss of saliva?

1. Less than once a day.
2. Occasionally: on average, once or twice a day.
3. Frequently: 2 to 5 times a day.
4. Very often: 6 to 10 times a day.
5. Almost constantly.

III. Do you experience loss of saliva during the night?

1. I do not experience loss of saliva during the night at all.
2. My pillow sometimes gets wet during the night.
3. My pillow regularly gets wet during the night.
4. My pillow always gets wet during the night.
5. Every night my pillow and other bedclothes get wet.

IV. Does your (loss of) saliva impair your eating and drinking?

1. No, my (loss of) saliva does not impair my eating or drinking.
2. Yes, my (loss of) saliva occasionally impairs my eating or drinking.
3. Yes, my (loss of) saliva frequently impairs my eating or drinking.
4. Yes, my (loss of) saliva very often impairs my eating or drinking.
5. Yes, my (loss of) saliva always impairs my eating or drinking.

V. Does your (loss of) saliva impair your speech?

1. No, my (loss of) saliva does not impair my speech.
2. Yes, my (loss of) saliva occasionally impairs my speech.
3. Yes, my (loss of) saliva frequently impairs my speech.
4. Yes, my (loss of) saliva very often impairs my speech.
5. Yes, my (loss of) saliva always impairs my speech.

VI. What do you have to do to remove saliva?

1. I do not have to remove saliva.
2. I always carry a handkerchief to remove possible saliva.
3. I daily use 1 or 2 handkerchiefs to remove some saliva.
4. I daily need more than 2 handkerchiefs to remove saliva.
5. I need to remove saliva so frequently that I always keep tissues near me or use a towel to protect my clothes.

VII. Does the loss of saliva limit you in contacts with others?

1. My loss of saliva does not limit me in contacts with others.
2. I have to pay attention, but that does not bother me.
3. I have to pay more attention because I know that others could see me losing saliva.
4. I try to avoid contact when I know that I lose saliva.
5. I notice that others avoid having contact with me because I lose saliva.

VIII. Does your loss of saliva limit you in doing activities inside or outside your home (work, hobbies)?

1. My (loss of) saliva does not limit me in activities.

2. I have to pay attention when I am busy, but that does not bother me.
3. I have to pay more attention, which is rather effortful.
4. My loss of saliva limits me in being active.
5. Due to my loss of saliva, important activities are no longer possible for me.

IX. How bothered are you as a result of your (loss of) saliva?

1. I hardly notice loss of saliva.
2. Feeling more saliva or losing it bothers me a little.
3. I am bothered by my loss of saliva, but it is not my priority concern.
4. My loss of saliva bothers me a lot because it is very limiting.
5. Losing saliva is the worst aspect of my disease

Italian Version
ROMPT-SALIVA
(Kalf et al. 2011)

I. Perde di saliva durante il giorno?

1. Non perdo la saliva durante il giorno e non sento l'accumulo di saliva nella mia bocca.
2. Non perdo la saliva, ma sento accumulo di saliva nella mia bocca.
3. Perdo un po 'di saliva agli angoli della bocca o sul mento.
4. Perdo la saliva sui miei vestiti. 5. Perdo la saliva sui miei vestiti, ma anche sui libri o sul pavimento.

II. Con quale frequenza sente di perdere maggiori di saliva?

1. Meno di una volta al giorno.
2. Occasionalmente: in media, una o due volte al giorno.
3. Frequentemente: da 2 a 5 volte al giorno.
4. Molto spesso: da 6 a 10 volte al giorno.
5. Quasi costantemente.

III. Perde la saliva durante la notte?

1. Non avverto la perdita di saliva durante la notte.
2. Il mio cuscino a volte si bagna durante la notte.
3. Il mio cuscino si bagna regolarmente durante la notte.
4. Il mio cuscino si bagna sempre durante la notte.
5. Ogni notte il mio cuscino e le altre lenzuola si bagnano.

IV. La sua (perdita di) saliva compromette il suo modo di mangiare e bere?

1. No, la mia (perdita di) saliva non pregiudica il mio mangiare o bere.
2. Sì, la mia (perdita di) saliva altera di tanto in tanto il mio mangiare o bere.
3. Sì, la mia (perdita di) saliva frequentemente mi impedisce di mangiare o bere.
4. Sì, la mia (perdita di) saliva molto spesso mi impedisce di mangiare o bere.
5. Sì, la mia (perdita di) saliva mi impedisce sempre di mangiare o bere.

V. La sua (perdita di) saliva altera il tuo modo di parlare?

1. No, la mia (perdita di) saliva non pregiudica il mio modo di parlare.
2. Sì, la mia (perdita di) saliva altera di tanto in tanto il mio discorso.
3. Sì, la mia (perdita di) saliva frequentemente altera il mio discorso.
4. Sì, la mia (perdita di) saliva molto spesso mi impedisce di parlare.
5. Sì, la mia (perdita di) saliva limita sempre il mio discorso.

VI. Cosa deve fare per rimuovere la saliva?

1. Non devo rimuovere la saliva.
2. Ho sempre un fazzoletto per rimuovere la possibile saliva.
3. Uso quotidianamente 1 o 2 fazzoletti per rimuovere un po 'di saliva.
4. Ho bisogno giornalmente di più di 2 fazzoletti per rimuovere la saliva.
5. Ho bisogno di rimuovere la saliva così frequentemente da tenere sempre i tessuti vicino a me o usare un asciugamano per proteggere i miei vestiti.

VII. La perdita di saliva limita i contatti con gli altri?

1. La mia perdita di saliva non mi limita nei contatti con gli altri.
2. Devo prestare attenzione, ma questo non mi infastidisce.
3. Devo prestare più attenzione perché so che altri potrebbero vedermi perdere la saliva.
4. Cerco di evitare il contatto quando so che perdo la saliva.
5. Ho notato che altri evitano il contatto con me perché perdo la saliva.

VIII. La sua perdita di saliva limita lo svolgere attività dentro o fuori casa (es: lavoro, hobby)?

1. La mia (perdita di) saliva non mi limita nelle attività.
2. Devo prestare attenzione quando sono occupato, ma ciò non mi infastidisce.
3. Devo prestare più attenzione, che è piuttosto impegnativo.
4. La mia perdita di saliva mi limita ad essere attivo.
5. A causa della mia perdita di saliva, non sono più possibili attività importanti per me.

IX. Quanto infastidisce la perdita di saliva?

1. A malapena noto la perdita di saliva.
2. Sentire più saliva o perdere mi infastidisce un po'.
3. Sono infastidito dalla mia perdita di saliva, ma non è la mia preoccupazione prioritaria.
4. La mia perdita di saliva mi infastidisce molto perché è molto limitante.
5. La perdita di saliva è l'aspetto peggiore della mia malattia.

APPENDIX C

The appreciation/discomfort of the sEMG procedure.

1) How do you rate the sEMG procedure?

1	2	3	4
_____	_____	_____	_____
Very uncomfortable	Uncomfortable	Comfortable	Very comfortable

2) Would you like to recommend any changes?

Italian Version

1) Come valuta la procedura con sEMG?

1	2	3	4
_____	_____	_____	_____
Molto spiacevole	Spiacevole	Confortevole	Molto confortevole

2) Vorrebbe consigliare eventuali modifiche ?

APPENDIX D



Coláiste na Tríonóide, Baile Átha Cliath
Trinity College Dublin
Ollscoil Átha Cliath | The University of Dublin

Irene Battel
School of Linguistic Speech & Communication Sciences
7-9 South Leinster St,
Dublin 2
Ireland

Date: 14th May 2018

Ref: 180304

Title of Study: Biofeedback to improve swallowing function in persons with
Dysphagia and Parkinson Disease: A feasibility study.

Dear Irene,

Further to a meeting of the Faculty of Health Sciences Ethics Committee held in
March 2018. We are pleased to inform you that the above project has ethical
approval to proceed.


As a researcher you must ensure that you comply with other relevant regulations,
including DATA PROTECTION and HEALTH AND SAFETY.

Yours sincerely,

A handwritten signature in black ink that reads "Prof. Brian O'Connell".

Prof. Brian O'Connell
Chairperson
Faculty Research Ethics Committee

APPENDIX E

<p>REGIONE DEL VENETO</p>  <p>ULSS3 SERENISSIMA</p>	<p>Documento di Supporto</p>	
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Oggetto: Studio di fattibilità sul trattamento mediante Biofeedback in pazienti con Parkinson Disease

Il Comitato Etico per la Sperimentazione Clinica (CESC) della provincia di Venezia e Irccs San Camillo, riunitasi il 13 Settembre 2018, in relazione alla richiesta di studio sperimentale effettuata da Irene Battel, ricercatrice e logopedista presso la stessa azienda,


conferma la presa visione dell'approvazione allo studio sperimentale con titolo " Biofeedback to improve swallowing function in persons with Dysphagia and Parkinson Disease: A feasibility study", approvato presso il Trinity College Dublin (N°180403).

Numero di Riferimento: 130918

Cordialmente

Rosco Quattre
Direttore del Comitato Etico

Con Delibera del Direttore Generale dell'ULSS12 n.1803 del 25/09/2013 è stato istituito il Comitato Etico per la Sperimentazione Clinica (CESC) della Provincia di Venezia e IRCCS San Camillo ai sensi del DM 8/2/2013 e della DGRV 1066 del 28/6/2013. Con decreto 150 del 31 ottobre 2013 il CESC è stato convalidato dalla Regione Veneto. Con Delibera del Direttore Generale dell'ULSS12 n.2665 del 25/10/2016 il CESC è stato rinnovato ,modificato con decreto del Direttore Generale AULSS3 n.810 del 7/4/2017

 <p>Sistema Gestione Qualità Azienda Ulss Serenuissima</p>	<p>File: Appendix E Comitato etico</p>	<p>Pagina 1 di 1</p>
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APPENDIX F

TRINITY COLLEGE DUBLIN SCHOOL OF LINGUISTIC SPEECH AND COMMUNICATION SCIENCES Participant Information Leaflet

To whom it may concern,

We are conducting a research on a swallowing treatment for persons with Parkinson Disease. This research project is carried out by Irene Battel, PhD student Trinity College Dublin and Italian SLT working at Hospital Sant'Angelo ULSS 3, Department of Neurology, Venice, Italy. This project is conducted under the supervision of Dr Margaret Walshe, Associate Professor, Trinity College Dublin.

What is the purpose of our research?

The aim of this study is to verify the effects of a specific swallowing treatment in order to improve swallowing function in patients with Idiopathic Parkinson Disease (IPD). Swallowing impairments are one the main causes of malnutrition, dehydration and chest infections in PD.

What will your involvement entail?

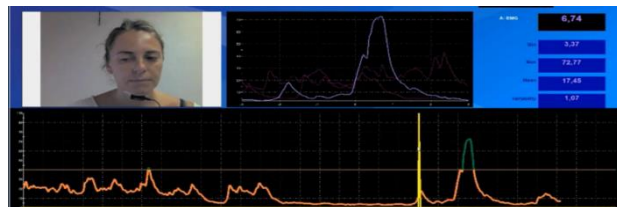
We want to invite you to take part in this research, which include a protocol of assessment and treatment.

The swallowing assessment consists on the fiber-endoscopic swallowing assessment (FEES) and questionnaires in order to examine different aspects of eating and drinking modality. The FEES consists a small tube with a camera and it is inserted through the nose and place down in the throat, allowing to see how people swallow and if there are food residues in the throat. The assessment will be completed four times: (1) at the onset of the study (4 weeks prior to treatment), (2) before the begging of treatment(4 weeks after the first assessment), (3) after 4 weeks of treatment and (4) at 3 months follow-up.

The treatment protocol involves a specific swallowing exercise using surface electromyography biofeedback (sEMG). The sEMG consists on surface electrodes which are place under the chin (see the imagine 1), they registered the contraction of the muscles under the chin who are responsible for swallowing. In this way, the persons is able to control and monitoring the muscle contraction (Imagine 2). The treatment lasts 1 hour a day, 4 days a week, for 4 weeks.



Imagine 1: Electrodes palced under chin



Imagine 2: Example of sEMG swallowing treatment

The benefit and risk of participating?

This treatment has been already described in previous study, no potential risks or adverse outcomes have been reported. In the unlikely event that you feel tired or uncomfortable, please do not hesitate to contact Irene Battel.

The major value of this study is to contribute the understanding of rehabilitation in persons with PD. The research of the swallowing rehabilitation is lacking and so this research will furnish important contribute in order to increase swallowing function in PD.

The information or data which can be identified with me will be changed in a reference code to ensure the anonymous identities.

If you have any questions about this research you can contact Irene Battel email batteli@tcd.it and mobile number (+39 3395317706). You are also free, to contact the research supervisor Dr Margaret Walshe on walshema@tcd.ie

Foglio Informativo del Paziente

Titolo: L'uso del Biofeedback per migliorare la deglutizione in persone affette da disfagia e malattia di Parkinson: uno studio di fattibilità.

Stiamo conducendo una ricerca sul trattamento di deglutizione per le persone con malattia di Parkinson. Questo progetto di ricerca è condotto da Irene Battel, una studentessa di dottorato Trinity College di Dublino e logopedista che lavora presso l'Ospedale Sant'Angelo ULSS 3, Dipartimento di Neurologia, Venezia, Italia. Questo progetto è condotto sotto la supervisione di Dr Margaret Walshe, Professore associato, Trinity College di Dublino.

Qual è lo scopo di questa ricerca?

Lo scopo di questo studio è di verificare gli effetti di un trattamento di deglutizione specifico al fine di migliorare la funzione di deglutizione nei pazienti con malattia di Parkinson idiopatico (PD). I deficit deglutitori sono una delle principali cause di malnutrizione, disidratazione e infezioni al torace nella malattia di Parkinson.

Quale sarà il suo coinvolgimento?

In questa ricerca le verrà richiesto di completare un protocollo di valutazione e trattamento. La valutazione consiste in un esame endoscopico. Questo è un esame di routine e consiste nel posizionare un tubo nel naso per vedere se il suo modo di deglutizione è sicuro. Ci sono anche tre test specifici che saranno completati al fine di esaminare la sua deglutizione. Queste valutazioni saranno completate quattro volte: (1) all'inizio dello studio (4 settimane prima del trattamento), (2) prima dell'inizio del trattamento (4 settimane dopo la prima valutazione), (3) dopo 4 settimane di trattamento e (4) a 3 mesi di follow-up.

Il protocollo di trattamento prevede uno specifico esercizio di deglutizione mediante biofeedback di elettromiografia di superficie (sEMG). Il sEMG è costituito da elettrodi di superficie posti sotto il mento (vedere l'immagine 1). Registra la contrazione dei muscoli sotto il mento che sono responsabili della deglutizione. In questo modo, le persone sono in grado di controllare e monitorare la contrazione muscolare (immagine 2). Il trattamento dura 1 ora al giorno, 4 giorni a settimana, per 4 settimane. Ci sono alcune prove per dimostrare che questa tecnica può migliorare la deglutizione.

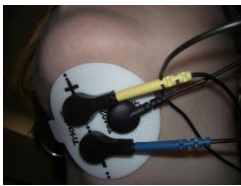


Immagine 1: Elettrodi posti sotto il mento

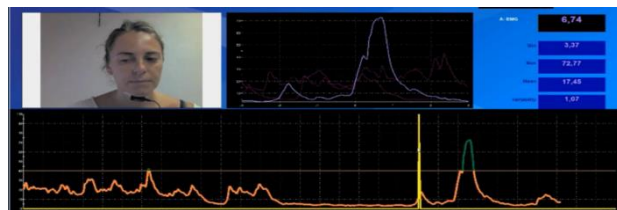


Immagine 2: Esempio del trattamento deglutitorio con sEMG

Quali sono i benefici e i rischi della partecipazione?

In studi precedenti, non sono stati segnalati rischi potenziali o eventi avversi. Nell'improbabile eventualità che ti senta stanco o a disagio, la sessione verrà interrotta. Il principale vantaggio è quello di contribuire alla comprensione di quali trattamenti siano efficaci per le persone con PD e problemi di deglutizione. Inoltre, questo ti consentirà di svolgere anche di un intenso programma di logopedia.

Dove è la sede?

La sede è l'ufficio del discorso e della terapia del linguaggio (n ° 6) presso il piano terra dell'Ospedale Sant'Angelo a Venezia (Italia).

La mia identità rimane segreta?

La tua identità rimarrà confidenziale. Le informazioni o i dati, che possono essere identificati, verranno modificati in un codice di riferimento per garantire le identità anonime.

I risultati saranno pubblicati?

I risultati saranno pubblicati in una rivista internazionale scientifica al fine di condividere le nostre scoperte e migliorare la conoscenza dei disturbi della deglutizione nelle persone con PD. Non verranno riportate informazioni sulla tua identità al fine di garantire l'anonimato.

Posso recedere dallo studio?

Se decidi di fare volontariato per partecipare a questo studio, puoi ritirarti in qualsiasi momento. Se decidi di non partecipare, o se ti ritiri, non sarai penalizzato e non rinuncerai ai benefici che avevi prima di entrare nello studio.

Questo è stato approvato dal Comitato Etico del Trinity College di Dublino (Irlanda) e dall'Ospedale Sant'Angelo di Venezia (Italia).

Per eventuali domande su questa ricerca è possibile contattare Irene Battel tramite email batteli@tcd.it e numero di cellulare (+39 3395317706). Sei anche libero di contattare il supervisore della ricerca Dr Margaret Walshe su walshema@tcd.ie.

Se il gruppo di studio verrà a conoscenza di informazioni che potrebbero influenzare il suo desiderio di rimanere nello studio, sarai informato immediatamente.

APPENDIX H

**TRINITY COLLEGE DUBLIN
SCHOOL OF LINGUISTIC SPEECH AND COMMUNICATION SCIENCES
RESEARCH ETHICS APPLICATION FORM**

Consent Form

Title: Biofeedback to improve swallowing function in persons with dysphagia and Parkinson
Disease: A feasibility study.
PI: Irene Battel,
Supervisor: Dr Margaret Walshe

I am invited to participate in this research project, which is being carried out by Irene Battel, PhD student Trinity College Dublin and Italian SLT working at Sant'Angelo Hospital ULSS 3, Department of Neurology, Venice (Italy). This project is conducted under the supervision of Dr Margaret Walshe, Associate Professor, Trinity College Dublin. My participation is voluntary. Even if I agree to participate now, I can withdraw at any time without any consequences of any kind.

If you agree to participate, firstly this will involve you completing instrumental swallowing evaluations and nutritional and functional oral intake assessments. These assessments will be complete at the onset of the study, after a 1 month prior to start the treatment, after 1 month of therapy sections at after 3 months post treatment.

The treatment consists sEMG biofeedback swallowing programme in order to increase the deglutition functions.

I understand that any information or data which can be identified with me will be changed in a reference code to ensure the anonymous identities. At the end of the study, the anonymous data will be stored in the Department of Clinical Speech and Language Studies TCD. Dr Margaret Walshe will be responsible for data storage and for destroying data after 5 years.

If I have any questions about this research I could contact Irene Battel email batteli@tcd.it and mobile number (+39 3395317706). I am also free, to contact the research supervisor Dr Margaret Walshe on walshema@tcd.ie

I understand what is involved in this research and I agree to participate in the study. [*I have been given a copy of the Participant Information Leaflet and a copy of this consent form to keep.*]

Signature of participant

Date

Signature of researcher

I believe the participant is giving informed consent to participate in this study.

Modulo di consenso

Titolo: Biofeedback per migliorare la funzione di deglutizione nelle persone con disfagia e morbo di Parkinson: studio di fattibilità.

PI: Irene Battel,

Supervisore: Dr Margaret Walshe

Sono invitato a partecipare a questo progetto di ricerca, condotto da Irene Battel, studente di dottorato Trinity College di Dublino e SLT italiano che lavora presso l'Ospedale Sant'Angelo ULSS 3, Dipartimento di Neurologia, Venezia (Italia). Questo progetto è condotto sotto la supervisione di Dr Margaret Walshe, Professore Associato, Trinity College di Dublino. La mia partecipazione è volontaria. Anche se accetto di partecipare, posso ritirarmi in qualsiasi momento senza conseguenze di alcun tipo.

Se accetto di partecipare, questa comporterà il completamento di una valutazione di deglutizione. Queste valutazioni saranno complete all'inizio dello studio, dopo 1 mese prima di iniziare il trattamento, dopo 1 mese di terapia a 3 mesi dopo il trattamento. Il trattamento consiste nel programma di deglutizione biofeedback al fine di migliorare le funzioni di deglutizione.

Comprendo che qualsiasi informazione o dato che possa essere identificato con me verrà modificato in un codice di riferimento per garantire l'anonimità. Alla fine dello studio, i dati in formato anonimo saranno archiviati nel Dipartimento di Discorso Clinico e Studi di Lingue TCD. Dr Margaret Walshe sarà responsabile per l'archiviazione dei dati e per la distruzione dei dati dopo 5 anni.

Se avessi qualche domanda su questa ricerca potrei contattare l'email di Irene Battel batteli@tcd.it e il numero di cellulare (+39 3395317706). Sono anche libero di contattare il supervisore alla ricerca Dr Margaret Walshe su walshema@tcd.ie. Capisco cosa è coinvolto in questa ricerca e sono d'accordo a partecipare allo studio. [Ho ricevuto una copia del volantino informativo del partecipante e una copia del presente modulo di consenso da conservare.]

Firma del partecipante

Data

Firma del ricercatore

Credo che il partecipante stia dando il consenso informato a partecipare a questo studio.

APPENDIX I



Trinity College Dublin
Coláiste na Tríonóide, Baile Átha Cliath
The University of Dublin

18th July, 2016

Application TT65 Academic Year 2015/16

Applicant: Irene Battel

Title of Research: Validation of the Functional Oral Intake Scale into Italian

Dear Irene,

Your submission for ethics approval for the research project above was considered by the Research Ethics Committee, School of Linguistic, Speech and Communication Sciences, Trinity College Dublin, on Monday, 18 July 2016, and has now been approved in full. We wish you the very best in your research activities.

Best wishes,



Dr Lorna Carson
Chair, Research Ethics Committee
School of Linguistic, Speech and Communication Sciences



APPENDIX J

Functional Oral Intake Scale (FOIS)

TUBE DEPENDENT (levels 1-3)	
1	No oral intake
2	Tube dependent with minimal/inconsistent oral intake
3	Tube dependent with consistent oral intake of food or liquid.
TOTAL ORAL INTAKE (levels 4-7)	
4	Total oral intake of a single consistency
5	Total oral intake of multiple consistencies requiring special preparation
6	Total oral intake with no special preparation, but must avoid specific foods or liquid items
7	Total oral intake with no restrictions

Functional Oral Intake Scale in Italian (FOIS-It)

NUTRIZIONE ENTERALE/PARENTERALE (Livelli 1 -3)	
1	Nessuna assunzione di alimenti per via orale
2	Nutrizione per via enterale/parenterale con minime quantità per via orale
3	Sistematiche quantità assunte per via orale integrate da nutrizione enterale/parenterale
NUTRIZIONE ORALE COMPLETA (Livelli 4-5)	
4	Nutrizione orale completa con cibi di una sola consistenza
5	Nutrizione orale completa con cibi a diversa consistenza in cui viene richiesta una preparazione specifica
6	Nutrizione orale completa con cibi senza la necessità di preparazione specifica, con esclusione di alcuni cibi o liquidi
7	Nutrizione orale completa senza restrizioni

APPENDIX K

It shows the questionnaire containing the same information of FOIS. The first is the English Versions the second is the Italian version of the questionnaire

Client ID:

Questionnaire Form in English

	Yes	No
Is the person fed only by enteral/parental tube and has no food or liquid by mouth?		
Is the person fed by enteral/parental tube but has minimal amounts of food by mouth?		
Is the person fed by enteral/parental tube but has regular and great amount of food by mouth?		
Is the person fed only per mouth with single consistency food?		
Is the person fed by a full oral diet but has many different foods that require specific preparation for swallowing safety?		
Is the person fed by a full oral normal diet but some foods/liquids must be avoided?		
Is the person by a full oral normal diet with no limitations?		

Questionnaire Form in Italian

	SI	No
La persona viene alimentata solo mediante nutrizione enterale/parenterale e non assume cibi o liquidi per bocca?		
La persona viene alimentata mediante nutrizione enterale/parenterale ma assume minime quantità di cibo per bocca?		
La persona viene alimentata mediante nutrizione enterale/parenterale ma assume regolari e maggiori quantità di cibo per bocca?		
La persona si alimenta esclusivamente per via orale con cibi di una sola consistenza?		
La persona si alimenta con una dieta completa per via orale ma diversi alimenti richiedono una preparazione appropriata per deglutirli in modo sicuro?		
La persona si alimenta con dieta completa per via orale ma alcuni cibi devono essere evitati?		
La persona si alimenta per via orale con una dieta normale senza restrizioni?		

La preghiamo di allegare questa scheda alla FOIS e restituire entrambe le schede insieme

APPENDIX L

Patient ID	Week 1	Week 2	Week 3	Week 4
1	no..easy for me	I will dream the plane after this treatment ...it was better to swallow with the yogurt	Do you think that I am better? As I am not sure if this game will help	It was fun I think I am more focus when I am swallowing, and I did not lost so much saliva I could read the newspaper without make it wet
2	I enjoy and I hope to be good	No all good. Instead of yogurt could you use prosecco. I am joking. Just to say that yogurt is nothing special for me	I feel good and I am happy to come. I'll try my best to get stronger	I have never thought that it was important to train the swallowing, I know that my voice is weak, but I have not imagined that also my swallowing were weak. I think that my fellow with PD should know that it is important and do the exercise. Since I have started, I pay attention on my swallow and sometimes I ask myself like how did you swallow? like you asked me during therapy
3	I try to swallow properly and after that treatment sometimes I feel tired, but I do not why as I have only swallow	For me it is better to swallow with the yogurt as I could feel it going down...I am mean I have more sensation of what I am doing not sure if I am clear	Well, it is not really easy ...it is nothing with you I mean yor really kind but I haven't thought it was so intense. Honestly, I am looking forward to finish it.	I was really happy that I have managed to complete it...well it help me to be able to drink the water. I mean to increase the numbers of glass of water.
4	It is good treatment for me as I have told I am scared to suffocate during	Something I do not understand how to control the signal on the screen, but I am happy to	You know that I went to the restaurant for my daughter birthday and I had a pizza...I managed to eat the	It was intense and demanding treatment ...incredible just for swallowing but

	meals. I hope to be able to swallow good and to not be scared while I am eating	come and I feel a little more confident while I am having some food	whole margherita without choking...although I was a little bit slow as I did small bites and I pay attention to the swallowing.	eventually I did not have any panic attacks since I am coming...finger crossed...something the saliva goes sideways and I cough but I am ok...I cough and swallow again
5	I didn't know that I could swallow so many times. I told my nephew that I am playing video game with you...	I could say that after coming here I do not lose saliva...maybe because I am swallowing and swallowing.	I like coming here...I think that this treatment helps me a lot...I must be concentrated to swallow. It seems easy but it is not	It was hard treatment and I like it as I like the challenges... I learn that I have to swallow often and if I see that the saliva fell out, I need to swallow it several times
6	it like a game...it is fun	(3 rd day of the week): Today I was really tired but now that the session is finished I am happy	My husband noticed that I was eating not really slow at usual and no cough episode...usually I am really slow more than 20 minutes to have a first plate	I am not coughing so often and I think I have a better voice. Maybe it is more clear... I hope to remember what I have learned because with this disease I forget everything
7	Day 1 I keep thinking that it is impossible that I could not drink the pill well...I hope that this treatment will help, Day 5: I enjoy ..it's like a game and the time goes by quickly	You know that I understand how to make a sharp hill. I concentrate and swallow it as strong and fast I could	this is a great news, I was a coffee lover but since I have the PD I could not drink an espresso without scaring the bar staff and my wife. Seriously once they were about to call the ambulance because I was coughing, and my face turned into red-blue. I scared my wife I decided to quit drinking coffee...but yesterday I wanted to try to hold the coffee in the mouth and drink it as I was doing the plane exercise I	It was an intense exercise...I was so glad to have done it not only because of the espresso but because I think more of how I am swallowing

			did not cough... (Me Really? Can you show me?)	
9	it is ok...nothing to say	I am good and sometimes I have lost the signal...I hope that it is worth it	2 [^] day. You know that I do not have a lot of problems on swallowing, so do you think that it is worth to come here? Because I thought that you were working also on voice	Good that it is finished...I did not that I could re-learn how to swallow, my daughter said that I do not loose saliva so often and she thinks that I am speaking better...(me: and you what are you think?) well I can see that I do not loose saliva like I used to be, but I think that I speak as usual I do not feel any changes.
10	I feel my mouth dry after the treatment, which it is a weird sensation (Me: do you feel that it is to dry? Do you want a sip of water?) No it is a feeling that it is difficult to describe, it is like that it is not wet as usual	I feel that the saliva it is not thick and sticky as it used to be	I feel good	I am pleased as I am swallowing with attention also at home .. I learn to be concentrate and most of all I know that I have to learn to swallow saliva
11	At the beginning I did not understand what I have to do because I was scared that it was difficult and I don't like electrodes	It is a little bit exhausting to come every day	3 day: yesterday I had a fry breast chicken ...it was almost 2 year I hadn't ...I am so happy as It was one of my favourite meal but I felt it stocking in the throat...terrible but yesterday I had some pieces a little with the mayonnaise and they went down incredibly	Although it was not easy for me to come I happy for the treatment. It pushes to swallow and swallow and maybe this make a sense for PD persons.