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Development of a Core Outcome Set for Dysphagia Interventions in Parkinson's Disease: the COS-DIP Study

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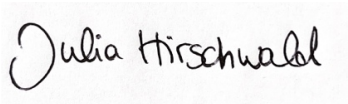
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Summary

"We need less research, better research, and research done for the right reasons" (Altman, 1994, p. 283). Although this statement was made over 30 years ago, it remains highly relevant, underscoring the need for high-quality research that minimises research waste and maximises benefits for patients. Core outcome sets (COS) are central in this goal. A COS is a standardised set of outcomes that should be measured and reported, at a minimum, in all clinical trials for a given health condition. COS aim to reduce heterogeneity in outcome measurement, ensure relevance to all key stakeholders, and mitigate selective outcome reporting bias - three key challenges in health research. Addressing these challenges helps reduce avoidable research waste and facilitate high-quality research.

One area where COS development is still limited is oropharyngeal dysphagia (OD). OD can have serious physiological as well as psychosocial effects, including malnutrition, dehydration, pneumonia and reduced quality of life. Parkinson's disease (PD) is the fastest growing neurological condition globally and has a prevalence of OD of up to 80%. Hence, there is an urgent need for high-quality, evidence-based OD interventions for people with PD.

The aim of this study was to develop an agreed, standardised COS for OD interventions in PD (COS-DIP). Following COS development guidelines, five studies were conducted. Study 1 established the protocol, including the development of a long list of outcomes - a standard term in COS development for the initial comprehensive set of outcomes - based on key stakeholders' perspectives. It also comprises the identification and agreement of the core outcomes, their definitions, outcome measurement instruments (OMIs) and timepoints of measurement through a Delphi survey and consensus meetings.

Study 2 was a scoping review identifying 39 outcomes measured across 19 OD intervention studies in PD. All 39 outcomes were added to the long list of outcomes. Definitions, OMIs, timepoints of measurement of outcomes were also identified and varied widely across included studies highlighting the need for the COS-DIP.

Study 3 involved an online survey to assess swallowing outcomes important to people with OD in PD, family members and family caregivers. To validate the survey results and clarify inconclusive findings, we conducted focus groups with previous survey participants. The outcome 'fear of choking' was identified by participants as important but missing and was therefore added to the long list of outcomes.

To finalise the long list of outcomes, an online survey of dysphagia clinicians' perspectives on important OD intervention outcomes in PD was conducted as part of Study 4. Five new outcomes ('independent eating/drinking', 'social aspect of eating/drinking', 'mealtime duration', 'respiratory swallow coordination' and 'hydration') were identified as missing but important by respondents and were added to the long list of outcomes.

In Study 5, an international three round online Delphi survey and two online consensus meetings were conducted with people with OD in PD, family members/caregivers, healthcare professionals and researchers to agree the core outcomes, definitions, OMIs and timepoints of measurement. The six core outcomes agreed were 'penetration/aspiration', 'aspiration pneumonia', 'choking', 'OD severity', 'swallowing related quality of life' and 'post swallow pharyngeal residue'. Consensus was reached for definitions and OMIs for each core outcome. Timepoints of measurement were agreed to be a minimum pre, immediately post and 3-months post completion of the intervention. In addition, advisable timepoints of measurement were agreed to be 1-month pre and 6- and 12-months post completion of the intervention.

Regular evaluation of the COS-DIP is crucial to ensure its continued validity and relevance. This ongoing assessment will determine whether the selected outcomes remain important to all key stakeholders and will provide opportunities to refine the COS-DIP through addition or removal of outcomes, OMIs or revised definitions. Monitoring the uptake and reporting of the COS-DIP in relevant studies will be essential to assess its impact on research quality.

The consistent use of the COS-DIP in OD intervention studies in PD will help ensure that these studies are measuring the aspects of OD in PD most important to all key stakeholders and to allow for more accurate comparisons between future studies. This, in turn, will improve the quality of evidence available to inform clinical decision-making and ultimately lead to better outcomes for people with OD in PD.

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List of abbreviations

ACCORD	Accurate Consensus Reporting Document
CCT	Controlled Clinical Trial
COMET	Core Outcome Measures in Effectiveness Trials
COREQ	Consolidated Criteria for Reporting Qualitative Research
COS	Core Outcome Set
COS-DIP	Core Outcome Set for Dysphagia Interventions in Parkinson's disease
COSMIN	COnsensus-based Standards for the selection of health Measurement INstruments
COS-STAD	Core Outcome Set-STandards for Development
COS-STAP	Core Outcome Set-STAndardised Protocol
COS-STAR	Core Outcome Set–STAndards for Reporting
CROSS	Checklist for Reporting of Survey Studies
DIGEST	Dynamic Imaging Grade of Swallowing Toxicity
DOSS	Dysphagia Outcome and Severity Scale
DSFS	Drooling Severity and Frequency Scale
DSS	Dysphagia Severity Scale
eDelphi	online Delphi
EMST	Expiratory Muscle Strength Training
FEES	Fibreoptic Endoscopic Evaluation of Swallowing
FOIS	Functional Oral Intake Scale
GRADE	Grading of Recommendations Assessment, Development and Evaluation
HRB	Health Research Board
ICA	Inductive Content Analysis
IROM	Investigator Reported Outcome Measure
JBI	Joanna Briggs Institute
LSVT	Lee Silverman Voice Treatment
MBSImP	Modified Barium Swallow Impairment Profile
MDADI	MD Anderson Dysphagia Inventory
N/S	not specified
OD	Oropharyngeal Dysphagia

OMERACT	Outcome Measures in Rheumatology
OMI	Outcome Measurement Instrument
OR	Odds Ratio
PAS	Penetration-Aspiration-Scale
PD	Parkinson's Disease
POE	Pleasure Of Eating
PRISMA-ScR	Preferred Reporting Items for Systematic Reviews and Meta-analysis extension for Scoping Reviews
PROM	Patient Reported Outcome Measure
RCT	Randomised Controlled Trial
rTMS	repetitive Transcranial Magnetic Stimulation
SDQ	Swallowing Disturbance Questionnaire
SWAL-QOL	Swallowing Quality of Life Questionnaire
SLT	Speech and Language Therapy/Therapist
SSC	Study Steering Committee
UES	Upper Esophageal Sphincter
VASES	Visual Analysis of Swallowing Efficiency and Safety
VDS	Videofluoroscopic Dysphagia Scale
VFS	Videofluoroscopy of Swallowing

List of publications and conference proceedings

Published peer-reviewed journal articles

Hirschwald, J., Mooney, L., Battel, I., Riera, S., & Walshe, M. (2025). What is measured in dysphagia interventions in Parkinson's disease: An international online survey of dysphagia clinicians. *International Journal of Speech-Language Pathology.* 1-10. https://doi.org/10.1080/17549507.2025.2555252

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Hirschwald, J., Mooney, L., Wolf, M., Boyle, G., Warnecke, T., & Walshe, M. (2025). 'Without the Data You're Not Going to Know If It's Important or Not': A Mixed Methods Study on What Dysphagia Intervention Outcomes are Important to People with Parkinson's Disease and Family Members. *Dysphagia*, *40*(4), 902–919. https://doi.org/10.1007/s00455-024-10788-x

Hirschwald, J., Duncan, S., Warnecke, T., Boyle, G., Regan, J., & Walshe, M. (2024). Development of a Core Outcome Set for Dysphagia Interventions in Parkinson's disease (COS-DIP): Study protocol. *BMJ Open*, *14*(2), e076350. https://doi.org/10.1136/bmjopen-2023-076350

Battel, I., Arienti, C., Del Furia, M. J., **Hirschwald, J.**, Lazzarini, S. G., & Walshe, M. (2024). Rehabilitation interventions for oropharyngeal dysphagia in people with Parkinson's disease. *Cochrane Database of Systematic Reviews*, *2024*(1). https://doi.org/10.1002/14651858.CD015816

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Published peer-reviewed abstracts

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Hirschwald, J., Wolf, M., Boyle, G., Warnecke, T., & Walshe, M. (2024, April 3-5). *Relevante Outcomes in Interventionsstudien aus Sicht von Menschen mit Dysphagie und Parkinson, Betreuenden und Angehörigen: Eine Mixed Methods Studie* [Relevant outcomes in intervention studies from the perspective of people with dysphagia and Parkinson's disease, carers and family members: A mixed methods study]. [Oral presentation]. Abstract from the 13th Congress of the German interdisciplinary Dysphagia Society, Berlin, Germany. *Endoscopy Campus.* https://www.endoscopy-campus.com/wp-content/uploads/ec-magazin-2024-01.pdf

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Hirschwald, J., Hofacker, J., Duncan, S., Warnecke, T., Boyle, G., & Walshe, M. (2023, March 1-3). *Entwicklung von Core Outcome Sets für die Dysphagieforschung am Beispiel des idiopathischen Parkinson-Syndroms* [Development of core outcome sets for dysphagia research using the example of Parkinson's disease]. [Oral presentation]. Abstract from the 12th Congress of the German interdisciplinary Dysphagia Society, Cologne, Germany. *Endoscopy Campus.* https://www.endoscopy-campus.com/wp-content/uploads/ec-magazin-2023-01.pdf

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Hirschwald, J. (2025, March 28). *Studien zu Dysphagietherapien für Menschen mit Parkinson: Wird die Forschung unseren klinischen Anforderungen gerecht?* [Clinical trial design in Parkinson's disease: Does research meet our clinical needs?]. [Oral presentation]. LSVT LOUD Workshop, Osnabrück, Germany.

Hirschwald, J. (2024, April 27). *Studien zu Dysphagietherapien für Menschen mit Parkinson: Wird die Forschung unseren klinischen Anforderungen gerecht?* [Clinical trial design in Parkinson's disease: Does research meet our clinical needs?]. [Oral presentation]. German Congress for Parkinson's and Movement Disorders, Rostock, Germany.

Hirschwald, J. (2023, December 7). *Clinical trial design in Parkinson's disease: Does research meet our clinical needs?* [Oral presentation]. Trinity College Dublin Study Day, Dublin, Ireland.

Hirschwald, J., Hofacker, J., Duncan, S., & Walshe, M. (2022, September 12-16). '*You can't improve what you don't measure': A scoping review of outcomes in clinical trials on dysphagia in Parkinson's disease* [Oral presentation]. 12th Congress of the European Society for Swallowing Disorders, Leuven, Belgium.

Poster presentations

Mooney, L., **Hirschwald, J.**, Battel, I., Riera, S., Zhu, H., & Walshe, M. (2024, September 25-27). *Surveying International Clinicians' Perspectives on Important Dysphagia Intervention Outcomes in Parkinson's Disease* [Poster presentation]. 14th Congress of the European Society for Swallowing Disorders, Münster, Germany.

Battel, I., Arienti, C., Del Furia, M. J., **Hirschwald, J.**, Lazzarini, S. G., & Walshe, M. (2024, September 25-27). *Rehabilitation interventions for oropharyngeal dysphagia in people with Parkinson's disease: A Cochrane Systematic Review* [Poster presentation]. 14th Congress of the European Society for Swallowing Disorders, Münster, Germany.

Chapter 1: Introduction

The aim of this project is to develop a Core Outcome Set for Dysphagia Interventions in Parkinson's disease (COS-DIP). The first chapter provides an overview of what Core Outcome Sets (COS) are. This is followed by consideration of three key challenges in health research and how these can be addressed by developing and using COS. Thereafter, barriers and facilitators for the uptake of COS in clinical trials are discussed. The importance of the development and use of a COS in dysphagia research, particularly in Parkinson's disease (PD), is then highlighted. Next, a brief overview and guideline for representing and reporting of participants in research studies is provided in order to be adhered to in this research project. This is followed by the research aims, objectives and questions that will be guiding this research project. The chapter concludes with an overview of the individual thesis chapters and a summary of this introduction.

1.1 Core outcome sets

A COS is a standardised set of outcomes that should be measured and reported in all clinical trials for a specific health condition and intervention. It should be devised by key stakeholders, including people with the health condition, healthcare professionals and researchers. This is usually achieved by applying a consensus process. At first, it should be agreed on *what* to measure. Once these core outcomes are established, it should be agreed on *how* to measure them. Ultimately, a COS comprises a minimum of outcomes considered to be important and relevant to all key stakeholders (Gargon, Williamson, et al., 2014; Williamson et al., 2017).

To establish a common understanding of what and how to measure, it is important to define two key concepts. First, what is understood by an outcome:

> "An outcome refers to what is being measured. It is also referred to as a construct or domain. In the context of a clinical trial, it refers to what is being measured on trial participants to examine the effect of exposure to a health intervention." (Prinsen et al., 2016a, p. 3)

Second, the meaning of an outcome measurement instrument (OMI):

> "An OMI refers to how the outcome is being measured. It is a tool to measure a quality or quantity of the outcome. The tool can be a single question, a questionnaire, a score obtained through

physical examination, a laboratory measurement, a score obtained through observation of an image, etcetera." (Prinsen et al., 2016a, p. 3)

Throughout this thesis, the term outcome will be used when discussing *what* is being measured. The term OMI will be used when referring to *how* to measure this outcome, i.e. the specific tool employed.

To support the development and use of COS, the Core Outcome Measures in Effectiveness Trials (COMET) Initiative was launched in 2010. Seven years later, it published its first handbook (Williamson et al., 2017). The document provides guidance on the development, implementation, evaluation and updating of COS. Additionally, the COMET website (www.comet-initiative.org) contains a database, where COS trials can be registered by researchers developing a COS and where people can search for the availability of a specific COS (Williamson et al., 2017).

Up to 2021, 589 COS in more than 29 different health conditions were available with approximately 500 additional, ongoing COS studies (Dodd et al., 2023; Kirkham & Williamson, 2022). This highlights the continuous increase of COS being developed and becoming available for use in research. Overall, I believe that there are three key challenges in health research that can be addressed by developing and using COS. These are described in the following two sections.

1.2 Three key challenges in health research

Heterogeneity in what and how outcomes are measured in health research studies. Researchers often investigate different interventions for the same health condition and each study measures different outcomes or uses different OMIs to measure the same outcome. Moreover, the quality of used OMIs often differs in terms of validity and reliability (Prinsen et al., 2016b). This variability in choice of outcomes and OMIs makes it challenging to compare and synthesise the results of different studies in systematic reviews and meta-analyses (Williamson et al., 2017), which in turn limits our ability to generate high-quality treatment evidence.

Lack of inclusion of all key stakeholders in the research process. In formulating a research question and choosing outcomes, researchers usually prioritise their own perspectives and interests. This may result in research questions or outcomes that have no or only low relevance to healthcare users, particularly people with the health condition and clinicians. Consequently, the treatment evidence may have a reduced impact on clinical practice and perceived benefits. Furthermore, studies have demonstrated that people

with the health condition and clinicians are more interested in functional, social and emotional wellbeing, as well as adverse outcomes and long-term outcomes, than researchers, but due to their lack of involvement, these outcomes are measured less often (Chalmers et al., 2014; Chalmers & Glasziou, 2009).

More than ten years later, this challenge remains, with only 1-18% of clinical trials reporting that they have involved people with the health condition and/or the public in their research process (Nicholls & Campbell, 2024). However, simply including these groups as a tick box exercise is posing an additional challenge. Their perspectives should truly be incorporated into every step of the research, such as developing the research question, designing the trial and deciding what to measure (Nicholls & Campbell, 2024; Richards et al., 2023).

Selective outcome reporting bias. This type of bias is defined as the practice of researchers reporting only a subset of the initially measured outcomes in a published study (Dwan et al., 2013; Hutton & Williamson, 2000). A systematic review found that a comparison of trial publications with their respective protocols showed that between 40-62% of trials had changed, added or omitted at least one primary outcome. Particularly, outcomes with a statistically significant difference had a greater likelihood of being completely presented than outcomes that were not statistically significant (Dwan et al., 2013). Another review found that of the 29 randomised controlled trials (RCTs) that assessed behavioural health interventions and had a study protocol or were registered, 27 (93.1%) had outcomes in the results section of the published manuscript that differed to the outcomes in the study protocol/registration. Of these, 21 studies (72.41%) omitted outcomes in their publication (Matvienko-Sikar, O'Shea, et al., 2024). Selective outcome reporting may result in overestimation of the effect of the evaluated intervention (Dwan et al., 2013) and may consequently compromise the validity of systematic reviews and meta-analyses (Kirkham et al., 2010).

These three challenges ultimately lead to an additional challenge: avoidable research waste. By conducting research studies with high heterogeneity of outcomes and OMIs, lack of stakeholder involvement in the research process, and selective outcome reporting bias, researchers contribute to ongoing but avoidable research waste (Khalil et al., 2022). Research waste is broadly defined as research results without societal benefits (Grainger et al., 2020). In 2009, Chalmers and Glasziou estimated that globally 85% of research is avoidably wasted. Considering that $200 billion is spent on health and medical research worldwide each year, this means that $170 billion is wasted annually (Chalmers & Glasziou,

2009; Glasziou & Chalmers, 2016). This holds still true for today as Pirosca and colleagues (2022) calculated that 88% of randomised trials included in Cochrane reviews of multiple health areas had a high or uncertain risk of bias and thus the authors classified the trials' funding wasted.

Research waste based on these three challenges may result in an evidence base that is both weakened and misleading, which in turn hinders people with the health condition, families and clinicians from informed decision-making (Yordanov et al., 2018). In other words: "poor evidence leads to poor clinical decisions" (Heneghan et al., 2017, p. 120). It is argued that the development and use of COS play a key role in improving the evidence and, consequently, clinical decision making and reducing research waste (Gargon, Williamson, et al., 2014; Williamson et al., 2017).

1.3 How COS address key challenges in health research

The use of COS facilitates higher quality meaningful trials and a reduction of research waste (Gargon, Williamson, et al., 2014; Williamson et al., 2017). This is achieved by specifically addressing the beforementioned three key challenges in health research.

Reduction of heterogeneity of what and how outcomes are measured in clinical trials. COS comprise a standardised, agreed minimum set of outcomes, with many COS also providing agreed definitions and suggestions for high quality OMIs. This leads to greater consistency in the outcomes used, so that trial results can be included in systematic reviews and meta-analyses and conclusions on the safety and effectiveness of interventions can be reached (Williamson et al., 2017).

Study outcomes are relevant to all key stakeholders. In the establishment of the COS all key stakeholders were involved and consequently the core outcomes are of importance to them. Which groups are considered key stakeholders depends on the scope of the COS. In particular, people with the health condition and healthcare professionals should be involved, as their perspectives have been shown to be different from those of researchers (Treweek et al., 2022; Williamson et al., 2012).

Risk of selective outcome reporting bias in studies is being reduced. The COS should be measured and reported as a minimum in all clinical trials (Williamson et al., 2017). It is important to note that a COS does not prevent or restrict researchers from investigating other, additional outcomes. However, the aim is to ensure that the core outcomes are always measured and reported, thereby reducing the risk of selective reporting bias for these core outcomes (Williamson et al., 2012).

By developing and using COS, the most important and relevant outcomes to key stakeholders are included in studies, measured consistently across studies, and reported in a standardised way, making it easier to compare results and synthesise evidence. This leads to more efficient and reliable research, an increase in relevance and applicability of research in clinical practice and helps to inform clinical decision-making, healthcare policy, and the development of guidelines for the management of these specific health conditions. Ultimately, the development and use of COS facilitates the reduction of research waste (Williamson et al., 2017).

However, if the core outcomes are not being measured and reported in clinical trials, they do not fulfil their aim and will not reduce but rather add to research waste (Williamson et al., 2022). Therefore, it is important to support the uptake of COS in health research and to also understand the barriers associated with it (Hughes et al., 2021). Barriers and facilitators to the uptake of COS in clinical trials are outlined in the following section.

1.4 Barriers and facilitators to the uptake of COS

Of 95 RCTs published in five large medical journals in 2019 and 2020, only two trials (2%) used some of the core outcomes relevant to the condition being researched and no trials used all of the core outcomes, despite a COS being available for all trials (Matvienko-Sikar et al., 2022). This raises the question of why COS uptake is so low, given its great potential to improve the quality of clinical trials, and what can facilitate COS uptake (Williamson et al., 2022).

The main suggested barrier to the uptake of COS is that many researchers, funders, and publishers are unaware of the existence and importance of COS, and how to identify, select and apply a COS. As a result, they may not prioritise the use of COS in their research, funding, or publishing decisions. In some cases, clinical trialists may be aware of an existing COS, but have their own outcome preferences and therefore simply do not measure the core outcomes. Another barrier is a lack of recommendations in the COS on *how* to measure the core outcomes. In addition, a lack of validated OMIs for a core outcome may reduce the effectiveness of a COS and therefore be less likely to be adopted in clinical trials. Further, COS comprising a larger number of outcomes may be perceived as an increased expense and participant burden by clinical trialists and are therefore less likely to be adopted. Limited stakeholder engagement, particularly lack of involving people with the health condition, poses another potential barrier to the

uptake of COS, as the core outcomes may be less relevant to key stakeholders. Furthermore, COS that were too broad in scope were also perceived as a barrier to uptake (Hughes et al., 2021; Matvienko-Sikar, Hussey, et al., 2024; Williamson et al., 2022).

Rhodes et al. (2024) reviewed practice guidelines from six countries and the World Health Organisation, covering ten disease groups. None of the guidelines in this study directly recommended the use of COS, and the consistency between the outcomes in the included guidelines and the applicable COS was low. However, in these six countries, one guideline producer (NICE in the UK) recommends the use of COS when developing guidelines. The authors conclude that more research is needed to better understand the barriers and facilitators to implementing COS recommendations in guidelines (Rhodes et al., 2024).

Conversely, it is well established that clinical trialists are more likely to adopt COS when there is an understanding of what COS are and of how to identify, select and apply COS. In addition, adoption is facilitated when clinical trialists recognise the advantages of using COS, feel responsible for adhering to recommendations, and when other clinical trialists in the field endorse and support the use of COS (Matvienko-Sikar, Hussey, et al., 2024).

Trial funders also play a key role in raising awareness of COS among clinical trialists and stimulating the search for and adoption of COS. Beecher et al. (2023) found that after the Irish Health Research Board (HRB) guidance in 2020 not only invited applicants to devise and adopt COS, but also queried whether they had searched the COMET database, this led to an increase in searches of the COMET database for applicable COS among clinical trialists applying for funding from the HRB. While overall the availability of COS is still low in many health research areas, this highlights the important role of funders in increasing clinical trialists' knowledge about COS (Beecher et al., 2023) and ultimately, facilitate uptake of COS.

Overall, trialists, funders, guidelines and others, that endorse the use of COS, are listed on the COMET website (www.comet-initiative.org). For example, the SPIRIT (Standard Protocol Items: Recommendations for Interventional Trials) explanation and elaboration guidance for clinical trial protocols states that "trial investigators are encouraged to ascertain whether there is a core outcome set relevant to their trial and, if so, to include those outcomes in their trial" (Chan et al., 2013, p. 12), when deciding on what to measure in their study. To date, the impact of these recommendations remains partly unknown, but based on the barriers identified, it can be hypothesised that the more funders, guidelines, journals and others recommend COS, the greater the chances that COS uptake will continue to increase.

The barriers of adopting COS primarily apply to health conditions, where COS are available. If no COS has been developed so far, it is even more challenging to decide which outcomes are most important to key stakeholders. One field where development of COS is still scarce is oropharyngeal dysphagia (OD). OD can affect both children and adults and can be caused by many different health conditions. OD can lead to severe medical consequences, such as malnutrition, dehydration, pneumonia, and psychosocial consequences, like a reduced quality of life. Due to OD being very heterogenous, OD interventions need to be specific targeting the underlying pathophysiology (Groher & Crary, 2016). Therefore, specific COS that are more likely to be used in future clinical trials and ultimately improve the evidence base, are needed.

1.5 The role of COS in clinical trials for OD interventions

The first COS for OD interventions was developed in intensive care by Duncan and colleagues (2022). The core outcomes that are included comprise aspiration, severity of aspiration, swallow function, efficiency of cough, and mortality. Definitions and guidance on OMIs and timepoints of measurement are provided as well (Duncan, 2022).

In addition, a COS for patient reported dysphagia interventions for the use in head and neck cancer clinical trials has been developed. The following seven core outcomes are included: difficulty swallowing (in general), difficulty swallowing liquids, coughing when swallowing, choking when swallowing, pain when swallowing, difficulty maintaining weight due to swallowing difficulty and difficulty swallowing impacting quality of life (Manduchi et al., 2024). So far, there is no guidance on how to measure these core outcomes.

One other COS study in OD is currently ongoing. Fan et al. (2022) registered a COS for clinical trials on Traditional Chinese Medicine in OD in the COMET database. At the time of writing, no publications regarding this COS were available.

To date, no COS for OD in a neurological population exists. This is despite the fact that neurological conditions are a leading cause of disability and mortality (Feigin et al., 2019). Furthermore, OD is a highly prevalent symptom in people with neurological conditions, increasing the risk for disability and mortality (Dziewas et al., 2021).

In OD research post stroke, a scoping review and qualitative analysis of patient reported outcome measures (PROMs) identified 30 different outcomes in the included RCTs (Moloney et al., 2023). Besides this large variety of outcomes, only two PROMs with robust psychometric properties were identified. Only 10% of the included studies used one of these two validated PROMs and 17% of studies used any PROM regardless of the psychometric properties. In addition, the two validated PROMs are not specific for OD post stroke but were developed for OD in general. This highlights two important limitations in RCT design and conduct in OD interventions post stroke. First, the significant heterogeneity of OMIs makes synthesising and comparing study results in systematic reviews and meta-analyses challenging and therefore limits the ability to direct treatment for patients. Second, due to the lack of validated PROMs and use thereof, patients' perspectives and experiences cannot be truly captured and measured in OD interventions post stroke, possibly making study results less relevant to patients. The authors therefore suggest considering the development and use of a COS for OD interventions post stroke (Moloney et al., 2023). This challenge is not unique to clinical trials including people post stroke but also affects OD interventions for people with PD.

1.6 The role of COS in clinical trials for OD interventions in PD

PD is the fastest growing neurological condition in terms of prevalence, disability and mortality in the world. In 2016, an estimated 6.1 million people were living with PD globally (Dorsey et al., 2018; Feigin et al., 2019). OD can affect more than 80% of people living with PD, depending on the OMI used (Kalf et al., 2012) and pneumonia is a leading cause of death in people with PD (Hobson & Meara, 2018; Hoehn & Yahr, 1967). As the prevalence of PD increases, more people will be living with OD in PD, thereby amplifying the need for evidence-based OD interventions in PD.

An initial search of the COMET database (www.comet-initiative.org) was conducted on 03 February 2021 and revealed no planned, ongoing, or published COS for OD intervention studies in PD. Two published COS studies on PD in general, but not on OD specifically, were identified (de Roos et al., 2017; Sjödahl Hammarlund et al., 2012). Both PD COS studies comprise outcomes related to swallowing but are not specific to OD and both lack the involvement of experts in OD, such as speech and language therapists (SLTs). However, the two studies provide preliminary evidence that OD in PD is relevant but a specific COS for OD in PD with involvement of all key stakeholders is needed.

1.7 Research aims, objectives and questions

The aim of this research study is to develop an agreed standardised COS measured and reported as a minimum in all clinical trials for OD in PD. The objectives of this study are:

1. To establish a long list of outcomes based on identified outcomes in the literature in a scoping review and survey and focus groups with people with OD in PD, family members, family caregivers and dysphagia clinicians.
2. To determine core outcomes for the COS-DIP through a three round online Delphi survey with international key stakeholders.
3. To achieve consensus on the core outcomes included in the COS-DIP, their definitions, OMIs and timepoints of measurement through two online consensus meetings with representative international key stakeholders.

Based on these objectives the following research questions are sought to be answered:

1. What outcomes are measured in clinical trials on OD interventions in PD?
    1.1. How are the outcomes in these clinical trials defined?
    1.2. How are the outcomes in these clinical trials measured?
    1.3. At which timepoints and at which frequency are the outcomes in these clinical trials measured?

2. What are the outcomes of interest to people with OD in PD, family members and caregivers when deciding if a swallowing treatment works?

3. What are the outcomes of interest to dysphagia clinicians working with people with OD in PD?
    3.1. How do dysphagia clinicians measure these outcomes?
    3.2. When do dysphagia clinicians believe should these outcomes be measured?

4. What are the core outcomes of interest to measure in clinical trials in OD interventions in PD for healthcare professionals, researchers, people with OD in PD, family members and caregivers?
    4.1. How are these core outcomes defined?
    4.2. How should these core outcomes be measured?
    4.3. At which timepoints should these outcomes be measured?

Each of the research question and its according sub-questions will be addressed in an individual chapter in this thesis. This is outlined in the following section.

1.8 Overview of thesis

This thesis comprises of eight chapters with the first one being the introduction. Chapter 2 to 8 are briefly described in the following:

- Chapter 2: introduces the guidelines and recommended and commonly applied methods for developing a COS, including the preparatory stage, finding consensus on what core outcomes to measure and how to measure them.
- Chapter 3: describes the methods used to develop the COS-DIP study protocol.
- Chapter 4: aims to answer research question 1 "What outcomes are measured in clinical trials on OD interventions in PD?". A scoping review synthesising the currently measured OD intervention outcomes, their definitions, OMIs and timepoints and frequencies of measurement in OD intervention studies in PD was conducted. This is the first step in developing a long list of outcomes for the COS-DIP.
- Chapter 5: aims to answer question 2 "What are the outcomes of interest to people with OD in PD, family members and family caregivers when deciding if a swallowing treatment works?". An online survey and focus groups with people with OD in PD, family members and caregivers were conducted to obtain their perspectives and gain a deeper understanding in why these outcomes are important. The findings complement the long list of outcomes from chapter 4.
- Chapter 6: aims to answer research question 3 "What are the outcomes of interest to dysphagia clinicians working with people with OD in PD?". An online survey collecting dysphagia clinicians' views was conducted. The findings complete the long list of outcomes for the following study.
- Chapter 7: aims to answer research question 4 "What are the core outcomes of interest to measure in clinical trials in OD interventions in PD for key stakeholders?". A Delphi survey and subsequent consensus meetings to agree on the core outcomes, their definitions, OMIs and timepoints of measurement were conducted with international key stakeholders.

- Chapter 8: concludes the thesis by including a discussion of the studies and their results, an evaluation of the research's strengths and limitations, and implications for clinical practice and future research in this field.

1.9 Summary

In summary, COS are standardised sets of outcomes that should be measured and reported in all clinical trials for a specific health condition and intervention. They are devised by key stakeholders, such as people with the health condition, healthcare professionals, and researchers, to ensure that the core outcomes are relevant to all stakeholders. COS are required in health research to ensure consistency and standardisation in the selection and measurement of outcomes across studies. Through decreasing heterogeneity of outcomes and selective outcome reporting, study results can be compared in systematic reviews and meta-analyses, and the evidence base can be evaluated more reliably. Ultimately, this will reduce research waste. Although a large number of COS are available, only one each for OD interventions in intensive care and head and neck cancer but none for PD have been developed so far.

The development of the COS-DIP will help ensure that clinical trials on OD interventions in PD are measuring the aspects of OD in PD most important to all key stakeholders and will allow for more accurate comparisons between studies. This, in turn, will improve the quality of evidence available to inform clinical decision-making and ultimately lead to better outcomes for people with OD in PD.

Chapter 2: Methods

This chapter starts by introducing the available guidance documents for developing COS. This is followed by a detailed discussion of the individual stages for developing a COS. In the preparation stage of a COS, it is essential to define the scope of the COS, draft a study protocol and register the study. As part of this process, the key stakeholders, who should be included in the COS study, are to be identified. The next step is to evaluate what should be measured, i.e. to determine the core outcomes. This involves developing a long list of outcomes that includes all key stakeholder perspectives. This long list is then reduced to the most important outcomes (core outcomes) through applying a consensus process involving all key stakeholders. The final step is to determine how the core outcomes will be measured. This consists of identifying existing OMIs, assessing their quality and then building consensus among key stakeholders on the appropriate OMI for each core outcome.

2.1 Guidance on COS development and reporting

Several guidance documents on COS development and reporting are available. Table 2.1 provides an overview of these.

Table 2.1: Guidance documents for COS development and reporting

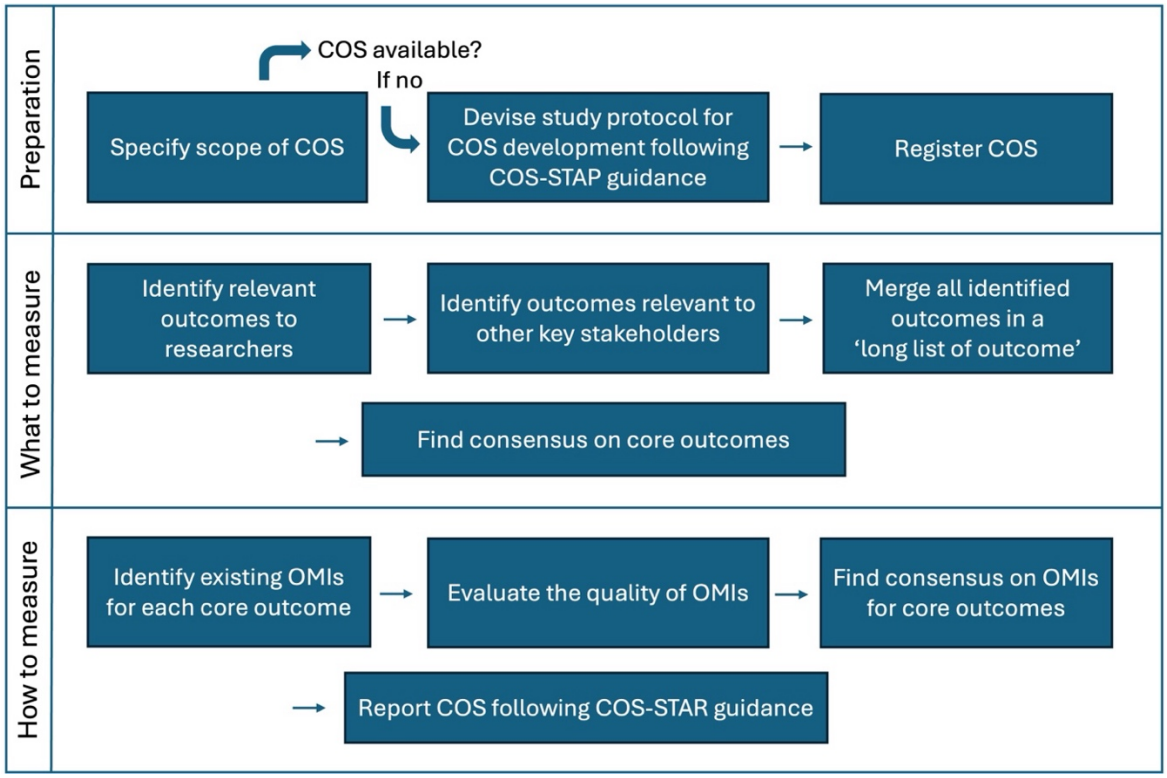
Guidance name	**Primary area addressed by guidance**
COMET handbook (Williamson et al., 2017)	COS development
COS-STandards for Development (COS-STAD) recommendations (Kirkham et al., 2017)	Minimum standard in COS development
How to select OMIs for outcomes included in a COS – a practical guideline (Prinsen et al., 2016b)	How to select OMIs in COS studies
COS-STAndardised Protocol (COS-STAP) statement (Kirkham et al., 2019)	Reporting of COS study protocols
COS-STAndards for Reporting (COS-STAR) statement (Kirkham et al., 2016b)	Reporting of COS studies
OMERACT handbook (Beaton et al., 2021)	COS development in rheumatology

COMET: Core Outcome Measures in Effectiveness Trials, COS: Core Outcome Set, OMERACT: Outcome Measures in Rheumatology, OMI: Outcome Measurement Instrument

One of the largest and longest established collaborations to standardise outcomes is the Outcome Measures in Rheumatology (OMERACT) initiative (Beaton et al., 2021; Williamson et al., 2017). OMERACT describes itself as a leader in COS methodology and has developed its own manual containing practical recommendations for conducting COS (Beaton et al., 2021)[1]. Many COS have been published in the field of rheumatology and the uptake of COS ranges from 0% (gout) up to 82% (rheumatoid arthritis) (Hughes et al., 2021). It is hypothesised that the rheumatoid arthritis COS is adopted so successfully due to its incorporation into existing policies and funding frameworks (Kirkham et al., 2013).

There are no standardised methods in developing a COS, yet. A variety of methods have been used by COS developers and it remains to be investigated which methods are the most effective and efficient (Williamson et al., 2017). However, to maintain a minimum standard both the COMET handbook (Williamson et al., 2017) as well as the Core Outcome Set-STandards for Development (COS-STAD) recommendations (Kirkham et al., 2017) list several stages for developing a COS. These are combined and depicted in Figure 2.1 and explained in more detail in the following sections. Where applicable, the recommendations will be complemented by suggested methods from the OMERACT handbook (Beaton et al., 2021).

[1] An updated version has been announced on their website (www.omeract.org) for May 2025, but was not available at the time of writing.



COMET: Core Outcome Measures in Effectiveness Trials, COS: core outcome set, COS-STAR: Core Outcome Set-STAndards for Reporting, OMI: outcome measurement instrument

Figure 2.1: Recommended core outcome set development process by Williamson et al. (2017) and Kirkham et al. (2017)

2.2 Preparation

Before consensus can be sought for the core outcomes, preparatory work should be completed. This comprises specifying the scope of the COS, writing a study protocol and making it publicly available, registering the COS study and deciding on the stakeholders to be involved throughout the study (Kirkham et al., 2017; Williamson et al., 2017).

2.2.1 Scope of the COS

The scope of a COS should cover all sections of the PICO (Population, Intervention, Comparator, Outcome) question format for clinical trials, except for the outcomes (Williamson et al., 2017), as determining these is the aim of the COS. The first four standards of the COS-STAD recommendations further refine this and suggest to specify the scope regarding "the research or practice settings (standard 1) in which the COS is to be applied, health conditions (standard 2), target populations (standard 3), and interventions (standard 4)" (Kirkham et al., 2017, p. 5). A clearly defined scope at the beginning will enable COS developers to ascertain whether an existing COS is available and will assist future COS users

in determining whether the COS is suitable for their requirements (Kirkham et al., 2017; Williamson et al., 2017).

In general, the overall scope or their specifications may be either broad or narrow. For example, the COS for PD by de Roos et al. (2017) encompasses a broad range of health conditions (all motor and non-motor PD symptoms) and interventions (all common treatment options for PD, i.e. pharmacological, surgical and behavioural). However, it is narrow in terms of the population (only idiopathic PD). In contrast, the COS-Speech by Mitchell et al. (2023) is designed for a broad population (adults post-stroke), but covers a smaller range of conditions (only dysarthria, no other symptoms such as dysphagia) and consequently, a narrower range of interventions (dysarthria treatment only). However, the setting is broad (research and clinical practice) (Mitchell et al., 2023).

A broader scope increases the probability of a greater number of outcomes being included into the COS. As stated in section 1.4, this can subsequently impede the adoption of the COS. Paul et al. (2023) have identified that numerous outcomes of the PD COS are underreported, and since the publication of the COS, there has been no significant increase in reporting of the core outcomes. One potential explanation for the low uptake may be that the COS encompasses 32 outcomes (de Roos et al., 2017), due to the extensive range of interventions included.

2.2.2 Study protocol and registration

Once the scope of the COS has been defined, the COMET database should be used to search whether a relevant COS already exists. If no relevant COS is available, then a study protocol should be written, and the study should be registered (Williamson et al., 2017).

It should be noted that neither a study protocol nor pre-registering the study were voted in the COS-STAD recommendations during the Delphi study (Kirkham et al., 2017). Particularly, the stakeholder group "COS developers" rated these two items the least often *important* whereas the stakeholder group "patient representatives" voted the two items the most often *important* (S2 Table in Kirkham et al., 2017).

Nevertheless, both the COMET and the OMERACT handbook recommend writing a study protocol following the Core Outcome Set-STAndardised Protocol (COS-STAP) statement (Kirkham et al., 2019) before commencing the COS study and making it publicly available (Beaton et al., 2021; Williamson et al., 2017). In addition, it is advised to register the COS study as this will help to avoid duplicates and to

identify planned, ongoing and completed COS more easily. This can be done on the COMET database via the COMET website (www.comet-initiative.org) (Beaton et al., 2021; Williamson et al., 2017).

2.2.3 Stakeholder involvement

As part of the study protocol, the key stakeholders to be involved in the COS developing process need to be specified. As a minimum, the COS-STAD suggests including people who will apply the COS in the future (e.g. researchers, industry), healthcare professionals in the field of the COS, people with the health condition and/or their advocates (Kirkham et al., 2017).

Although it may appear self-evident to include researchers who will subsequently utilise the COS in their clinical trials, it is also essential to incorporate the perspectives of healthcare professionals who have direct experience of delivering the intervention in clinical practice. Treweek and colleagues (2022) found that people living with the health condition and healthcare professionals, only agree 28% of the time with researchers when choosing the primary outcome in breast cancer and nephrology trials. While people with the health condition generally ranked the primary outcome higher in breast cancer trials than healthcare professionals did, the opposite was true in nephrology trials. This finding underscores the necessity for a more inclusive approach in the selection of outcomes, one that incorporates a wider range of stakeholders, including, but not limited to, researchers.

The inclusion of people with the health condition and/or their advocates is becoming more prevalent in COS studies (Biggane et al., 2018). However, some COS developers perceive the inclusion of this stakeholder group as challenging, noting that lay people encounter more difficulties in comprehending the prioritisation process to the same extent as researchers and healthcare professionals (Gargon et al., 2017).

Despite this, a recent systematic review demonstrated that the inclusion of people with the health condition is critical as their participation in COS development significantly influences outcome selection. When people with the health condition are involved, there is a higher likelihood of including outcomes from Dodd et al.'s (2018) "Life impact" core area. This comprises outcomes related to the impact of the disease on physical, social, and emotional functioning, quality of life and the perceived health status. This finding highlights the critical role of people with the health condition in ensuring that COS capture outcomes directly relevant to their experiences and priorities, which might not be prioritised by clinicians

or researchers alone (Dodd et al., 2023). Ultimately, this will facilitate the uptake of the COS in future clinical trials (see section 1.4).

2.3 What to measure

To find consensus on the core outcomes, it is recommended to first develop a long list of outcomes comprising the perspectives of all key stakeholders, often completed through literature reviews and qualitative interviews (Kirkham et al., 2017). To find consensus on the core outcomes within the long list of outcomes, Delphi surveys and consensus meetings are commonly used methods (Biggane et al., 2018; Cao et al., 2024).

2.3.1 Relevant outcomes to researchers

Most COS developers start with conducting a systematic review to identify what outcomes researchers prioritise when assessing an intervention in relevant clinical trials (Cao et al., 2024). This is in line with recommendations in the COMET handbook (Williamson et al., 2017). Systematic reviews have a high level of evidence and should be conducted with rigorous methodology (Cao et al., 2024). For example, to develop the COS for OD in intensive care, a systematic review was conducted. The objective was to evaluate the effectiveness of OD interventions in comparison to standard care in enhancing oral intake and reducing the incidence of aspiration in adults receiving acute and critical care (Duncan et al., 2020).

Cao et al. (2024) assessed the quality of 175 systematic reviews conducted as part of developing a COS using the AMSTAR 2 tool (Shea et al., 2017). The overall confidence in the included systematic reviews was found to be “critically low” or “low” for more than 90% of the studies as per AMSTAR 2. The authors conclude that this could affect the overall quality of the COS (Cao et al., 2024).

This is problematic, as systematic reviews with poor methodology add to ongoing research waste instead of facilitating its reduction (Khalil et al., 2022). In light of the shortcomings in the quality of systematic reviews for COS development, it is worth reconsidering the overall suitability of this particular method of knowledge synthesis in developing a long list of outcomes.

An alternative to systematic reviews are scoping reviews. While systematic reviews often focus on assessing the effectiveness of interventions and primarily include RCTs, scoping reviews are rather used to systematically map the research done in an area, as well as to identify existing gaps in knowledge. It is

not uncommon for other clinical trial designs, in addition to RCTs, to be included (Arksey & O'Malley, 2005; Munn et al., 2018). This can result in a greater number of outcomes being included in the long list, due to the inclusion of a greater variety of studies. Scoping reviews have also shown to be useful in mapping outcomes, their definitions and OMIs to inform the establishment of a COS (Khalil et al., 2022) and are recommended by the OMERACT handbook (Beaton et al., 2021). Clearly, also scoping reviews need to follow rigorous methodology, such as guidance from the Joanna Briggs Institute (JBI) (Peters et al., 2021), to contribute to the knowledge base effectively. Despite the possibility of identifying more outcomes, it is anticipated that the choice of a systematic or scoping review will not have any other implications for the COS study. However, the most suitable review type and its impact on subsequent COS development stages still need to be systematically evaluated.

2.3.2 Relevant outcomes to people with the health condition and healthcare professionals

Regardless of whether a systematic or scoping review is conducted, the identified outcomes are likely to reflect the perspectives of the researchers conducting the studies, particularly when there is limited or no involvement of people with the health condition and the public. It is therefore recommended that the perspectives of healthcare professionals and people with the health condition be assessed in order to add relevant outcomes to the long list of outcomes (Kirkham et al., 2017; Williamson et al., 2017).

A common approach is to conduct qualitative interviews and/or focus groups with these stakeholder groups. This allows for the collection of information regarding which outcomes are considered important and are not currently measured in research studies, the reasons why these outcomes are important, and the most appropriate language to use in order to ensure the future steps of the research are accessible to all stakeholders (Keeley et al., 2016). Questions that have been asked in previous qualitative research to identify important outcomes were both broader in nature, e.g. "What is important in terms of your health?" (Sanderson et al., 2010, p. 642), and more specific, e.g. "What are your goals in living with your symptoms?" (Retzer et al., 2020, see supplemental material 1) or "How do you know when a treatment is working?" (Sanderson et al., 2010, p. 642). The OMERACT handbook recommends including a minimum of 30 participants in qualitative studies (Beaton et al., 2021). Although there is no standard approach to analysing qualitative data for COS development, it has been recognised that coding, labelling and categorising data supports the aim of determining important outcomes for subsequent stages of the COS

project. While qualitative research adds in depth insights, it may be that less additional outcomes are identified by participants and overall, it can be more time consuming (Keeley et al., 2016).

In addition to this approach, few studies also incorporate quantitative surveys instead or in addition to qualitative research to identify important outcomes to these stakeholder groups (Jones et al., 2017; Sinha et al., 2012). Using a survey, a broader range of perspectives can be gathered and the burden on participants can be reduced (Sinha et al., 2012). For example, Sinha et al. (2012) received responses through their survey from 49 parents of children with asthma. In addition, researchers could conduct subsequent interviews or focus groups to validate the survey results and clarify inconclusive results (Pope & Mays, 2020). However, to date no study has been found which applied such a mixed methods approach, i.e. survey and interviews/focus groups, when developing a COS.

2.3.3 Long list of outcomes

In a next step, all identified outcomes from the different stakeholders (sections 2.3.1 and 2.3.2) should then be combined in a long list of outcomes. Different methods for categorising outcomes have been proposed. One outcome taxonomy has been devised by the COMET team particularly for developing COS in medical research. It comprises five core areas and 38 outcome domains into which outcomes can be classified (Dodd et al., 2018).

While it is important to include all key stakeholder perspectives when devising the long list of outcomes (section 2.2.3), the list should not become overly long. Gargon and colleagues (2019) found that Delphi surveys that initially included more outcomes had significantly lower response rates. Specifically, for every ten additional outcomes, the response rate decreased by 1.4% (Gargon et al., 2019). For this reason, the OMERACT handbook recommends that a maximum of 70 outcomes, preferably fewer, should be included in the initial long list of outcomes. If there are more than 70 outcomes, COS developers are advised to revise their long list of outcomes, for example by removing duplicates (Beaton et al., 2021).

In addition to the technical outcome terms on the long list, lay descriptions that are accessible and understandable to people with the health condition should be provided (Williamson et al., 2017). However, clear guidance on how to create such lay terms is limited. One option is to base lay terms on language commonly used by people with the health condition and family members as identified in qualitative research (see section 2.3.2). These lay descriptions are then oftentimes further refined through

discussion within the study steering committee (SSC), particularly in collaboration with patient partners[2] (e.g. Katiri et al., 2022; Mitchell et al., 2023). Once agreed upon within the SSC, the wording can be piloted or pretested using cognitive or think aloud interviews to assess the comprehensibility and allow further refinement, if needed (Williamson et al., 2017).

2.3.4 Consensus on core outcomes

In the next step, the aim is to reduce the long list of outcomes to the core outcomes that are important to all key stakeholders. Different methods exist and have been used in previous COS studies, for example expert panel, nominal group technique, Delphi survey and consensus meeting (Williamson et al., 2017). A Delphi survey alone or with subsequent consensus meetings is the most commonly applied method (Biggane et al., 2018) and recommended by both the COMET and OMERACT handbook (Beaton et al., 2021; Williamson et al., 2017). For this reason, these two approaches are explained in more detail in the following sections.

2.3.4.1 Delphi survey

In a Delphi survey to develop a COS, participants are presented with the long list of outcomes and are asked to rate or rank the importance of each outcome (Alkhaffaf et al., 2021). A minimum of two rounds is required to constitute a Delphi survey, with two to three rounds being the most common in COS studies. At the beginning of the second and, if applicable, third round, participants receive feedback on the results of the previous round and are asked to re-rate or re-rank the importance of each outcome (Williamson et al., 2017). This gives participants the opportunity to reconsider their decision based on their own rating/ranking as well as that of other stakeholder groups (Alkhaffaf et al., 2021). Due to Delphi surveys being anonymous the influence of dominant participants can be avoided. Oftentimes Delphi surveys are conducted online which enables the involvement of a large number of international stakeholders from a wide geographical area (Williamson et al., 2017).

The COMET initiative has developed the web-based system DelphiManager specifically designed to find consensus on core outcomes through online Delphi surveys. Access can be purchased through the

[2] A patient partner is defined as a person "with their own health issues and experiences and includes caregivers, family members, and friends, who actively contribute to research […]. [It] does not refer to participants in research studies […]." (Richards et al., 2023, p. 2).

University of Liverpool (www.comet-initiative.org/delphimanager). Use of this platform automatically comes with some default settings, such as that outcomes are rated instead of ranked regarding their importance. While both rating and ranking in Delphi surveys likely lead to similar prioritisation of outcomes, ranking is perceived as more difficult by participants and the time taken to complete each round increases (Del Grande & Kaczorowski, 2023). Nevertheless, OMERACT has entered into an agreement with COMET for the adaptation of their DelphiManager. Consequently, the *DelphiManager modified for OMERACT* employs a ranking methodology and can be used as an alternative to the original DelphiManager. No justification for choosing ranking over rating is provided in the OMERACT handbook (Beaton et al., 2021).

In DelphiManager, outcomes are rated on a 9-point Likert scale as recommended by the Grading of Recommendations Assessment, Development and Evaluation (GRADE) Working Group (Crew & Williamson, n.d.; Guyatt et al., 2011). The most commonly used scales in Delphi surveys are 5-point and 9-point scales (Niederberger & Spranger, 2020). There is conflicting evidence whether the use of a 9-point scale compared to a 5-point scale reduces the total number of items that reach consensus (Remus et al., 2021) or whether it increases it (Lange et al., 2020). It most likely impacts the final results, but the direction of impact remains inconclusive. In DelphiManager, the scales 1 to 3 are labelled as *not important*, 4 to 6 *important but not critical* and 7 to 9 *critical*. A button "unable to rate" and the option to provide feedback is also available (Crew & Williamson, n.d.).

When the long list of outcomes is uploaded to DelphiManager, each outcome is accompanied by a definition called a "help text". This is in line with COS-STAD recommendations to provide clear language for all participants, both professionals and lay people. Ideally, the terms are piloted with stakeholders prior to the Delphi survey to ensure comprehensibility (Kirkham et al., 2017).

In accordance with the COMET handbook, various aspects need to be agreed upon before starting a Delphi survey, as all of them could potentially influence the results. For a comprehensive overview, the reader is referred to the COMET Handbook, section 2.7.6.1. (Williamson et al., 2017). The following section describes the group size, additional outcomes, feedback type, retaining or dropping outcomes and consensus criteria.

Group size. Determining the number of people to involve in the Delphi survey is not a question of statistical power but is usually a practical decision. Generally, the more participants the better. It is of

particular importance that each stakeholder group is well represented (Williamson et al., 2017). While the COMET handbook does not provide a recommended range, the OMERACT handbook suggests including at least 100 participants per stakeholder group in the first round. Although there are strategies to reduce attrition, it is not possible to eliminate it entirely. This approach ensures that at least 30-50 participants in each stakeholder group will complete all Delphi rounds (Beaton et al., 2021).

Additional outcomes. At the beginning of the first round, participants can be asked to list the outcomes that are most important to them. This will verify that no key outcome was omitted. Alternatively, participants can be asked at the end of the first round whether any important outcomes were missing in the long list of outcomes and if so, to specify them in a provided comment box. This ensures that a comprehensive long list of outcomes important to each stakeholder group is included (Sinha et al., 2011; Williamson et al., 2017). Criteria for when to include additional outcomes should be set a priori. This could mean that any proposed outcome not yet included in the long list of outcomes will be added in the second round or, for example, only if it has been suggested by at least two participants (Williamson et al., 2017).

Feedback type. Feedback on ratings from the preceding round is typically provided for each outcome in the subsequent round. This enables participants to consider diverse perspectives and ultimately achieve consensus on the core outcomes. The type of feedback can comprise individual ratings, ratings of the according stakeholder group (peer group) or across all stakeholder groups (Williamson et al., 2017). Brookes et al. (2016) found that consensus was greater when participants were given feedback from all stakeholder groups instead of only their peer group. Barrios et al. (2021) demonstrated that perspectives shifted towards those where strong consensus (≥ 70%) was achieved in one group and moved away from this perspective if weak consensus (< 70%) was achieved, thereby impeding the process of reaching consensus.

Retaining or dropping outcomes. Between Delphi rounds, all outcomes may be retained, or some may be dropped based on predetermined criteria (Williamson et al., 2017). While the COMET handbook does not provide clear recommendation for one or the other, the OMERACT handbook suggests dropping outcomes between rounds (Beaton et al., 2021). As the aim is to reduce the long list of outcomes to the most important ones, it seems reasonable to drop outcomes between rounds when developing a COS. This

also reduces the time needed to complete each round, thereby reducing the participants' workload (Williamson et al., 2017).

Consensus criteria. Different criteria to drop and retain outcomes are being used to ultimately find consensus on the core outcomes. Many COS studies include outcomes into the next round if they are rated 7 to 9 on a 9-poin Likert scale by ≥ 70% of participants and 1 to 3 by ≤ 15% of participants, e.g. in the field of SLT Mitchell et al. (2024) and Katiri et al. (2022). This ensures that the majority of participants in all stakeholder groups perceives this outcome to be critically important and only a very small proportion believes it is not important (Williamson et al., 2017). The OMERACT handbook only uses the ≥ 70% of participants rated 7 to 9 criterion (Beaton et al., 2021).

The disadvantage of this approach is that too strict criteria in the first round can lead to outcomes being dropped too soon and participants do not have the opportunity to reassess their importance. But given that an extensive, long list of outcomes has been developed beforehand, applying overall more inclusive criteria would result in rating and re-rating all outcomes to be time-consuming and may increase attrition between rounds (Williamson et al., 2017). Therefore, Blazeby et al. (2015) used broader inclusion criteria after Round 1 with > 50% of participants rating an outcome 7 to 9 and < 15% rating 1 to 3, and stricter inclusion criteria after Round 2 with > 70% of participants rating an outcome 7 to 9 and < 15% rating 1 to 3.

There is still a lack of evidence on the best method to use, although this is a critical decision as it most likely affects the final core outcomes. The choice of specific criteria by COS developers may depend on the initial length of the long list of outcomes. Whichever method COS developers choose, it should be specified a priori in a study protocol (Williamson et al., 2017). As per COS-STAD recommendations this includes a minimum of pre-specifying the consensus method and criteria for adding, retaining and dropping outcomes (Kirkham et al., 2017).

2.3.4.2 Consensus meeting

It is recommended that a consensus meeting be held after the Delphi survey to agree on the final core outcomes. For this, most COS developers use an informal approach with pre-specified consensus criteria. An important decision to be made is who to invite to these meetings. Some COS developers conduct the consensus meeting only with members of their SSC, others with previous Delphi survey participants, including people with the health condition (Williamson et al., 2017). Recommendations for including

people with the health condition and the public are available on the COMET website (www.comet-initiative.org/Patients/Consensus).

Particularly before the COVID-19 pandemic, meetings were often face-to-face and lasted about half a day (Williamson et al., 2017). During and after the pandemic, consensus meetings are now also held online, facilitating the participation of international stakeholders, and last about 2 to a maximum of 4 hours (Gorst et al., 2021).

This section 2.3 has described the recommended and commonly used methods for agreeing *what* to measure in clinical trials for a particular health condition, i.e. the core outcomes. However, as highlighted in section 1.4, one barrier to the uptake of a COS is the lack of recommendations on *how* to measure these core outcomes, i.e. how to define core outcomes, which OMIs to use and when to use them. This is addressed in the following section.

2.4 How to measure

In 2016, the COMET and COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) groups published recommendations on deciding how to measure the core outcomes (Prinsen et al., 2016b). Consensus was achieved through a Delphi survey on four key steps in the selection of OMIs to be included in COS (Prinsen et al., 2016b). These are depicted in Figure 2.2.



OMI: outcome measurement instrument

Figure 2.2: Recommended steps in selecting outcome measurement instruments for core outcome sets by Prinsen et al. (2016b)

2.4.1 Defining the concept

This comprises defining the construct (i.e. outcome or domain) to be measured and the target population to be addressed (Prinsen et al., 2016b). As part of section 2.3, if agreement on the core outcomes and a clear definition of the scope was achieved, this constitutes the completion of this initial step.

2.4.2 Identifying existing OMIs

The next step is identifying existing OMIs. It is recommended to first search for a good quality systematic review on OMIs for the specific health condition in the target population. If none is available, it is recommended to conduct one (Prinsen et al., 2016b). As discussed in the COMET handbook, one option is to extract OMIs when identifying outcomes that are currently measured in published research studies (section 2.3.1) (Williamson et al., 2017). The COMET and COSMIN guideline suggests conducting a separate systematic review with the primary aim of identifying all OMIs and using appropriate search terms (Prinsen et al., 2016b). Recommended methods on how to conduct these systematic reviews are provided in the OMERACT handbook (Beaton et al., 2021), JBI reviewer's manual[3] (Stephenson et al., 2019) or COSMIN guideline[4] (Mokkink et al., 2024). It is now possible to access reporting guidelines in the form of the PRISMA-COSMIN for OMIs, which should be adhered to (Elsman et al., 2024).

2.4.3 Evaluating the quality of OMIs

Once the OMIs have been identified in step 2, their methodological quality should be evaluated. In order to do so, it is recommended that the nine measurement properties be assessed (Prinsen et al., 2016b) in accordance with the COSMIN taxonomy (Mokkink et al., 2010b). The content validity (including face validity) of the OMI should first be assessed. If it is not possible to identify what the OMI measures (unknown content validity) or if the content validity is poor, the OMI should be excluded, and the remaining eight measurement properties do not require evaluation. If the OMI presents good content validity, the structural validity and internal consistency should be assessed. Similarly, if it is found to be unknown or poor, the OMI should be excluded, and the remaining six measurement properties can be disregarded for this OMI (Prinsen et al., 2016b). In case the OMI also presents good structural validity and internal consistency, the remaining measurement properties should be assessed (reliability,

[3] In the updated JBI manual for evidence synthesis (Aromataris et al., 2024) chapter 12 on systematic reviews of measurement properties was removed. Instead, the manual refers the reader to the COSMIN guidelines (www.cosmin.nl/tools/guideline-conducting-systematic-review-outcome-measures).

[4] This guideline is specifically for conducting systematic reviews for PROMs. However, it was amended to other types of OMIs, such as clinician reported outcome measures (www.cosmin.nl/tools/guideline-conducting-systematic-review-outcome-measures).

responsiveness, measurement error, hypothesis testing, criterion validity, cross-cultural validity) (Prinsen et al., 2016b).

In addition to assessing the quality of the OMI itself, the quality of the validation study of the specific OMI should be evaluated (Prinsen et al., 2016b) following the COSMIN checklist (Mokkink et al., 2010a). As a third step, researchers should ensure that the OMI is feasible to be used in clinical trials. This comprises aspects such as ease of administering the OMI, length and completion time of the OMI and cost and required equipment of the OMI (Prinsen et al., 2016b).

2.4.4 Following recommendations for choosing OMIs

As the final step, the COMET and COSMIN guidance offers general recommendations for selecting OMIs in COS. As a first principle, only one OMI should be specified for each core outcome. This OMI should demonstrate at least good content validity and good internal consistency and should be feasible to utilise in clinical trials. Finally, consensus should be reached among all key stakeholders on the OMIs for the core outcomes (Prinsen et al., 2016b).

If no OMI meets the specified criteria for a core outcome, e.g. poor or unknown content validity, the OMERACT handbook advises that this should be regarded as a research gap. In such instances, COS developers should highlight the gap and encourage the development and validation of a suitable, high-quality OMI for that outcome (Beaton et al., 2021).

2.5 Report of COS

Once all steps have been completed and consensus has been achieved on what outcomes to measure and how to measure them, the COS study should be reported following the COS-STAndards for Reporting (COS-STAR) statement (Kirkham et al., 2016b). In 2014, Gargon et al. identified inconsistent reporting of COS studies, leading to a lack of transparency in the methods used (Gargon, Gurung, et al., 2014). As a result, end users, such as clinical trialists and guideline developers, face difficulties in identifying appropriate COS (Kirkham et al., 2016b). This may ultimately impede COS uptake.

2.6 Summary

This chapter provides principles and procedures for developing a COS. This comprises agreeing on what to measure as well as how to measure it. Clear guidance on the best methods is still missing. However, adhering to the available recommendations and following reporting guidelines will ensure that rigorous methodology is transparently applied, and COS are of good quality. Ultimately, this will increase the likelihood of its uptake in clinical trials.

The COS-STAD recommendations did not agree on a study protocol as a minimum standard. However, having such a protocol can still enhance the transparency of a research project. Following open research guidelines, it should be written prior to initiating the COS development and should be accessible to the public (Kirkham et al., 2017). For this reason, a study protocol for developing the COS-DIP was written and published. This is reported on in the next chapter.

Chapter 3: Development of a core outcome set for dysphagia interventions in Parkinson's disease (COS-DIP): study protocol[5]

3.1 Introduction

This chapter describes the study protocol to devise an agreed, standardised COS-DIP measured and reported as a minimum for all clinical trials for OD in PD. The objectives of the COS-DIP study are: (1) To establish the long list of outcomes by identifying OD in PD intervention outcomes that are measured in OD intervention studies in PD through a scoping review and outcomes that are important to people with OD in PD, family members and caregivers and dysphagia clinicians through online surveys and focus groups. (2) To determine core outcomes for the COS-DIP through a three round online Delphi survey with international key stakeholders. (3) To achieve consensus on the core outcomes included in the COS-DIP, their definitions, OMIs and timepoints of measurement through two online consensus meetings with representative international key stakeholders.

3.1.1 Scope of the COS

The COS-DIP is devised primarily for use in future clinical trials. The health conditions that will be covered by the COS are all types and severities of OD and the population all individuals with idiopathic PD regardless of disease severity and duration. The COS will focus on interventions that directly improve swallowing difficulties, and/or reduce specific physiological risks associated with OD, such as pneumonia, malnutrition, or choking.

3.2 Methods

This study is developed in line with guidance from the COMET Handbook (Williamson et al., 2017), the COS-STAD recommendations (Kirkham et al., 2017) and the guideline on how to select OMIs for outcomes included in a COS by the COSMIN and COMET initiative (Prinsen et al., 2016b). The study

[5] Adapted from: Hirschwald, J., Duncan, S., Warnecke, T., Boyle, G., Regan, J., & Walshe, M. (2024). Development of a Core Outcome Set for Dysphagia Interventions in Parkinson's disease (COS-DIP): study protocol. *BMJ Open*, *14*(2), e076350. https://doi.org/10.1136/bmjopen-2023-076350

was registered prospectively with the COMET Database on 24 September 2021 (www.comet-initiative.org, registration number: 1942). The study protocol is written in accordance with the COS-STAP statement (Kirkham et al., 2019). Ethical approval for all stages of the project was obtained from the local ethics committee (Appendix 3.1).

3.2.1 Study oversight

An SSC was established that will lead and conduct the development of the COS-DIP. The SSC is a multidisciplinary, international group and comprises experts from the field of SLT, COS methodology, neurology, clinical trials in OD in PD and a patient partner with PD. The SSC will be responsible for planning, advising and implementing each stage of the process: the study goals and timeline, study protocol, access to participants for the study, designing focus groups interviews and surveys, analysing results, reporting findings, writing and publishing manuscripts and finally, disseminating the COS-DIP.

3.2.2 Patient and public involvement

As part of the SSC, a patient partner with PD is included. The individual was diagnosed with early-onset PD and at the time of beginning the project he had been living with PD for more than 12 years and is in his 50s. He experiences voice and speech difficulties and is receiving voice and speech treatment, whereas he does not experience swallowing difficulties. His wife is an SLT.

The patient partner with PD was identified through MW's network, given his previous involvement in the department's research and his active engagement in the PD community as a patient advocate. He is also the vice-president of an international PD organisation. Unfortunately, due to a lack of additional financial resources, it will not be possible to reimburse him for his work as a patient partner in this study.

He will be primarily involved in the studies presented in chapters 5 and 7. Chapter 5 describes an online survey with people with OD in PD and family members/caregivers. In this study, the individual with PD will be involved in developing and reviewing the survey questionnaire and the interview guide for the focus groups to improve applicability and credibility. Chapter 7 describes the Delphi survey and consensus meetings with all key stakeholders. Here, the patient partner will be involved in the design and review of the Delphi surveys, the consensus meetings, recruitment of participants, and dissemination of results, thereby enhancing the accessibility, understandability, and validity of this study. He will be a co-author of manuscripts related to these studies.

3.2.3 Stakeholder involvement

Throughout this study three key stakeholder groups will be included. The definition of each group and planned ways of recruitment is outlined in Table 3.1. Every attempt will be made to recruit a diverse range of participants to ensure that outcomes generated represent a wide range of perspectives.

Table 3.1: Stakeholder group, definition, and planned recruitment for the development of the Core Outcome Set for Dysphagia Interventions in Parkinson's disease

Stakeholder	Definition	Planned recruitment
People living with OD in PD, family members and caregivers	People diagnosed with OD in PD, people with experience of providing care for a family member and/or living with a family member with OD in PD	Through national and international public research partners, relevant organisations, societies, and patient support groups.
Healthcare professionals	People with ≥ 3 years of experience of providing care in a health setting for people with OD in PD, e.g. SLTs	Through national and international multidisciplinary professional dysphagia associations/networks, trial registries and central PD websites. Authors of included studies in the scoping review will be invited directly.
Researchers	People with ≥ 3 years of experience of undertaking research in the field of OD in PD	

OD: oropharyngeal dysphagia, PD: Parkinson's disease, SLT: speech and language therapist

The development of the COS-DIP comprises five stages. These are displayed in Figure 3.1 and explained in detail in the following sections.

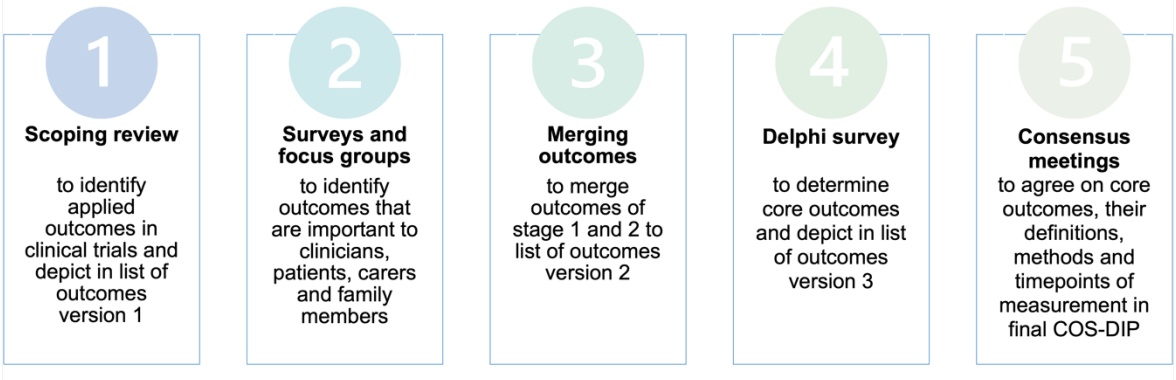


Figure 3.1: Stages during the development of the Core Outcome Set for Dysphagia Interventions in Parkinson's disease

3.2.4 Stage 1 – scoping review

In a scoping review, we will identify all swallowing outcomes, their definitions, OMIs and timepoints of measurement applied in clinical trials in OD in PD. The outcomes will be mapped to the taxonomy by Dodd et al. (2018) and merged into a long list of outcomes version 1. A separate study protocol for the scoping review will be devised and published before conducting the scoping review.

3.2.5 Stage 2 – online surveys and focus groups

This stage will be a sequential explanatory mixed methods design comprising an online survey with dysphagia clinicians and an online survey with people with OD in PD, family members and caregivers. To understand the perspectives of people with OD in PD, family members and caregivers in more detail, focus groups will be conducted after the online survey.

Both the perspectives of people with OD in PD, their family members and caregivers as well as of dysphagia clinicians are important. Researchers need to be able to trust that all stakeholders' views have been heard and included in the Delphi and later in the final COS-DIP (Keeley et al., 2016; Williamson et al., 2017). However, these views are typically not represented or insufficiently considered in OD intervention studies in PD. As they might be different to those of researchers, it is beneficial to expand the long list of outcomes version 1 with outcomes that are particularly important to clinicians, people with OD in PD, family members and caregivers. Furthermore, this provides the opportunity to amend wording of outcomes for the Delphi survey by using patient and carer language and hence, make the Delphi survey more accessible to all stakeholder groups (Keeley et al., 2016).

Therefore, an international online survey will be designed based on the list of outcomes version 1 to understand the perspectives of people living with OD in PD, family members and caregivers on relevant outcomes that should be measured in clinical trials in OD in PD. The survey will be conducted online via Qualtrics (www.qualtrics.com). If considered appropriate, outcomes may be reworded with lay terms to increase the comprehensibility participants (Williamson et al., 2017). Decisions about rewording will be discussed with the SSC's patient partner, who will advise on whether the alternative phrasing would be more accessible and meaningful to lay people. Although this process is not based on a rigid framework, rewording will be guided by the principle of enhancing clarity and inclusiveness (e.g. applying more commonly used terms such as "voice box" instead of "larynx"). Agreement will be reached within the SSC before changes will be finalised. The survey will be piloted by lay people and SLT students and is estimated to take less than 10 minutes to complete. Data will be analysed in R descriptively (rating of importance of outcomes in %) and inferentially using Pearson's χ^2 test to determine if the rating of importance (important/not important) of each outcome is significantly different between the two groups (people with OD in PD and family members/caregivers) (Ravid, 2020).

After the online survey with people with OD in PD, family members and caregivers, focus groups with participants from the previous survey will be conducted. The aim is to validate the findings of the previous survey and to clarify uncertainties that might have emerged from the survey and ultimately gain a deeper understanding of the participants' perspectives (Pope & Mays, 2020). Approximately 30 participants will be involved in four focus groups with 7–8 participants each (Pope & Mays, 2020). At the end of the online survey, participants will be asked if they would like to take part in subsequent focus groups. Those who signal interest, will be contacted via email. Each focus group will last approximately 60 min to keep the time demand on participants as low as possible. A semi-structured interview guide will be designed in accordance with recommendations by Keeley et al. (2016) and based on the survey results and in discussion with the SSC. All focus groups will be conducted virtually using Zoom platform to increase accessibility for participants. The interviews will be conducted by JH and MW and will be audio recorded, transcribed verbatim, pseudonymised and entered into NVivo V.12 Plus (QSR International, USA). Data will be analysed following Reflexive Thematic Analysis by JH (Braun & Clarke, 2006).

To assess dysphagia clinicians' views, an online survey based on the previous online survey with people with OD in PD and family members/caregivers will be conducted. The procedure, questions and items will be identical. Instead of lay terms the technical terms identified in stage 1 will be used. The survey will be translated from English into different languages (Chinese, Italian, Spanish and German). This will allow us to send the survey to a wider community and to obtain a comprehensive understanding of the clinicians' perspectives globally while also following the COS-STAD recommendations (Kirkham et al., 2017). These languages were chosen because of the well-developed services for people with dysphagia in the countries where these languages are spoken. The resources available to the research team were also a factor in choosing these languages. Translators will be encouraged to cross-check their translations with another colleague who is proficient in both English and their relevant language. The survey will be piloted on two dysphagia clinicians and is estimated to take less than 15 min to complete. Data will be analysed descriptively in R using the same tests as in the previous survey.

3.2.6 Stage 3 – merging outcomes

The list of outcomes version 1 will be supplemented with outcomes that emerged in stage 2 through the surveys and focus groups. Both technical terms and lay terms from stages 1 and 2 will be used to ensure comprehensibility for all stakeholder groups in the next stage.

Following discussion and agreement within the SSC, outcomes will be combined or dropped if they are considered to be overlapping in content or repetitious, subdomains may be added for better categorisation of outcomes and will be merged into an adapted list of outcomes version 2.

3.2.7 Stage 4 – Delphi survey

A three round online Delphi (eDelphi) survey will be conducted to obtain participants' views on the importance of each outcome of the list of outcomes version 2. The Delphi technique will be applied as this allows us to collect responses anonymously and objectively, to prevent the impact of dominant participants and to include a large number of international stakeholders from a wide geographical range (Williamson et al., 2017).

Participants in this stage will be from all stakeholder groups listed in Table 3.1. Using as large a panel as is practical, it is anticipated that at least 150–200 participants will be recruited for valid consensus with a comprehensive range of perspectives achieved.

All three round eDelphi surveys will be conducted through the online DelphiManager platform (www.comet-initiative.org/delphimanager) using the list of outcomes version 2. The survey will be piloted to ensure face validity (Williamson et al., 2017). This will also inform the time needed to complete each eDelphi round.

In each round participants will be asked to rate every outcome according to their perceived importance on a 9-point scale as recommended by the GRADE Working Group (Guyatt et al., 2011). The scales 1 to 3 represent limited importance, 4 to 6 importance but not critical and 7 to 9 critical importance. A button 'unable to score' and a text field for additional comments will be available (Sinha et al., 2011; Williamson et al., 2017). Each outcome will be displayed using the technical term with the lay terms available within the 'help text' as preset by DelphiManager. This 'help text' can be accessed through clicking or hovering over the outcome with the mouse or finger. Each survey round will be open for 3 weeks with an email informing the participants about the survey opening. Reminder emails will be sent to all participants who have not completed the survey after two weeks.

Participants who agree to take part in the eDelphi will be assigned a code and therefore data will be pseudonymised. At the beginning of Round 1 participants will be asked to provide some general personal data (gender, age, nationality, stakeholder group, for people with the health condition: number of years

since PD diagnosis). Participants will then be informed about the survey procedure. Participants will then be asked to rate the importance of each outcome. At the end of Round 1, participants will be asked whether any important outcomes were missing and if so, to specify them in a provided comment box. This ensures that no key outcome important to each stakeholder group is omitted (Sinha et al., 2011; Williamson et al., 2017).

In Round 2 the remaining outcomes and new outcomes identified from Round 1 will be included. Participants will be shown their own score from Round 1 with feedback on the percentage of scores of each outcome for each stakeholder group. They will then be asked to re-rate the outcome, taking the feedback from Round 1 into consideration (Williamson et al., 2017).

Round 3 will include remaining outcomes and new outcomes identified from Round 2. Participants will again be asked to re-rate the outcomes based on the feedback from Round 2 (Williamson et al., 2017). At the end of Round 3, participants will be asked if they are willing to participate in a consensus meeting in the next step. Figure 3.2 provides an overview of the process of the three round eDelphi survey.

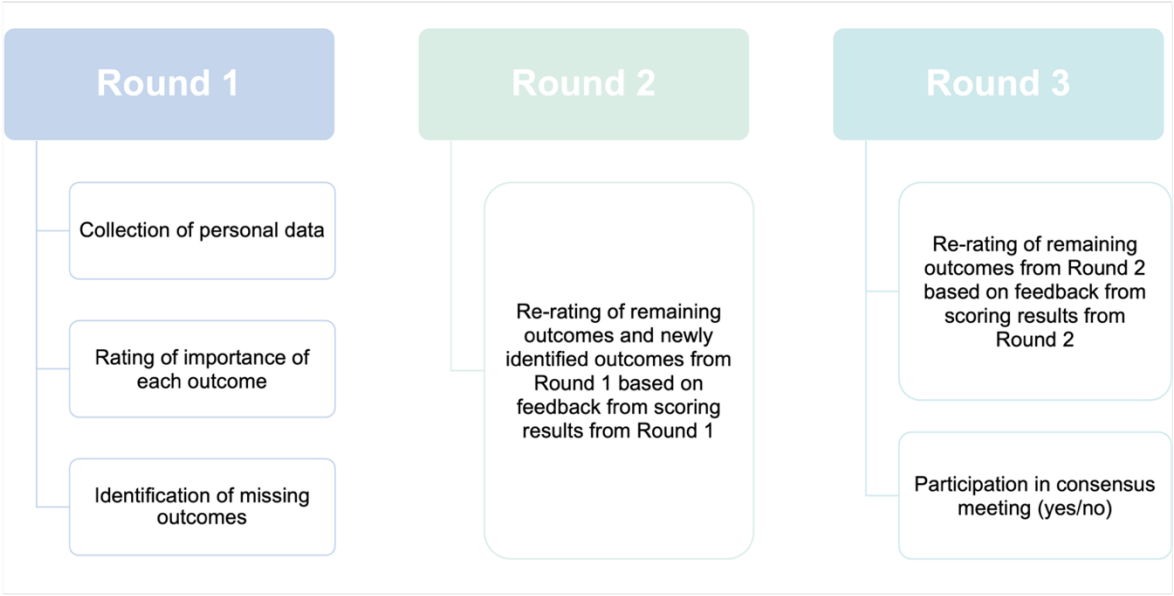


Figure 3.2: Steps for each round of the online Delphi survey

3.2.8 Adding/retaining/dropping outcomes

New outcomes will be added to the list of outcomes if they are suggested by at least two participants. Outcomes will be retained after each round if they meet the according *consensus in* or *no consensus* criteria as defined in Table 3.2. All outcomes that will reach *consensus out* will be dropped. Agreed

outcomes and outcomes without consensus will form the list of outcomes for the following round (Blazeby et al., 2015; Williamson et al., 2017).

This approach will ensure that the burden on participants is kept to a minimum due to shortening the list of outcomes from one round to another. At the same time, starting with less rigorous criteria in Round 1 will prevent outcomes dropping too early and will give participants the opportunity to re-rate an outcome based on the feedback provided from the stakeholder groups (Williamson et al., 2017).

Table 3.2: Definition of consensus in accordance with Blazeby et al. (2015) and Harman et al. (2018)

Consensus classification	**Description**	**Definition**	
		Round 1	**Round 2 + 3**
Consensus in	Outcome should be included in COS	≥ 50% scored 7-9	≥ 70% scored 7-9
Consensus out	Outcome should not be included in COS	< 50% scored 7-9	
No consensus	Uncertainty about outcome importance		≥ 50% and < 70% scored 7-9

3.2.9 Missing data

To facilitate retention of participants in general only participants who replied to a pre-Delphi invitation will take part in the Round 1, contact details of JH will be provided, participants are offered to be acknowledged in the published manuscript and the eDelphi will be concise, user friendly and not overly time-consuming to complete (Sinha et al., 2011; Williamson et al., 2017).

A common challenge in Delphi surveys is that participants with minority opinions might drop out of the study, which can lead to overestimation of the level of consensus. In case attrition is higher than 20% personalised reminder emails about survey completion and current response rates will be sent. Only participants who completed the survey will be invited to take part in the following rounds to minimise attrition bias (Sinha et al., 2011; Williamson et al., 2017).

From this stage, the list of outcomes version 3 will result and will be extended with definitions that achieved agreement and suggested definitions for those that did not achieve consensus.

3.2.10 Stage 5 – consensus meetings

Two consensus meetings will be held virtually to facilitate attendance of participants from different time zones (Gorst et al., 2021). The aims are to discuss the results of the eDelphi survey, outcomes that have

reached *no consensus* and to ultimately verify the core outcomes to devise a final list of outcomes including definitions of outcomes, OMIs and timepoints of measurement of outcomes for the COS-DIP.

Attendees at this meeting will comprise a 20% selection of all eDelphi participants. Only those who indicated to be willing to participate at the end of stage 4, will be randomly and equally selected from each stakeholder group. Prior to the meeting participants will be sent a reminder email with their individual eDelphi scoring from Round 3. The meeting will be led by a moderator with experience in conducting consensus meetings.

3.2.11 Agreeing on outcomes included in the COS-DIP

The moderator will inform the participants about the results of the eDelphi and will elicit feedback on the preliminary COS-DIP and on outcomes with *no consensus*. Final voting on including or excluding outcomes (yes/no) will be carried out by using an electronic voting system. Outcomes that are voted ≥ 70% 'yes' will be included in the final COS-DIP.

3.2.12 Agreeing on definitions, OMIs and timepoints of measurement of outcomes

The list of outcomes version 3 with definitions used in the eDelphi will be extended by the OMIs and timepoints of measurement from the scoping review in stage 1 before the consensus meeting. The moderator will inform the participants and elicit feedback about the definitions of outcomes, OMIs and timepoints of measurement. Final voting on agreement (yes/no) will be carried out by using the same voting system and criterion (≥ 70%) as for the agreement on outcomes. In the end, an agreed set of core outcomes, definitions, OMIs and timepoints of measurement for the COS-DIP will be generated.

3.3 Dissemination

Promoting the COS-DIP and recommended measurements will be critical to adoption in clinical OD trials in PD. The awareness of the COS-DIP project will be enhanced through submissions for scientific presentations at national and international multidisciplinary conferences and through peer-reviewed, open access publications of related manuscripts. Lay and professional information sheets and infographics will be circulated through relevant patient and professional organisations and networks.

Furthermore, a social media X (formerly Twitter) account (Research in Dysphagia and Parkinsonian Syndromes, @DysphagiaPhDs) was established through which the COS-DIP will be further disseminated. Additionally, a page on the Swallowing and Voice Centre website of Trinity College Dublin (TCD) was created to inform about the COS-DIP project in general and provide specific information for each stage of the project (www.tcd.ie/slscs/research/research-projects/cos-dip-research). Animated explainer videos were created using the online platform Vyond (www.vyond.com) to provide easy to understand and accessible information on the project. This will especially be helpful for lay people, such as people living with PD and OD and their family members and caregivers. These videos are linked into the above-mentioned TCD website and will also be linked into emails to gatekeepers and potential participants and shared on X to raise awareness of and to inform about the COS-DIP research project.

3.4 Summary

So far, there is no published COS for OD interventions in PD. The development of the COS-DIP will improve the interpretation and comparison of future studies and therefore enhance comparative effectiveness research to direct treatment for people with OD in PD. In the next chapter, the first step of devising the long list of outcomes through a scoping review, is presented.

Chapter 4: Swallowing outcomes in dysphagia interventions in Parkinson's disease: a scoping review[6]

4.1 Introduction

The systematic reviews by Gandhi and Steele (2021) and López-Liria et al. (2020) concluded that there is an ongoing significant lack of fundamental scientific evidence on the treatment of OD in PD. In an attempt to address this issue, Schindler and colleagues (2021) established a consensus on the treatment of OD in PD. One conclusion made by the authors is that in neurodegenerative conditions such as PD, longer-term treatment effects and adverse events must also be assessed. These three published reports demonstrate not only a deficiency of evidence on OD interventions in PD but argue that longer-term treatment effects and adverse events should be assessed consistently. Despite being intricately linked, these studies focus on the efficacy and efficiency of OD interventions themselves rather than on the outcomes that are targeted by the interventions.

A solution to these challenges is the development and use of an agreed standardised COS-DIP devised by key stakeholders including people with the health condition, healthcare professionals and clinical trialists. In order to establish the COS-DIP, the first step is to identify currently measured outcomes in published clinical trials in OD in PD.

This chapter describes the scoping review on swallowing outcomes in OD intervention studies in PD. The objective was to report on all applied outcomes, their definitions, OMIs and timepoints and frequency of measurement in (quasi-) RCTs, controlled clinical trials (CCTs) and pilot/feasibility studies with control groups in OD in PD.

4.2 Methods

This scoping review is reported in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-analysis extension for Scoping Reviews (PRISMA-ScR) checklist (Tricco et al., 2018)

[6] Adapted from: Hirschwald, J., Hofacker, J., Duncan, S., & Walshe, M. (2023). Swallowing outcomes in dysphagia interventions in Parkinson's disease: a scoping review. *BMJ Evidence-Based Medicine*, *28*(2), 93–100. https://doi.org/10.1136/bmjebm-2022-112082

(Appendix 4.1). A review protocol was devised beforehand and published online (http://hdl.handle.net/2262/97652) (Appendix 4.2).

A scoping review was chosen as the most appropriate method to systematically map the research done in this area, as well as to identify any existing gaps in knowledge (Arksey & O'Malley, 2005). This allowed to include all types of clinical trials with control groups, rather than just RCTs as is commonly done in systematic reviews, and consequently a larger number of outcomes was expected to be identified. In addition, duplicate efforts were prevented, i.e. in critiquing the quality of the included studies and evaluating the best available evidence, as these had already been conducted in previous systematic reviews (see section 4.1).

4.2.1 Searches

A comprehensive search strategy, including two search strings (1) dysphagia and (2) Parkinson's Disease, was devised with the assistance of a subject librarian. The databases AMED, CINAHL, EMBASE, MEDLINE, Web of Science, ProQuest Dissertations & Theses and trial registry Clinicaltrials.gov were searched from inception to 3 December 2021 and updated on 10 March 2022 (Appendix 4.3). The reference lists of all included studies were screened for additional studies and study authors were contacted for additional information, if required.

4.2.2 Study inclusion and exclusion criteria

Studies were included if (1) participants had a diagnosis of OD and PD, (2) clinical interventions were aimed at improving swallowing or feeding difficulties, (3) at least one swallowing (related) outcome was measured and (4) the study design included at least one intervention and one control group (RCT, quasi-RCT, CCT or feasibility/pilot study with control group). Only studies with control groups were included as ultimately the COS-DIP will inform clinical trial design with a focus on RCTs so that meta-analysis of clinical trials in OD and PD will be feasible in the future. In accordance with the patient, concept and context framework, the following was applied:

- Patient: OD in idiopathic PD ≥ 18 years.
- Concept: any clinical intervention in OD in PD.

- Context: any clinical context (all countries and healthcare settings, e.g. acute care, primary healthcare and community setting).

Studies were excluded if they did not fit into the conceptual framework of the study or if a heterogeneous participant population including people with PD was studied but data could not be extracted for the PD subgroup solely. Finally, studies were excluded if no full text was available, e.g. a conference abstract only, and authors were unable to provide sufficient information. No date or language restrictions were applied.

4.2.3 Study selection

Following the search, all identified citations were collated and uploaded to an online platform (www.covidence.org), where duplicates were removed automatically. To improve reliability, JHi and JHo discussed the eligibility criteria for studies to be included and then carried out a pilot test of a random sample of 25 titles and abstracts independently. The cut-off agreement typically varies between 70% and 80% in the literature (Tricco et al., 2018), and therefore the cut-off for this scoping review was preset at 75%. An agreement of 96% was achieved between both reviewers. Following, all abstracts and titles and thereafter full texts, were screened independently by JHi and JHo against the inclusion criteria. Any disagreement that arose between the reviewers at each stage of the selection process was either resolved through discussion or with an additional reviewer (MW).

4.2.4 Data extraction strategy

Data was extracted independently from papers included in the scoping review by JHi and JHo using a data extraction tool developed previously and applied by Hofacker (2021). The extracted data includes details about the first author's name, year of publication, country of origin, study design, population, sample size and applied outcomes. The data extraction form was trialled independently on the first three included sources by JHi and JHo. As a result of discussion together with MW, further parameters were added to the data extraction form: number of participants and dropouts, intention-to-treat analysis, age, gender, PD severity, OD severity, intervention, comparator, OMIs, timepoints of measurement and frequency of measurement (Appendix 4.4). Any disagreements that arose were resolved through discussion between JHi and JHo, or with MW in addition.

4.2.5 Data analysis and presentation

As no taxonomy for categorisation of outcomes in OD interventions specifically exists, a widely used taxonomy by Dodd et al. (2018) was applied. This taxonomy was designed for trial outcomes and is applicable to all fields within medical research (Katiri et al., 2021; Williamson et al., 2017). It includes five core areas (death, physiological/clinical, life impact, resource use and adverse events) and 38 outcome domains (Dodd et al., 2018).

The outcomes used in the included studies were extracted as verbatim following COS methodology as described in the COMET Handbook (Williamson et al., 2017). All extracted data were categorised in an Excel spreadsheet also previously developed and applied by Hofacker (2021) and further adapted for the purpose of this study. It comprises information regarding core area of the outcome and outcome domain in accordance with the taxonomy by Dodd et al. (2018), outcome description as reported verbatim by study authors, definition of outcome, OMIs and timepoints and frequency of measurement. Furthermore, the amount and percentages of used outcomes within the respective core area and outcome domain were calculated.

4.3 Results

4.3.1 Protocol deviations

In addition to (quasi-) RCTs and CCTs also, feasibility/pilot studies with control groups were included in this scoping review. This allows for inclusion of all clinical trial designs. No other deviations were made from the study protocol.

4.3.2 Search results

The literature search identified 2,587 studies. After removal of duplicates, further 2,328 records were excluded during title/ abstract screening. Of the remaining 54 reports, nine could not be retrieved as full texts and 27 did not meet the inclusion criteria in the full-text review (Appendix 4.5). Additionally, two records were identified through citation searching, of which one was included. In total, 19 studies were included in this scoping review. Of these, 18 were in English and one in Chinese. The Chinese study was translated with the help of a translator and Chinese speaking SLT. The results of the search and the study

inclusion process are outlined in the PRISMA-ScR flow diagram in Figure 4.1 (Page et al., 2021; Tricco et al., 2018).

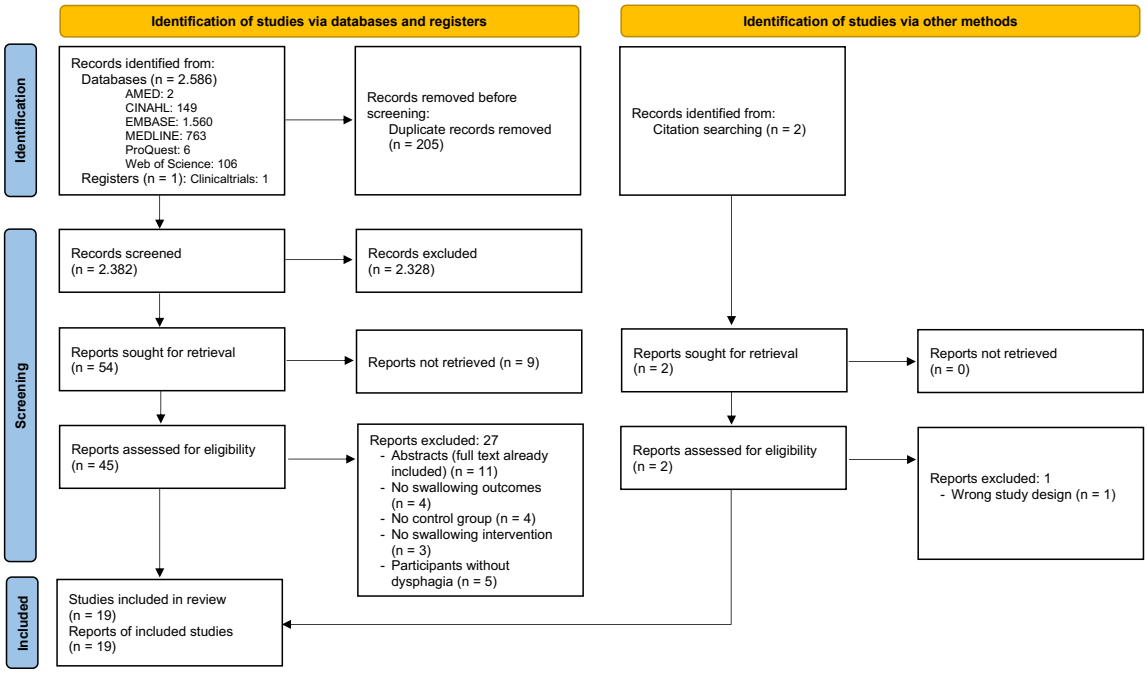


Figure 4.1: Preferred Reporting Items for Systematic Reviews and Meta-analysis extension for Scoping Reviews flow diagram.

The included studies comprised ten RCTs, two quasi-RCTs, four CCTs and three feasibility/pilot studies with control groups. Overall, 2,124 participants were included with studies being published between the years 2000 and 2021, although all years were included in the search. Assessed interventions in the included studies were surface electrical stimulation (n=4), postural swallowing techniques (n=3), Expiratory Muscle Strength Training (EMST) (n=3), application of biofeedback (n=2), repetitive transcranial magnetic stimulation (rTMS) (n=2), standardised swallowing training (n=2), deep brain stimulation (n=1), vocal training (n=1) and aural stimulation with capsaicin (n=1) (see Appendix 4.4 for the detailed characteristics of the included studies).

Within the 19 studies, 180 outcomes were identified. Of these, 46 outcomes were excluded for different reasons: (1) the outcomes were only assessed for determining whether the participants met the inclusion criteria for study participation but not the effect of the intervention, (2) neither the specific OMI nor results were reported or (3) due to missing information it was unclear what the outcome referred to (Appendix 4.6).

4.3.3 Outcome areas

The majority of the 134 included outcomes belong to the outcome areas physiological/clinical (n=112; 83.58%) or life impact/functioning (n=19; 14.18%). One outcome was categorised to each of the outcome areas death, resource use and adverse events (n=1; 0.75% respectively).

4.3.4 Outcome domains

As the taxonomy by Dodd et al. (2018) was developed for clinical trials in medical research in general but not specifically for OD interventions, the outcome domains were adapted through an in-depth discussion between JHi, SD and MW for the purpose of this scoping review. Figure 4.2 depicts the outcome domains from the included studies and the number of outcomes mapped accordingly.

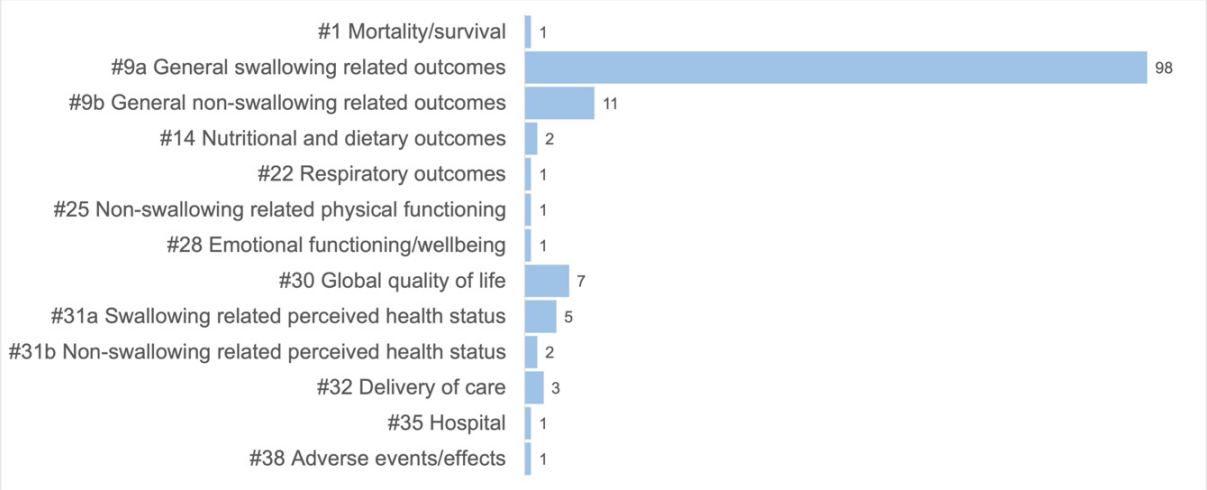


Figure 4.2: Outcome domains used in clinical trials in oropharyngeal dysphagia in Parkinson's disease

4.3.5 Outcome subdomains

The outcome domain #9a comprises almost three quarters of all outcomes and the outcome domain #9b includes with over 8% the second most outcomes. In order to categorise these general outcome domains more precisely, re-categorising according to subdomains was necessary. Outcome domain #9a was divided into the following five subdomains: (1) saliva management, (2) swallowing related physiology, (3) swallow efficiency, (4) swallow safety and (5) neurological status. The outcome oropharyngeal dysphagia severity remained as was. Outcome domain #9b was split into two subdomains: (1) neurological findings and (2) voice.

4.3.6 Research question 1: what are the outcomes of interest in clinical trials in OD in PD?

The 134 included outcomes were merged to 39 outcomes due to overlaps or being identical. Table 4.1 presents the long list of outcomes including the outcomes, subdomains, domains and outcome areas accordingly.

Table 4.1: Long list of outcomes included in clinical trials in oropharyngeal dysphagia in Parkinson's disease

Outcome area	Outcome domain	Outcome sub-domain	Outcome
Death	1. Mortality/ survival		Death
Physiological/ clinical	9a. General swallowing related outcomes		Oropharyngeal dysphagia severity
		Saliva management	Drooling
			Salivary pooling
		Swallowing related physiology	Orolingual bolus control
			Oral bolus transport
			Swallowing related lingual movement
			Timing of oropharyngeal swallow components
			Laryngeal elevation
			Laryngeal sensation
			Swallowing related hyoid bone movement
			Initiation of pharyngeal swallow
		Swallow efficiency	Post swallow oral residue
			Post swallow pharyngeal residue
			Post swallow pharyngeal pooling
			Piecemeal deglutition
		Swallow safety	Penetration/aspiration
		Neurological status	Cortical reorganization
			Motor evoked potential
	9b. General non-swallowing related outcomes	Neurological findings	Overall motor symptoms
			Tremor
			Rigidity
			Bradykinesia

			Axial symptoms
			Freezing of gait
		Voice	Phonation
			Loudness
	14. Nutritional and dietary outcomes		Level of oral intake
	22. Respiratory outcomes		Aspiration pneumonia
Life impact/ functioning	25. Non-swallowing related physical functioning		Activities of daily living
	28. Emotional functioning/wellbeing		Pleasure of oral intake
	30. Global quality of life		Swallowing related quality of life
	31a. Swallowing related perceived health status		Self-perception of swallowing
	31b. Non-swallowing related perceived health status		Self-perception of walking
			Self-perception of activities of daily living
	32. Delivery of care		Participant's satisfaction with intervention
			Participant's adherence to intervention
Resource use	35. Hospital		Hospitalisation
Adverse events	38. Adverse events/effects		Adverse events

The outcome of most interest was 'penetration/aspiration' measuring the depth of the entry of food and fluid into the larynx and airway. This was measured in 10 of the 19 studies. 'Oropharyngeal dysphagia severity' was the second most often measured outcome (n=9). Figure 4.3 depicts the eight most commonly reported outcomes, while outcomes measured only once or twice across studies were grouped together into combined categories to allow clearer depiction.

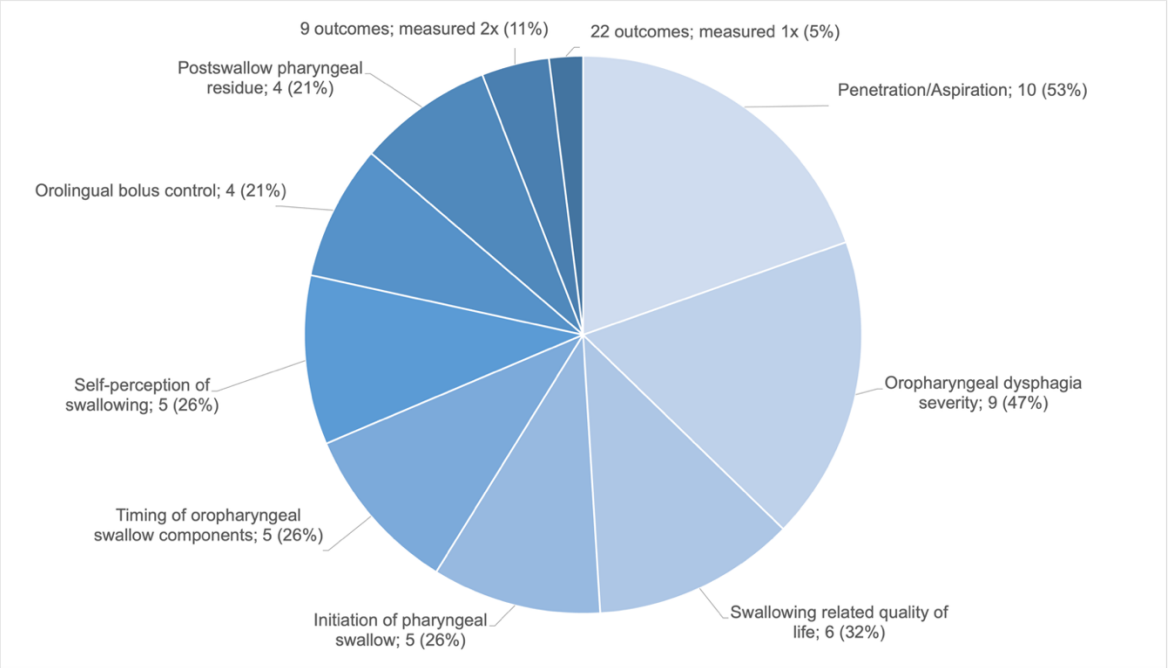


Figure 4.3: Number and percentage of studies measuring each outcome in clinical trials in oropharyngeal dysphagia in Parkinson's disease

Of these top eight outcomes, six belong to the outcome domain #9a general swallowing related outcomes, whereas the other two belong to the outcome domain #30 global quality of life and #31a swallowing related perceived health status.

4.3.7 Research question 2: how are the outcomes in these clinical trials defined?

Most definitions of the outcomes in the included studies vary widely. Thereof, 17 outcomes were not defined at all in any study, and 13 outcomes were defined by some studies but not by others. The outcomes 'swallowing related hyoid bone movement' and 'timing of oropharyngeal swallowing components' were the most diverse defined outcomes. Each study used different parameters within these outcomes with either very specific or no definition. Outcomes that were assessed by using a scale or questionnaire were oftentimes not defined at all. Instead, the numerical scores from the scale or questionnaire were provided without explanation of what they related to. The outcome 'penetration/aspiration' was defined in accordance with the definition by Rosenbek et al. in 9 of 10 studies: 'Penetration is defined […] as passage of material into the larynx that does not pass below the vocal folds. Aspiration is defined as passage of material below the level of the vocal folds.' (1996, p. 93). There was no matching definition for any other outcome by at least two studies with different authors. The definitions of outcomes reported by Baijens

et al. (2012, 2013) were unsurprisingly in agreement given that both studies were conducted by the same main author group (see Appendix 4.7 for all definitions of outcomes).

4.3.8 Research question 3: how are the outcomes in these clinical trials measured?

Overall, the applied OMIs show high variability. Most of the outcomes in the outcome domain #9a were measured using instrumental assessments and either validated scales or scales designed for the purpose of the according study during Videofluoroscopy of Swallowing (VFS) or Fibreoptic Endoscopic Evaluation of Swallowing (FEES) or measuring it in milliseconds during electromyography.

The outcomes 'penetration/aspiration' and 'swallowing related quality of life' were measured the most consistently. Eight of the ten studies used the Penetration-Aspiration-Scale (PAS) by Rosenbek et al. (1996) during VFS and/or FEES. One study (Logemann et al., 2008) did not report the scale that was used to measure the outcome during VFS and another study (Manor et al., 2013) used a self-designed 4-point scale. The outcome 'swallowing related quality of life' was measured through the Swallowing Quality of Life (SWAL-QOL) Questionnaire (McHorney et al., 2002) in six studies, whereas one study (Heijnen et al., 2012) additionally assessed this outcome through the MD Anderson Dysphagia Inventory (Chen et al., 2001). The outcome 'self-perception of swallowing' was measured by the Swallowing Disturbance Questionnaire (Manor et al., 2007) three times and the Dysphagia Severity Scale (DSS) (Tohara et al., 2003) and Arabic Dysphagia Handicap Index (Farahat et al., 2014) once each.

The outcome 'oropharyngeal dysphagia severity' was the outcome measured most differently across all included studies. In order to assess the outcome either validated scales or self-designed scales for VFS and/or FEES, the Standardized Swallow Assessment (L. Perry, 2001) or a Clinical Swallow Evaluation were conducted. The OMIs of 'death' and 'hospitalisation' were not described and while it may seem self-explanatory, their methods of recording were not clear (see Appendix 4.8 for the applied OMIs).

4.3.9 Research question 4: at which timepoints and at which frequency are the outcomes in these clinical trials measured?

In the outcome domains #1, #22, #35 and #38 only one study (Robbins et al., 2008) assessed the outcomes 'death', 'aspiration pneumonia', 'hospitalisation' and 'adverse events', respectively. These were measured continuously over a period of three months without further details provided by the authors. All remaining outcomes in the outcome area physiological/clinical were measured in the included studies one

to five times either during or pre to post and/or with one to two follow-ups. The timepoints post intervention ranged from five minutes to six months.

The outcomes in the outcome area life impact/functioning were measured one to 36 times during, pre and/or post with follow-up assessments two weeks to six months post intervention. The outcome 'participant's adherence to intervention' was assessed weekly over a period of three months, hence 36 times in one study (Robbins et al., 2008). The outcome 'self-perception of swallowing' was measured differently at varying timepoints and frequency across the included studies. For example, one study (Heijnen et al., 2012) assessed the outcome through the DSS (Tohara et al., 2003) after each session, hence 13 to 15 times, whereas the other four studies assessed it two, three or five times. Only the outcome 'participant's satisfaction with the intervention' was measured once during or two weeks post intervention.

Overall, most outcomes were measured at least pre and post intervention with frequencies from one to four times. If the outcome was measured only once this was usually during the intervention, for example, rTMS (see Appendix 4.9 for all timepoints and frequency of measurements).

4.4 Discussion

In this scoping review, 19 clinical trials that investigated OD interventions in PD with 134 outcomes were included. Outcomes were merged to 39 final outcomes in 13 outcome domains. Outcomes of interest, definitions, OMIs, timepoints and frequency of measurement varied highly across the included studies. This scoping review identified relevant challenges within the included studies.

One major challenge in the included studies is the lack of information on outcomes, their definitions, and OMIs and omitted outcomes. For example, three studies did not report on outcomes in detail if there were no differences between the intervention and the control group (Ayres et al., 2017; Baijens et al., 2012, 2013). In two other studies, the authors did not report why outcomes were omitted (Heijnen et al., 2012; Silva-Arone et al., 2021). Incomplete reporting of research methods (e.g. what was measured and how was it measured) and selective reporting of findings (e.g. omitted outcomes) decrease the transparency of the research studies and raise questions about the applicability of the findings and study reporting practices. This heightens the risk that results lack credibility and studies are not easy to replicate and reproduce (El Amin et al., 2022; John et al., 2012; Samsa & Samsa, 2019). This is especially problematic

in healthcare research involving OD interventions in PD where the outcomes of these clinical trials are essential for decision-making, such as the safety and effectiveness of the intervention (Williamson et al., 2017).

Another identified challenge in the included studies is that some of the outcomes that were previously identified in the literature as relevant for people with OD and PD were not or rarely assessed. Only one study (Feng et al., 2019) included the outcome voice as they assessed the effect of vocalisation training in improving drooling in people with PD. However, three pilot studies have previously shown that cross-system effects through an intensive, evidence based voice and speech treatment (Lee Silverman Voice Treatment, LSVT LOUD) in people with PD can improve swallowing (El Sharkawi et al., 2002; Miles et al., 2017; Park et al., 2022). These studies suggest that training voice and speech may improve swallow function as well. Concurrently, dysphagia therapy might affect voice and speech in PD. Therefore, voice as an outcome in clinical trials in OD in PD might be relevant to assess.

Just six of the 19 included studies assessed 'swallowing related quality of life', and five studies included 'self-perception of swallowing' as patient-reported outcomes. This is a relatively small number, as patient-reported outcomes are considered critical for evidence-based practice and for assessing treatment effectiveness (Watts & Zhang, 2022). It should be noted, that specifically in people with PD the self-assessment may differ from objective or investigator-reported outcomes due to sensorimotor deficits (Kalf et al., 2012; Pflug et al., 2018) and hence, both types of assessments should be included.

Outcomes that were not typically measured but may be considered relevant included parameters associated with cough, hydration and nutrition. People with OD in PD are at high risk of developing malnutrition and dehydration. This can further impair swallow function and delay the rehabilitation process. It can also increase the risk of medical complications or even mortality (Wirth et al., 2016). Furthermore, impaired cough (dystussia) reduces airway protection as material entering the airway might not be expelled effectively. Dystussia often coexists with OD in people with PD and therefore the risk of aspiration and pneumonia is increased, but also quality of life can be decreased (Troche et al., 2014, 2016). This is not surprising as both coughing and swallowing are sensorimotor behaviours that overlap in anatomy and neuroanatomical substrates (Troche et al., 2014). Therefore, outcomes associated with hydration, nutrition and cough might be relevant to assess in future OD interventions in PD.

A further interesting finding in this review is that only one study (Robbins et al., 2008) assessed outcomes pertaining to the outcome areas of death, resource use and adverse events. Assessing and reporting adverse events in RCTs is crucial for determining the safety of an intervention but is less focused on than assessing and reporting efficacy and effectiveness in these trials (Ioannidis, 2004). The outcome area adverse events might comprise numerous outcomes, which typically are not predefined as they are usually unknown before commencing a study. Furthermore, only assessing if adverse events are present or absent is regarded as insufficient. Additional information on the severity, timing, duration and number of occurrences of the events is required and thus, making the assessment, reporting and analysis of these outcomes more laborious and possibly inconsistent (Phillips et al., 2019). Death might be incorporated as an outcome of adverse events. In OD interventions in PD, the assessment and reporting of death might be more relevant than currently considered. As stated before, dysphagia and dystussia increase the risk of aspiration and hence pneumonia, which is a leading cause of death in people with PD (Hobson & Meara, 2018; Hoehn & Yahr, 1967). Therefore, addressing this outcome area in future clinical trials in addition with adverse events might be relevant. The reporting of adverse events in clinical trials might further be improved by the adoption of the Consolidated Standards of Reporting Trials harm extension guideline (Ioannidis, 2004).

Following the Dodd et al. (2018) taxonomy, the outcome area resource use comprises the outcome domains economic, hospital, need for further intervention and societal/carer burden. Of these, only hospital in terms of length of hospital stay was assessed in one of the included studies. This outcome area might be underrepresented in OD in PD studies. A recent systematic review showed that the presence of dysphagia increases the hospital length of stay, regardless of admission cause. Furthermore, this also increases the monetary costs by over 40% in patients with dysphagia compared with non-dysphagic patients. In addition, pneumonia is one of the most common reasons for emergency hospital admission in people with PD (Woodford & Walker, 2005), making people with OD in PD more likely to be admitted to hospital and increase overall healthcare costs. Additionally, Perry and colleagues (2022) found that providing care for a person with OD in PD reduces the carer's quality of life due to an increased burden. In future studies on OD in PD it might be important to assess outcomes related to carer burden as ultimately, a less burdened carer might improve health outcomes and quality of life of a person with PD (S. E. Perry et al., 2022).

Lastly, the use of unvalidated OMIs (in general or for the specific patient population) in the included studies comprised a further challenge (Byeon, 2016; Heijnen et al., 2012). Validated OMIs are important to ensure that the tool is measuring what it is supposed to measure, and hence, that the results are valid.

Strengths and limitations of the study

In order to categorise the outcomes, the taxonomy devised by Dodd et al. (2018) was applied as recommended by the COMET Handbook. This is widely implemented in outcome research and facilitates consistent use of clinical outcome terms (Dodd et al., 2018; Williamson et al., 2017). However, the taxonomy was devised largely for medical research, making it less specific to OD interventions. Furthermore, a possible limitation in this review is the restriction of the review to include clinical trials only, but this was based on the fact that the focus of the COS-DIP is a COS for clinical trial design.

4.5 Summary

This scoping review has systematically extracted and categorised all outcomes in clinical trials in OD interventions in PD. We identified high variability in included outcomes in addition to outcomes that were rarely measured or not measured at all. Furthermore, a lack of information on outcomes, such as definitions, OMIs and timepoints of measurement, was identified which can affect a study's replicability, credibility and decrease its validity. Additionally, in some of the included studies, outcomes mentioned as part of the study's research question were later omitted and hence pose a risk of selective outcome reporting bias and skewing individual study results.

The identified outcomes form the first version of the long list of outcomes. This will further be expanded in the next chapter through outcomes important to people living with OD in PD, family members and caregivers.

Chapter 5: 'Without the data you're not going to know if it's important or not': A mixed methods study on what dysphagia intervention outcomes are important to people with Parkinson's disease and family members[7]

5.1 Introduction

In the previous scoping review on clinical trials on OD interventions in PD, presented in chapter 4, none of the included studies had stated that patient involvement was incorporated when designing and implementing the study. Thus, it remains unclear if the outcomes being used in these trials are relevant to all key stakeholders or primarily to researchers. One important factor that may contribute to differing perspectives between these two groups, is that OD is less often recognised subjectively when compared to objective assessments (van Hooren et al., 2021).

The aim of this study was to obtain perspectives on intervention outcomes important to people living with OD in PD, family members and caregivers. The objectives of this study were: (1) to identify OD in PD intervention outcomes that are important to people living with OD in PD, family members and caregivers through an online survey, and (2) to validate the survey findings and clarify inconclusive survey results with previous survey participants in focus group interviews.

5.2 Methods

This study is a two-phase explanatory-sequential mixed methods study with a cross-sectional online survey followed by online focus groups with people with OD in PD, family members and caregivers. The study is reported in accordance with the Checklist for Reporting of Survey Studies (CROSS) (Sharma et al., 2021) (Appendix 5.1) and the Consolidated Criteria for Reporting Qualitative Research (COREQ)

[7] Adapted from: Hirschwald, J., Mooney, L., Wolf, M., Boyle, G., Warnecke, T., & Walshe, M. (2025). 'Without the Data You're Not Going to Know If It's Important or Not': A Mixed Methods Study on What Dysphagia Intervention Outcomes are Important to People with Parkinson's Disease and Family Members. *Dysphagia*, *40*(4), 902–919. https://doi.org/10.1007/s00455-024-10788-x

(Tong et al., 2007). Completing the online survey was taken as consent. Written and verbal informed consent was obtained from participants prior to the focus groups (Appendix 5.2).

5.2.1 Stage 1: survey

Study design

An anonymous, international online survey was designed based on the list of outcomes identified in chapter 4 to understand the perspectives of people living with OD in PD, family members and caregivers on important outcomes that should be measured in OD intervention studies in PD. The outcomes were adapted by JH using lay terms (Appendix 5.3) and subsequently discussed and further refined together with the patient partner from the SSC. Adaptations followed several principles to ensure comprehensibility and consistency for participants. First, to emphasise that outcomes capture changes over time, most adaptations began with the phrase "changes in…" (e.g. 'self-perception of swallowing' was amended to 'changes in overall awareness of swallowing function'). Second, technical terminology was replaced with more familiar everyday language (e.g. 'piecemeal deglutition' was adapted to 'changes in need to break down food or liquids in the mouth in order to swallow'). Third, when outcomes were considered to be unfamiliar or abstract, clarifying examples were added to aid comprehension (e.g. 'level of oral intake' was adapted to 'changes in how food and liquids are taken, e.g. via mouth, feeding tube, pureed diet, thickened liquids'). Finally, outcomes related to symptoms or events were expressed in terms of frequency and/or severity depending on how they are typically measured in the literature (chapter 4) (e.g. 'penetration/aspiration' was adapted to 'changes in frequency of food or liquids going the wrong way – into the airway/windpipe'). All adaptations were discussed in-depth within the SSC and agreed on by all SSC members.

In the online survey by Sinha et al. (2012) the aim was to identify important outcomes through open-ended questions rather than identifying which outcomes are *important* and which are *not important* and thus, outcomes were not adapted using lay terms. Adapting terms is often only done after the long list of outcomes has been developed, rather than during the process, as most studies do not use quantitative surveys to assess the perspectives of different stakeholders in the preparatory stages of COS development. Nevertheless, choosing this informal but practical approach is consistent with that adopted in other COS studies (e.g. Katiri et al., 2022; Mitchell et al., 2023), and it reduced the burden on the patient partner. At

the same time, we ensured his full and equal involvement in reviewing, modifying, or rejecting terms in collaboration with the SSC.

Part one of the questionnaire aimed at obtaining demographic information of the participants (role, gender, age category, primary residency, age at time of PD diagnosis and year of PD diagnosis). In the second part, participants were asked to indicate from a choice of two answer options which of the 34 items on the long list of outcomes were *important* and which were *not important* by ticking the according box. All items needed to be rated in order to be able to proceed and complete the survey. Lastly, participants were asked if there was anything that was not listed but that is important to them when deciding if a swallowing treatment works. If the same outcome was mentioned by at least two participants, it was incorporated for discussion in the subsequent focus groups.

The questionnaires were the same for all participants, with only the wording of the questions and instructions differing. Using the display logic function in Qualtrics, people with OD in PD were asked, for example, “What age were you when you were diagnosed with Parkinson’s disease?” whereas family members and caregivers were asked “What age was the person with Parkinson’s disease at the time of diagnosis?”. The complete questionnaire can be found in Appendix 5.4.

Data collection methods

The survey was conducted online via Qualtrics (www.qualtrics.com) and was open for three months in the beginning of 2023. The survey was piloted by the SSC’s patient partner, lay people and SLT students. Based on their feedback, minor changes were made to wording. It was estimated to take less than 10 minutes to complete.

Eligibility criteria

People living with OD in idiopathic PD and family members and caregivers of people living with OD in idiopathic PD ≥ 18 years were eligible to take part in the survey. Because the survey was conducted anonymously online, the presence of PD and OD could not be independently verified by the researchers. Therefore, both conditions could be either self-reported by the participant or reported as having been diagnosed by a clinician/physician.

Sample characteristics

Snowball sampling was used by which relevant English-speaking PD organisations, associations, and societies, and clinicians working with people with OD in PD were emailed and asked to act as gatekeepers and disseminate the online survey to their members/people with the health condition. An initial call for participants was posted on X (www.x.com) in January 2023 (Appendix 5.5). Additional tweets and retweets were shared by all members of the SSC. An animated explainer video was designed to provide easy to understand and accessible information on the online survey for lay people. The link to the video was also available in the emails to potential gatekeepers. The aim was to achieve a sample size of ≥ 100 participants, involving both people with OD in PD as well as family members and caregivers. This was calculated based on feasibility within the research project and based on sample sizes in previous surveys related to COS development including people with the health condition and/or caregivers (Jones et al., 2017).

Data analysis

Data was analysed in R (The R Foundation for Statistical Computing, version 4.3.0) descriptively (rating of importance of outcomes in %). To further differentiate important outcomes, they were categorised into most important (≥ 90% of participants in both groups rated outcome as *important*), very important (80-89% of participants in both groups rated outcome as *important*) and important (60-79% of participants in both groups rated outcome as *important*). Data was also analysed inferentially using Pearson's Chi-squared test to determine if there was a significant relationship between the two categorical variables importance and group membership (Bruce et al., 2020). The significance level was set at $p < 0.05$. In case of a statistically significant difference, the strength of association was assessed calculating Cramer's V with values of > 0.1 to 0.3 indicating a small, > 0.3 to 0.5 a medium, and > 0.5 a large effect size (Sheskin, 2011). Odds ratio (OR) was calculated for all statistically significant differences to determine effect sizes. OR > 1 indicates that people with PD are more likely to rate an outcome important, whereas OR < 1 indicates that people with PD are less likely to rate an outcome important compared to family members and caregivers (Field, 2005).

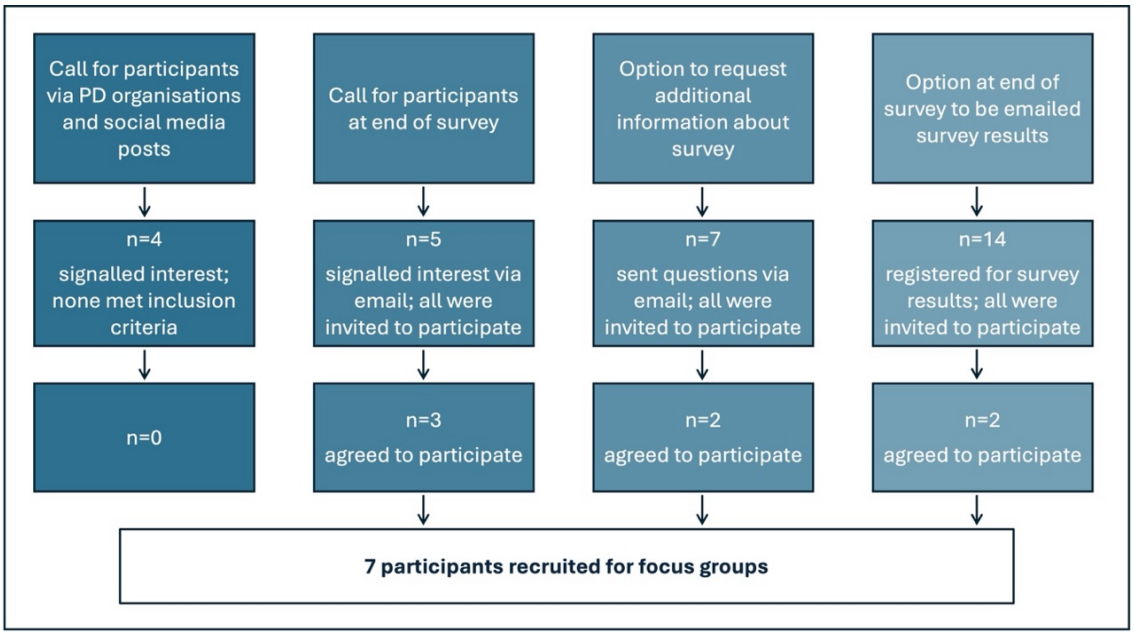
5.2.2 Stage 2: focus groups

Study design, research team and reflexivity

International, online focus groups were conducted to validate the survey findings and clarify inconclusive survey results with participants from the online survey described in section 5.2.1. All focus groups were conducted by JH, who is a PhD candidate, SLT and has been trained in qualitative methodology, and a senior researcher (MW), PhD supervisor, SLT, who is highly experienced in qualitative research. No prior relationship with the participants was established before the commencement of the study. As all participants had completed the online survey, some knowledge about the research project was already established. Before beginning the interviews, the researchers introduced themselves and informed the participants about their professions and roles. LM, a final year undergraduate SLT student, was involved as a second coder in the analysis of the focus group data.

Participant recruitment

Participants were recruited via different strategies. Each way of recruitment and the number of participants recruited via this strategy are outlined in Figure 5.1. The recruitment poster and participant information leaflet can be found in Appendix 5.6 and 5.7. In total, seven participants agreed to take part in the focus groups.



PD: Parkinson's disease

Figure 5.1: Participant recruitment path for focus groups

Focus group setting

All focus groups took place online via Zoom and were audio recorded using the Zoom record function for ease of transcription. All participants agreed to the interviews being audio-recorded in the consent forms and verbally at the beginning of the focus groups. Only JH and MW were present in the virtual meeting room together with the participants. Initially, two focus groups, with four and three participants each, were planned. However, one participant did not show up to the first focus group meeting and due to different time zones could not participate in the second focus group. Therefore, a third focus group was conducted. Two participants each took part in the first two focus groups and three participants in the third. None of the seven participants refused to participate or dropped out.

Data collection

The semi-structured interview guide was designed by JH based on recommendations by Keeley et al. (2016) and the results of the previous survey and was discussed with MW. Afterwards, the interview guide was discussed with the SSC's patient partner to increase comprehensibility and relevance for people with PD, family members and caregivers. The interview guide included three sections: (1) questions on participants' demographics (name, stakeholder group, time of PD diagnosis, country of residence and age category), (2) open-ended questions exploring the experiences, impacts and expectations of swallowing difficulties and treatments in PD, and (3) outcome specific questions aimed at clarifying inconclusive results (outcomes rated statistically significant between people with OD in PD and family members/caregivers and outcomes rated *important*/*not important* by 50% of all survey participants) and to validate survey findings (most important outcomes, outcomes identified as missing but important by survey respondents) (Appendix 5.8).

The interview guide was not shared with the participants before the focus groups to avoid practiced responses. During the focus groups, the questions were shared with the participants via PowerPoint slides using the share screen function on Zoom. JH took notes during and after the focus groups. All participants except one lived in an English-speaking country. To ensure that the participant from Germany understood the questions correctly and could communicate his responses flawlessly, verbal translation was provided when needed by JH, who is a native German speaker.

Data analysis

After the completion of the focus groups, the interviews were transcribed verbatim into Microsoft Word by JH. Any identifying information (such as name, specific age, or hometown) was removed, and all participants were assigned a pseudonym (capital letter P with sequential numbering). All participants were emailed and asked if they would like to read the transcripts. Four participants replied and were then sent the transcripts. Of these, three responded but none asked for any corrections or clarifications.

All three focus group transcripts were uploaded to NVivo (QSR International Pty Ltd., version 12.7.0). Data on participants' demographics were synthesised using case classifications. Data from part two and three were first analysed using Inductive Content Analysis (ICA) following the work steps outlined in Table 5.1. To increase the internal validity, LM independently coded the transcripts using the codes previously created by JH (Elo & Kyngäs, 2008), with any discrepancies discussed together with MW, until consensus was reached.

Although the study protocol had initially specified the use of thematic analysis, ICA was ultimately chosen as a more appropriate analytic method for this study. The rationale for this decision lies in the nature and purpose of the focus group questions, which were relatively specific and directive rather than broad and exploratory. Further, ICA is well suited for our aim to achieve practical answers to inform, e.g. the next stages of the COS project. On the other hand, thematic analysis is typically more theory-oriented, aiming to capture participants' broader worldviews and situate findings within wider conceptual or theoretical frameworks (Vears & Gillam, 2022).

The framework approach (also called framework method) was also considered as an alternative as this is often used for mixed-methods studies where qualitative work follows quantitative work and interview guides are usually more structured (Pope & Mays, 2020). While the analytical process is similar to that in thematic analysis, the key difference with the framework approach is that a matrix is produced for each theme. In each matrix, sub-themes are listed in the columns and participants in the rows (Pope & Mays, 2020). This allows researchers to analyse the data by (sub-)themes and participants. However, the framework approach is better suited for homogenous data with similar topics so that categorising and producing the matrix is facilitated (Gale et al., 2013). While this would have applied to the outcome specific responses in the focus group interviews, this may have been more difficult with responses related

to experiences, impacts and expectations of swallowing difficulties and treatments in PD. Therefore, ICA was chosen as the best available analytical approach.

Choosing ICA over thematic analysis or the framework approach has implications for the study's findings: rather than producing broad thematic narratives, the analysis generated a more focused categorisation of participants' views, prioritising practical applicability over interpretive depth. This methodological decision is consistent with the pragmatic orientation of the study, where the central goal was to obtain actionable evidence to guide consensus-building in the COS project.

Table 5.1: Work steps as part of inductive content analysis adapted from Vears and Gillam (2022)

#	**Work step**	**Conducted by**
1	Read and familiarise	JH
2	First round coding: identifying big picture meanings	JH
3	Second round coding: developing subcategories and fine-grained codes	JH
4	Independent coding by second researcher	LM
5	Merging codes from both researchers	JH
6	Discussion on conflicts with third researcher	JH, MW
7	Refining the fine-grained subcategories	JH
8	Synthesis and interpretation	JH

Part three of the transcripts comprised the outcome specific information. In addition to the inductive coding described above, this part was also coded using a deductive content analysis approach. The aim was to validate the survey findings and clarify inconclusive survey results. Therefore, the deductive approach, where existing theories are tested, was chosen (Vears & Gillam, 2022). First, a categorisation matrix was developed (Table 5.2) (Elo & Kyngäs, 2008). Each outcome was further subdivided into *important* and *not important*.

Table 5.2: Categorisation matrix as part of deductive content analysis

Type of outcome	Outcome
Statistically significantly different outcomes	Changes in how food and liquids are being taken
	Changes in frequency of chest infections
	Changes in number of hospital admissions related to swallowing difficulties
	Other unfavourable changes or events that occur due to swallowing treatment
Most important outcomes	Changes in feeling of food and liquids being stuck in the throat
	Changes in frequency of food and liquids going into the airway
Inconclusive outcomes	Changes in overall speed of swallowing
	Changes in 'tongue pumping' or 'rocking' movements when eating or drinking
	Changes in speed of food/liquids passing through the mouth
Missing outcomes	Changes in fear of choking

Then, the data was coded according to this matrix (Elo & Kyngäs, 2008) by JH. Additionally, a table listing which participant rated an outcome *important*, *not important* or *inconclusive* and, where available, listing the supporting statements for this decision was created in an Excel spreadsheet by JH. LM provided feedback on this table indicating agreement or providing rationale when in disagreement. In cases of uncertainty, MW was included in the discussion.

5.3 Results

5.3.1 Protocol deviations

In total, we planned for ≥ 100 participants in the online survey and seven to eight participants each in four focus groups. However, we were unable to recruit the planned sample size. After careful consideration, we used a content analysis approach instead of thematic analysis for the analysis of the focus group data for the reasons outlined in section 5.2.2. Using content analysis is consistent with another qualitative research study in the field of SLT to develop a COS (Wallace et al., 2017).

5.3.2 Stage 1: online survey

Demographic data

The online survey was completed by 62 participants. Of these, 39 (62.9%) were people living with OD in PD, eight (12.9%) were family caregivers and 15 (24.2%) were family members. In the following analysis of the survey results, we have merged family caregivers and family members to one group (n = 23, 37.1%). Therefore, approximately two thirds of participants were people living with PD and one third were family members/caregivers. All but five participants (8.1%) indicated that they completed the survey on their own. One person with PD completed the survey with the help of someone, and four family members/caregivers completed the survey on behalf of a person with OD in PD. Most participants (n = 39, 62.9%) identified as female, 22 (35.5%) as male, and one (1.6%) identified as non-binary.

Of the 62 participants, 27 (43.5%) were in the age group 61-75 years, with more people with PD than family members/caregivers. The second most often represented age group was 46-60 years with 20 (32.3%) participants. Figure 5.2 shows the number of participants in each age group and gender category.

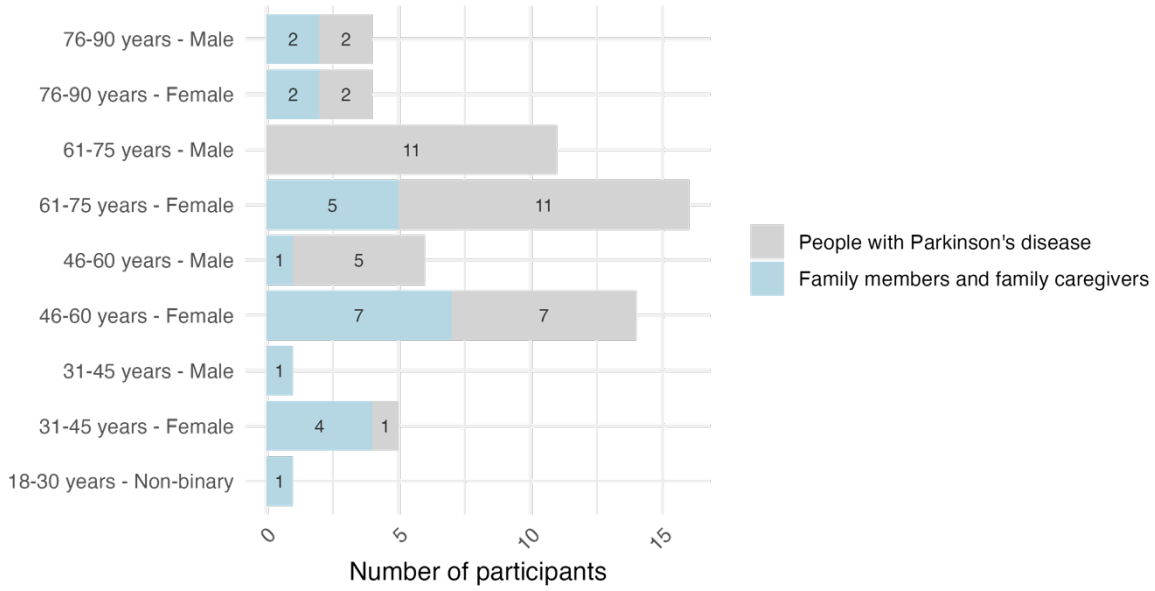


Figure 5.2: Number of participants in each age group and gender category

One participant did not indicate the country of residency. Of the remaining 61 participants, the majority came from Ireland (n = 17, 27.9%), the UK and USA (each n = 12, 19.7%) and New Zealand (n = 10, 16.4%). Four participants (6.6%) were from Canada and two (3.3%) from Germany. One participant each (1.6%) was from Belgium, Croatia, France, or Italy.

The majority of people with PD were ≥ 50 years old when being diagnosed with PD (n = 50, 80.6%), with a range of 35 to 85 years and a mean age of 61.5 years. This was stated by both the people with PD as well as family members/caregivers indicating the age of the person with PD when receiving the diagnosis (Figure 5.3).

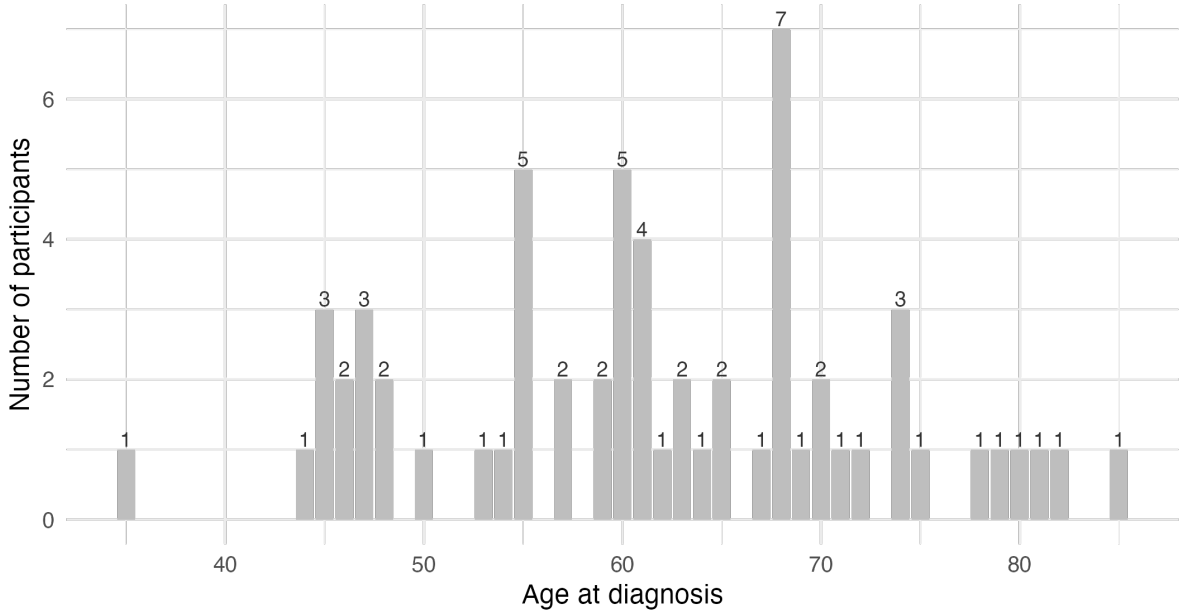


Figure 5.3: Age of people with Parkinson's disease when receiving the Parkinson's disease diagnosis

The number of years since PD diagnosis ranged from one to 21 years. The mean time since diagnosis was 9.52 years. Individual data are depicted in Figure 5.4.

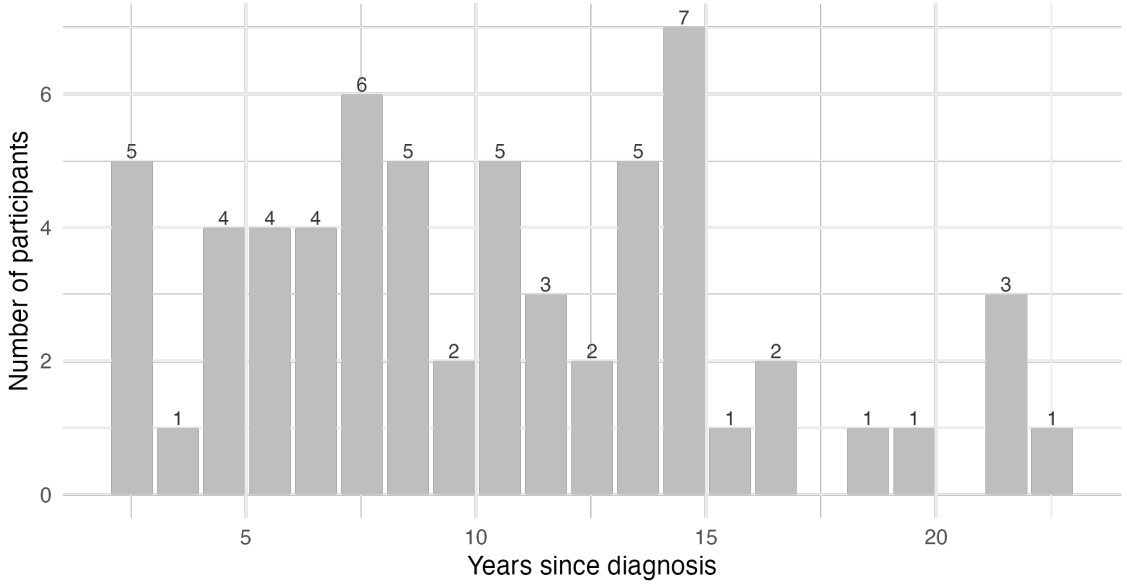


Figure 5.4: Number of years since the Parkinson's disease diagnosis

Outcome specific data

In the second part of the survey, the importance of outcomes was rated by the participants. Table 5.3 depicts all outcomes with the lay terms presented to the participants and the percentage of participants rating each outcome as *important* for each group. A chi-squared test was calculated for all outcomes to determine whether there was a statistically significant difference between the importance ratings of both groups. OR was calculated for all statistically significant differences.

All outcomes were rated as *important* by ≥ 50% of family members/caregivers. All but three outcomes (#22 'Changes in body weight', #24 'Changes in frequency of chest infections', and #25 'Changes in number of hospital admissions') were rated as *important* by ≥ 50% of people with PD. For this reason, we further differentiated important outcomes into most important (≥ 90% of participants in both groups rated outcome as *important*), very important (80-89% of participants in both groups rated outcome as *important*) and important (60-79% of participants in both groups rated outcome as *important*). The two outcomes rated as *most important* were #13 'Changes in frequency of food or liquids going into the airway' and #14 'Changes in feeling of food or liquids being stuck in the throat'. The outcomes #3 'Changes in overall speed of swallowing', #8 'Changes in tongue "pumping" or "rocking" movements' and #11 'Changes in speed of food or liquids passing through the mouth' were inconclusive, which was defined as 40-59% of participants in both groups rating an outcome as *important*. The outcomes #4 'Changes in how food and liquids are taken', #24 'Changes in frequency of chest infections', #25 'Changes in number of hospital admissions' and #32 'Other unfavourable changes or events' were rated statistically significantly different ($p < 0.05$) between participants in both groups in a chi-squared test. However, the OR was 0 for outcome #4, indicating no difference in the likelihood of people with PD rating this outcome more often as *important* than family members/caregivers. On the other hand, outcome #24 has an OR of 0.193, indicating that people with PD are almost one fifth less likely to rate 'Changes in frequency of chest infections' as *important* than family members/caregivers. In other words, family members/caregivers are almost five times more likely to rate this outcome as *important*.

As the recruited sample size was smaller than the target, a post hoc power analysis was conducted for the chi-square tests with p values of < 0.05. Using Cramer's V, the achieved sample size of 62 participants provided values in the range of upper small to lower medium effect sizes.

Table 5.3: Rating of importance of outcomes by people with Parkinson's disease, family members/caregivers and interpretation of results

	Survey item	PwPD	Fm/c	p (OR, V)	Interpretation
1	Changes in overall swallowing severity scores	82%	78%	0.974	important
2	Changes in overall awareness of swallowing function	92%	87%	0.807	very important
3	Changes in overall speed of swallowing	56%	52%	0.952	inconclusive
4	Changes in how food and liquids are taken (e.g. via mouth, feeding tube, pureed diet, thickened liquids)	77%	100%	**0.034*** (0, 0.269)	important, different
5	Changes in how the mouth feels in terms of dryness	67%	65%	>0.999	important
6	Changes in frequency and severity of saliva leaking out of the mouth	67%	87%	0.143	important
7	Changes in amount of saliva accumulated in the mouth or throat	77%	78%	>0.999	important
8	Changes in tongue "pumping" or "rocking" movements when eating or drinking	51%	48%	>0.999	inconclusive
9	Changes in control over food or liquids in the mouth	82%	78%	0.974	important
10	Changes in need to break down food or liquids in the mouth in order to swallow	85%	96%	0.362	very important
11	Changes in speed of food or liquids passing through the mouth	51%	57%	0.892	inconclusive
12	Changes in frequency of food or liquids going the wrong way - into the nose	77%	96%	0.114	important
13	Changes in frequency of food or liquids going the wrong way - into the airway/windpipe	90%	100%	0.292	most important
14	Changes in feeling of food or liquids being stuck in the throat	92%	96%	>0.999	most important
15	Changes in frequency of choking episodes	79%	100%	0.053	important
16	Changes in food or liquids left in the mouth after swallowing	67%	74%	0.754	important
17	Changes in food or liquids left in the throat after swallowing	79%	87%	0.689	important

18	Changes in cough strength so the airway/windpipe can be cleared easier	82%	100%	0.082	very important
19	Changes in coughing as a response of feeling food or liquids going down the wrong way	85%	96%	0.362	very important
20	Changes in strength of tongue muscles	64%	57%	0.747	important to people with PD only
21	Changes in strength of throat muscles	87%	83%	0.904	very important
22	Changes in body weight related to swallowing difficulties	41%	70%	0.056	important to fm/c only
23	Changes in swallowing Parkinson's medication	77%	83%	0.835	important
24	Changes in frequency of chest infections	41%	78%	**0.01*** (0.193, 0.328)	important to fm/c only, different
25	Changes in number of hospital admissions related to swallowing difficulties	41%	83%	**0.003*** (0.146, 0.371)	important to fm/c only, different
26	Changes in quality of life related to swallowing	87%	96%	0.519	very important
27	Changes in caregiver's quality of life related to swallowing in person with Parkinson's	62%	87%	0.066	important
28	Changes in fear or anxiety related to eating and drinking	82%	87%	0.881	very important
29	Changes in pleasure of eating and drinking related to swallowing	92%	74%	0.107	important
30	Changes in avoidance of food or liquids related to swallowing	82%	87%	0.881	very important
31	Changes in other areas related to swallowing muscles, e.g. voice or speech	87%	96%	0.519	very important
32	Other unfavourable changes or events that occur due to swallowing treatment	54%	91%	**0.006*** (0.111, 0.351)	important to fm/c only, different
33	Person with Parkinson's following the intervention as prescribed	77%	83%	0.835	important
34	Opportunity to give feedback on overall swallowing programme	82%	87%	0.881	very important

Fm/c: family member/caregiver, OD: oropharyngeal dysphagia, OR: odds ratio, PwPD: people with Parkinson’s disease, V: Cramer’s V

Most important: ≥ 90% of participants in both groups rated outcome as important; very important: 80-89% of participants in both groups rated outcome as important; important = 60-79% of participants in both groups rated outcome as important; inconclusive = 40-59% of participants in both groups rated outcome as important; different: outcome was rated statistically significantly different between groups, when $p < 0.05$ using chi square test, the odds ratio was calculated

Of the 62 participants, eleven provided an answer to the question if things were not listed in the long list of outcomes, but they felt were important when deciding if a swallowing treatment works (Appendix 5.9). Three of these answers were feedback in general, two comprised outcomes that were already included in the long list and twelve referred to new outcomes. Of these, 'changes in fear of choking' was the most often and the only outcome being listed by at least two participants. For this reason, 'changes in fear of choking' was included for discussion in the subsequent focus groups.

5.3.3 Stage 2: focus groups

Demographic data

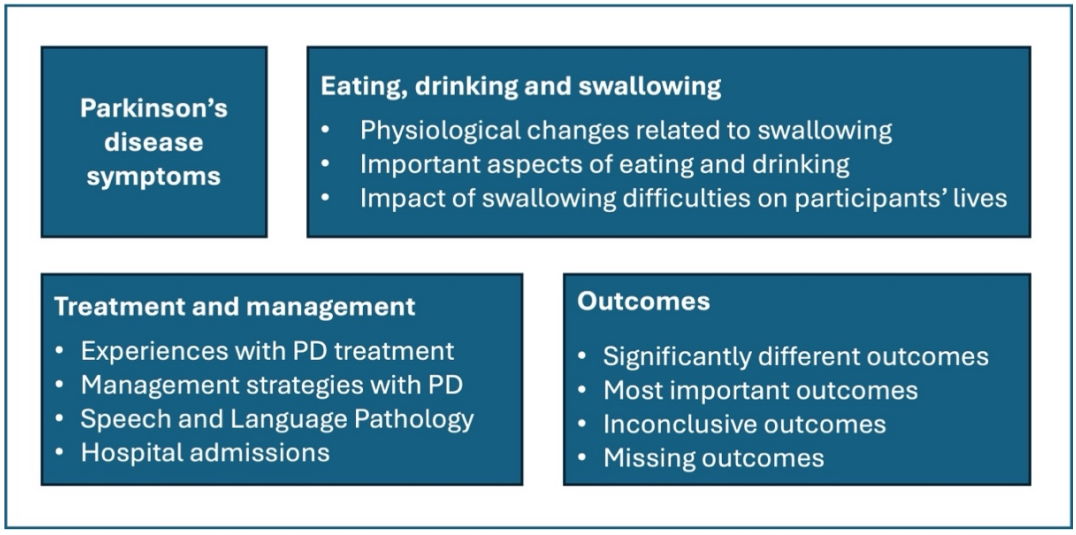
Three focus groups with seven participants were conducted. The duration of the focus groups varied between 55-65 minutes. Table 5.4 provides an overview of the demographic data of the seven participants. In terms of P3 (the family caregiver) the age group, years since PD diagnosis and received swallowing treatment refer to the person with OD in PD, her father. P1 was originally from the Netherlands and P4 from the USA. All participants, except for P2, were fluent English speakers.

Table 5.4: Demographic data of focus group participants

Group	ID	Role	Country of residency	Gender	Age range (years)	Years since PD diagnosis	Swallowing treatment
1	P1	PwPD	New Zealand	Female	76-90	< 1	No
1	P2	PwPD	Germany	Male	61-75	10	Yes
2	P3	FC	Ireland	Female	31-45	1	Yes
2	P4	PwPD	New Zealand	Male	61-75	5	No
3	P5	PwPD	Canada	Female	61-75	< 1	No
3	P6	PwPD	UK	Female	76-90	10	Yes
3	P7	PwPD	USA	Female	61-75	6	Yes

PD: Parkinson's disease, PwPD: person with Parkinson's disease, FC: family caregiver

In addition to the outcome category from the deductive approach, three further content categories were generated through ICA. These comprise seven sub-categories and are depicted in Figure 5.5.



PD: Parkinson's disease

Figure 5.5: Content categories and sub-categories of focus group interviews

Category 1: Parkinson's disease symptoms

Participants were not explicitly asked about PD symptoms but did refer to some symptoms throughout the interviews. Therefore, this does not represent a comprehensive list of all PD symptoms that participants might experience. P1 and P5 were living with the condition for the shortest time (less than 1 year). P1 did not report any specific PD symptoms besides swallowing difficulties, whereas P5 used to experience dyskinesia due to medication intake which has now improved. P4 received the PD diagnosis 5 years ago and recognized that "Everything takes me longer than it used to. The whole life has slowed down" (P4). P7 received the PD diagnosis 6 years ago and is rather experiencing internal than external tremors "even down to intestinal, rectal issues" (P7). She also reported to have had increasing dyskinesia due to medication intake which has resolved. P2 and P6 have both been living with the PD diagnosis for ten years. P2 only reported that his SLT was helpful as she could also advise him on issues such as rigidity. P6 stated that she is a wheelchair user as she has spinal stenosis. Additionally, she reported not having tremors but dystonia as "my feet, my toes curl under all the time" (P6). P3 was the primary caregiver of her father who had been diagnosed one and a half years ago but had passed away half a year prior to the interview. According to P3 he had a very late diagnosis and therefore his symptoms were already more advanced. He spent most of the time in bed and needed to use a wheelchair outside of bed.

Category 2: Eating, drinking, and swallowing

Physiological changes related to swallowing difficulties. This sub-category is further refined into overall changes and changes in the pre oral, oral, oropharyngeal and pharyngeal phase (Table 5.5). Additionally, one participant (P5) described that she has noticed difficulties with singing which she associates with her swallowing difficulties. Overall, 18 different symptoms that participants reported during the focus groups were identified.

Table 5.5: Physiological changes related to swallowing difficulties and exemplary quotes from participants

Swallowing phase	**Changes related to swallowing**	**Quotes**
Changes in pre oral phase	Difficulties holding cutlery	“It takes me longer because I can’t use a knife or fork anymore. I can’t hold them” (P6)
	Loss of sense of smell	“Because of the Parkinson’s, I have also lost a lot of my sense of smell” (P1)
Changes in oral phase	Change in taste	“I need to feed myself to stay alive, but a lot of the fun, of the flavour is gone” (P1) “He enjoyed it still. Not as much as he would have before, because I think his taste had changed. Food that he used to like, he didn’t like anymore“ (P3) “One weekend I was eating a steak and it was wonderful. And the next weekend I ate another, or tried to eat another steak, and the taste; it tasted rotten. Like I couldn’t eat it. My taste buds change, they fluctuate on me” (P5)
	Drooling	“I have noticed on occasion that I drool every once in a while. It doesn’t happen often” (P5)
	‘Tightening’ of orofacial muscles	“My biggest problem is the muscle in my jaw. Which I used to feel like tightened up. Now it’s kind of improved. With that new medication. But I wonder if it will come back. But basically, it’s the jaw, it’s spasming” (P7)
	Oromotor tremors	“My jaw moves constantly. I can’t keep it still”, “My tongue moves all the time” (P6)
	Chewing difficulties	“I’m having to chew and chew and chew” (P5)
	Tongue pumping	“There was the tongue pumping and rocking, he didn’t notice he was doing that” (P3) “My tongue will pump and rock when I can’t get stuff down, but I don’t have a constant problem with my tongue moving” (P5)
	Piecemeal deglutition	“Dad was holding on to a little bit of, so it was like he was swallowing, but then bits, he wasn’t actually swallowing the whole amount” (P3)
	Difficulties with bolus transport	“When I get to dryer meat, I have to swallow and swallow and swallow and swallow” (P5)
Changes in oropharyngeal phase	Difficulties with swallowing specific consistencies	“I have it mainly with drinks, not really with food yet“ (P1) “I don’t have a problem with liquids, but I have problems with different meats, like pork and beef, that are more dry than chicken or fish” (P5)

	Difficulties with swallowing pills	“Liquids don’t seem to pose a problem unless I’m trying to swallow 15 pills” (P5)
	Reduced swallowing speed	“You don’t swallow so quickly anymore” (P1) “He didn’t really notice that it was taking a long time for him to swallow something” (P3) “My speed of swallowing has changed a lot. I used to eat very quickly” (P6)
	Odynophagia	“Occasionally it can even be a bit painful to swallow”, “Sometimes it can be sore” (P1)
Changes in pharyngeal phase	Food stuck in the throat	“Sometimes you have things stuck in your throat” (P1) “When it sticks around in the throat and doesn't want to go forward or backward“ (P2)
	Penetration/aspiration of food/liquids and reflexive coughing	“Especially when I drink, I very easily inhale the drink and start coughing” (P1) “My biggest problem is the food and liquids going down the airway” (P4) “He was coughing, yes, there was a lot of coughing” (P3)
	Choking	“If I’m choking, it’s because the food went down the wrong tube and then I have to cough it back up” (P4)

Important aspects of eating and drinking. This sub-category includes seven important aspects of eating and drinking identified by the focus group participants. Table 5.6 provides an overview of these aspects with corresponding quotes from the participants.

Table 5.6: Important aspects of eating and drinking and exemplary quotes from participants

Important aspects of eating and drinking	Quotes
Appearance	"We went back to giving him food that was softer, but resembled the actual food", "Seeing the food as food was really important to him" (P3)
Taste	"I like to eat things that taste good, and I like to drink things that taste good" (P4)
Texture	"For me, it makes a difference whether I can eat food the way it is served or whether it is crushed, for example, or not. So, I don't do purees" (P2) "I know he was swearing a lot at mealtimes. He was saying 'this is shit, I can't eat this'. He'd have it unpureed", "He did become quite depressed about the texture of the food" (P3)
Pleasure	"I'm going to think what he would have said would be pleasure. To enjoy food, to enjoy mealtimes. I think if he could have had; like he didn't want to have necessarily more time, just a good time" (P3) "It's important what we eat and if we enjoy it. Parkinson's can take a lot of things away, but one of the things that I enjoy is food. I intend to keep on enjoying it with help, with exercise" (P5)
Eating and talking	"Eat in such a way that you can still participate in the conversation to some extent" (P2)
Same as before	"I'd rather eat food the way I've eaten it all my life" (P4)
Same as everyone else	"I want to eat the same as everyone else" (P6), "YES!" (P5) "I want to eat the same way as the others. The same food, if possible." (P2), "Yea, I agree" (P1)

Impact of swallowing difficulties on participants' lives. This sub-category was further subdivided into impacts on physical functioning and medical status, behaviour, emotional functioning and wellbeing, and on social functioning (Table 5.7).

Table 5.7: Impacted aspects of participants' lives and exemplary quotes from participants

Aspects of participants' lives	**Changes related to swallowing**	**Quotes**
Impact on physical functioning and medical status	Pneumonia	"It was an increase in chest infections, they were just coming much closer together and then it was aspiration pneumonia in the end that he couldn't recover from" (P3)
	Loss of appetite	"There are times when I sit down at the table, and I just choke a lot and I'm having to chew and chew and chew and I lose my appetite. I'm not interested" (P5)
		"I used to love French bread and big sandwiches, all this just kind of looks intimidating and like I'm just going to lose my appetite" (P7)
	Dehydration	"To make sure that you drink enough during the day, it's not that easy" (P1)
		"I was told I need to drink more. I don't drink enough. Typical dehydration" (P2)
	Change in weight	"Probably because of this slow eating, I have trouble keeping my weight" (P2)
		"Every time he went to the hospital, he lost weight, he deteriorated, and then he'd come home, and we really had to spend time on building him back up again" (P3)
		"I've lost about twenty pounds and continue to lose" (P5)
Impact on behaviour	Alternating liquid solid swallows	"I find that drinking often or regularly makes food easier to swallow. Probably because it is softer then" (P2)
	Changing specific consistencies	"So, we did have to give him pureed food near the end" (P3)
		"My favourite is chicken. Just because it goes down so easily" (P5)
		"When I go out, I just order sandwiches now. I find that easier. I eat sandwiches more than anything else" (P7)
	Coughing to clear the airway	"Eventually I had to cough it up. I couldn't get it down. It was wedged. I could hardly breathe" (P5)
	Eating and drinking sufficiently	"I have now decided to add weight gainer to my diet again in the near future, so that I can at least regain a little more weight" (P2)
		"Anything that would have discouraged my dad from eating obviously was really a thing because calories were so important to him" (P3)
	Reducing bolus volume	"I'm taking much longer to eat. Because I'm taking smaller bites, especially meat" (P5), "Like P5, I take small bites" (P7)
	Reducing the speed	"I know that I slow down my speed of food and liquids through my mouth. Not necessarily because I have to, but because I want to. Because I want to avoid what P5 is talking about. We drink carefully now, not to choke or anything. And that's not a big deal for me, doing a little slower and drinking slower. It just reminds you that you have Parkinson's" (P7)
	Taking pills in yoghurt	"I take my pills in yoghurt. […]. Because it's easier" (P6)
	Using drinking aids	"I bought these sippy cups with straws and lid so I wouldn't spill everything all over me" (P5)
		"I use a straw" (P6)

	Being careful and paying attention	"I have to pay attention to my eating" (P2) "I'm a little more careful, a little more aware" (P4) "We drink carefully now, not to choke or anything" (7)
	Seeking help and advice	"The most useful person in dad's circle of care that he had, was the speech and language therapist. She was very good" (P3) "Get someone else to cut it [the food] for me" (P6)
	Waiting and getting over it	"I choke when I swallow sometimes. […] I just get over it. […] People, they want to tap you on the back and stuff like that. No, it's alright. I just be alright in a minute" (P4)
Impact on social functioning	Longer mealtime durations	"So, it takes me a long time to eat, and there is my husband sitting there. and he's finished" (P5) "I'm pretty much always the last one to finish eating. I wouldn't say it's problematic" (P7)
	Going out for food and drinks	"No, not really. I'm still going out as often" (P6), "We're not. And that's just me." (P5)
	Participating in conversations during mealtimes	"I don't get to talk much because I have to pay attention to my eating. Chewing well, swallowing small pieces. […] That's the disadvantage for me, it's also a burden, that I can hardly take part in conversations. I become more and more silent, so to speak, pushed to the edge a bit" (P2)
Impact on emotional functioning and wellbeing	Continuing to enjoy	"He really enjoyed being with us all at the table even though he wasn't taking part in the conversation. He clearly enjoyed it. That was quality of life to him" (P3)
	Loss of enjoyment	"It is less fun to go out to eat with others" (P7)
	Discomfort and fear	"It was harder to find someone who would be comfortable with sitting with dad if he's like coughing while he is eating. They were scared that if something happened that they would be responsible for him" (P3) "The biggest fear is, of course, choking" (P2)
	Frustration	"I would be lying if I said that there wasn't frustration sometimes" (P3)
	Worries	"I have a tendency to worry more about making an idiot of myself in a restaurant, choking and needing all this attention, instead of just eating a quiet dinner and enjoying myself" (P5)
	Uncertainties	"What is this? Is this normal? Is it something different? Does it belong to the Parkinson's?" (P1)

Category 3: Treatment and management

Experiences with general PD treatment. Some participants asked each other questions about access to neurologists and about their current medication intake. In focus group 3, participants talked about their experiences with medication doses resulting in secondary symptoms, e.g. dyskinesia.

> "I started to get dyskinesia. I mean, it was horrendous. I now understand what Michael Fox goes through […]. It's just a constant shaking and it lasts for hours and it's because I was taking too much levodopa/carbidopa" (P5)

Management strategies with PD. One approach was accepting the condition and not looking too far ahead. Another one was staying independent for as long as possible. Exercising and staying active was important to most participants.

> "If we don't exercise, we're like in a canoe without a paddle" (P5)

At the same time, one participant emphasized that exercise can only slow down the progression of PD but not stop it.

> "Exercise has been crucial for me being stable, I think. And of course it's still downhill" (P7)

Speech and Language Pathology. Experiences with SLT and swallowing treatment in particular varied largely between participants. Three participants have not accessed SLT for their PD while two participants did in the past but were not anymore at the time of the interviews. P4 was not aware that swallowing treatment for OD in PD existed. P7 was involved in research on OD in PD and received OD treatment at a university centre. P3 reported that an SLT came to their home and mainly provided information and advice. The expectations of a swallowing treatment varied among participants. The majority emphasized that they wished for a swallowing treatment to improve the physical aspects of swallowing.

> "It would make it so that when I eat the food it goes into my oesophagus and not my lung" (P4)

This would then facilitate or resolve other difficulties associated with OD, such as choking, inability to participate in communication during mealtimes, pharyngeal residue, pneumonia and hospital admissions related to OD in PD.

Hospital admissions. This sub-category was further divided into 'Experiences with hospital admissions' and 'Feelings towards being in hospital'. All participants felt that they wanted to avoid going to hospital as much as possible and that if they had to go, it would be a serious situation for them. Only P3 had experiences with hospital admissions, as her father was having recurring pneumonias that needed to be treated in hospital. She outlined the differences between the care her father received at home compared to when being in hospital, the difficulties of hospitals being understaffed, not being able to meet their patients' needs and that her father had to be on a modified diet when being in hospital, which he would not eat.

> "Every time he went to the hospital, he lost weight, he deteriorated and then he'd come home, and we really had to spend time on building him back up again" (P3)

Category 4: Outcomes

Significantly different outcomes. The majority of participants considered the outcomes that were rated statistically significantly different in the online survey (Table 5.3) as important during the focus group interviews. Outcome #4 'Changes in how food and liquids are taken' was *important* to all but one participant due to their perspectives on important aspects of eating and drinking. P4 considered this outcome as *not important* as he does not need to modify his diet, and he prefers to eat the same as before.

Outcome #24 'Changes in frequency of chest infections' was *important* to five participants. Especially for P3, whose father had recurring pneumonias, this was an important indicator of her father's OD.

> "So, that I guess, like was a huge measure of his swallowing difficulty at the very late stage; was the frequency of the chest infections" (P3)

Other participants were aware of chest infections being a possible complication and therefore considered it to be *important*. P4 initially did not understand the term 'chest infection'. P1 and P4 considered this outcome *not important* as they were not experiencing any chest infections.

Outcome #25 'Changes in number of hospital admissions' was *important* to all participants based on the described experiences with feelings towards being in hospital.

Outcome #32 'Other unfavourable changes or events that occur due to swallowing treatment' was an outcome that was difficult to comprehend for many of the participants and hence, the decision whether this was considered *important* or *not important* seemed to be more challenging for some of them. The term 'adverse events' with examples of potential adverse events due to a swallowing treatment seemed to facilitate the comprehensibility of this outcome. Overall, five participants considered this outcome to be *important*. Most of them explained that they did not envision any adverse events and if there were any, most likely they would be minor and acceptable to them.

> "A lot of times the unfavourable side effects are going to be small, and you can discard them later, but get the data. Without the data you're not going to know if it's important or not" (P4)

Participants' reasons for rating this outcome as *not important* were either that they did not want to learn about potential adverse events or that they had not experienced any relevant adverse changes due to a swallowing treatment. Therefore, they would not expect to avoid a swallowing therapy due to adverse events and hence, did not consider it important.

Most important outcomes. All participants agreed to outcome #13 ‘Changes in frequency of food or liquids going the wrong way - into the airway/windpipe’ and all but one participant agreed that outcome #14 ‘Changes in feeling of food or liquids being stuck in the throat’ are the two most important outcomes. P7 did not clearly indicate whether she thought only outcome #13 or both outcomes #13 and #14 were *important*. P2 added that ideally the frequency of food and liquids going into the airway should be ‘zero’ and that not only the feeling of food and liquids being ‘stuck in the throat’ should change but that no residue should remain in the pharynx at all. P3 explained that to her those outcomes are important as they would affect outcome #24.

Inconclusive outcomes. The outcomes that were not clearly either *important* or *not important* in the online survey mostly remained inconclusive in the focus groups. Outcome #3 ‘Changes in overall speed of swallowing’ was *important* to four participants, *not important* to two and was not clearly rated by one participant. Reasons for considering it *important* were that it increases mealtime duration and affects the social aspect of eating and drinking. P3 did not particularly refer to the overall speed of swallowing but to a prolonged mealtime duration only. P5 added that not only speed was important but also efficiency, whereas P7 responded that the effectiveness of the swallow was important. P4 considered this as *not important* as he was not affected by a change of his swallowing speed.

Outcome #8 ‘Changes in tongue ‘pumping’ or ‘rocking’ movements when eating or drinking’ was considered *important* only to P6. She reported that her tongue moves all the time regardless of whether she is eating and drinking which leads to a permanent rubbing of her tongue on her teeth. This eventually makes her tongue sore. It remains unclear whether the reported symptom is primarily a tongue pumping movement or rather a lingual tremor. P3 reported that she observed that her father’s tongue was pumping when eating and drinking but he himself did not seem to notice it. Likewise, P5 noticed that her tongue is pumping when eating. Both, P3 and P5, did not clearly indicate whether this was an important outcome to measure or not. P7 did not express any opinion. P1, P2 and P4 considered this outcome *not important* as they were not experiencing any tongue pumping.

Outcome #11 ‘Changes in speed of food and liquids passing through the mouth’ was rated important by P1 and P6 as both were experiencing this change.

> “Yes. Because that’s one change, an enormous one” (P6)
>
> “In which way?” (R1)

"I'm much slower now" (P6)

P2, P4 and P7 considered this outcome as *not important* as again, they were not experiencing this change. P7 added that she deliberately slowed down the rate at which food and liquids passed through her mouth to make it safer to swallow.

"And that's not a big deal for me, doing a little slower and drinking slower. It just reminds you that you have Parkinson's" (P7)

P3 and P5 did not clearly categorise this outcome as *important* or *not important*.

Missing outcomes. Lastly, participants were asked if they agreed that it was important to measure 'Changes in fear of choking' as an outcome in studies on OD interventions in PD. Five participants agreed to it being *important*. P1 indicated that to her it was not fear but rather being aware of the possibility to choke on food or liquids. P4 considered this outcome to be *not important*.

"I never really thought of it as an anticipatory fear of choking. I don't anticipate any fear until it starts happening" (P4)

5.3.4 Comparison of important and most often measured outcomes

The following step is to evaluate whether the outcomes selected by researchers in OD intervention studies in PD align with the outcomes identified very important by individuals with PD and family members/caregivers. To achieve this, outcomes that are measured in ≥ 2 studies included in the scoping review in chapter 4 are compared with outcomes rated as *important* by ≥ 80 participants in the online survey as shown in Figure 5.6. To facilitate comparison, technical terms instead of lay terms are being used.

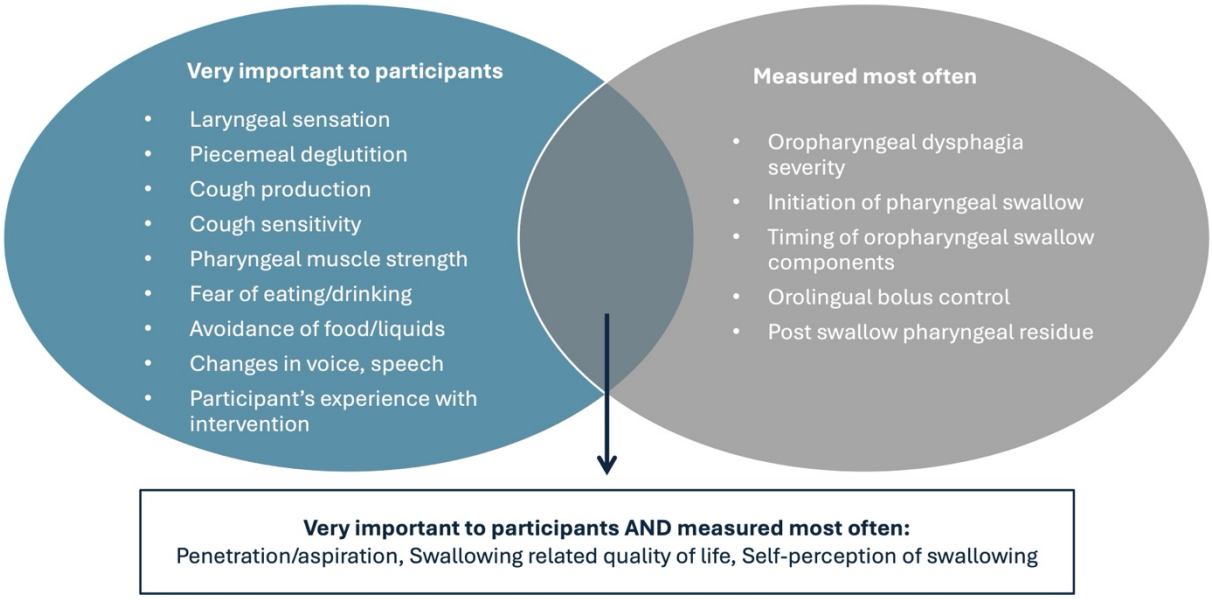


Figure 5.6: Comparison between outcomes that are very important to participants in the online survey and outcomes measured most often in research studies with overlap of outcomes

Figure 5.6 highlights that only the outcomes 'penetration/aspiration', 'swallowing related quality of life', and 'self-perception of swallowing' are the three outcomes considered very important by people with PD and family members/caregivers as well as measured most often in research studies.

5.4 Discussion

In this mixed methods study, we identified intervention outcomes that are important to people living with OD in PD and family members/caregivers for measurement in OD studies. Differences between the perspectives of people with PD, family members/caregivers, and what researchers measure in clinical trials were evident in this study. This discrepancy aligns with findings from other COS studies, underscoring a recurring gap between clinical research priorities and the lived experiences of patients and family members/caregivers (Dodd et al., 2023).

In total, twelve outcomes were classified as important to survey participants. Of these, only three are being measured by researchers on a regular basis, four are measured rarely and five are not measured by researchers at all (chapter 4) (Hirschwald et al., 2023). The two most important outcomes identified through the online survey and validated by focus group participants were 'penetration/aspiration' and 'laryngeal sensation'. 'Penetration/aspiration' was also the most frequently measured outcome in research studies on OD interventions in PD (in ten of the 19 studies) identified in chapter 4. In contrast, 'laryngeal sensation' was only measured in one of the 19 included studies. It should be noted, however, that when

measuring ‘penetration/aspiration’, ‘laryngeal sensation’ is often measured indirectly, for example when the widely used PAS (Rosenbek et al., 1996) is applied as an OMI.

Of the ten outcomes classified as very important in the survey, only ‘self-perception of swallowing’ and ‘swallowing related quality of life’ were among the most frequently measured outcomes found in the earlier scoping review in chapter 4. Of the remaining eight outcomes rated as very important by survey participants, five were not measured in any of the included studies in the scoping review. ‘Cough production’ and ‘pharyngeal muscle strength’ were measured twice and once respectively in uncontrolled studies only (Curtis et al., 2020; Hirschwald et al., 2023; Pitts et al., 2009) but not in controlled studies in chapter 4.

Three outcomes not measured in published OD in PD intervention trials but considered as *important* by the SSC were ‘cough sensation‘, ‘fear of eating/drinking‘ and ‘avoidance of food/liquids‘. These outcomes were added to the long list of outcomes in the survey. Even though they did not feature in any OD intervention study in PD, they were classified as very important to survey participants. The inclusion of these three outcomes in the survey may introduce a potential bias, stemming from the possibility that participants were influenced by their presence, potentially elevating their perceived importance. The SSC’s decision to add these outcomes was based on expert opinion, as well as the recognition that these factors are significant in clinical practice, despite their absence in prior research. While these outcomes were rated as very important by the majority of survey participants, it is important to acknowledge that their inclusion could have skewed the results toward recognising these specific aspects.

Additionally, ‘changes in fear of choking’ was suggested as a so far missing but important outcome by participants in the survey and validated by focus group participants. Therefore, this outcome will be added to the long list of outcomes and incorporated into the next stages of the COS project.

Previous studies found that people with the health condition review the importance of outcomes in the perspective of their own life (Hewlett, 2003; Sathanapally et al., 2020). This is true also in this study. During the focus groups it became evident that participants assessed the importance of outcomes in the context of their own experiences. Outcomes experienced as people with PD or noticed as family caregiver, were rated particularly important to measure in OD intervention studies in PD. Therefore, if, for example, someone did not notice a symptom, it was more likely to be not important to them. This might suggest that especially OD severity and the specific symptoms influence the choice of outcomes by people with

the health condition and family members/caregivers. Additionally, this is further supported by the feedback of one survey participant stating that (s)he only described the symptoms when rating the importance of outcomes.

Strengths and limitations

A key strength is that, to our knowledge, this is one of the first studies investigating perspectives of people living with the condition on what outcomes are important to measure in OD intervention studies in PD. Despite this study being conducted in English only, we received responses to the online survey from countries where English is not the first language. However, particularly perspectives from Asia, Africa, and South America were lacking in both the survey and the focus groups. Conducting the study in English and online only may have been a barrier for people with OD in PD from these continents in particular, but not exclusively, to participate in this study. This may have led to a smaller sample size than the planned ≥ 100 participants. Particularly the statistically significantly different outcome ratings between groups need to be interpreted with caution as the effect sizes were small to medium.

Furthermore, the use of snowball sampling may have introduced a potential sampling bias. However, by asking a diverse group of societies and organisations to act as gatekeepers, an attempt was made to minimise this bias. This is reflected in a wide range of participant ages and years since PD diagnosis, suggesting a mitigated sampling bias. In addition, women, who have been shown to be underserved in previous OD in PD research studies (Hirschwald et al., 2024), were well represented in our sample. Nevertheless, multiple participation by the same person could not be avoided due to the anonymity of the survey. The number of focus groups as well as the number of participants in each focus group were small, and it remains unclear if data saturation was reached.

One unanticipated challenge in this study was that some focus group participants did not fully understand the outcomes ‘other unfavourable changes or events that occur due to swallowing treatment’ and ‘changes in the frequency of chest infections’, and additional explanations during the focus groups were needed. This was despite the fact that guidance provided by Gargon et al. (2017) and Keeley et al. (2016) was followed and a short video explaining the concept of a COS and the aims of the online survey using lay terms was developed together with the SSC’s patient partner. Therefore, there is a risk that participants in the survey did not fully or correctly understand the meaning of these outcomes either. One strategy to address this issue could have involved the preliminary testing of the items’ wording through cognitive or

think aloud interviews with individuals with the health condition, as suggested in the COMET handbook (Williamson et al., 2017). While such methods can provide detailed insights, they are resource-intensive and may place a considerable burden on people with OD in PD. Since the COS project was still in the preparatory stage, a more pragmatic approach was adopted, with informal feedback being sought from the SSC's patient partner, lay people, and SLT students. This approach yielded a range of perspectives on comprehensibility and usability, while concurrently minimising participant burden and ensuring the process remained proportionate to the study stage. For the next stage of the COS project, we will amend the wording of the two outcomes 'other unfavourable changes or events that occur due to swallowing treatment' and 'changes in the frequency of chest infections' to optimise comprehensibility for people with OD in PD and family members/caregivers.

5.5 Summary

The assessment of the choice of outcomes by individuals living with OD in PD and family members/caregivers ensures the inclusion of perspectives from this stakeholder group in the long list of outcomes. This list will be further expanded through the views on important OD intervention outcomes in PD by dysphagia clinicians in the next chapter.

Chapter 6: What is measured in dysphagia intervention studies in Parkinson's disease: an international online survey with dysphagia clinicians[8]

6.1 Introduction

In the previous chapter 5 we identified similarities and differences between the perspectives of people with PD, family members/caregivers, and what researchers measure in clinical trials (chapter 4). This suggests that there may also be discrepancies between perceived importance of outcomes between dysphagia clinicians and researchers. A dysphagia clinician is defined as a person implementing OD interventions in clinical practice. The aim of this study was to identify dysphagia clinicians' views on important outcomes, OMIs and timepoints of measurement in OD interventions in PD.

6.2 Methods

The reporting of this study follows CROSS (Sharma et al., 2021) (Appendix 6.1). Completing the online survey was taken as consent.

6.2.1 Study design

An international, cross-sectional, anonymous online survey was conducted. The survey was divided into three main sections with 18 questions. The first section aimed to identify participants' perspectives on OMIs used in clinical practice, relevant timepoints of measurement, and important outcomes.

First, participants (i.e. dysphagia clinicians working with people with OD in PD) were asked to choose all OMIs they use in clinical practice with people with OD in PD from a predefined list and to add further OMIs, if applicable. Next, participants were asked to indicate at which timepoints they believed outcomes should be measured from a predefined list (ranging from 4 weeks before start of intervention to 24 months

[8] Adapted from: Hirschwald, J., Mooney, L., Battel, I., Riera, S., & Walshe, M. (2025). What is measured in dysphagia intervention studies in Parkinson's disease: An international online survey of dysphagia clinicians. *International Journal of Speech-Language Pathology.* 1-10. https://doi.org/10.1080/17549507.2025.2555252

after completion of intervention) and to add further timepoints, if applicable. Then, participants were asked what outcomes are important to them when deciding if a swallowing intervention for people with OD in PD works by rating each item as *important* or *not important*. The long list of outcomes was devised based on outcomes currently being measured in OD intervention studies in PD (chapter 4). Participants were then asked to add outcomes if they considered them to be important but missing in the long list of outcomes provided.

In the second section, information was collected on the profile of participants' clients with OD in PD, including age, ethnicity, gender, co-occurring cognitive impairment, and dysarthria. In the final section, demographic data on participants themselves was obtained, including profession, current employment status, work setting and country of employment, years of work experience (as a clinician in general and with people with OD in PD specifically), age group and gender (Appendix 6.2).

The questionnaire was piloted by a dysphagia clinician experienced in providing OD interventions in PD, thus a potential participant. This test demonstrated some limitations due to the lack of a multiple-choice option. Subsequently, the necessary adjustments were made. Pilot testing helped establish the ecological validity of the survey. The survey was shared with the study translators for review and translation. One of the translators raised concerns about the flow of questions. The feedback was discussed and agreed on between LM and MW. As a result, the order of questions was further improved based on the feedback. Pilot testing improved respondent reliability by ensuring that there was no ambiguity in the survey questions and that the survey followed a similar presentation throughout. However, formal reassessment of reliability and validity was not conducted after the revisions. The survey was estimated to take less than 15 minutes to complete.

6.2.2 Data collection

The survey was conducted using the online platform Qualtrics (www.qualtrics.com) and was open for five weeks in July and August 2023. The questionnaire was translated from English into German, Italian, Spanish, and Mandarin Chinese (Simplified) by four translators. All translators were native speakers of their relevant language. These languages were selected due to the well-developed dysphagia services in the countries where these languages are spoken and ready access to translators. This allowed for the survey to be disseminated internationally, increasing the sample size and enhancing the sample's

representativeness of the study population. Relevant settings in Qualtrics were amended to prevent multiple participation by respondents.

6.2.3 Data sampling

Dysphagia clinicians working with people with idiopathic PD, regardless of country of practice, setting, or years of experience were included. In addition, for eligibility, participants needed to be proficient in either English, German, Italian, Spanish, or Mandarin Chinese (simplified).

Purposive, non-probability snowball sampling was applied by contacting national and international professional SLT organisations, national and international dysphagia special interest groups, and contacts of the authors. A second email was sent to the same distribution list two to three weeks after the initial email, as a reminder to disseminate the survey and to reduce non-response error. The survey was also disseminated via the authors' social media accounts (Appendix 6.3).

As no reference number of clinicians working with people with OD in PD internationally exists, the sample size was calculated based on an unknown population, using an online sample size calculator (www.calculator.net/sample-size-calculator.html). A minimum of 385 survey respondents were needed to achieve a confidence level of 95%.

6.2.4 Data analysis

All data were analysed within Microsoft Excel (Version 16.79). Quantitative data was analysed using descriptive statistics (absolute numbers, percentages). Qualitative data was translated to English (where applicable), and any identifying information was removed. OMIs listed by participants were first differentiated between 'swallowing related OMI' or 'unable to classify' (e.g. too global, not swallowing specific or not an OMI but rather an outcome). Duplicates within the swallowing related OMIs were removed and all remaining OMIs classified to one of the following categories: (1) protocols or scales used with instrumental assessment, (2) investigator reported outcome measures (IROMs), (3) PROMs, or (4) screening tools.

Likewise, responses regarding additional important but so far missing outcomes were first differentiated between 'swallowing related outcome' or 'unable to classify' (e.g. too broad, not swallowing related, not an outcome but rather an OMI). If it was identified as a swallowing related outcome, duplicates both

within the participants' responses as well as regarding the long list of outcomes were removed. Based on the previous survey with people with OD in PD and family members/caregivers on relevant outcomes in OD intervention studies in PD (chapter 5), important outcomes were further differentiated into most important (≥ 90% of participants rated outcome as *important*), very important (80-89% of participants rated outcome as *important*), important (60-79% of participants rated outcome as *important*), inconclusive (40-59% of participants rated outcome as *important*) and not important (≤ 40% of participants rated outcome as *important*).

6.3 Results

From a total of 337 responses, 123 (36.5%) responses were partially completed, and therefore were excluded from further analysis. With the remaining 214 responses, the required sample size of n = 385 to reach a confidence level of 95% was not achieved. The response rate cannot be calculated due to the use of snowball sampling and the number of dysphagia clinicians working with people with PD being unknown.

6.3.1 Participants

Most participants identified as female (n = 191, 89.3%) and all participants were ≤ 65 years. Participants were from five continents and 28 different countries, with a third being from Germany (n = 71, 33.2%). Half of the participants (n = 107, 50%) completed the survey in English. Most participants were working clinically at the time of completing the survey and 87 (40.7%) participants indicated to be working in more than one setting. Table 6.1 summarises the demographics of survey participants.

Table 6.1: Participants' demographics (n = 214)

Demographic characteristics		**n**	**%**
Gender (n=214)	Woman	191	89.3%
	Man	18	8.4%
	Non-binary	1	0.5%
	Prefer not to answer	4	1.9%
Age groups in years (n= 214)	18-25	9	4.2%
	26-33	68	31.8%
	34-41	55	25.7%
	42-49	27	12.6%
	50-57	31	14.5%
	58-65	24	11.2%

Continent and country of practice (n=212)	North America (n=7)	USA	6	2.8%
		Canada	1	0.5%
	South America (n=1)	Brazil	1	0.5%
	Africa (n=12)	South Africa	9	4.2%
		Namibia	2	0.9%
		Rwanda	1	0.5%
	Europe (n=145)	Germany	71	33.2%
		Italy	26	12.1%
		Ireland	22	10.3%
		UK	11	5.1%
		Slovenia	4	1.9%
		Spain	2	0.9%
		Switzerland	2	0.9%
		Austria	1	0.5%
		Czech Republic	1	0.5%
		Denmark	1	0.5%
		France	1	0.5%
		Greece	1	0.5%
		Sweden	1	0.5%
		The Netherlands	1	0.5%
	Europe/Asia (n=9)	Turkey	8	3.7%
		Russia	1	0.5%
	Asia (n=13)	China	5	2.3%
		Israel	4	1.9%
		India	2	0.9%
		Thailand	2	0.9%
	Oceania (n=25)	Australia	22	10.3%
		New Zealand	3	1.4%
Language which participant completed the survey in (n=214)		English	107	50.0%
		German	74	34.6%
		Italian	27	12.6%
		Mandarin Chinese (Simplified)	4	1.9%
		Spanish	2	0.9%
Profession (n= 214)		SLT	204	95.3%
		Occupational therapist	2	0.9%
		Physiotherapist	2	0.9%
		Rehabilitation doctor/physician	2	0.9%
		Dietitian-Nutritionist	1	0.5%
		Medical doctor	1	0.5%
		Neurologist	1	0.5%
		Phoniatrician	1	0.5%
Currently working clinically (n= 214)		Yes	198	92.5%

		No	16	7.5%
Setting of clinical practice (326 responses from n=214)		Rehabilitation Setting	90	27.5%
		Community	88	26.9%
		Hospital Setting (e.g. outpatient clinic, acute care)	83	25.4%
		Residential Setting	48	14.7%
		Research Clinic or University	6	1.8%
		Private Practice	6	1.8%
		Specialised Unit/Ward	3	0.9%
		Hospice or Palliative Care	1	0.3%
		Mental Health Service	1	0.3%
		Not specified	1	0.3%
Number of years of experience as a dysphagia clinician (n=211)		< 1	5	2.4%
	1-5 (n=47)	1	5	2.4%
		2	10	4.7%
		3	11	5.2%
		4	10	4.7%
		5	11	5.2%
	6-10 (n=57)	6	10	4.7%
		7	7	3.3%
		8	11	5.2%
		9	7	3.3%
		10	22	10.4%
		≥ 11	102	48.3%
Number of years of experience as a dysphagia clinician working with people with OD in PD (n=209)		< 1	3	1.4%
	1-5 (n=60)	1	8	3.8%
		2	11	5.3%
		3	13	6.2%
		4	13	6.2%
		5	15	7.2%
	6-10 (n=57)	6	13	6.2%
		7	7	3.3%
		8	11	5.3%
		9	4	1.9%
		10	22	10.5%
		≥ 11	89	42.6%

OD: oropharyngeal dysphagia, PD: Parkinson's disease, SLT: Speech and Language Therapist

6.3.2 Participants' clients with OD in PD

The most common gender of participants' clients with OD in PD was indicated as male (n = 185, 86.4%).

The majority of participants also responded that their clients had co-occurring cognitive impairment

(n = 116, 54.2%) and dysarthria (n = 182, 85%). Full details of the demographics of participants' clients with OD in PD are depicted in Table 6.2.

Table 6.2: Demographics of participants' clients

Demographic characteristics		n	%
Gender (n=214)	Male	185	86.4%
	Female	14	6.5%
	Other	15	7%
Age groups in years (448 from n=214)	< 50	11	2.5%
	50-59	40	8.9%
	60-69	124	27.7%
	70-79	181	40.4%
	≥ 80 years	92	20.5%
Ethnicity (216 from n=214)	White	185	85.6%
	Mixed/multiple ethnic groups	14	6.5%
	Other	9	4.2%
	Asian	8	3.7%
Co-occurring cognitive impairment (n=214)	Yes	116	54.2%
	No	73	34.1%
	Unsure	25	11.7%
Co-occurring dysarthria (n=214)	Yes	182	85.0%
	No	15	7.0%
	Unsure	17	7.9%

6.3.3 Outcome measurement instruments

Overall, the 214 participants provided a total of 475 OMI selections, indicating that dysphagia clinicians use two to three OMIs on average. The most frequently used OMIs were PAS (Rosenbek et al., 1996), reported by 132 participants (61.7%) and accounting for 27.7% of total selections. The Functional Oral Intake Scale (FOIS) (Crary et al., 2005) was reported by 100 participants (46.7%) and constituted 21.1% of total selections, while the SWAL-QOL questionnaire (McHorney et al., 2002) was reported by 68 participants (31.8%) and made up 14.3% of total selections.

The option 'other' was chosen 101 times by participants with many participants suggesting multiple OMIs within one response. A total of 194 suggested OMIs were identified. Of these, 39 were categorised as 'unable to classify' (Appendix 6.4). After further removing OMIs that were duplicates of the prespecified OMIs in the survey, identical OMIs were merged, which resulted in 40 different additional OMIs.

Both the prespecified OMIs as well as the additional OMIs were analysed and categorised together. Of the 46 OMIs, 16 were classified as protocols or scales used with instrumental assessment (Table 6.3), twelve as IROMs (Table 6.4), eleven as PROMs (Table 6.5) and seven as screening tools (Appendix 6.5).

Table 6.3: Outcome measurement instruments classified as protocols or scales used with instrumental assessment

Outcome being measured	**Scale used with instrumental assessment**	**n**
Overall OD severity	Dysphagia Severity Rating Scale (DSRS) (Gramigna, 2006; Waxman et al., 1990)*	46
Overall OD severity	Dysphagia Outcome and Severity Scale (DOSS) (O'Neil et al., 1999)	5
Overall OD severity	Videofluoroscopic Dysphagia Scale (VDS) (Kim et al., 2012)	1
Overall OD severity	Dysphagia Severity Scale (DSS) (Tohara et al., 2003)	1
Swallowing safety and efficiency	Visual Analysis of Swallowing Efficiency and Safety (VASES) (Curtis et al., 2022)	5
Overall OD severity, swallowing safety and efficiency	Dynamic Imaging Grade of Swallowing Toxicity (DIGEST) , DIGEST-FEES (Labeit et al., 2023; Starmer et al., 2021)	4
Swallowing safety	Penetration-Aspiration Scale (Rosenbek et al., 1996)*	132
Swallowing efficiency	The Yale Pharyngeal Residue Severity Rating Scale (Gerschke et al., 2019; Neubauer et al., 2015)	10
Swallowing efficiency	Bolus Residue Scale (Rommel et al., 2015)	1
Swallowing efficiency	Pooling Score (Farneti et al., 2014)	2
Saliva management	Radboud Oral Motor Inventory for Parkinson's Disease (ROMP) Saliva (Kalf et al., 2011)*	13
Secretion management	Murray Secretion Scale (Murray et al., 1996; Pluschinski et al., 2014)	8
Secretion management	Oral Secretion Scale (Cazzolli et al., 2020)	1
Pill swallowing	FEES based graduation of ability to swallow pills (Buhmann, Bihler, et al., 2019)	1
Type of instrumental assessment	**Protocol used with instrumental assessment**	**n**
Videofluoroscopy	MBS Measurement Tool for Swallow Impairment (MBSImP) (Martin-Harris et al., 2008)	2
Videofluoroscopy	Analysis of Swallowing Physiology: Events, Kinematics and Timing (The ASPEKT Method) (Steele et al., 2023)	2

OD: Oropharyngeal dysphagia

* This outcome measurement instrument was listed in the survey.

Table 6.4: Outcome measurement instruments classified as investigator reported outcome measures

Category	Investigator reported outcome measure	n
Global OMI	AusTOMs Swallowing scale (A. Perry et al., 2004)	3
Global OMI	American Speech-Language-Hearing Association (ASHA): NOMs - Functional Communication Measures - Swallowing (FCMs) (American Speech-Language-Hearing Association (ASHA), 2020)	1
Clinical swallow evaluation tool	The Test of Masticating and Swallowing Solids (TOMASS) (Huckabee et al., 2018)	1
Clinical swallow evaluation tool	Mann Assessment of Swallowing Ability (MASA) (Mann, 2002)	5
Clinical swallow evaluation tool	Kölner Befundsystem für Schluckstörungen (Kö.Be.S) (Birkmann, 2017)	5
Clinical mealtime evaluation tool	Minimal Eating Observation Form-Version II (MEOF-II) (Westergren, 2019)	1
Scale for level of oral intake	Functional Oral Intake Scale (FOIS) (Crary et al., 2005; Hamzic et al., 2021)*	100
Scale for level of oral intake	Bogenhausener Dysphagia Score (BODS 1 and 2) (Starrost et al., 2012)	11
Scale for level of oral intake	Royal Brisbane Hospital Outcome Measure for Swallowing (RBHOMS) (Ward & Conroy, 1999)	4
Scale for overall OD severity, level of oral intake, saliva swallowing	Munich Swallowing Score (MUCSS) (Bartolome et al., 2021)	4
Scale for level of oral intake	Schluckbeeinträchtigungsskala (SBS) (Prosiegel et al., 2002)	1
Scale for drooling	Drooling Severity and Frequency Scale (DSFS) (Evatt et al., 2009)	2

OD: oropharyngeal dysphagia, OMI: outcome measurement instrument,

* This outcome measurement instrument was listed in the survey.

Table 6.5: Outcome measurement instruments classified as patient or carer reported outcome measures

Outcome being measured	Patient reported outcome measure	n
Swallowing related quality of life	SWAL-QOL questionnaire (McHorney et al., 2002)*	68
Swallowing related quality of life	MD Anderson Dysphagia Inventory (MDADI) (Chen et al., 2001)*	15
Self-perception of swallowing	Eating Assessment Tool (EAT-10) (Belafsky et al., 2008)	19
Self-perception of swallowing	Swallowing Disturbance Questionnaire (SDQ) (Manor et al., 2007)	7
Self-perception of swallowing	Munich Dysphagia Test – Parkinson's disease (MDT-PD) (Buhmann, Flügel, et al., 2019)	4
Self-perception of swallowing	The Dysphagia Handicap Index (Silbergleit et al., 2012)	1
Self-perception of swallowing	Self-report of swallowing symptoms by person with PD	1
Self-perception of swallowing	Swallowing diary	1

Self-perception of sialorrhea	Sialorrhea Clinical Scale for PD (SCS-PD) (Perez Lloret et al., 2007)	2
Self-perception of reflux	Reflux Symptom Index (RSI) (Belafsky et al., 2002)	1
Outcome being measured	**Carer reported outcome measure**	**n**
Swallowing related carer burden	Caregiver Analysis of Reported Experiences with Swallowing Disorders (CARES) Screening Tool (Shune et al., 2020)	1

*This outcome measurement instrument was listed in the survey.

6.3.4 Timepoints of measurement

Table 6.6 summarises the timepoints presented to participants and the frequency at which they were selected. Overall, 816 choices were recorded from 214 participants. Thus, participants selected three to four measurement timepoints on average. Assessments immediately before and after completion of the intervention were chosen the most frequently by participants (n = 205, 96%; n = 177, 83% respectively).

Table 6.6: Timepoints of suggested measurement of outcomes by participants

Chronological order*	Timepoints of measurement	n	%
2	At start of intervention	205	96%
3	At end of intervention	177	83%
6	6 months after intervention	92	43%
8	12 months after intervention	89	42%
5	3 months after intervention	69	32%
4	4 weeks after intervention	58	27%
11	24 months after intervention	43	20%
N/A	Other	31	14%
10	18 months after intervention	24	11%
7	9 months after intervention	14	7%
1	4 weeks before start of intervention	7	3%
9	15 months after intervention	7	3%

N/A: not applicable

* This represents the order of choices according to timepoints related to the intervention, with (1) being the earliest and (11) the latest timepoint.

Twelve participants responded through the option ‘other’ that the timepoint of measuring outcomes depends on the client and their individual treatment plan. A further six participants responded that they believe OMIs should be used after a clinical change or a change in a client’s condition. Five participants indicated that they consider outcomes to be measured “as required”, three participants responded with

"every visit", two "every six to twelve months or at regular review sessions", one suggested at "commencement and close of an episode of care" and another participant responded "when videofluoroscopy or fibreoptic endoscopic evaluation of swallowing is used". One participant ticked this option but did not provide a response.

6.3.5 OD intervention outcomes in PD

All outcomes were considered *important* by ≥ 60% of participants (Table 6.7). Of these, 14 were classified as most important, nine as very important and eleven as important. Nearly a fifth (n = 40, 18.7%) of participants rated all outcomes *important*, whereas none of the participants rated all outcomes *not important*. The outcomes 'penetration/aspiration' and 'swallowing related quality of life' were most frequently (n = 209, 98%; n = 208, 97% respectively) rated as *important*.

Table 6.7: Participants' ratings of importance of outcomes and classification

Outcome	n	%	Classification
Penetration/aspiration	209	98%	Most important
Swallowing related quality of life	208	97%	Most important
Avoidance of food/liquids	205	96%	Most important
Fear of eating/drinking	202	94%	Most important
Pneumonia	202	94%	Most important
Weight loss	202	94%	Most important
Level of oral intake	200	93%	Most important
Oropharyngeal dysphagia severity	198	93%	Most important
Pleasure of oral intake	199	93%	Most important
Post swallow pharyngeal residue	200	93%	Most important
Cough production	194	91%	Most important
Participant's adherence to intervention	195	91%	Most important
Self-perception of swallowing	194	91%	Most important
Orolingual bolus control	193	90%	Most important
Medication intake	191	89%	Very important
Post swallow oral residue	191	89%	Very important
Participant's experience with intervention	186	87%	Very important
Oral bolus transport	183	86%	Very important
Adverse events	181	85%	Very important
Changes in Voice, and/or Speech	178	83%	Very important
Initiation of pharyngeal swallow	175	82%	Very important
Choking	173	81%	Very important
Laryngeal sensation	172	80%	Very important
Carer burden	170	79%	Important
Pharyngeal muscle strength	169	79%	Important
Drooling	166	78%	Important
Hospitalisation	165	77%	Important
Cough sensitivity	161	75%	Important
Nasal regurgitation	158	74%	Important
Salivary pooling	157	73%	Important
Swallowing related orolingual movement	156	73%	Important
Lingual muscle strength	154	72%	Important
Dry mouth	141	66%	Important
Piecemeal deglutition	128	60%	Important

Most important: ≥ 90% of participants rated outcome as important; very important: 80-89% of participants rated outcome as important; important = 60-79% of participants rated outcome as important

In total, 59 participants used the open text box within the survey and entered a total of 83 potential outcomes. After removing duplicates and responses that could not be classified, 14 outcomes remained. Table 6.8 lists the additional outcomes and the number of participants that suggested each additional outcome.

Table 6.8: Additional relevant outcomes identified by participants

Outcome	n
Independent eating/drinking	4
Social aspect of eating/drinking	4
Mealtime duration	3
Respiratory swallow coordination	2
Hydration	2
Nutrition	1
Eating and drinking preferences	1
Efficiency of oral intake	1
Effort of eating and drinking	1
Infrahyoid and suprahyoid muscle plasticity	1
Lingual tremor	1
Opening of the upper oesophageal sphincter	1
Fatigue levels at mealtime	1
Patient's level of swallowing related education	1

6.4 Discussion

This research study explored dysphagia clinicians' perspectives on clinically relevant outcomes, OMIs used and timepoints of measurement in OD interventions in PD. A total of 46 different OMIs were used by participants in clinical practice with people with OD in PD with PAS (Rosenbek et al., 1996), FOIS (Crary et al., 2005) and SWAL-QOL (McHorney et al., 2002) being used most often. All 35 swallowing related outcomes were considered *important* by most participants and five new outcomes ('independent eating/drinking', 'social aspect of eating/drinking', 'mealtime duration', 'respiratory swallow coordination' and 'hydration') were identified. Participants considered it important to measure outcomes before the start and up to 24 months after completion of the intervention.

The current study found that dysphagia clinicians use a wide range of OMIs and consider a wide range of outcomes to be important to measure when working with people with OD in PD. This is unsurprising given the individuality of patient care and the international work context of the respondents. However, we have found similar findings in OD intervention studies in PD in chapter 4, where a wide range of outcomes and OMIs are also used. In OD intervention studies in PD the outcomes 'penetration/aspiration' and 'swallowing related quality of life' were measured most consistently using PAS (Rosenbek et al., 1996) and SWAL-QOL (McHorney et al., 2002), which is consistent with the common use of these OMIs in clinical practice. However, 'level of oral intake' is rarely measured in OD intervention studies in PD

and consequently FOIS is not an OMI regularly used in the studies in chapter 4, whereas one fifth of participants in this study use the OMI in clinical practice. This might emphasise the importance of monitoring the change in level of oral intake in clinical practice.

Despite the large number of OMIs used, we still lack validated OMIs for outcomes that are considered important by respondents, such as 'choking'. This outcome is currently not measured in OD intervention studies in PD in chapter 4 but is considered important not only by participants in this study, but also by people living with the condition in chapter 5. In addition, some of the OMIs lack good reliability and validity for the PD population. When developing the COS, only reliable OMIs validated for PD will be considered, if available.

The outcomes reported as most important to dysphagia clinicians in this survey, i.e. 'penetration/aspiration' and 'swallowing related quality of life', overlap with the outcomes commonly measured in OD intervention studies in PD. However, other outcomes important to dysphagia clinicians, such as 'avoidance of food/liquids', 'fear of eating/drinking', 'pneumonia' and 'weight loss', are rarely measured or not measured at all in OD intervention studies in PD in chapter 4. It is interesting to note that the outcomes considered important by participants reflect difficulties in both the oral and pharyngeal phases of swallowing. However, few of the OMIs used by participants comprehensively assess the oral phase of swallowing. This may indicate a discrepancy between what is considered important and what is measured.

Participants considered for outcomes to be measured up to 24 months post completion of the intervention. This is contrary to current practice in OD intervention studies in chapter 4, where primarily short-term effects are being assessed. This limits the possibility of assessing maintenance effects of the intervention, particularly important in neurodegenerative conditions like PD. Thus, the consideration of assessing long-term intervention effects aligns with the call by Battel and Walshe (2023) for outcomes to be measured at 6, 12, 18 and 24 months after completion of the intervention.

While more than half of the participants reported that their clients have co-occurring cognitive impairment, it is interesting to note that in research more than half of OD intervention studies in PD explicitly excluded participants with cognitive impairment. Other studies either do not report the cognitive status of participants or only include people with PD who have mild cognitive impairment (Hirschwald

et al., 2024). This highlights the need to include all clinically present characteristics of clients with OD in PD in interventions studies for this population.

Strengths and limitations

This study is one of the first to assess dysphagia clinicians' perspectives on what outcomes are important to measure in OD interventions for people with PD. It also explored how and when dysphagia clinicians measure these outcomes. A key strength of the study was that the survey was available in five different languages, allowing for a broad international perspective on this topic. It should be noted that the target sample size was not reached and although participants represented 28 different countries, more than half were from Germany, Ireland and Italy. Particularly, one third was from Germany indicating that sampling bias may be a potential limitation of our study. This imbalance likely reflects the use of snowball sampling and JH's strong professional network in Germany, which facilitated more effective distribution in that context. While this increased participation enhanced response rates, it may limit the representativeness of the sample and the generalisability of findings to other cultural or linguistic groups.

The inclusion of only six OMIs in the survey may have led to a potential bias in participants reporting that these OMIs are more commonly used in clinical practice than OMIs that were not listed. However, an open text field was provided, which was used extensively by participants minimising this bias.

6.5 Summary

The findings highlight not only the overlaps between clinical practice and research but also the important gaps between both areas. While clinicians employ a diverse range of OMIs and consider numerous outcomes important, many clinically relevant outcomes, such as 'choking', and 'fear of eating/drinking', remain underrepresented in intervention studies and validated OMIs are lacking.

The disparity between the follow-up period clinicians consider important (up to 24 months post completion of intervention) and the predominantly short-term focus of current research underscores the need for longer-term follow-ups, particularly given the neurodegenerative nature of PD. Furthermore, the exclusion of participants with cognitive impairment from many intervention studies is in contrast to clinical practice, where co-occurring cognitive impairment is common.

This study completes the development of the long list of outcomes. In the next chapter, the results on establishing the COS-DIP are presented.

Chapter 7: Developing a core outcome set for dysphagia interventions in Parkinson's disease: an international Delphi consensus study[9]

7.1 Introduction

This study describes the development of the COS-DIP involving people living with OD in PD, family members and family caregivers, healthcare professionals working with people with OD in PD and researchers working in this field. The aim was to achieve agreement on *what* outcomes should be measured and *how* and *when* these outcomes should be measured. We devised the COS-DIP primarily for use in future clinical trials. The health conditions covered by the COS are all types and severities of OD and the population all individuals with idiopathic PD regardless of disease severity and duration. The COS focusses on interventions that directly improve swallowing difficulties, and/or reduce specific physiological risks associated with OD, such as pneumonia, malnutrition, or choking.

7.2 Methods

The COS-STAR (Kirkham et al., 2016a) and the Accurate Consensus Reporting Document (ACCORD) (Gattrell et al., 2024) guidelines were followed in this manuscript to report the study. Participation in the eDelphi was taken as consent. Written and verbal consent was obtained from all participants prior to consensus meetings (Appendix 7.1).

7.2.1 Participants

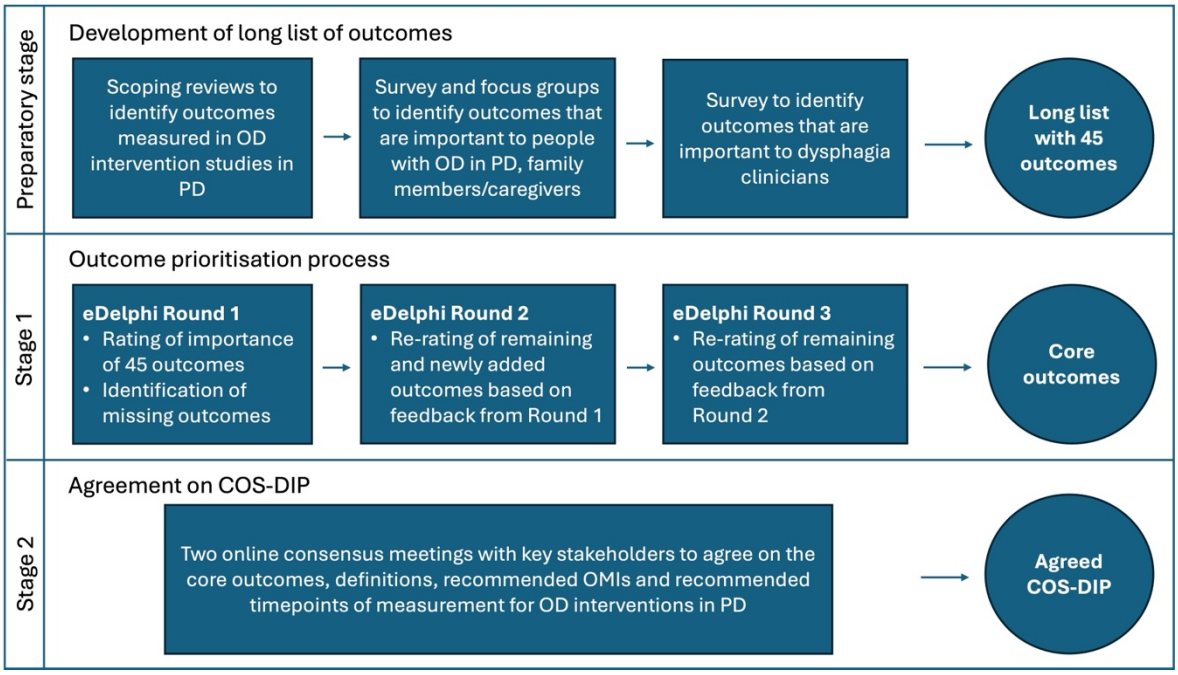
Key stakeholder groups included in this study were people living with OD in PD, family members/caregivers with experience of providing care for a family member and/or living with a family member with OD in PD, healthcare professionals with experience of delivering OD interventions for people with PD, and researchers with experience of undertaking research on OD interventions in PD. Participants needed to be proficient in English and ≥ 18 years old. We applied a maximum variation

[9] Adapted from: Hirschwald, J., Conroy, P., Boyle, G., Warnecke, T., & Walshe, M. (2025). Developing a core outcome set for dysphagia interventions in Parkinson's disease: An international Delphi consensus study. *BMJ Evidence-Based Medicine*, bmjebm-2024-113543. https://doi.org/10.1136/bmjebm-2024-113543

sample through purposive and snowball sampling through relevant organisations, societies, support groups, personal contacts and social media posts (Appendix 7.2). Authors of published OD in PD trials were contacted directly. In total, it was planned to recruit 150-200 participants for the Delphi survey. Consensus meeting participants were people who completed all three rounds of the online Delphi (eDelphi) survey and agreed to take part in such a meeting at the end of Round 3 of the eDelphi survey. Participant information leaflets for people with OD in PD and family members/caregivers as well as for healthcare professionals and researchers can be found in Appendix 7.3 and 7.4.

7.2.2 Development of long list of outcomes

The development of the COS-DIP involved several stages as depicted in Figure 7.1.



COS-DIP: Core Outcome Set for Dysphagia Interventions in Parkinson's disease, OD: oropharyngeal dysphagia; PD: Parkinson's disease, OMI: outcome measurement instrument

Figure 7.1: Overview of stages involved in developing the Core Outcome Set for Dysphagia Interventions in Parkinson's disease

In the preparatory stage, we generated a long list of outcomes for the use in the eDelphi survey through two scoping reviews. In these, we identified 26 OD related intervention outcomes currently measured in OD studies with control groups in PD (chapter 4) and three additional OD related intervention outcomes in OD studies without control groups in PD (Hirschwald et al., 2023). Based on discussion within the SSC, we added ten further OD-related intervention outcomes.

To identify outcomes that are important to people with OD in PD, family members/caregivers and dysphagia clinicians, we conducted online surveys and subsequent focus groups. Based on the results of these studies, we added six additional outcomes (chapters 5 and 6). The final long list comprised 45 intervention outcomes related to OD in PD (Appendix 7.5).

7.2.3 Stage 1: outcome prioritisation process

We conducted a three-round eDelphi survey to identify the core outcomes. The long list of outcomes was translated into lay terms with guidance from a lay person living with PD using the same approach as in section 5.2.1. All outcomes and lay terms were uploaded to DelphiManager platform (www.comet-initiative.org/delphimanager). Round 1 was piloted by two SLTs and two lay persons. Based on the received feedback, we amended wording of the instructions to increase comprehensibility. To mitigate potential attrition bias, we sent regular personalised reminder emails about survey completion and current response rates to participants who had not yet completed the current eDelphi round.

7.2.3.1 Outcome scoring

We asked participants to rate the importance of each outcome on a 9-point Likert scale as recommended by the GRADE Working Group (Guyatt et al., 2011). As preset by DelphiManager, scores of 1 to 3 indicate *not important*, 4 to 6 *important but not critical* and 7 to 9 *critically important*. A score of 10 was *unable to rate* and a text field was provided for additional comments. At the end of Round 1, we asked participants to list any missing but important outcomes. Outcomes suggested by ≥ 2 participants were added as new outcomes to Round 2.

In Rounds 2 and 3, we asked participants to re-rate the importance of the outcomes based on the feedback provided. This included the participant's own rating from the previous round and the percentage of each stakeholder group's rating for each outcome from the previous round. Participants were anonymous to each other. Only participants who rated all outcomes in one round were invited to take part in the following round.

7.2.3.2 Consensus definition

Consensus in after Rounds 1 and 2 was defined as ≥ 70% of all participants scoring 7 to 9. After Round 3, *consensus in* was set at ≥ 60% of all participants scoring 7 to 9. All outcomes that did not reach the

consensus in criterion were dropped. Outcomes that reached the *consensus in* criterion formed the list of outcomes for the following round or after Round 3 for the consensus meetings.

7.2.4 Stage 2: agreement on COS-DIP

We conducted two online consensus meetings using the videoconferencing platform Zoom (www.zoom.com) to find consensus on the core outcomes, suggested definitions, OMIs and timepoints of measurement. Suggested definitions of outcomes were based on previously published definitions and suggested OMIs were based on validated OMIs for OD in PD, either commonly used in research studies in OD in PD (chapter 4) or in clinical practice by dysphagia clinicians (chapter 6) as recommended by Prinsen et al. (2016b). Timepoints of measurement were suggested based on commonly used timepoints in research studies (chapter 4) as well as through the discussion within the SSC. We asked participants whether they agreed (yes/no/unsure) with the final core outcomes, suggested definitions, OMIs and timepoints of measurement by voting anonymously using Zoom's polling function. Consensus was defined as ≥ 70% of participants voting 'yes'.

Due to conducting two different consensus meetings, voting on agreement could vary between meetings. To confirm participants' perspectives, we conducted an anonymous follow-up survey on Qualtrics (www.qualtrics.com) and all previous consensus meeting participants were invited by email to complete the survey. Participants were asked whether they agreed (yes/no/unsure) with the final COS-DIP (Appendix 7.6). We applied the same consensus definition of ≥ 70% of participants voting 'yes'.

7.2.5 Data analysis

Data on participant demographics and ratings of each outcome in each eDelphi round were analysed descriptively in R (The R Foundation for Statistical Computing, version 4.3.0). We included only data from participants who rated all outcomes in one round. Participants who rated all outcomes identically to each other in one round were excluded. A rating of 10 (unable to rate) was excluded from data analysis for individual outcomes. Feedback on results from previous eDelphi rounds was calculated and displayed in the following round automatically by DelphiManager. Data of the consensus meetings were analysed descriptively in Excel (Version 16.87).

7.3 Results

7.3.1 Protocol deviations

Deviations made from the study protocol and the corresponding reasons for these changes are displayed in Table 7.1.

Table 7.1: Deviations from study protocol with reasons

Study protocol	Deviation	Reason
Stakeholder definition: people *diagnosed* with OD in PD; healthcare professionals and researchers with ≥ *3 years of experience*	Self-perceived OD or diagnosed OD; any time period for which healthcare professionals and researchers worked in this field	To reach as large as a panel as possible, to facilitate a wide range of participants
Timeline: eDelphi survey rounds to be open for *three weeks* each, reminder emails to be sent to participants who have not completed current round after *two weeks*	Deadline for eDelphi survey closure extended by two to five days depending on response rate, reminder emails were sent weekly	To minimise attrition rate/increase response rate as suggested by COMET handbook (Williamson et al., 2017)
Definitions of outcomes in eDelphi: N/S	Definitions were amended between rounds based on participants' feedback	Suggestions for changing definitions were not anticipated by SSC, but considered important and relevant
Provided feedback in eDelphi Round 2: participants re-rated importance of outcomes based on their individual ratings and overall ratings *of each stakeholder group* from Round 1	Participants were provided their own ratings and the ratings of their own stakeholder group from Round 1, not of all stakeholder groups, to re-rate importance of outcomes	Due to a technical issue with the DelphiManager platform that was not within the SSC's control
Consensus in criteria in eDelphi: Round 1: ≥ *50%* of participants rated an outcome 7-9 Round 3: ≥ *70%* of participants rated an outcome 7-9	Round 1: ≥ 70% of participants rated an outcome 7-9 Round 3: ≥ 60% of participants rated an outcome 7-9	Round 1: burden for participants was reduced as less outcomes needed to be re-rated Round 3: aim was to agree on 5-6 core outcomes
Exclusion of participants from data analysis: N/S	Participants who rated all outcomes identically in one round were excluded from analysis	Participants most likely did not carefully consider the importance of each outcome individually or they did not incorporate the provided feedback from previous rounds
Invite to take part in consensus meetings: *random sample of 20% of participants* who expressed willingness to participate in consensus meeting at end of Delphi Round 3 was selected and invited via email	All participants who agreed to take part in a consensus meeting were invited via email	Given the smaller number of overall participants who completed Round 3 randomisation was not practical

Consensus meetings: voting options 'yes' and 'no'	Additional voting option 'unsure'	As it was recognised that particularly for people with OD in PD and family members/caregivers it was difficult to vote on OMIs and timepoints of measurement.
Follow-up survey: N/S	Anonymous survey with previous consensus meeting participants in which participants were asked whether they agreed (yes/no/unsure), conducted on Qualtrics, open for one week, reminder emails were sent after five days	As participants, particularly in second consensus meeting, suggested changes to definitions and OMIs, voting on agreement was then partly based on different definitions or OMIs compared to the first consensus meeting. This therefore made combining results of both meetings challenging.

COMET: Core Outcome Measures in Effectiveness Trials, N/S: not specified, OD: oropharyngeal dysphagia, OMI: outcome measurement instrument, PD: Parkinson's disease, SSC: study steering committee

7.3.2 Participants

One hundred thirteen participants registered via DelphiManager, 114 completed Round 1, 104 completed Round 2 and 90 completed Round 3. This represents a response rate of 91% between Rounds 1 and 2, and 87% between Rounds 2 and 3. Numbers and percentages of participants in each stakeholder group who dropped out are depicted in Appendix 7.7. One participant was excluded due to rating all outcomes in all rounds with a score of 9. The 90 participants were from 21 countries and represented all three stakeholder groups. Participants' demographics for each eDelphi round are displayed in Table 7.2.

Table 7.2: Characteristics of participants in each eDelphi round

	eDelphi Round	**1**	**2**	**3**
	Total number of participants	114	104	90
Stakeholder group, n (%)	Person with OD in PD	26 (22.8%)	22 (21.2%)	18 (20%)
	Family member/caregiver	9 (7.9%)	8 (7.7%)	7 (7.8%)
	HCP/Researcher	79 (69.3%)	74 (71.2%)	65 (72.2%)
Gender, n (%)	Male	31 (27.2%)	29 (27.9%)	27 (30%)
	Female	83 (72.8%)	75 (72.1%)	63 (70%)
Age group, n (%)	18-30	13 (11.4%)	13 (12.5%)	12 (13.3%)
	31-45	42 (36.8%)	40 (38.5%)	37 (41.1%)
	46-60	23 (20.2%)	20 (19.2%)	14 (15.6%)
	61-75	24 (21.1%)	20 (19.2%)	18 (20%)
	76-90	12 (10.5%)	11 (10.6%)	9 (10%)
Geographical area, n (%)	Asia	3 (2.6%)	2 (1.9%)	2 (2.2%)
	Europe	91 (79.8%)	84 (80.8%)	73 (81.1%)
	North America	11 (9.6%)	9 (8.7%)	8 (8.9%)
	South America	1 (0.9%)	1 (1%)	0 (0%)
	Oceania	6 (5.3%)	6 (5.8%)	5 (5.6%)
	Other	2 (1.8%)	2 (1.9%)	2 (2.2%)
Age at time of PD diagnosis in years, mean (range)		65.9 (41-88)	65.9 (41-88)	65.4 (41-88)
Time since PD diagnosis in years, mean (range)		9.2 (1-41)	8.9 (1-41)	8.6 (1-41)

HCP: healthcare professional, OD: oropharyngeal dysphagia, PD: Parkinson's disease

Of the 90 participants who completed Round 3, 50 agreed to participate in a consensus meeting. Of these, 35 attended one of the two subsequent consensus meetings (Table 7.3).

Table 7.3: Demographics of participants in each consensus meeting

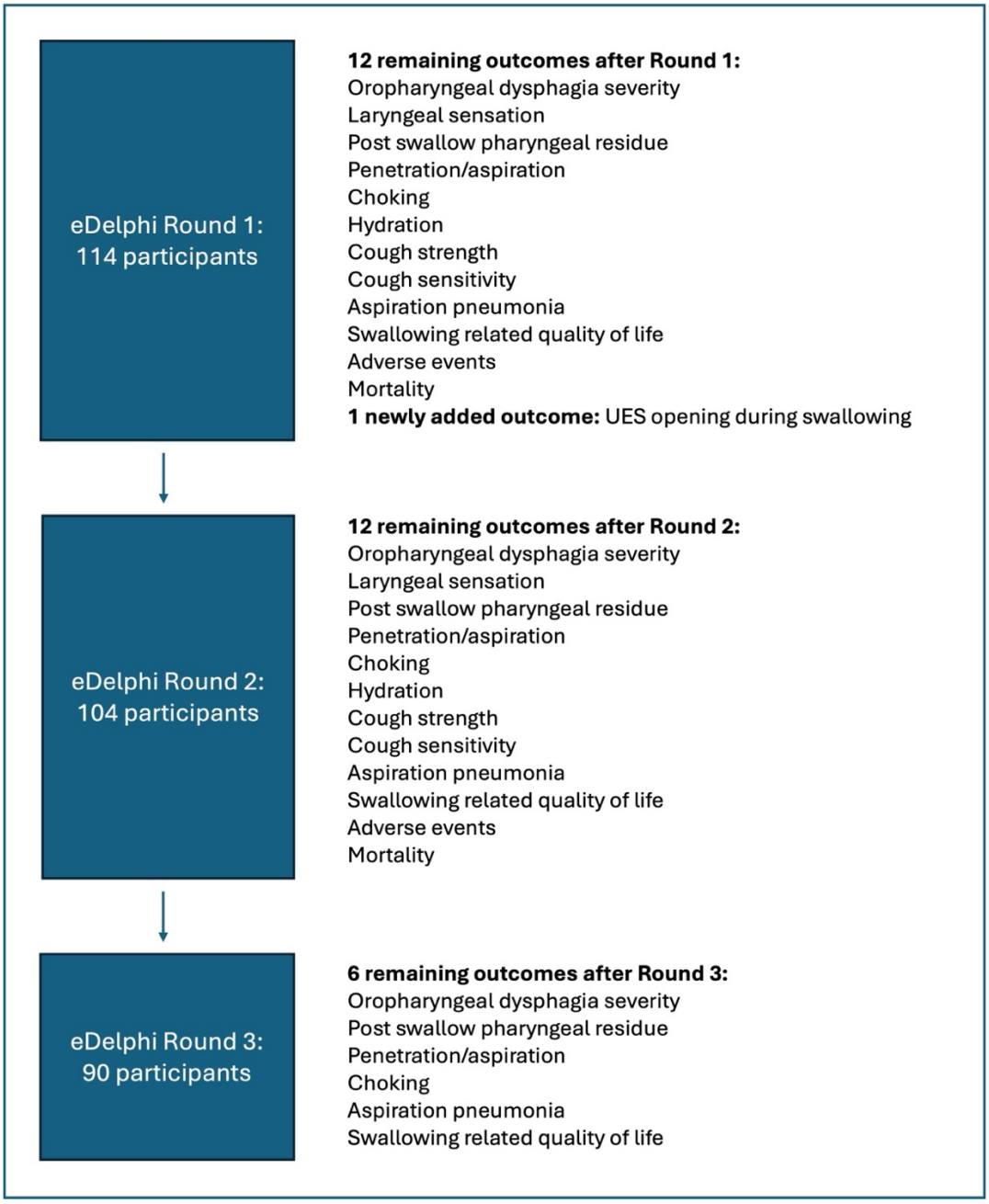
	Consensus meeting	**1**	**2**
	Total number of participants	21	14
Stakeholder group, n (%)	Person with OD in PD	4 (19%)	1 (7.1%)
	Family member/caregiver	2 (9.5%)	0 (0%)
	HCP/Researcher	15 (71.4%)	13 (92.9%)
Gender, n (%)	Male	5 (23.8%)	2 (14.3%)
	Female	16 (76.2%)	12 (85.7%)
Age group, n (%)	18-30	2 (9.5%)	3 (21.4%)
	31-45	11 (52.4%)	6 (42.9%)
	46-60	1 (4.8%)	4 (28.6%)
	61-75	7 (33.3%)	0 (0%)
	76-90	0 (0%)	1 (7.1%)
Geographical area, n (%)	Europe	14 (66.7%)	11 (78.6%)
	North America	1 (4.8%)	3 (21.4%)
	Oceania	4 (19%)	0 (0%)
	Other	2 (9.5%)	0 (0%)

HCP: healthcare professional, OD: oropharyngeal dysphagia, PD: Parkinson's disease

We invited all 35 consensus meeting participants to participate in the anonymous follow-up survey. Of these, 26 participants completed the survey. Sociodemographic data from survey participants was not collected to keep burden on respondents as low as possible.

7.3.3 Stage 1: outcome prioritisation process

Overall, participants chose 'unable to rate' (score of 10) in 4.6% of outcomes in Round 1, 2.4% in Round 2 and 1.6% in Round 3. Most participants did not provide a reason for choosing this score. Due to exclusion of these ratings, the overall number of participants' ratings differs between outcomes. Appendix 7.8 depicts the number of participants using a rating of 10 for each of the three rounds. In addition, we found that people with OD in PD and family members/caregivers requested more support via email to log in to DelphiManager and access the eDelphi rounds than healthcare professionals/researchers. Figure 7.2 outlines the prioritisation process through the three round eDelphi surveys with the numbers of participants per round and the retained outcomes per round.



eDelphi: online Delphi, UES: upper oesophageal sphincter

Figure 7.2: Outcome prioritisation process through three round eDelphi survey

Round 1 was open from January 10 to February 3, 2024, and 45 outcomes were included. Of these, 33 did not reach *consensus in* and were dropped after this round. No outcome was rated *critically important* (scores 7 to 9) by ≥ 70% of participants in each stakeholder group individually. The outcome 'laryngeal sensation' was rated *critically important* only by ≥ 70% of people with PD (n = 16, 76%). The outcomes rated most frequently *critically important* by family members/caregivers were 'choking' (n = 8, 89%) and 'penetration/aspiration' (n = 7, 88%). The outcomes rated most frequently *critically important* by

healthcare professionals/researchers were ‘penetration/aspiration’ (n = 78, 99%), ‘aspiration pneumonia’ (n = 72, 92%) and ‘OD severity’ (n = 72, 91%).

In total, participants provided 49 suggestions for additional outcomes in Round 1. Only ‘upper oesophageal sphincter (UES) opening during swallowing’ met the criterion for inclusion in the next round and was therefore added as an outcome to the remaining 12 outcomes from Round 1, resulting in 13 outcomes for Round 2.

Round 2 was open from February 14 to March 12, 2024. The newly added outcome ‘UES opening during swallowing’ did not reach *consensus in* and was dropped. All other outcomes were retained for Round 3. The outcomes ‘penetration/aspiration’ and ‘choking’ were rated as *critically important* by ≥ 70% of participants in each stakeholder group individually. To 70% (n = 14) of people with PD the outcome ‘mortality’ was also *critically important*. ‘OD severity’ (n = 7, 88%) and ‘laryngeal sensation’ (n = 5, 71%) were *critically important* to ≥ 70% of family members/caregivers. To healthcare professionals/researchers, all outcomes except for ‘UES opening during swallowing’ were *critically important* to ≥ 70%. Particularly ‘penetration/aspiration’ (n = 73, 99%), ‘aspiration pneumonia’ (n = 70, 96%) and ‘OD severity’ (n = 70, 95%) were rated most frequently as *critically important* by this stakeholder group.

Round 3 was open from March 27 to April 19, 2024. Of the 12 outcomes in Round 3, six outcomes were dropped due to not reaching *consensus in*. The six remaining outcomes were ‘penetration/aspiration’ (n = 75, 83%), ‘aspiration pneumonia’ (n = 65, 73%), ‘choking’ (n = 60, 67%), ‘OD severity’ (n = 57, 65%), ‘swallowing related quality of life’ (n = 56, 62%) and ‘post swallow pharyngeal residue’ (n = 54, 61%).

The outcomes ‘penetration/aspiration’ and ‘choking’ were rated as *critically important* by ≥ 60% of participants in all three stakeholder groups. No other outcome was rated as *critically important* by ≥ 60% of family members/caregivers, whereas the outcome ‘mortality’ was *critically important* to ≥ 60% of people with PD (n = 11, 65%). All six remaining outcomes were *critically important* to ≥ 60% of healthcare professionals/researchers. Appendix 7.9 provides all absolute numbers and percentages of 7 to 9 ratings for all three stakeholder groups individually and together. Appendix 7.10 comprises all outcomes and definitions that were uploaded to each round to DelphiManager.

7.3.4 Stage 2: Agreement on COS-DIP

We conducted two online consensus meetings in May 2024. All six core outcomes were agreed on by 82% of consensus meeting participants. Of the remaining participants, 9% voted ‘no’ or ‘unsure’ respectively. Definitions and OMIs were discussed within the consensus meetings and agreed on by participants. An overview of the definitions and OMIs presented for discussion by the SSC and the suggested changes made by consensus meeting participants is provided in Appendix 7.11.

Timepoints for measurement suggested by the SSC in the consensus meetings were as a minimum immediately before the start of the intervention, immediately after and 3-months after completion of intervention. Additionally, measurements 1-month before and 6- and 12-months after completion of the intervention were recommended. In the first consensus meeting, 83% (n = 15) of participants agreed on these timepoints of measurement. In the second consensus meeting, it was suggested to only have a pre and post assessment as a minimum and change the term ‘recommended’ to ‘advisable’. This was agreed on by 69% (n = 9) participants in this meeting. After discussion within the SSC, we amended the wording as suggested in the second meeting. However, as the absolute number of participants in the first meeting was higher than in the second meeting, no changes for the timepoints of measurement were made.

For the follow-up survey, we incorporated the suggested changes by consensus meeting participants into the definition of choking and OMIs for ‘penetration/aspiration’, ‘OD severity’ and ‘post swallow pharyngeal residue’. Agreement on six core outcomes, definitions, OMIs and timepoints of measurement was achieved. Table 7.4 depicts the agreed core outcomes, definitions and OMIs.

Table 7.4: Core outcomes with definitions and measurement(s) agreed on by consensus meeting and follow-up survey participants

Outcome	Definition	Suggested outcome measurement instrument(s)	Agreement
Penetration/aspiration	**Penetration:** Entry of material (e.g. food, fluids or saliva/secretions) into the larynx that does not pass below the vocal folds. **Aspiration:** Entry of material (e.g. food, fluids or saliva/secretions) into the larynx and below the level of the vocal folds.	**VFS:** e.g. Penetration-Aspiration Scale (Rosenbek et al., 1996), DIGEST safety rating (Hutcheson et al., 2017, 2022) **FEES:** e.g. Penetration-aspiration events (Warnecke et al., 2016), VASES safety rating (Curtis et al., 2022), DIGEST-FEES safety rating (Labeit et al., 2023)	**Definition:** 91% agreement in consensus meetings **Measurements:** 81% agreement in survey
Aspiration pneumonia	An acute respiratory infection likely caused by aspiration of material (e.g. food, fluids or saliva/secretions).	A clinical suspicion of a new or worsening respiratory infection in the context of at least 2 of the following: temperature <35°C or >38°C; abnormal chest x-ray (CT scan); a white cell count of <4x109/l or >11x109/l or purulent (tracheal) secretions.	**Definition and measurement:** 71% agreement in consensus meetings
Choking	A condition that occurs when breathing is perceived to be impeded through the inhalation or ingestion of material (e.g. food, fluids or saliva/secretions), which is felt as an obstruction of the respiratory tract.	e.g. report the number of choking events over or within a specific time period	**Definition:** 77% agreement in survey **Measurement:** 70% agreement in consensus meetings
Oropharyngeal dysphagia severity	The overall degree of impairment (i.e. swallowing safety and swallowing efficiency) in swallowing material (e.g. food, liquids or saliva/secretions).	**VFS:** e.g. overall DIGEST (Hutcheson et al., 2017, 2022), MBSImP (Martin-Harris et al., 2008) **FEES:** e.g. overall DIGEST-FEES score (Labeit et al., 2023), Endoscopic PD dysphagia score (Warnecke et al., 2016)	**Definition:** 86% agreement in consensus meetings **Measurements:** 92% agreement in survey
Swallowing related quality of life	An individual's perception of their ability to swallow within the context of their daily life, considering the cultural norms, values, and personal goals that influence their experience.	e.g. SWAL-QOL (McHorney et al., 2002)	**Definition and measurement:** 91% agreement in consensus meetings
Post swallow pharyngeal residue	The presence of material (e.g. food, fluids or saliva/secretions) remaining in the pharynx after swallowing.	**VFS:** e.g. Bolus Residue Scale (Rommel et al., 2015), DIGEST efficiency rating (Hutcheson et al., 2017, 2022) **FEES:** e.g. Yale Pharyngeal Residue Severity Rating Scale (Neubauer et al., 2015), VASES efficiency rating (Curtis et al., 2022), DIGEST-FEES efficiency rating (Labeit et al., 2023), Residue Scale (Warnecke et al., 2016)	**Definition:** 94% agreement in consensus meetings **Measurements:** 77% agreement in survey

DIGEST: Dynamic Imaging Grade of Swallowing Toxicity, FEES: Fibreoptic Endoscopic Evaluation of Swallowing, MBSImP: Modified Barium Swallow Impairment Profile, OMI: outcome measurement instrument, VASES: Visual Analysis of Swallowing Efficiency and Safety, VFS: Videofluoroscopy

The agreed 'minimum' timepoints of measurement are before the start and at the end of intervention and 3-months post completion of intervention. The agreed 'advisable', additional timepoints of measurement are 1-month before the start and 6- and 12-months post completion of intervention.

7.4 Discussion

In this study we developed a COS for OD interventions in people with PD. Consensus was reached on six core outcomes. Definitions, OMIs and timepoints of measurement were established and agreed for all six core outcomes.

7.4.1 Core outcomes

The six core outcomes agreed in this study were largely consistent with previous research. Of these, 'penetration/aspiration', 'OD severity', 'swallowing related quality of life', and 'post swallow pharyngeal residue' are outcomes previously considered important by all key stakeholder groups based on surveys (chapters 5 and 6) and are frequently measured by researchers in OD in PD studies (chapter 4). 'Penetration/aspiration' is the outcome that is clearly considered the most important outcome in OD intervention studies in PD by all three stakeholder groups and shows the largest agreement.

7.4.2 OMIs

There is a huge variety of OMIs to measure OD-related outcomes. However, some are not validated for PD specifically or are overly long and not user friendly (e.g. SWAL-QOL (McHorney et al., 2002)). Further, there is a lack of agreement on diagnostic criteria for pneumonia (Ferguson et al., 2018; Yoshimatsu et al., 2022). The suggested measurement for pneumonia was agreed on by consensus meeting participants in both COS studies on intensive care (Duncan, 2022) and this one for OD in PD as well as criteria suggested by Robbins et al. (2008).

In published research studies in OD in PD, 'penetration/aspiration' and 'swallowing related quality of life' were the outcomes measured most consistently using the PAS (Rosenbek et al., 1996) and SWAL-QOL (McHorney et al., 2002), respectively. In contrast, 'OD severity' is being measured differently (chapter 4), which needs to be addressed in future research. Scales used with instrumental assessment, such as Visual Analysis of Swallowing Efficiency and Safety (VASES) (Curtis et al., 2022) and Dynamic Imaging Grade of Swallowing Toxicity for FEES (DIGEST-FEES) (Labeit et al., 2023), have only been

developed or validated for PD after completion of the scoping review (chapter 4) and are therefore not yet used in the included OD intervention studies in PD. However, due to their established validity and comprehensive outcome assessment, we believe that they are likely to be useful OMIs in future clinical trials. The measurement of 'choking' poses a challenge as no validated OMI exists so far. It is, therefore, essential to develop a valid and reliable OMI for choking.

7.4.3 Timepoints of measurement

In a neurodegenerative condition like PD, long-term follow-up assessments are critical. The longest follow-up assessment in studies included in the scoping review was 6-months after completion of the intervention (chapter 4). Considering the core outcome 'aspiration pneumonia' and its delayed potential development of post aspiration events, longer-term follow-ups, as advised in the COS-DIP, are needed. The explicit specification of timepoints of measurement in this COS provides crucial guidance for future trials, further enhancing the comparability of results across studies and enabling more meaningful meta-analyses.

7.4.4 Challenges in COS research

It is important that a COS is widely adopted; otherwise it will add to research waste rather than facilitate its reduction (Khalil et al., 2022). In a systematic review on the uptake of the COS for PD by de Roos et al. (2017) in RCTs, the problems of multiple outcomes not being measured and a high variety of OMIs used were identified (Paul et al., 2023). This is in line with other studies that have identified barriers to the uptake of COS (section 1.4). Ongoing dissemination of the COS-DIP to raise awareness among clinical trialists, funders and people with the health condition is key to its adoption and integration into clinical trials. Additionally, the COS-DIP should be seen as a living document, that needs to be revised and updated once new information (e.g. experience with using the COS in research or new, validated OMIs) emerges.

7.4.5 Strengths and limitations

A strength of this study is the novel approach to developing the long list of outcomes by conducting online surveys with people with OD in PD, family members/caregivers and dysphagia clinicians. This ensured that all perspectives were included in the development of the long list of outcomes prior to the

eDelphi survey. An additional strength is the inclusion of key stakeholders from previously underserved groups, with a large geographical coverage, representing a wide range of different perspectives, making it more relevant to all stakeholder groups. This is despite the study being in English and online only. Furthermore, this COS achieved consensus on the OMIs and timepoints of measurement for the core outcomes. While many COS studies focus primarily on *what* to measure, this COS also addresses *how* and *when* to measure outcomes, providing comprehensive guidance for future clinical trials in this field.

One limitation of this study was the relatively small number of people with OD in PD and family members/caregivers compared to healthcare professionals/researchers. Due to the small sample size, the ratings per eDelphi round need to be interpreted with caution. One potential reason for this smaller sample size is both the online format as well as the use of the DelphiManager platform that might have posed challenges to these two groups. This became evident in the increased number of email requests for assistance from the research team. For future COS studies planning on using DelphiManager, it would be advisable to have the option of bigger font for people with visual impairment, to have adapted website formats to different devices (computer, tablet and smartphone) and to improve the log in process to the website. This would reduce burden of participants and potentially increase participation and retention rates.

Despite these challenges, proportionately more people with the health condition and family members/caregivers were included in this study compared to other COS studies in dysphagia. In the COS for patient reported dysphagia in head and neck cancer clinical trials, nine of the 45 participants were people with the health condition and family members (Manduchi et al., 2024). The COS for dysphagia intervention outcomes in intensive care included a total of 160 participants, with 18 people with the health condition and no family members/caregivers (Duncan, 2022).

Participant attrition between Delphi rounds is a common challenge that requires careful consideration (Williamson et al., 2017). Although we achieved response rates of 91% and 87% between Rounds 1 and 2, respectively, those who did not complete all rounds or submitted incomplete responses may have held different views. This potential selection bias could influence the final consensus if participants with certain perspectives were more likely to drop out. However, our analysis showed there was no particular stakeholder group that had a significantly higher attrition rate between the two rounds than any other

stakeholder group. Additionally, the strong consensus achieved in the final COS-DIP suggests that any impact of participant attrition was likely minimal.

7.5 Summary

This COS for OD interventions in PD was developed by key stakeholder groups, including people living with the health condition, and consists of six core outcomes. These comprise ‘penetration/aspiration’, ‘aspiration pneumonia’, ‘choking’, ‘OD severity’, ‘swallowing related quality of life’ and ‘post swallow pharyngeal residue’.

Chapter 8: Discussion

This chapter first synthesises the research process by examining the key findings of the studies conducted and then provides an interpretation of the findings in relation to the wider literature. This is followed by an analysis of the inclusion of people with OD in PD in the research process. The strengths and limitations that may affect the overall COS-DIP are then discussed. Building on this, the chapter then considers the uptake of the COS-DIP, implications for future research and clinical practice, developments in the wider field, and concludes with final reflections on this thesis.

8.1 Summary of key study findings

The aim of this study was to develop an agreed standardised COS for clinical trials of OD interventions in PD. Two COS for PD in general (de Roos et al., 2017; Sjödahl Hammarlund et al., 2012) and two COS in the field of OD interventions already existed (Duncan, 2022; Manduchi et al., 2024), but none specifically for OD in PD. This is despite the fact that PD is the fastest growing neurological condition in the world (Dorsey et al., 2018; Feigin et al., 2019) and OD can affect up to 80% of people with PD (Kalf et al., 2012). OD can have a significant impact on a person's quality of life and can lead to malnutrition, dehydration and pneumonia (Groher & Crary, 2016). In people with PD, pneumonia is a leading cause of death (Fernandez & Lapane, 2002; Hobson & Meara, 2018). Thus, as the prevalence of PD increases, more people will be living with OD in PD, driving the need for a COS for OD intervention studies in PD to facilitate the ability to generate high-quality treatment evidence.

There are no standardised methods for developing a COS, but a variety of methods have been used by COS developers and are described in COS development guidelines, such as the COMET handbook (Williamson et al., 2017). Chapter 2 described common principles and procedures for developing a COS, typically including three main steps. The first step is the preparatory phase. In this phase, the scope of the COS should be defined and if no available COS exists, the study protocol should be devised and published. The study should also be registered, e.g. on the COMET database. The next step is to develop a long list of outcomes, including key stakeholder perspectives on important outcomes, and to reach consensus on the most important core outcomes, most often using a Delphi survey. Having agreed on what to measure, the next step is to agree on the OMIs that will be used to measure these core outcomes.

Completing all three steps and following a rigorous methodology ensures that the COS will be of good quality, which then increases the likelihood that it will be used in future clinical trials.

Chapter 3 reported on the study protocol for developing the COS-DIP. The study was designed and reported following established guidelines highlighted in the previous chapter 2. All 13 elements that are required as a minimum by the COS-STAP guideline were met to ensure optimal study design and transparency (Kirkham et al., 2019).

The next step was to develop the long list of outcomes, which was initiated in chapter 4. First, we conducted a scoping review to identify outcomes that are currently measured in published OD intervention studies in PD. This choice of outcomes represents researchers' views on what is important to measure in these studies. In addition to the outcomes, we also extracted the outcome definitions used, OMIs used and the timepoints at which the outcomes were measured. In total, we identified 39 outcomes, with 'penetration/aspiration', 'OD severity' and 'swallowing related quality of life' being the most commonly measured outcomes in the included 19 studies. However, we also identified omitted outcomes suggesting selective outcome reporting bias in some of the included studies. As with the outcomes, the definitions, OMIs and timepoints of measurement varied widely across the included studies. 'Penetration/aspiration' and 'swallowing related quality of life' were most consistently measured using the PAS (Rosenbek et al., 1996) and SWAL-QOL (McHorney et al., 2002), respectively. In contrast, 'OD severity' was measured most differently. Outcomes were most commonly measured before and immediately after the intervention, with some studies conducting follow-up assessments up to 6 months after completion of the intervention.

To further develop the long list of outcomes, we then aimed to identify OD intervention outcomes in PD that are important to people with OD in PD and family members/caregivers through an online survey. To validate the survey findings and clarify inconclusive results, we conducted focus groups with seven previous survey participants. This study is reported in chapter 5. In total, twelve outcomes were considered important by ≥ 80% of survey respondents. The two outcomes considered important most often were 'penetration/aspiration' and 'laryngeal sensation'. The outcome 'fear of choking' was identified by participants as important but missing in the long list of outcomes. Survey results were validated by focus group participants. Therefore, 'fear of choking' was added to the long list of outcomes.

In chapter 6, an online survey with dysphagia clinicians' perspectives on important OD intervention outcomes in PD is presented. The results of this study completed the long list of outcomes with the views of this stakeholder group. 'Penetration/aspiration' and 'swallowing related quality of life' were the outcomes rated important most frequently by dysphagia clinicians. Further, more than 90% of dysphagia clinicians also considered outcomes like 'avoidance of food/liquids', 'pneumonia' and 'level of oral intake' as important. Five new outcomes ('independent eating/drinking', 'social aspect of eating/drinking', 'mealtime duration', 'respiratory swallow coordination' and 'hydration') were identified as missing but important by survey respondents and added to the long list of outcomes. OMIs used most commonly in clinical practice by dysphagia clinicians to measure OD intervention outcomes were PAS (Rosenbek et al., 1996), FOIS (Crary et al., 2005) and SWAL-QOL (McHorney et al., 2002). Most participants suggested a need to measure outcomes at least before and after completion of the intervention. Follow-up assessments were considered important up to 24 months post completion of the intervention.

The last study on agreeing the core outcomes, definitions, OMIs and timepoints of measurement for the COS-DIP is reported in chapter 7. We conducted an international three round eDelphi survey and two subsequent online consensus meetings with all key stakeholders. Six core outcomes were agreed on: 'penetration/aspiration', 'aspiration pneumonia', 'choking', 'OD severity', 'swallowing related quality of life' and 'post swallow pharyngeal residue'. Consensus was achieved on suggestions for OMIs for each core outcome. Timepoints for measurement were agreed to be a minimum before the start and immediately after and 3-months post completion of intervention. In addition, advisable timepoints of measurement were agreed to be 1-month before the start and 6- and 12-months post completion of the intervention.

8.2 Interpretation of study findings

8.2.1 Development of long list of outcomes: scoping reviews, surveys and focus groups

Most researchers develop the questionnaire for the Delphi study based on the available literature and in some cases also based on qualitative interviews or focus groups (Niederberger & Spranger, 2020). This study employed a novel approach in developing the long list of outcomes. I am not aware of any other COS study that has collected the perspectives of four key stakeholder groups through scoping reviews

including all types of intervention studies (not only RCTs), online surveys, and focus groups to devise the long list of outcomes. This approach allowed for a thorough assessment of perspectives to inform a comprehensive long list of outcomes. At the same time, this approach was more time and resource consuming, and it may be questioned whether this added valuable information and, if so, what this information was. Remus et al. (2021) acknowledge the potential value of, for example, qualitative interviews with people living with the health condition to develop the long list of outcomes. They also critically reflect on the increased workload associated with it, while clear evidence for the benefits is still lacking. In their study, they found that none of the outcomes derived from interviews with people with the health condition were later included in their COS or had a direct impact on it. They hypothesise that collecting perspectives of people with the health condition during the development phase of the long list of outcomes may be duplicate effort if they are included as a stakeholder group in the Delphi survey and consensus meetings (Remus et al., 2021).

A comparison of the outcomes identified in the scoping review in chapter 4 with the six core outcomes included in the COS-DIP in chapter 7 shows that all core outcomes except for 'choking' were already included in this first version of the long list of outcomes. The outcome 'choking' was added by the SSC to this first version of the long list of outcomes. Thus, all core outcomes were derived through the scoping review in chapter 4 and the SSC. Based on this, it could be argued that the second scoping review with intervention studies without control groups (Hirschwald et al., 2023), and the online surveys and focus groups with people with OD in PD, family members/caregivers in chapter 5 and dysphagia clinicians in chapter 6 did not strongly advance our understanding at that point in time.

While it may not be feasible to follow this approach in every COS development, I believe that valuable and more in-depth information for the COS and in addition to the COS was obtained through these studies. This is discussed in the following paragraphs.

Scoping review. The second scoping review included all OD intervention studies in PD without control groups and added three new, additional outcomes (Hirschwald et al., 2023). Of these, 'cough strength' was rated as *critically important* by 86% and 90% of healthcare professionals/researchers in Round 1 and 2 in the Delphi survey, respectively. This highlights the importance of 'cough strength' to this stakeholder group. One reason for this may be that healthcare professionals/researchers more likely acknowledge the clinical significance of coughing as a protective mechanism of the airway while people with OD in PD

and family members/caregivers may not be aware of the relationship between coughing and swallowing. However, it is interesting to note that in the scoping review in chapter 4, three studies were included that assessed the effects of EMST on swallowing function (Byeon, 2016; Claus et al., 2021; Troche et al., 2010). None of these studies measured cough related outcomes in addition to swallowing related outcomes, although EMST specifically focuses on strengthening the expiratory muscles and only indirectly affects swallowing function. Even though the scoping review only included trials that aimed to improve swallowing or feeding difficulties, this does not fully explain why these trials did not also measure cough related outcomes when assessing the effectiveness of a cough specific intervention. It is possible that there are differing opinions between researchers and healthcare professionals that are not reflected in the eDelphi ratings as these stakeholders were grouped together, or that the perceived importance of measuring 'cough strength' in OD interventions in PD is simply not represented in the included studies.

One of the author groups of the EMST trials recently conducted another RCT on the effectiveness of EMST. In this study, the researchers only measured cough related and no swallowing related outcomes (Troche et al., 2023). This discrepancy is similar to what was described in the scoping review in chapter 4. There we discussed that only one of the included studies assessed voice and speech outcomes, although the literature suggests that there is a cross-system effect in that intensive voice and speech treatment may also improve swallowing. This is also true for intensive expiratory muscle training, e.g. through EMST, which affects swallowing, even though it is not a swallowing specific intervention. The lack of measurement of these additional outcomes may lead to an incomplete understanding of the full therapeutic potential of interventions such as LSVT LOUD and EMST.

Study steering committee (SSC). Through discussion within the SSC, we added ten additional outcomes to the long list of outcomes. All outcomes except for 'choking' and 'cough sensitivity' were dropped after Round 1. While 'choking' became one of the six core outcomes, 'cough sensitivity' was dropped after Round 3. This outcome was considered *critically important* by 78% and 86% of healthcare professionals/researchers in Rounds 1 and 2, respectively. 'Cough strength' and 'cough sensitivity' are closely intertwined, and therefore the reasons for the critical importance for one group and the lower importance for the other two groups are most likely the same for 'cough sensitivity' as for 'cough strength'. The decrease in healthcare professionals/researchers rating these two outcomes as *critically*

important in Round 3 may be due to the change in instructions (to rate only five to six outcomes as *critically important*) in this round. Thus, for this stakeholder group, 'cough strength' and 'cough sensitivity' may be important outcomes, but not core outcomes to measure in OD intervention trials in PD.

Survey and focus groups with people with OD in PD and family members/caregivers. 'Fear of choking' emerged to be of particular importance to family members/caregivers. Based on the results of the online survey and focus groups with people with OD in PD and family members/caregivers in chapter 5, 'fear of choking' was added to the long list of outcomes. In Round 1 of the Delphi survey, this outcome was rated as *critically important* by 78% of family members/caregivers but was dropped as in total only 62% of all participants rated it as *critically important*. This may be due to family members/caregivers witnessing choking episodes first hand but may feel less control over the situation than other stakeholder groups. Healthcare professionals and researchers may also consider this outcome to be one of multiple aspects that can affect the outcome 'swallowing related quality of life'. For example, the SWAL-QOL (McHorney et al., 2002) comprises the questions "I fear I may start choking when I eat food" and "I am afraid of choking when I drink liquids" and therefore, may indirectly measure this outcome.

Survey with dysphagia clinicians. 'Hydration' proved to be an outcome particularly important to dysphagia clinicians. In the online survey with dysphagia clinicians in chapter 6, five additional outcomes were suggested and added to the long list of outcomes. Of these, all outcomes except for 'hydration' were dropped after Round 1 as they did not meet the *consensus in* criterion. 'Hydration' was maintained until Round 3 due to being of critical importance to 84% and 92% of healthcare professionals/researchers in Rounds 1 and 2, respectively. Dehydration poses a physiological risk caused by OD and is well described as such in the literature (Scorza et al., 2022; Wirth et al., 2016). Consequently, its relevance to this stakeholder group is to be expected. However, another common physiological risk due to OD in PD is malnutrition (Kacprzyk et al., 2022; Wirth et al., 2016). It is surprising that 'nutrition' was not suggested as an important but missing outcome at any stage of the research process. One potential explanation may be that SLTs perceive their responsibilities to be more in functional aspects of swallowing and immediate safety concerns (i.e. hydration and aspiration) and believe that nutrition is part of a dietician's responsibility. This would suggest that healthcare professionals in the Delphi survey were more likely to be represented by SLTs than dieticians. Unfortunately, the data do not provide information on the

professions of the eDelphi participants. Another reason may be that dehydration typically develops more rapidly than malnutrition and is therefore considered more relevant. Alternatively, the outcome 'level of oral intake', which was included in the long list of outcomes, may have been regarded as a proxy measure for nutrition and therefore, participants did not feel the need to add 'nutrition'. Finally, even very comprehensive approaches such as this one, can have blind spots and 'nutrition' may simply have been overlooked by both the SSC and the participants and was therefore not added.

In contrast, the outcome 'self-perception of swallowing' was measured more frequently in the studies included in the scoping review (in 5 of the 19 studies) in chapter 4 and was rated as *important* by the majority of participants in all stakeholder groups in the surveys in chapters 5 and 6 (people with OD in PD: 92%, family members/caregivers: 87%, dysphagia clinicians: 91%). However, in Round 1 of the Delphi survey in chapter 7, less than 70% of participants in all stakeholder groups individually and combined rated this outcome as *critically important*. While this was not anticipated, one explanation may be that the use of a 9-point scale in the eDelphi survey allows for better discrimination between generally important and critically important (core) outcomes than the 2-point scale used in the online surveys.

In addition, the outcome 'self-perception of swallowing' may be perceived of greater importance in clinical practice than in research studies. Both patient-reported perceptions and outcomes are important in designing patient-centred treatment plans (Francis et al., 2017; Watts & Zhang, 2022). Alternatively, participants may have felt that the patient perspective was already incorporated through the outcome 'swallowing related quality of life'. The SWAL-QOL (McHorney et al., 2002) also assesses the self-perceived frequency of OD symptoms, although it is a PROM designed to assess swallowing related quality of life.

Overall, an advantage of this approach to developing the long list of outcomes was that only one additional outcome was identified as important but missing in the long list of outcomes in Delphi Round 1 and was added in Round 2, with the outcome being dropped immediately after Round 2. This indicates that we had successfully captured almost all relevant outcomes in the long list of outcomes and suggests good content validity. Having a nearly complete long list of outcomes from the start can make the Delphi process more efficient. It allows Delphi participants to focus on prioritising rather than identifying new outcomes. It may also have helped to maintain participants' engagement during the Delphi, as the long list of outcomes did not lengthen, but became significantly shorter between rounds. This highlights a

strong advantage of a comprehensive approach including all key stakeholders' perspectives in developing the long list of outcomes. While some of the additional outcomes identified were mainly important for one stakeholder group and therefore did not qualify as a core outcome, they may still be important for specific research questions and individual stakeholder groups.

8.2.2 Agreeing on what, how and when to measure: Delphi survey and consensus meetings

In addition to the novel method adopted in developing the long list of outcomes, we also took a new approach to agreeing *what*, *how* and *when* to measure outcomes. Most previous COS studies have focused on agreeing *what* to measure. This is also reflected in the COMET handbook, which primarily provides guidance on this stage (Williamson et al., 2017). Where agreement is sought on *how* to measure core outcomes, this is often done in a separate study. Further, COS developers rarely seek consensus on *when* to measure the core outcomes. I am not aware of any COS study that has combined the *what*, *how* and *when* stages in one study through conducting a Delphi survey and subsequent consensus meetings. While this approach was less time-consuming and resource-intensive than conducting multiple Delphi surveys and consensus meetings, the question is whether it was as reliable and rigorous.

Delphi survey. The aim of the Delphi survey presented in chapter 7 was to reduce the long list of outcomes to the core outcomes. There are two important methodological factors that can impact which outcomes will be agreed on as core outcomes (Lange et al., 2020). First, the rating scale that is being used. Although there is conflicting evidence, it is acknowledged that the results differ depending on whether a 5-point or a 9-point Likert scale is being used (Lange et al., 2020; Remus et al., 2021). Having identified DelphiManager as the best platform available for conducting eDelphi studies to develop a COS for this study, the question of whether to use a 5-point or 9-point scale did not arise, as the 9-point scale is preset by DelphiManager and cannot be changed to a 5-point scale (Crew & Williamson, n.d.).

The other factor that can influence the results of a Delphi survey is the consensus criteria used (Lange et al., 2020). In the study protocol we specified to apply a more inclusive *consensus in* criterion after Round 1 (≥ 50% of participants rated an outcome 7 to 9) and a stricter criterion after Round 2 (≥ 70% of participants rated an outcome 7 to 9). While our long list of outcomes was not overly long compared to other COS studies, which initially had up to 70 outcomes (e.g. Mitchell et al., 2024), we were surprised to find that only eight of the 45 outcomes would have been excluded after Round 1 using the 50%

criterion. Through discussion within the SSC, we decided that minimising the burden for participants in Round 2 by applying a stricter *consensus in* criterion and thereby excluding more outcomes was our priority. This resulted in 33 of the 45 outcomes being excluded after Round 1. While we achieved what we set out to do, there is a risk in that we dropped outcomes too early. If outcomes are excluded between Delphi rounds, particularly after Round 1, certain excluded outcomes may have been critically important to some stakeholders but not to all. This in turn prevents other stakeholders from reassessing the importance of the outcomes based on feedback from the previous round (Williamson et al., 2017). As it has been shown that provided feedback from previous rounds influences participants' ratings (Barrios et al., 2021), this may have changed the final COS. This dilemma underscores the need for more research and subsequently clear evidence-based guidance on the most effective consensus criteria for Delphi surveys. The following section discusses outcomes that may have been dropped too early, based on individual stakeholder group ratings in the Delphi survey.

No outcomes rated as *critically important* by ≥ 70% of people with OD in PD were dropped after Round 1. In contrast, 'drooling', 'salivary pooling', 'swallowing related hyoid bone movement' and 'fear of choking' were rated as *critically important* by ≥ 70% of family members/caregivers in Round 1, but did not meet the *consensus in* criterion and were therefore dropped. No other stakeholder group rated outcomes related to saliva management as *critically important*. These outcomes are externally visible signs and family members/caregivers may be more aware of the potential social impact and stigma. Conversely, individuals with OD in PD may be less aware of these symptoms due to sensorimotor deficits associated with PD. 'Fear of choking' was of critical importance to 78% of family members/caregivers and for the reasons stated in the previous section this may have been an outcome that was dropped too early along with 'drooling' and 'salivary pooling'.

Ten of the outcomes dropped after Round 1 were rated as *critically important* by ≥ 70% of healthcare professionals/researchers in Round 1. These comprise 'orolingual bolus control', 'orolingual bolus transport', 'initiation of pharyngeal swallow' and 'post swallow pharyngeal pooling', 'medication intake', 'weight loss', 'level of oral intake', 'respiratory swallow coordination', 'participant's adherence to intervention' and 'hospitalisation'. The most striking outcome here is 'level of oral intake' as the FOIS (Crary et al., 2005), which measures this outcome, was among the OMIs used most frequently by

dysphagia clinicians in clinical practice in the survey in chapter 6. Therefore, there is a risk for having dropped this outcome too soon.

In summary, there is a risk that some outcomes were dropped too early. Two alternative options to prevent dropping outcomes too soon while keeping participant burden as low as possible may be considered for future COS studies. The first option would be to adapt the *consensus in* criterion, for example ≥ 60% of all participants rating an outcome as *critically important*. Niederberger and Spranger (2020) found that a *consensus in* criterion of 60% or higher is most frequently used in Delphi studies. In our study, 60% would be a compromise between 50% and 70%. By using this criterion, we would have maintained 14 additional outcomes but not all of the above listed outcomes that were of critical importance to one stakeholder group would have been included. Thus, particularly smaller stakeholder groups may still be disadvantaged.

Many COS studies use the *consensus in* criterion of < 15% of participants rating an outcome as *not important* (scores 1 to 3) in addition to the ≥ 70% criterion as also recommended in the COMET handbook (Williamson et al., 2017). This aims to balance majority support of an outcome with minority opposition against an outcome. However, this only works in one direction. If the minority favours an outcome but the majority does not, it may still be excluded, and the minority group is still being disadvantaged.

The alternative option is an iterative approach. In addition to retaining outcomes that were rated as *critically important* by ≥ 70% of all participants, group specific *consensus in* criteria can be applied. For example, outcomes that were rated as *critically important* by ≥ 70% of at least one stakeholder group could be maintained in addition. This criterion considers the findings from Barrios et al. (2021) that perspectives may shift towards those where strong consensus (≥ 70%) was achieved in one group. Mitchell and colleagues (2024) have defined this criterion as *consensus unclear*. This approach would provide the opportunity for smaller stakeholder groups to maintain outcomes of critical importance to them and allow the other participants to re-assess the importance of these outcomes based on the feedback provided in the next round.

Using the ≥ 70% *consensus in* criterion after Round 3, only two outcomes would have remained, and ten outcomes would have been dropped. One possible reason for this may be that, for participants in Round 3, our instructions specified that only five to six outcomes should be rated as *critically important*. This may have resulted in fewer outcomes being rated as *critically important* overall in Round 3. To not

exclude outcomes that may otherwise have been considered as *critically important*, we lowered the threshold to ≥ 60% resulting in the inclusion of six core outcomes. Applying the iterative consensus criteria approach described above would have changed the core outcomes, resulting in only five core outcomes. 'OD severity' would have been dropped, as 69% of healthcare professionals/researchers rated this outcome as *critically important*, missing the *consensus in* criterion by 1%. Exclusion of this outcome may be viewed critical if only 1% (n=1) is missing. In such an inconclusive case, it may have been an option to discuss the outcome 'OD severity' in the subsequent consensus meetings and seek participants' perspectives on whether or not to include it as a core outcome.

Overall, this highlights how differences in consensus criteria can affect the final COS. For this reason, transparent reporting of the criteria used is crucial, especially when criteria are changed during the course of the study. We have both published the study protocol (chapter 3) and provided a detailed explanation on our deviations from the study protocol in our report on the COS-DIP (chapter 7). Therefore, I believe that the process used during the Delphi survey is transparent and reliable. At the same time, this shows that it may not be ideal to have one set of pre-defined criteria, but rather that COS developers must adapt the criteria based on the composition of their stakeholder groups, the sample size and the individual results of each Delphi round. Although this carries a risk of bias, I believe that as long as the choices are transparent and justified, this may be a better alternative in the absence of clear, evidence-based guidelines.

Consensus meetings. We originally intended to seek consensus on the definitions of the core outcomes from all Delphi participants at the end of Round 3. We were kindly informed by the COMET team that this was not feasible and so we removed it from the study protocol. As a result, the only possibility of achieving consensus on definitions in this study was through the consensus meetings, together with the OMIs and timepoints of measurement. This increased the amount of consensus that we sought in these meetings, and 90 minutes per meeting may have been too short.

While this approach was very efficient, we faced two main challenges within these meetings. First, conducting two meetings allowed us to accommodate people with different (work) schedules and people from different time zones. On the other hand, based on participants' suggestions to change definitions or OMIs in the second consensus meeting, participants in that meeting voted on slightly different definitions and OMIs than those in the first meeting. Thus, the final COS-DIP would have been agreed by only half

of the consensus meeting participants. We therefore had to combine the results and seek consensus from participants on the combined results to maintain validity. The best available option in this case was to conduct a short follow-up survey. This is an unusual approach to the development of a COS and carries the risk of participants dropping out in between, but it may be generally necessary if consensus is to be reached not only on the core outcomes, but also on their definitions, OMIs and timepoints of measurement.

The second challenge was that while we also discussed the core outcomes, most of the time was spent discussing OMIs. For people with OD in PD and family members/caregivers, this was most likely a discussion they could not participate in the way that healthcare professionals and particularly researchers did. This may have decreased input from these stakeholder groups. One strength in our meetings was that we had participants who had already either self-developed or validated an OMI measuring one of the core outcomes and therefore added valuable knowledge to the discussion.

Reflecting on the consensus meetings, they seemed to provide an efficient approach in seeking consensus on definitions, OMIs and timepoints of measurement. To further improve this process, more time should be accounted for each meeting. Potentially 2 to 2.5 hours with a break in between may be better suited. As recommended by Gorst et al. (2021), it may be helpful to send the core outcomes, suggested definitions, OMIs and timepoints of measurement including lay terms to consensus meeting participants ahead of the meetings. This would allow participants to get familiar with the topics to be discussed and identify areas for change in advance.

In summary, I believe that the approach adopted was both rigorous and reliable. This is primarily due to the fact that throughout the entire research process, transparency was maintained, and clear justifications were provided for methodological choices and/or changes. Furthermore, the limitations were acknowledged and documented, and where possible, the identified issues were addressed (e.g. through the follow-up survey).

8.2.3 COS-DIP: agreed core outcomes, definitions, OMIs and timepoints of measurement

In the eDelphi survey and consensus meetings presented in chapter 7, six core outcomes were agreed. Participants also agreed on definitions, OMIs and timepoints of measurement for these core outcomes. In this section, the most important aspects of the agreed COS-DIP are discussed.

Core outcomes. Six outcomes were agreed on as the core outcomes. 'Penetration/aspiration' is the outcome that is consistently considered by all three stakeholder groups to be the most important outcome in OD intervention studies in PD and shows the highest level of agreement in the Delphi survey and consensus meetings. Conversely, 'choking' is on the opposite end of the spectrum. The outcome has not been measured in any of the included studies in the scoping reviews, and it may be considered as the most significant finding of the COS-DIP. It also emphasises the importance of including all key stakeholders; otherwise, this may likely not have been agreed upon as a core outcome.

The outcome **'**aspiration pneumonia' was particularly important to healthcare professionals/researchers and in comparison, less important to people with OD in PD and family members/caregivers in the eDelphi survey in chapter 7. In the previous surveys in chapters 5 and 6, it was the third most important outcome for dysphagia clinicians and rated as *important* by 78% of family members/caregivers but only 41% of people with OD in PD. In OD intervention studies in PD, 'aspiration pneumonia' is only measured in one of the 19 included studies in the scoping review in chapter 4 and hence rarely selected to be measured by researchers in studies. Thus, there was a divergence between the perceived importance of most stakeholder groups and the studies measuring this outcome. One reason for this may be that it is more resource-intensive to assess, as longer-term follow-up is required to reliably identify pneumonia.

Furthermore, the term *aspiration* pneumonia has been a controversial one in the literature. Yoshimatsu and colleagues (2022) argue that a causal relationship between aspiration and pneumonia is unclear given the delay between aspiration events and the development of pneumonia. At the same time, almost all pneumonias are due to aspiration and are therefore technically aspiration pneumonias. Because differentiating between the two is both difficult and of unclear benefit, it has been suggested that only the term pneumonia should be used instead of aspiration pneumonia (Ferguson et al., 2018; O'Keeffe, 2024). However, the term aspiration pneumonia was chosen for this study as it was the term used in the study included in the scoping review in chapter 4 that measured this outcome. Consideration should be given to rewording this outcome if the COS-DIP is updated in the future.

Definitions of outcomes. Hardly any commonly established and agreed definitions of the core outcomes existed in advance and thus, most definitions were based on previous research and amended by the SSC. Having a clear, agreed definition for each core outcome is crucial to standardising what and how we measure. Although we achieved consensus on definitions for all core outcomes, some definitions were

easier to develop and agree than others. The definitions for ‘penetration/aspiration’, ‘OD severity’, ‘swallowing related quality of life’ and ‘post swallow pharyngeal’ residue were agreed on by 86-91% of consensus meeting participants.

Defining the outcome ‘choking’ posed a challenge for both the SSC as well as consensus meeting participants. ‘Choking when swallowing’ was included as a core outcome in the COS for head and neck cancer PRO (Manduchi et al., 2024), but the COS did not seek consensus on how to measure the core outcomes, nor did it provide definitions for them. Particularly the understanding of the term choking seems to vary. While lay people mostly use choking and penetrating/aspirating material interchangeably, in clinical or research contexts choking is rather referred to material obstructing the airway. The definition was amended through discussion within the consensus meeting. Participants highlighted the need for the definition to underline the subjective aspect of this outcome by adding the words “perceived” and “felt”.

The definition of aspiration pneumonia was critically discussed by consensus meeting participants as well. The main difficulty was, as outlined above, the lack of causal relation between aspiration and pneumonia. For this reason, the definition comprised the wording “*likely* caused by aspiration”. This part of the definition may be omitted, should the wording of the core outcome be changed to ‘pneumonia’ in an update of the COS-DIP.

Outcome measurement instruments (OMIs). Lack of guidance on how to measure outcomes in a COS has been identified as a barrier to its uptake (e.g. Hughes et al., 2021). It is therefore advisable to seek consensus on how to measure core outcomes. This should be done by following the steps outlined in section 2.4. Due to the limited time and resources available to us in our study, it was not possible to conduct a systematic review of the measurement properties of all identified OMIs for the core outcomes, as was done, for example, by Davies and colleagues (2023).

It is recommended that only one OMI be chosen for each core outcome. While this is likely to improve the comparability of study results in systematic reviews and meta-analyses, it may potentially reduce the uptake of COS if researchers prefer an alternative OMI to the recommended one. Future research should include a more in-depth, systematic analysis of the measurement properties of the OMIs according to established guidelines. This could then be incorporated into a possible update of the COS-DIP. In the meantime, I would argue that having a range of OMIs agreed by consensus meeting participants will

facilitate uptake of the COS-DIP, even though guidance on how to measure the core outcomes has not yet been finalised.

Most importantly, an OMI to reliably measure 'choking' needs to be established. Based on the agreed definition in chapter 7, this OMI could be a PROM, where people with OD in PD are asked to report the frequency and severity of choking. This could be in accordance with the commonly used Drooling Severity and Frequency Scale (DSFS) (Evatt et al., 2009). With this OMI, drooling severity is measured on a 5-point Likert scale from 'never drools' (score 1) to 'profuse' (score 5) (Evatt et al., 2009). In terms of choking this could be a scale from 'never chokes' to 'life-threatening choking episodes'. With the DSFS, the drooling frequency is measured on a 4-point Likert scale from 'no drooling' (score 1) to 'constant drooling' (score 4) (Evatt et al., 2009). Translated to choking, this could be 'never chokes' to 'daily choking episodes'. Despite this, choking frequency is being assessed through the SWAL-QOL (McHorney et al., 2002). Another option therefore would be to evaluate whether the SWAL-QOL reliably assesses choking and hence, could be used as an OMI for 'choking' in addition to 'swallowing related quality of life'.

'Aspiration pneumonia' may be a core outcome where researchers feel more hesitant or restricted to measure it as it requires longer-term follow-up assessments. However, we clearly need long-term follow ups in OD intervention studies in PD. The lack of funding for trials with an extended period can potentially hinder the implementation of such follow-ups, thereby contributing to the infrequent measurement of this outcome in current studies. It is noteworthy that in one of the included studies in the scoping review in chapter 4, the authors concluded with the following statement in the discussion section: "It is essential that empirical study be completed to assess whether EMST treatment effects generalise to reductions in aspiration pneumonia in PD, where pulmonary sequelae is considered the leading cause of death." (Troche et al., 2010, p. 1917). This call for further research remains relevant 15 years later and applies to all OD interventions in PD, not just EMST.

Timepoints of measurement. In a neurodegenerative disease such as PD, the timepoints of measurement are of great importance, especially when measuring outcomes such as 'aspiration pneumonia'. An RCT by Ramig and colleagues (2001) assessing the effects of LSVT LOUD in PD, included a 24-month follow-up. The authors found that treatment effects were maintained up to this point. Such maintenance effects may be missed in current OD intervention studies in PD, as the longest follow-up identified in chapter 4

is six months. El Hayek and colleagues come to the same conclusion for physiotherapy interventions in PD, although the longest follow-up in the studies included in their systematic review was 18 months after completion of the intervention (El Hayek et al., 2023). This highlights both the potential for treatment effects to be maintained over such a long period in people with PD and the need for long-term follow-up. While the agreed timepoints of measurement in the COS-DIP are a start towards standardising longer-term follow-up, I believe that this should be extended to 18 to 24 months. It is recognised that this will increase the resources required to conduct a study and may increase the risk of higher dropout rates among participants. Without follow-up in general, and long-term follow-up in particular, no conclusions can be drawn about potential maintenance effects.

8.3 Patient and public involvement in this study

As part of the SSC, one patient partner with PD contributed particularly to the studies presented in chapters 5 and 7. His lived experience of PD, active role in the PD community, and advocacy background meant that he was well positioned to provide critical insight into the research process. His contributions strengthened the credibility and applicability of the work, and his co-authorship of related manuscripts reflects genuine collaboration rather than tokenistic involvement.

We chose to involve only this patient partner because he was readily accessible through existing networks and no prior contacts with other potential patient partners had been established. It was also expected that the more patient partners we involved that this would further limit the already challenging recruitment pool of people with PD and family members/caregivers. As we were unable to reimburse the patient partner for his work, we decided to only include him in the research studies involving people with the health condition (chapters 5 and 7), thereby reducing his workload.

Involving only one patient partner does have limitations. While his input was highly valuable, the perspective of one person cannot capture the heterogeneity of experiences of people living with PD. Including only one patient partner is not unusual (Richards et al., 2023), however, two patient partners are recommended as a minimum (Arumugam et al., 2023). Importantly, the patient partner of this study does not experience OD himself and is living with early onset PD. This means that while he could comment on broader PD-related issues (e.g. lived experiences with PD, patient priorities, accessibility and comprehensiveness of materials), he could not directly represent the lived experience of people with

OD in PD, idiopathic PD, older people and/or women living with PD. Particularly women with PD have been shown to experience different motor and non-motor symptoms than men (Subramanian et al., 2022) and therefore their perspectives may be different. In addition, it became evident throughout the study, that family members/caregivers hold different perspectives than people living with OD in PD. Including representatives from this group as patient partners could have added further valuable perspectives.

The dual role of the patient partner's wife, a family member of a person with PD and an SLT, may also have influenced his perspective. While this may have enriched discussions, it also risks that his expertise is not purely based on patient experience but also incorporates professional aspects. This may differ to people living with PD who do not have direct relationships with healthcare professionals.

Overall, having a patient partner as part of the SSC was a clear strength in enhancing accessibility and clarity, but it should be acknowledged that the involvement of a single individual - particularly one without personal experience of OD - may have introduced bias and limited representativeness. Future work should aim to include a more diverse group of people with PD, including those with lived experience of OD in PD.

8.4 Strengths and limitations of the study

The strengths and limitations of each individual study are described in chapters 4 to 7. In the following, the most important strengths and relevant limitations that may impact the overall COS-DIP are discussed.

As stated above, the main strengths of this research project are the very comprehensive approach adopted in developing the long list of outcomes and in seeking agreement not only on what outcomes to measure, but also on how and when to measure them. In doing so, we have met all eleven minimum COS-STAD standards, although the authors of the guideline acknowledge that "it is clear that few existing published COS would meet all 11 standards" (Kirkham et al., 2017, p. 7). This may make the COS-DIP one of the few COS studies to meet and even exceed the minimum standards.

One of the key limitations is our sampling approach and sample size, particularly in the Delphi survey. While the overall number of participants can be considered sufficient in the eDelphi survey, people with OD in PD and family members/caregivers were underrepresented in comparison to healthcare professionals and researchers. As discussed, this may have influenced the results of the eDelphi survey,

particularly with the used *consensus in* criterion. Despite our broad recruitment strategy through both dysphagia and PD organisations, societies and social media, the use of snowball sampling may have introduced potential sampling bias. Those who did participate may represent more engaged and research-minded individuals; and we may have missed individuals from commonly underserved groups and those who are not connected to support networks or social media. Additional barriers to participation included the English-language requirement and online-only format of the eDelphi survey. Another potential barrier, particularly for people with OD in PD and family members/caregivers, may have been difficulty in navigating the DelphiManager platform. While every effort was made to recruit a diverse sample, we have not fully achieved this.

In addition, our approach to determining what to measure was comprehensive, and in comparison, the identification of and agreement on valid and reliable OMIs was less exhaustive. Although the recommended OMIs provide a valuable starting point, future studies should conduct a more thorough evaluation of their measurement properties and agree on one single OMI for each core outcome that is feasible to implement and is reliable and validated as recommended by the COMET and COSMIN guidance (Prinsen et al., 2016b).

8.5 Uptake of the COS-DIP

The successful implementation of a COS in clinical trials faces several barriers, but there are also factors that can facilitate its adoption, as outlined in section 1.4. In developing the COS-DIP, we have addressed and mitigated many of these potential barriers. For example, the COS-DIP is rather focused than broad in scope, and, with six core outcomes, the COS is rather precise than overly long. We have ensured inclusivity by involving all key stakeholders and have provided recommendations for valid and reliable OMIs for each core outcome, apart from 'choking' for which no suitable OMI currently exists. This lack of a valid OMI for 'choking' should be addressed in the near future to further facilitate the uptake of the COS-DIP.

To promote awareness and understanding of the COS-DIP, we have developed and disseminated explainer videos, presented our findings at relevant national and international conferences, conducted a webinar for the European Society of Swallowing Disorders, and published our manuscripts predominantly open-access. At the time of writing the study protocol was viewed online > 2,600 times and the scoping review

> 3,700 times over a period of 1 and 2 years, respectively, suggesting an interest and awareness in the COS-DIP. We will further continue disseminating the results.

It is to be encouraged that the COS-DIP be included in relevant guidelines and recommendations by trial funders for future clinical trials on OD interventions in PD, a measure which would further increase the uptake of the COS-DIP. Endorsement by key journals such as Dysphagia and Movement Disorders, e.g. by requesting authors to state whether they have considered an appropriate COS and, if not, to justify this, would further promote its implementation. Similarly, endorsement and promotion by professional organisations such as the Dysphagia Research Society, the European Society for Swallowing Disorders and Movement Disorders, for example through their websites, could provide additional support for the dissemination and use of COS in the field.

Despite these efforts, it is reasonable to expect that the degree of openness to adopting the COS-DIP will vary among clinical trialists. Howarth and colleagues for example described that they "were met with sustained resistance from intervention developers" saying that "it would not be possible to 'prescribe' a set of outcomes that could be relevant to all interventions" (Howarth et al., 2024, p. 2). When presenting the results of the scoping review (chapter 4) and highlighting the need for the COS-DIP at an interdisciplinary dysphagia conference, I was met with similar reactions from some people in the audience. However, most of the people who were initially sceptical participated in the eDelphi survey two years later. This highlights the need for early and continued efforts to inform relevant stakeholders about what COS are and how they can contribute to improving the evidence base. It is also important to reiterate that a COS represents a *minimum* set of outcomes that should be measured and reported, and that it is not expected that an intervention will show a change in all core outcomes (Howarth et al., 2024).

8.6 Implications for future research

As per the definition of a COS, the core outcomes in the COS-DIP should be measured and reported as a minimum in future OD intervention studies in PD. To optimise the utility and uptake of the COS-DIP, several key research priorities have been identified in the previous sections. Firstly, there is an urgent need to develop and validate a standardised OMI for 'choking', which is currently lacking. In addition, the development and validation of a shorter version of the SWAL-QOL (McHorney et al., 2002) could improve its feasibility in clinical trials. Once these methodological gaps have been addressed, a

comprehensive assessment of the measurement properties of the recommended OMIs for all six core outcomes should be conducted to identify and agree on one robust and user-friendly OMI for each core outcome.

Implementation in future clinical trials is crucial, and feedback from clinical trialists should be systematically collected, e.g. through qualitative interviews. A systematic assessment of the uptake should be undertaken in approximately five years' time, similar to the systematic review of the uptake of the PD COS by Paul et al. (2023). Based on these findings, the COS-DIP should be regularly updated and refined. New evidence or interventions, such as those exploring cross-system effects of voice, cough, and swallowing, may warrant the addition of new core outcomes. Likewise, newly developed or validated OMIs could be incorporated, or used to replace existing ones if they are of higher quality. Conversely, outcomes that prove unfeasible to measure in clinical trials, or that no longer hold relevance for key stakeholders, may need to be removed. Finally, if conceptual understanding evolves, e.g. regarding the outcome 'choking', or if trialists consistently report that definitions are unclear or difficult to interpret, further adaptation of outcome definitions may be required.

Future research is also needed to determine whether the COS can be applied in settings that are culturally different from Europe, North America and Oceania. Translating OMIs for core outcomes into languages other than English, where they are not yet available, would increase the applicability of the COS-DIP in non-English-speaking countries.

Given the comprehensive methodology employed, the COS-DIP may serve as a transferable framework, either for the development of new COS or for adapting the COS-DIP to related clinical populations. One possible application could be individuals with OD in Atypical Parkinsonian Syndromes. However, it is important to acknowledge that OD manifests differently in these populations (Warnecke et al., 2020). For example, Progressive Supranuclear Palsy or Multisystem Atrophy, particularly their parkinsonian subtypes, tend to have greater impairment in the oral than in the pharyngeal phase of swallowing (Flynn et al., 2024; Vogel et al., 2021). Due to the COS-DIP primarily addressing pharyngeal swallowing impairments, modifications may be necessary to better accommodate these populations. Additionally, validated OMIs specific to these populations should be agreed and proposed for measuring the core outcomes, where available. Overall, adapting the COS-DIP to other populations would support

methodological consistency and resource efficiency by building on established work, rather than initiating developments de novo.

8.7 Implications for clinical practice

The COS-DIP is designed to address critical methodological weaknesses in current OD intervention studies in PD. By improving the comparability of study results, increasing the relevance of outcomes to all stakeholders, and mitigating selective outcome reporting bias, the COS-DIP will facilitate more robust systematic reviews and meta-analyses. This will lead to a stronger and more truthful evidence base for OD interventions in PD, enabling clinicians to make more informed, evidence-based decisions and provide more accurate recommendations to people with OD in PD and their family members and caregivers. This reverses the quote from Heneghan, Mahtani and colleagues (2017) referenced at the beginning of the thesis: Having a good evidence base leads to good clinical decisions. This premise will only hold true if the COS-DIP is consistently adopted in future clinical trials, as discussed in section 8.5 on the importance of uptake of the COS-DIP.

Although primarily designed for clinical trials, the COS-DIP offers value for use in clinical practice. Its use could promote standardised assessments including outcomes, particularly the PROMs 'swallowing related quality of life' and 'choking', relevant to people with the health condition and comparison of these outcomes at different timepoints and between different clinical settings, for example, when patients transition between wards or healthcare institutions. Furthermore, by providing a standardised set of outcomes to measure, the COS-DIP can support clinicians in documenting and communicating patients' changes in OD more effectively across multidisciplinary teams, potentially improving coordination of care for people with OD in PD. Ultimately, measuring a minimum of the same outcomes in clinical practice as in research studies could support clinicians in more evidence-based treatment decisions.

8.8 Development in the wider fields

Since the development of the COS-DIP began in 2021, other COS have been developed and published in the field of dysphagia (e.g. Manduchi et al., 2024) as well as in SLT (e.g. Mitchell et al., 2024). These parallel developments reflect a growing recognition of the importance of COS in rehabilitation research.

Of particular relevance to our work is the uptake analysis of the PD COS by Paul and colleagues (2023), as they showed no statistically significant increase in reporting of core outcomes after the publication of the COS, with some core outcomes remaining largely underreported. The authors attribute this limited uptake in part to the high proportion of PROMs, which is likely to increase participant burden (Paul et al., 2023). In addition, I would hypothesise that the broad scope of the PD COS, covering all motor and non-motor symptoms across different intervention types (i.e. pharmacotherapy, surgery and behavioural rehabilitation), the large number of outcomes and the lack of inclusion of all key stakeholders may compromise its practical utility and relevance in clinical trials. These findings reinforce key principles that guided the development of the COS-DIP: maintaining a focused scope, limiting the number of core outcomes, and including all key stakeholders to potentially facilitate better uptake in future clinical trials.

In the field of dysphagia and dystussia rehabilitation in PD, new studies focusing on skill training using biofeedback have emerged. Troche et al. (2023) demonstrated in an RCT that sensorimotor training for airway protection (skill based) was more effective than EMST (strength based) in improving reflex cough outcomes up to five weeks after completion of the intervention. Although the inclusion criterion specified participants with dysphagia in PD (and not necessarily dystussia), no swallowing related outcomes were assessed, as discussed in section 8.2.1. Battel and Walshe (2023) conducted a feasibility study using a skill based training approach with surface electromyography biofeedback, showing improvements in swallowing related outcomes up to three months after completion of the intervention. Due to the different outcomes, OMIs and timepoints of measurement used, it is difficult to compare the study results. It is acknowledged that skill training with biofeedback is not new to OD and PD rehabilitation, but it suggests an emerging area of research where implementing the COS-DIP may help to improve the comparability of study results.

A further advancement is the recent development and validation of scales used with instrumental assessment in PD, such as VASES (Curtis et al., 2022) and DIGEST-FEES (Labeit et al., 2023). Given their established validity and comprehensive outcome assessment, it is to be anticipated that they are likely to be useful OMIs in future clinical trials and clinical practice.

8.9 Conclusions

This thesis has documented the development of a Core Outcome Set for Dysphagia Interventions in Parkinson's disease (COS-DIP). The COS-DIP comprises the six core outcomes ‘penetration/aspiration’, ‘aspiration pneumonia’, ‘choking’, ‘OD severity’, ‘swallowing related quality of life’ and ‘post swallow pharyngeal residue’. It was established through inclusion of key stakeholders, i.e. people with OD in PD, family members/caregivers, healthcare professionals, and researchers. The applied methodology extended beyond traditional COS development by incorporating all key stakeholder perspectives in developing the long list of outcomes. Furthermore, we sought consensus not only on which core outcomes to measure but also on their definitions, OMIs, and timepoints of measurement.

Regular evaluation of the COS-DIP will be crucial to ensure its continued validity and relevance. This ongoing assessment will determine whether the selected outcomes remain important to all key stakeholders and will provide opportunities to refine the COS through addition or removal of outcomes, OMIs or revised definitions. Monitoring the uptake and reporting of the COS-DIP in OD intervention studies in PD will be essential to assess its impact on research quality.

The consistent use of the COS-DIP in clinical trials in OD interventions in PD has the potential to strengthen the evidence base in this field. This will help identify the most effective interventions for OD in PD, ultimately leading to more informed clinical decision-making and improved patient care for this growing population.

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Appendix 3.1: Ethical approval



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

Application	Academic Year 2021/22
Applicant Code	MT9
Applicant/Supervisor Name	Julia Hirschwald / Dr Margaret Walshe
Title of Research	COS-DIP Study
Date of this letter	08/12/2022

Dear Julia,

Your submission for ethical approval for the research project above was considered by the Research Ethics Committee, School of Linguistic, Speech and Communication Sciences, Trinity College Dublin on 08/12/2022 and has been approved in full.

Please note:
(i) On completion of research projects, applicants should complete the *End of Project Report Form* (which can be found at: https://www.tcd.ie/slscs/research/ethics/) and submit one electronic copy (to slscs@tcd.ie)
(ii) The REC requests, in particular, that you attend to your commitments regarding the storage and destruction of data arising from this research, in keeping with REC policy and General Data Protection Regulation (GDPR) guidelines.

We wish you every luck with your research.

Best wishes,

Ciarán Kenny

Dr Ciarán Kenny
Chair, Research Ethics Committee
School of Linguistic, Speech and Communication Sciences

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Application	Academic Year 2022/23
Applicant Code	HT27
Applicant/Supervisor Name	Julia Hirschwald / Prof Margaret Walshe
Title of Research	The COS-DIP Study
Date of this letter	15/05/2023

Dear Julia,

Your amended submission (dated 21/04/2023) for ethical approval for the research project above was considered by the Research Ethics Committee, School of Linguistic, Speech and Communication Sciences, Trinity College Dublin and has been approved in full.

Please note:
(i) On completion of research projects, applicants should complete the *End of Project Report Form* (which can be found at: https://www.tcd.ie/slscs/research/ethics/) and submit one electronic copy (to slscs@tcd.ie)
(ii) The REC requests, in particular, that you attend to your commitments regarding the storage and destruction of data arising from this research, in keeping with REC policy and General Data Protection Regulation (GDPR) guidelines.

We wish you every luck with your research.

Best wishes,

Ciarán Kenny

Dr Ciarán Kenny
Chair, Research Ethics Committee
School of Linguistic, Speech and Communication Sciences

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Application	Academic Year 2022/23
Applicant Code	TT80
Applicant/Supervisor Name	Lauren Mooney / Prof Margaret Walshe
Title of Research	Examining Dysphagia Clinicians' Views on Core Outcomes in Interventions for people with Parkinson's Disease
Date of this letter	14/06/2023

Dear Lauren,

Your submission for ethical approval for the research project above was considered by the Research Ethics Committee, School of Linguistic, Speech and Communication Sciences, Trinity College Dublin on 13/06/2023 and has been approved in full.

Please note:
(i) On completion of research projects, applicants should complete the *End of Project Report Form* (which can be found at: https://www.tcd.ie/slscs/research/ethics/) and submit one electronic copy (to slscs@tcd.ie)
(ii) The REC requests, in particular, that you attend to your commitments regarding the storage and destruction of data arising from this research, in keeping with REC policy and General Data Protection Regulation (GDPR) guidelines.

We wish you every luck with your research.

Best wishes,

Ciarán Kenny

Dr Ciarán Kenny
Chair, Research Ethics Committee
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Appendix 4.1: Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews (PRISMA-ScR) Checklist

SECTION	ITEM	PRISMA-ScR CHECKLIST ITEM	REPORTED ON PAGE #
TITLE			
Title	1	Identify the report as a scoping review.	1
ABSTRACT			
Structured summary	2	Provide a structured summary that includes (as applicable): background, objectives, eligibility criteria, sources of evidence, charting methods, results, and conclusions that relate to the review questions and objectives.	1-2
INTRODUCTION			
Rationale	3	Describe the rationale for the review in the context of what is already known. Explain why the review questions/objectives lend themselves to a scoping review approach.	3-5
Objectives	4	Provide an explicit statement of the questions and objectives being addressed with reference to their key elements (e.g., population or participants, concepts, and context) or other relevant key elements used to conceptualize the review questions and/or objectives.	5
METHODS			
Protocol and registration	5	Indicate whether a review protocol exists; state if and where it can be accessed (e.g., a Web address); and if available, provide registration information, including the registration number.	6
Eligibility criteria	6	Specify characteristics of the sources of evidence used as eligibility criteria (e.g., years considered, language, and publication status), and provide a rationale.	6-7
Information sources*	7	Describe all information sources in the search (e.g., databases with dates of coverage and contact with authors to identify additional sources), as well as the date the most recent search was executed.	6
Search	8	Present the full electronic search strategy for at least 1 database, including any limits used, such that it could be repeated.	Additional file 2
Selection of sources of evidence†	9	State the process for selecting sources of evidence (i.e., screening and eligibility) included in the scoping review.	7
Data charting process‡	10	Describe the methods of charting data from the included sources of evidence (e.g., calibrated forms or forms that have been tested by the team before their use, and whether data charting was done independently or in duplicate) and any processes for obtaining and confirming data from investigators.	8
Data items	11	List and define all variables for which data were sought and any assumptions and simplifications made.	8
Critical appraisal of individual sources of evidence§	12	If done, provide a rationale for conducting a critical appraisal of included sources of evidence; describe the methods used and how this information was used in any data synthesis (if appropriate).	Not applicable

Synthesis of results	13	Describe the methods of handling and summarizing the data that were charted.	8-9
RESULTS			
Selection of sources of evidence	14	Give numbers of sources of evidence screened, assessed for eligibility, and included in the review, with reasons for exclusions at each stage, ideally using a flow diagram.	9
Characteristics of sources of evidence	15	For each source of evidence, present characteristics for which data were charted and provide the citations.	9
Critical appraisal within sources of evidence	16	If done, present data on critical appraisal of included sources of evidence (see item 12).	Not applicable
Results of individual sources of evidence	17	For each included source of evidence, present the relevant data that were charted that relate to the review questions and objectives.	10-16
Synthesis of results	18	Summarize and/or present the charting results as they relate to the review questions and objectives.	10-16
DISCUSSION			
Summary of evidence	19	Summarize the main results (including an overview of concepts, themes, and types of evidence available), link to the review questions and objectives, and consider the relevance to key groups.	16-18
Limitations	20	Discuss the limitations of the scoping review process.	18
Conclusions	21	Provide a general interpretation of the results with respect to the review questions and objectives, as well as potential implications and/or next steps.	19
FUNDING			
Funding	22	Describe sources of funding for the included sources of evidence, as well as sources of funding for the scoping review. Describe the role of the funders of the scoping review.	21

JBI = Joanna Briggs Institute; PRISMA-ScR = Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews.
* Where *sources of evidence* (see second footnote) are compiled from, such as bibliographic databases, social media platforms, and Web sites.
† A more inclusive/heterogeneous term used to account for the different types of evidence or data sources (e.g., quantitative and/or qualitative research, expert opinion, and policy documents) that may be eligible in a scoping review as opposed to only studies. This is not to be confused with *information sources* (see first footnote).
‡ The frameworks by Arksey and O'Malley (6) and Levac and colleagues (7) and the JBI guidance (4, 5) refer to the process of data extraction in a scoping review as data charting.
§ The process of systematically examining research evidence to assess its validity, results, and relevance before using it to inform a decision. This term is used for items 12 and 19 instead of "risk of bias" (which is more applicable to systematic reviews of interventions) to include and acknowledge the various sources of evidence that may be used in a scoping review (e.g., quantitative and/or qualitative research, expert opinion, and policy document).

From: Tricco AC, Lillie E, Zarin W, O'Brien KK, Colquhoun H, Levac D, et al. PRISMA Extension for Scoping Reviews (PRISMAScR): Checklist and Explanation. Ann Intern Med. 2018;169:467–473. doi: 10.7326/M18-0850.

Note: indicated page numbers refer to pages on manuscript submitted to journal

Appendix 4.2: Scoping review protocol

Swallowing Outcome Measures in Dysphagia Interventions in Parkinson's Disease: Protocol for a Scoping Review

Julia Hirschwald*, Jule Hofacker, Sallyanne Duncan and Margaret Walshe

November 2021

Abstract

Background: Combining trial results to direct people with oropharyngeal dysphagia (OD) in Parkinson's disease (PD) for treatment is difficult because of a high variability of how outcomes are being defined and measured, missing information on adverse events and possible outcome-reporting bias. Therefore, as a first step in the establishment of a Core Outcome Set for Dysphagia Interventions in Parkinson's Disease (COS-DIP), a scoping review will be conducted to systematically map the research done in this area and to identify any existing gaps in knowledge.

Objective: This scoping review aims to report on swallowing outcomes, their assessments and applied timepoints in (quasi-) Randomised Controlled Trials (RCTs) and Controlled Clinical Trials (CCT) in people with OD in PD and ultimately develop a COS-DIP.

Methods: All clinical interventions with a (quasi-) RCT and CCT design for people of 18 years or older with OD in PD will be included. Key electronic databases and trial registries without any date or language restrictions will be searched. Two independent reviewers will assess articles for eligibility. The outcome measures applied in the included studies will be extracted following the taxonomy by Dodd et al. (2018). Additionally, information on the chosen verbatim and definition of the outcome, the applied assessments, frequency and timepoints of measurement will be charted.

Conclusions This scoping review will inform the establishment of the COS-DIP as a first step. Ultimately this has immense potential to change how trials are conducted for OD in PD worldwide as it will translate to higher quality meaningful trials and increase consistency among trials.

Keywords: Dysphagia, Parkinson's Disease, Clinical Interventions, Scoping Review

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Introduction

Oropharyngeal dysphagia (OD) is a common and clinically significant symptom in people with Parkinson's disease (PD) [9]. The prevalence varies between 11-81% according to severity of the disease, definition of dysphagia and assessment tools used [10]. Nearly 50% of people with PD experience aspiration [12], increasing the risk of developing aspiration pneumonia, which is a leading cause of death in people with PD [3, 6, 7].

Dysphagia interventions aim for safe, efficient, and sufficient intake of food and fluids while maximizing quality of life (QoL) for the patient, their carers and their family [3]. For people with PD, dysphagia therapy comprises surgical, pharmacological, rehabilitative, compensatory and adaptive approaches [2]. Overall, clinical decisions on the safety and efficacy of interventions are based on selected outcomes, thus the choice of outcomes to be measured and reported in clinical trials is critical [13].

Furthermore, comparing, and synthesizing study results to direct treatment for people with OD in PD is necessary. So far, no systematic review in this research field was carried out. This is, to our understanding, because combining trial results for systematic reviews is still challenging as applied outcomes are heterogenous between the trials. Some trials focus directly on medical aspects of OD while others ignore them. Outcomes are also measured differently using partly methods of poor validity with vague descriptions of treatment effects. Additionally, in a neurodegenerative condition such as PD the longer-term treatment effects, including adverse events, must be known. For example, an intervention for OD may not work beyond the 4-week trial but outcomes might be assessed only at the immediate end of the experimental period. Lastly, in clinical trials on OD in PD the patients themselves and their carers are rarely consulted on what outcomes matter to them in these trials most.

In summary, a solution to the depicted challenges is the development and use of an agreed standardised Core Outcome Set for Dysphagia Interventions in Parkinson's Disease (COS-DIP) devised by key stakeholders including patients, carers and healthcare professionals amongst others. This will result in higher quality meaningful trials, increase the consistency across trials which will then enhance combining individual study results to reach conclusions on the safety and effectiveness of the interventions. It will not prevent or restrict researchers from examining other outcomes [5, 13].

In order to establish a COS-DIP the first step is a scoping review of the literature on the applied outcome measures in clinical trials in OD and PD. A scoping review is chosen as the most appropriate method to systematically map the research done in this area, as well as to identify any existing gaps in knowledge [1]. A preliminary search of MEDLINE, the Cochrane Database of Systematic Reviews and JBI Evidence Synthesis was conducted and no current or underway scoping reviews on the topic were identified.

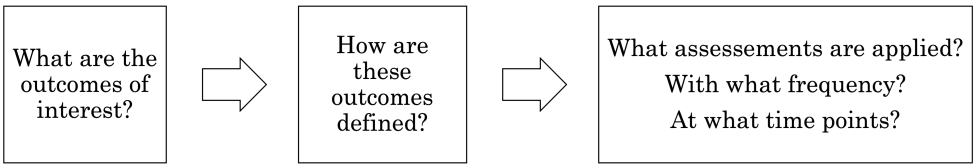


Figure 1: Overview of the research questions that will be addressed by the scoping review

The objective of this scoping review is to report on the swallowing outcomes, their definitions, assessments, timepoints and frequency of assessment that are applied in all (quasi-) RCTs and CCTs in OD in PD. See Figure 1 for an overview of the research questions. Ultimately the aim is to develop a COS-DIP.

Methods

The proposed scoping review will be conducted in accordance with the Preferred Reporting Items for Systematic Reviews and Meta-analysis extension for Scoping Reviews (PRISMA-ScR) checklist [11].

Table 1 provides an overview of the eligibility criteria in accordance with the patient, concept and context framework. The types of sources that will be included are all RCTs, quasi-RCTs and CCTs. An additional scoping review including all study designs except those included in this study will be conducted in adjunction by Karir and colleagues from University College London, UK.

Search strategy

The text words contained in the titles and abstracts of relevant articles, and the index terms used to describe the articles will be used to develop a full search strategy for relevant databases and trial registries with the assistance of a subject librarian. The search strategy (see Appendix I for an example), including all identified keywords and index terms, will be adapted for each included database. The reference lists of all included sources of evidence will be screened for additional studies. It is planned to contact the study authors for additional information, if required. Studies published in any language from inception will be included as this will increase the diversity of information obtained. The databases to be searched include AMED, CINAHL, EMBASE, MEDLINE, Web of Science and ProQuest Dissertations & Theses. Additionally, ClinicalTrial.gov will be searched for unpublished studies.

Study selection

Following the search, all identified citations will be collated and uploaded to an online platform (www.covidence.org) and duplicates will be removed. Following a pilot test of a random sample of 25 titles and abstracts with an agreement of at least 75%, all titles and abstracts will be screened by two independent reviewers (JAH, JMH) for assessment against the inclusion criteria for the review. The full text of selected citations will be assessed in detail against the inclusion criteria by the same two independent reviewers.

Table 1: Eligibility criteria for study inclusion in scoping review

Patient	Concept	Context
All types of OD in idiopathic PD of 18 years or older	Any clinical interventions in OD in PD	Any clinical context

Reasons for exclusion of sources of evidence at full text that do not meet the inclusion criteria will be recorded and reported in the scoping review. Any disagreements that arise between the reviewers at each stage of the selection process will be resolved through discussion, or with an additional reviewer (MW). The results of the search and the study inclusion process will be reported in full in the final scoping review and presented in a PRISMA-ScR flow diagram [11].

Data Extraction

Data will be extracted from studies included in the scoping review by two independent reviewers using a data extraction tool developed previously and applied by Hofacker et al. [8] (see Appendix II). The data extracted will include details about the first author's name, year of publication and country of origin, study design, population and sample size and applied outcome measures. The draft data extraction form will be trialed independently on 3 included sources by JAH and JMH and will be modified and revised as necessary during the process of extracting data from each included evidence source. Modifications will be detailed in the scoping review. Any disagreements that arise between the reviewers will be resolved through discussion, or with an additional reviewer (MW). If appropriate, authors of papers will be contacted to request missing or additional data.

Data Analysis and Presentation

The outcome measures used in the studies will be extracted as verbatim as suggested by the Core Outcome Measures in Effectiveness Trials Handbook (COMET Initiative) [13]. The COMET initiative also recommends using the taxonomy proposed by Dodd et al. [4] as it provides the granularity needed for the development of a COS later on. Therefore, this taxonomy will be used to classify the outcomes in different thematic groups. All extracted data will be categorized in an excel spreadsheet also previously developed and applied by Hofacker et al. [8] (see Appendix III) comprising information regarding core area of the outcome and outcome domain, number and verbatim of chosen outcome, definition of outcome, way of measurement, timepoints of measurement and frequency of measurement. Furthermore, the amount and percentages of used outcomes within the respective core area will be calculated and listed in a table. Despite the numerical presentation of the results, a narrative summary of the results will accompany the tabulated and charted results and will describe how the results relate to the reviews objective and questions.

Discussion

This scoping review aims at systematically mapping the research done on clinical dysphagia interventions in people with OD in PD and to

identify any existing gaps in knowledge. More specifically, outcomes that are being applied in all (quasi-) RCTs and CCTs will be extracted and their definitions, assessments, timepoints and frequency of assessment will be charted. This will allow to further on establish a COS-DIP. The availability of the COS-DIP has immense potential to change how trials are conducted for OD in PD worldwide. It will translate to higher quality meaningful trials, make it easier to combine individual study results to direct decision-making about interventions. It will not prevent or restrict researchers from examining other outcomes.

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Declaration of Interest

The corresponding author JH undertakes this research project as part of a Provost's PhD Award funded by Trinity College Dublin, Ireland. The funding body does not have any role in the study design or the writing of this protocol and will not have any role in the data collection and analysis, interpretation of data, or decision to publish the results of the study.

Abbreviations

CCT = Controlled Clinical Trial, COS = Core Outcome Set, COS-DIP = Core Outcome Set in Dysphagia Interventions in Parkinson's Disease, OD = Oropharyngeal Dysphagia, PD = Parkinson's Disease, QoL = Quality of Life, RCT = Randomised Controlled Trial

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Appendices

Appendix I: Exemplary search strategy for PubMed

1. "Deglutition Disorders"[Mesh]
2. dysphagi*[Title/Abstract]
3. deglutition[Title/Abstract]
4. swallow*[Title/Abstract]
5. OR/1-4
6. "Parkinson Disease"[Mesh]
7. idiopathic parkinson's disease[Title/Abstract]
8. idiopathic parkinsons disease[Title/Abstract]
9. idiopathic parkinsons' disease[Title/Abstract]
10. idiopathic parkinson disease[Title/Abstract]
11. OR/6-10
12. 5 AND 11

Appendix II: Data extraction form

Included Study: 1st author's name, year, country	Study design, population, sample size	Applied outcomes

Appendix III

CORE AREA	DEATH		PHYSIOLOGICAL/ CLINICAL		LIFE IMPACT		RESOURCE USE		ADVERSE EVENTS	
Outcome domain			Physiological/clinical		Functioning		Resource use			
	n°	verbatim	n°	verbatim	n°	verbatim	n°	verbatim	n°	verbatim
1st author's name, year Item Definition How measured? Time point Frequency										
Item Definition How measured? Time point Frequency										
1st author's name, year Item Definition How measured? Time point Frequency										
Item Definition How measured? Time point Frequency										

Appendix 4.3: Search strategy

#	Search strategy	Results
AMED		
S3	S1 AND S2	2
S2	SU parkinson disease OR TI (idiopathic parkinson's disease OR idiopathic parkinsons disease OR idiopathic parkinsons' disease OR idiopathic parkinson disease) OR AB (idiopathic parkinson's disease OR idiopathic parkinsons disease OR idiopathic parkinsons' disease OR idiopathic parkinson disease)	80
S1	SU dysphagia OR TI (dysphagi* OR deglutition OR swallow*) OR AB (dysphagi* OR deglutition OR swallow*)	1.019
CINAHL		
S8	S5 AND S6 (Limiters - Exclude MEDLINE records)	149
S7	S5 AND S6	388
S6	S3 OR S4	24.108
S5	S1 OR S2	20
S4	T1 (idiopathic parkinson's disease OR idiopathic parkinsons disease OR idiopathic parkinsons' disease OR idiopathic parkinson disease) OR AB (idiopathic parkinson's disease OR idiopathic parkinsons disease OR idiopathic parkinsons' disease OR idiopathic parkinson disease)	860
S3	(MH "Parkinson Disease")	
S2	TI (dysphagi* OR deglutition OR swallow*) OR AB (dysphagi* OR deglutition OR swallow*)	17.089
S1	(MH "Deglutition Disorders"")	
EMBASE		
8	#7 AND (embase|/lim NOT ((embasel/lim AND [medline]/lim)	1.560
7	#3 AND #6	2.796
6	#4 OR #5	174.509
5	idiopathic parkinson/s disease':ab.ti OR 'idiopathic parkinsons disease':ab.ti OR 'idiopathic parkinsons/ disease':ab.ti OR "idiopathic parkinson disease':ab.ti	4.939
4	parkinson disease/exp	174.081
3	#1 OR #2	126.373
2	dysphagi*:ab.ti OR deglutition:ab,ti OR swallow*:ab,ti	92.668
1	'dysphagia"/exp	85.162
ProQuest		
	noft(dysphagi* OR deglutition OR swallow*) AND noft(idiopathic parkinson's disease OR idiopathic parkinson disease OR idiopathic parkinson' disease OR idiopathic parkinson disease)	6
PubMed		
#7	(("Parkinson Disease" [Mesh]) OR (idiopathic parkinson's disease[Title/Abstract] OR idiopathic parkinsons disease[Title/Abstract] OR idiopathic parkinsons' disease[Title/Abstract] OR idiopathic parkinson disease[Title/Abstract])) AND (("Deglutition Disorders"[Mesh]) OR (dysphagi*[Title/Abstract] OR deglutition[Title/Abstract] OR swallow*[Title/Abstract]))	763
#6	("Parkinson Disease" [Mesh]) OR (idiopathic parkinson's disease[Title/Abstract] OR idiopathic parkinsons disease[Title/Abstract] OR idiopathic parkinsons' disease[Title/Abstract] OR idiopathic parkinson disease[Title/Abstract])	74.422
#5	idiopathic parkinson's disease[Title/Abstract] OR idiopathic parkinsons disease[Title/Abstract] OR idiopathic parkinsons' disease[Title/Abstract] OR idiopathic parkinson disease[Title/Abstract]	336
#4	Parkinson Disease [Mesh]	73.538
#3	("Deglutition Disorders" [Mesh]) OR (dysphagi*[Title/Abstract] OR deglutition[Title/Abstract] OR swallow*[Title/Abstract])	94.511
#2	dysphagi*[Title/Abstract] OR deglutition[Title/Abstract] OR swallow*[Title/Abstract]	58.181
#1	Deglutition Disorders[Mesh]	55.955
Web of Science		
3	(#1) AND #2	106
2	TS=(idiopathic parkinson's disease OR idiopathic parkinsons disease OR idiopathic parkinsons' disease OR idiopathic parkinson disease)	7.220
1	dysphagi* OR deglutition OR swallow* (Topic)	60.163
Clinicaltrials.gov		
	dysphagia | Interventional Studies | Parkinson Disease	24

Appendix 4.4: Data extraction chart

Complete data extraction

1st author's name, year	Country	Study design	N (dropouts)	Intention to treat analysis	Age	Gender	PD severity	OD severity	Intervention	Comparator	Applied outcome measures + way of measurement	Timepoints	Frequency
Ayres, 2017	Brazil	CCT	32 (8)	No	IG: 62.0 (SD±11.5) CG1: 62.8 (SD±6.2) CG2: 64.5 (SD±5.6)	m=18, f=6	IG: 2.8 (SD±0.8) CG1: 2.5 (SD±0.7) CG2: 2.5 (SD±0.8)	FOIS: IG: 5.9 (SD ±1.3) CG1: 6.8 (SD±0.5) CG2: 6.8 (SD±0.4)	Chin-down posture maneuver	CG1: no intervention CG2: swallowing orientations	> FEES: - prior state of secretion in nasopharyngeal structures, oropharynx, laryngopharynx - presence/absence of: thickening on the posterior laryngeal wall, tremor in structures (BOT, vallecula), early escape, vallecular stasis in glossoepiglottic folds and pyriform sinus, penetration, tracheal aspiration, cough reflex > CSE (presence/absence) - history of aspiration pneumonia - alert state, interaction attention/ability, postural control, fatiguability - awarness of the swallowing problem & of secretion - ability to manipulate flows - anatomy and oral, pharyngeal and laryngeal physiology - orofacial tones & sensitivity - oral apraxia, swallowing apraxia - gag pharyngeal contraction, cough and hawk - saliva swallowing - oral residue - delayed swallowing reflex, multiple swallowing - reduction in laryngeal elevation - wet voice > FOIS > SWAL-QOL	pre, post (4w)	2
Baijens, 2012	Netherlands	Feasibilit/pilot study with control group	20 (N/S)	No	65.5 (46-81)	m=14, f=6	median: 2 (1-3)	mild to severe	SES (VitalStim), 3 different electrode positions applied in random order per subject	Healthy controls with same intervention	VFS: - moment of opening and closing of: glossopalatal junction, velopharyngeal junction, laryngeal vestibule, upper esophageal sphincter - PAS - movement patterns of hyoid bone - extent of movement of hyoid bone - pre-swallow anterior spill - preswallow posterior spill - lingualpumping - swallow hesitancy - piecemeal deglutition - delayed initiation of the pharyngeal reflex - postswallow oral oral residue - postswallow vallecular pooling - postswallow pyriform sinus pooling - PAS	during	1
Baijens, 2013	Netherlands	quasi-RCT	109 (19)	No	median: 68	m=66, f=24	median: 2(1-4)	mild to severe	**IG1:** traditional logopedic dysphagia treatment + SES of submental region; motor-level stimulation; **IG2:** traditional logopedic dysphagia treatment + SES of submental region; sensory-level stimulation	Traditional logopedic dysphagia treatment	> FEES: - Preswallow posterior spill - Piecemeal deglutition - Delayed initiation pharyngeal reflex - Postswallow vallecular pooling - Postswallow pyriform sinus pooling - PAS > VFS: - Preswallow anterior spill - Preswallow posterior spill - Lingual pumping - Swallow hesitancy - Piecemeal deglutition - Delayed initiation pharyngeal reflex - Postswallow oral residue - Postswallow vallecular pooling - Postswallow pyriform sinus pooling - PAS	pre, post (15d)	2

Byeon, 2016	Korea	RCT	33 (N/S)	No	IG: 63.8 (SD ± 8.2) CG: 65.1 (SD ± 9.5	m=31, f=2	IG: 15 ≤ H&Y 3; 3 > H&Y 4 CG: 11 ≤ H&Y 3; 4 > H&Y 4	N/S	Postural techniques + EMST	EMST	> VFS: - Functional Dysphagia Scale (FDS)	pre, post (4w)	2
Claus, 2021	Germany	RCT	53 (8)	No	IG: 67.3 (54-83; SD ± 9.5) CG: 67.1 (49-82; SD ± 7.7)	IG: m=19, f=5 CG: m=18, f=3	IG: 2.5 (2-4), CG: 2.6 (2-4)	N/S	EMST (calibrated)	EMST (sham)	> FEES (5-point scales): - premature spillage - penetration-aspiration events - residue - total FEES score (0-108) > SWAL-QOL (German) > Swallowing Disturbance Questionnaire (SDQ) (German) > MEG (only 22 patients, sub-group): - frequency bands: theta(4–8 Hz), alpha (8–13 Hz), beta (13–30 Hz), low-gamma, (30–60 Hz), and high-gamma (60–80 Hz) - in all frequency bands: source localization of each subject's swallowing-associated event-related desynchronization (ERD) of cortical rhythms	pre, post (4w, 3m)	3
Feng, 2019	China	CCT	60 (N/S)	No	IG: 65.20 (SD ± 6.84) (54-84) CG: 64.66 (SD ± 5.27) (56-75)	m=38, f=22	N/S	>18 on the SSA (inclusion criterium)	Vocal training + conventional swallowing treatment	Conventional swallowing treatment	> SSA > Evaluation of salivation: - UPDRS II - Drooling Severity and Frequency Scale (DSFS) > Voice evaluation - Maximum phonation time (MPT) - Maximum phonation decibel	pre, post (4w)	2
Heijnen, 2012	Netherlands	quasi-RCT	109 (21)	No	Median 68 (42-81)	m=65, f=23	Median 2 (1-4)	FOIS: median 7 (1-7)	Traditional logopedic dysphagia treatment + NMES (VitalStim) of the supra hyoid musculature; Group 2 (NMES-M): motor level, Group 3 (NMES-S): sensory level	Group 1: traditional logopedic dysphagia treatment	> FOIS > SWAL-QOL (Dutch) > MD Anderson Dysphagia Inventory (MDADI) (Dutch) > Dysphagia Severity Scale (DSS) > FEES > VFS	pre, post, 3m (DSS post each treatment session)	3 (15)
Khedr, 2019	Egypt	RCT	33 (3)	No	IG: 60.7 (SD ± 8.8) CG: 57.4 (SD ± 10.0)	N/S	IG: 3.1 (SD ±1.1) CG: 3.5 (SD ±1.0)	SDQ: IG: 17.4 (SD ±6.1) CG: 16.2 (SD±5.8)	Repetitive Transcranial Magnet Stimulation	Sham	> H&Y > UPDRS III > Instrumental Activities of Daily Living (IADL) > Self-assessment scale of swallowing > SDQ > Arabic DHI > VFS (for 9 in IG, 6 in CG, pre, post) - pharyngeal transit time (PTT) - time of first superior-anterior movemnt of hyoid bone H1 - time when hyoid bone reached its max elevation H2 - time required for max elevation of the hyoid bone H2-H1 - PAS - postswallow residue	pre, post (2w, 1m, 2m, 3m)	5
Kondo, 2017	Japan	RCT	20, thereof 3 with PD (N/S)	No	IG: 80.4 (SD ±9.5) CG: 80.1 (SD ±5.9)	m=19, f=1	N/S	N/S	Aural stimulation with capsaicin ointment	Aural stimulation with placebo ointment	> Endoscopic swallowing scoring: - Salivary pooling in vallecula and pyriform sinuses - The response of glottal closure reflex induced by touching epiglottis with endoscope - The location of the bolus at the time of swallow onset assessed by endoscopic whiteout - The extent of pharyngeal clearance after swallowing of blue-dyed water - Total swallowing function > Sensory-Motor-Reflex-Clearance (SMRC) scale: - Sensory: Initiation of swallowing reflex as assessed by endoscopic whiteout - Motion: holding bolus in oral cavity and inducing laryngeal elevation according to instructions - Reflex: glottal closure and cough reflexes induced by touching epiglottis or arytenoids with endoscope - Clearance: pharyngeal clearance of bolus after swallowing	pre, post (5, 30, 60 min)	4
Logemann, 2008	United States	RCT	742, thereof 360 with PD (31)	No	range: 50-95	m=498, f=213	N/S	Aspiration of water on VFS (inclusion criterium)	Chin-down posture while consuming thin liquids	No postural adjustment during swallows of nectar and honey-thickened liquids	> VFS: - Aspiration Preference for different interventions	during	1

Manor, 2013	Israel	RCT	42 (N/S)	No	68.8 (SD ±8.1)	m=24, f=18	IG: 2.21 (SD ±0.79) CG: 2.19 (SD ±0.84)	N/S	Video assisted swallowing therapy (VAST) with conventional therapy	Conventional therapy	> SDQ > FEES: - bolus flow time - bolus location when the swallowing reflex is triggered - residue location - penetration before/after swallowing - aspiration before/after swallowing > SWAL-QOL > SWAL-CARE > Pleasure of Eating (POE) Scale	pre, post (2w, 4w, 6 m)		4
Nagaya, 2000	Japan	CCT	24, thereof 10 with PD (N/S)	No	IG: 70.5 (53-80) CG: 72 (47-93)	m=8, f=14 IG: m=5, f=5 CG: m=3, f=6	H&Y III = 8x, H&Y IV = 2	N/S	Swallowing training in PwPD	Swallowing training in HC	> Electromyography in submental muscles - premotor time (PMT)	pre, post		2
Park, 2018	Korea	RCT	18 (0)	No	IG: 63.44 (SD ±13.55) CG: 54.67 (SD ±13.82)	m=8, f=10	< H&Y III	N/S	NMES (VitalStim) with effortful swallowing + conventional therapy	Sham NMES (VitalStim) with effortful swallowing + conventional therapy	> VFS: - horizontal displacement of the hyoid bone - vertical displacement of the hyoid bone - Videofluoroscopic dysphagia scale (VDS) total - VDS-oral phase - VDS-pharyngeal phase - PAS	pre, post (4w)		2
Robbins, 2008	United States	RCT	515, thereof 255 with PD (213)	Yes	IG: 81 CG: 80	m=359, f=156	N/S	Aspiration of water on VFS (inclusion criterium)	Chin-down posture with thin liquids	Head neutral position with thickened liquids (nectar thick or honey thick)	> Pneumonia > Death > Adverse events > Hospitalization > Adherence to intervention	continuously during 3m, intervention adherence weekly	continuously, 36	
Sasegbon, 2021	UK	Feasibilit/pilot study with control group	12 (N/S)	No	70 (SD ±8)	m=10, f=2	1 Hz: 2.9 (±0.3) 5 Hz: 2.1 (±0.6) PES: 1.8 (±0.3)	PAS ≥ 2	1 Hz rTMS, 5 Hz rTMS, PES	Sham	> VFS: - PAS - Oral transit time (OTT) - Pharyngeal transit time (PTT) Pharyngeal responese time (PRT) > EMG: - Pharyngeal motor evoked potential (MEP) amplitudes - MEP latencies	pre, post (0min; for MEP 30min)	2 to 3	
Silva-Arone, 2021	Brazil	Feasibilit/pilot study with control group	6 (N/S)	No	73.1 (64-83) (SD ±6.2)	m=6, f=0	2.3 (2-3)	FOIS: 6.5 (6-7)	Prophylactic speech-language therapy associated with EMG biofeedback	Same treatment without EMG biofeedback	> VFS: - Dysphagia Outcome and Severity Scale (DOSS) > SWAL-QOL > FOIS	pre, post (3m, 6m)		3
Troche, 2010	United States	RCT	68 (8)	Yes	IG: 66.7 (SD ±8.9) CG: 68.5 (SD ±10.3)	m=47, f=13	range II-IV	mild to moderate	EMST (calibrated)	EMST (sham)	> VFS: - PAS - duration of hyoid movement - onset of bolus transit - UES-opening - UES-widset - UES-closure - laryngeal closure - max laryngeal closure - laryngeal opening > SWAL-QOL	pre, pre, post (4w)	up to 3	
Wei, 2017	China	CCT	217 (N/S)	No	IG: 71.4 (SD ±12.7) CG: 69.3 (SD ±11.3)	m=130, f=87	N/S	Level 6-3 (unspecified scale)	Standardized out-of-hospital management: education, skill training (oral muscle exercises, effective cough training, pronunciation training, eating training, compensatory training with video and presentation)	Face and tongue training, eating considerations and dysphagia rehabilitation guidance	> Dysphagia > Mis-inhalation	post (6m)		1
Xie, 2018	United States	RCT	11 (1)	No	68.5 (SD ± 5.9)	m=9, f=2	N/S	N/S	3 DBS conditions (sequence random order of 130 Hz, 60Hz, DBS off)	crossover: 3 DBS conditions (sequence random order of 130 Hz, 60Hz, DBS off)	> VFS: - Aspiration > SDQ > UPDRS III > FOG spells > FOG time > Walking difficulty perception	V1: during, SDQ post 30 min V2 (at least 6m later): during, SDQ post 30 min		2

A-DHI: Arabic Dysphagia Handicap Index; CCT: Controlled Clinical Trial; CG: Control Group; CSE: Clinical Swallow Evaluation; DBS: Deep Brain Stimulation; DOSS: Dysphagia Outcome and Severity Scale; DSFS: Drooling Severity and Frequency Scale; DSS: Dysphagia Severity Scale; EMG: Electromyography; EMST: Expiratory Muscle Strength Training; FDS: Functional Dysphagia Scale; FEES: Fibreoptic Endoscopic Evaluation of Swallowing; f: female; FOG: Freezing of Gait; FOIS: Functional Oral Intake Scale; H&Y: Hoehn&Yahr; IADL: Instrumental Activities of Daily Living; IG: Intervention group; m: male; MDADI: MD Anderson Dysphagia Inventory; NMES: Neuromuscluar Electrical Stimulation; N/S: not specified; OD: Oropharyngeal Dysphagia; PAS: Penetration-Aspiration-Scale; PD: Parkinson's Disease; POE: Pleasure of Eating; rTMS: repetitive Transcranial Magnetic Stimulation; SDQ: Swallowing Disturbance Questionnaire; SES: Surface Electrical Stimulation; SSA: Standardized Swallowing Assessment; SWAL-CARE: Swallowing Quality of Care Questionnaire; SWAL-QOL: Swallowing Quality of Life Questionnaire; QoL: Quality of Life; RCT: Randomized Controlled Trial; UPDRS: Unified Parkinson's Disease Rating Scale; VDS: Videofluoroscopic Dysphagia Scale; VFS: Videofluoroscopy of Swallowing; V1 / V2: Visit 1 / Visit

Appendix 4.5: Excluded reports with reason

#	Study	Reason for exclusion
1	Antonello, N., & Grecchi, B. (2019). EFFECTIVENESS OF AN EXPIRATORY FLOW ACCELERATION DEVICE IN DYSPHAGIC PATIENTS WITH PARKINSON DISEASE. Chest, 156(4), A1784–A1785. https://doi.org/10.1016/j.chest.2019.08.1549	Congress abstract of Riboldazzi et al. 2020
2	Baijens, L., Speyer, R., & Pilz, W. (2011). The effect of surface electrical stimulation on swallowing in Parkinson's disease. Dysphagia, 26(4), 462. https://doi.org/10.1007/s00455-011-9345-1	Congress abstract on Baijens et al. 2012
3	Baijens, L. W. J., Speyer, R., Passos, V. L., Pilz, W., Van Der Kruis, J., Haarmans, S., & Desjardins-Rombouts, C. (2014). Surface electrical stimulation in dysphagic parkinson patients: A Randomized clinical trial. Dysphagia, 29(3), 404. https://doi.org/10.1007/s00455-014-9536-7	Comment on Baijens et al. 2013
4	Bird, M., Woodward, M., Gibson, E., Phyland, D., & Fonda, D. (1996). The effect of levodopa on swallowing in Parkinson's disease: Are there benefits in administration before meals? AUSTRALIAN JOURNAL ON AGEING, 15(1), 27–29. https://doi.org/10.1111/j.1741-6612.1996.tb00196.x	Wrong patient population (PD without OD)
5	Bushmann, M., Dobmeyer, S. M., Leeker, L., & Perlmutter, J. S. (1989). Swallowing abnormalities and their response to treatment in Parkinson's disease. Neurology, 39(10), 1309–1314. https://doi.org/10.1212/wnl.39.10.1309	No clinical intervention
6	Claus, I., Muhle, P., Suttrup, J., Schroeder, J. B., Suntrup-Krueger, S., Dziewas, R., & Warnecke, T. (2020). Expiratory muscle strength training (EMST) for treatment of pharyngeal dysphagia in Parkinson's disease: A randomized controlled trial. Dysphagia, 35(1), 143. https://doi.org/10.1007/s00455-019-10078-x	Congress abstract on Claus et al. 2021
7	De Graaf, L. (2007). Loss of saliva due to dysphagia: Patients with Parkinson's disease benefit from explanation and training. *Pharmaceutisch Weekblad*, *142*(18), 29–31.	No clinical intervention
8	FENG Qing-ling, CHENG Yuan-yuan, LIU Pei-pei, ZHOU Xiao-na, WANG Yue, CHEN Rong-jie, YU Yang, & WU Jia-ling. (2019). Effects of vocalization training on drooling severity in Parkinson's disease. *Chinese Journal of Contemporary Neurology & Neurosurgery*, *19*(11), 891–896.	Congress abstract on Feng et al. 2019
9	Jost, W. H., Michel, O., Oehlwein, C., Slawek, J., Bogucki, A., Ochudlo, S., Banach, M., Pagan, F., Flatau-Baqué, B., Csikós, J., & Blitzer, A. (2018). Efficacy of incobotulinumtoxinA in subjects with sialorrhea, assessed using the modified radboud oral motor inventory for Parkinson's disease (mROMP). Toxicon, 156, S53–S54. https://doi.org/10.1016/j.toxicon.2018.11.127	Congress abstract on Jost et al. 2019
10	Jost, W. H., Friedman, A., Michel, O., Oehlwein, C., Slawek, J., Bogucki, A., Ochudlo, S., Banach, M., Pagan, F., Flatau-Baqué, B., Dorsch, U., Csikós, J., & Blitzer, A. (2020). Long-term incobotulinumtoxinA treatment for chronic sialorrhea: Efficacy and safety over 64 weeks. Parkinsonism & Related Disorders, 70, 23–30. https://doi.org/10.1016/j.parkreldis.2019.11.024	Irrelevant outcomes not related to those specified in this review
11	Kitashima, A., Umemoto, G., Tsuboi, Y., Higuchi, M., Baba, Y., & Kikuta, T. (2013). Effects of subthalamic nucleus deep brain stimulation on the swallowing function of patients with Parkinson's disease. Parkinsonism & Related Disorders, 19(4), 480–482. https://doi.org/10.1016/j.parkreldis.2012.10.023	No control group
12	Kondo, E., Takeda, N., & Jinnouchi, O. (2017). Aural stimulation with capsaicin ointment improved the swallowing function in patients with dysphagia: A randomized, placebo-controlled, double-blind, comparative study. Dysphagia, 32(6), 817. https://doi.org/10.1007/s00455-017-9805-3	Congress abstract on Kondo et al. 2017
13	Manor, Y., Freud, D., Mootanah, R., Gurevich, T., Giladi, N., & Jacob, C. T. (2010). Video assisted swallowing therapy improved quality of life measures more than traditional swallowing therapy in patients with Parkinson's disease. Movement Disorders, 25, S427. https://doi.org/10.1002/mds.23162	Congress abstract on Manor et al. 2013
14	Marks, L., Turner, K., O'Sullivan, J., Deighton, B., & Lees, A. (2001). Drooling in Parkinson's disease: A novel speech and language therapy intervention. International Journal of Language & Communication Disorders, 36 Suppl, 282–287. https://doi.org/10.3109/13682820109177898	Wrong patient population (PD without OD)
15	Merello, M., Starkstein, S., Nouzeilles, M. I., Kuzis, G., & Leiguarda, R. (2001). Bilateral pallidotomy for treatment of Parkinson's disease induced corticobulbar syndrome and psychic akinesia avoidable by globus pallidus lesion combined with contralateral stimulation. Journal of Neurology, Neurosurgery, and Psychiatry, 71(5), 611–614. https://doi.org/10.1136/jnnp.71.5.611	Irrelevant outcomes not related to those specified in this review

16	Necati, E., Demir, N., Serel Arslan, S., Eker, A., Kaymakamzade, B., & Karaduman, A. A. (2020). Effect of kinesio taping on swallowing function in parkinson patients: A surface electromyographic swallowing study. Turkish Journal of Physiotherapy and Rehabilitation, 31(2), 202–209. Embase. https://doi.org/10.21653/tjpr.540557	Wrong patient population (PD without OD)
17	Nozaki, S., Matsui, T., Yoshikawa, H., Kaneto, T., Domen, K., & Daimon, T. (2013). Metronome therapy to treat dysphagia in patients with parkinson's disease. Dysphagia, 28(2), 298. https://doi.org/10.1007/s00455-013-9455-z	Full text not available (contact via the email address received through ESSD not possible, message is blocked; contaced Jglobal database, where article is listed; they have not been able to find a site where the full text of this paper can be accessed)
18	Nozaki, S., Sugishita, S., Imai, T., Oguro, D., Inokawa, M., Matsui, T., Umaki, Y., Kaneto, T., Domen, K., & Yoshikawa, H. (2011). Training for dysphagia with metronome improves swallowing function in parkinson disease. Dysphagia, 26(4), 443. https://doi.org/10.1007/s00455-011-9345-1	Full text not available (contacted DRS; no email address received)
19	Oh, E., Sookyong, B., Soyoung, A., & Sungjoo, J. (2019). Developing silver food which is easy to swallow in patients with Parkinson's disease. Journal of Parkinson's Disease, 9(1), 180–181. Embase. https://doi.org/10.3233/JPD-199900	Congress abstract on Oh et al. 2021
20	Oh, E., Jee, S., Kim, B. K., Lee, J. S., Cho, K., & Ahn, S. (2021). A new swallowing supplement for dysphagia in patients with Parkinson's disease. Neurological Sciences : Official Journal of the Italian Neurological Society and of the Italian Society of Clinical Neurophysiology, 42(5), 1949–1958. https://doi.org/10.1007/s10072-020-04730-w	No control group
21	Ondo, W. G., Hunter, C., & Moore, W. (2004). A double-blind placebo-controlled trial of botulinum toxin B for sialorrhea in Parkinson's disease. Neurology, 62(1), 37–40. https://doi.org/10.1212/01.wnl.0000101713.81253.4c	Irrelevant outcomes not related to those specified in this review
22	Pflug, C., Nienstedt, J. C., Gulberti, A., Müller, F., Vettorazzi, E., Koseki, J.-C., Niessen, A., Flügel, T., Hidding, U., Buhmann, C., Weiss, D., Gerloff, C., Hamel, W., Moll, C. K. E., & Pötter-Nerger, M. (2020). Impact of simultaneous subthalamic and nigral stimulation on dysphagia in Parkinson's disease. Annals of Clinical and Translational Neurology, 7(5), 628–638. https://doi.org/10.1002/acn3.51027	Wrong patient population (PD without OD)
23	Restivo, D. A., Palmeri, A., & Marchese-Ragona, R. (2002). Botulinum toxin for cricopharyngeal dysfunction in Parkinson's disease. The New England Journal of Medicine, 346(15), 1174–1175. https://doi.org/10.1056/NEJM200204113461517	No clinical intervention
24	Riboldazzi, G., Spinazza, G., Beccarelli, L., Prato, P., Grecchi, B., D'Abrosca, F., & Nicolini, A. (2020). Effectiveness of expiratory flow acceleration in patients with Parkinson's disease and swallowing deficiency: A preliminary study. Clinical Neurology and Neurosurgery, 199, 106249. https://doi.org/10.1016/j.clineuro.2020.106249	Irrelevant outcomes not related to those specified in this review
25	Saleem, A., Sapienza, C., Rosenbek, J., Musson, N., & Okun, M. (2005). The effects of expiratory muscle strength training program on pharyngeal swallowing in patients with idiopathic Parkinson's disease. *MOVEMENT DISORDERS*, *20*, S73–S74.	Congress abstract, full text not available; most likely on Troche et al. 2010
26	Saleem, A., Sapienza, C., Rosenbek, J., Musson, N., & Okun, M. (2005). The effects of expiratory muscle strength training on pharyngeal swallowing in patients with idiopathic Parkinson's disease. *NEUROLOGY*, *64*(6), A397–A397.	Congress abstract, full text not available; most likely on Troche et al. 2010
27	Sapienza, C. M., Troche, M., Silverman, E. P., Rosenbek, J., & Musson, N. (2012). Strength training outcomes for airway protection in PD. Movement Disorders, 27, S139. https://doi.org/10.1002/mds.25051	Congress abstract, full text not available; most likely on Troche et al. 2010
28	Sapienza, C., Troche, M., Silverman, E., Rosenbek, J., Musson, N., & Okun, M. (2011). Strength training outcomes for airway protection in PD. Neurodegenerative Diseases, 8. https://doi.org/10.1159/000327701	Congress abstract, full text not available; most likely on Troche et al. 2010
29	Sordoni, E., Andrenelli, E., Di Biagio, L., Millevolte, M., Ceravolo, M. G., & Capecci, M. (2014). Comparative efficacy and safety of botulinum toxin type A and B in treating Parkinson's disease-related sialorrhea: A pre-test post-test study. Annals of Physical and Rehabilitation Medicine, 57, e363–e364. https://doi.org/10.1016/j.rehab.2014.03.1215	Congress abstract, full text not available, contacted first and second author, awaiting response
30	Sundstedt, S., Nordh, E., Linder, J., Hedström, J., Finizia, C., & Olofsson, K. (2017). Swallowing Quality of Life After Zona Incerta Deep Brain Stimulation. The Annals of Otology, Rhinology, and Laryngology, 126(2), 110–116. https://doi.org/10.1177/0003489416675874	Wrong patient population (PD without OD)
31	Tawadros, P., Cordato, D., Cathers, I., & Burne, J. (2012). An Electromyographic Study of Parkinsonian Swallowing and Its Response to Levodopa. MOVEMENT DISORDERS, 27(14), 1811–1815. https://doi.org/10.1002/mds.25262	No control group

32	Troche, M., Okun, M., Rosenbek, J., Musson, N., & Sapienza, C. (2009). Swallow outcomes following intervention with expiratory muscle strength training (EMST) in Parkinson's disease: Results of a randomized clinical trial. Dysphagia, 24(4), 455–456. https://doi.org/10.1007/s00455-009-9233-0	Congress abstract on Troche et al. 2010
33	Troche, M., Rosenbek, J., Okun, M., & Sapienza, C. (2011). Swallowing and breathing related outcomes following detraining in EMST. Dysphagia, 26(4), 443–444. Embase. https://doi.org/10.1007/s00455-011-9345-1	Congress abstract on Troche et al. 2010
34	Wu, M.-X., Wang, L.-G., Li, H.-P., & Zeng, X. (2021). [Acupuncture adjuvant treatment for dysphagia in patients with Parkinson's disease: A randomized controlled trial]. Zhongguo zhen jiu = Chinese acupuncture & moxibustion, 41(5), 485–488. https://doi.org/10.13703/j.0255-2930.20200724-0002	Abstract, full text not available
35	Xie, T., Bloom, L., Padmanaban, M., Bertacchi, B., Kang, W., Dachman, A., MacCracken, E., Zadikoff, C., Markopoulou, K., Warnke, P., & Kang, U. (2018). Long-term effect of low frequency stimulation of STN on dysphagia, freezing of gait and other motor symptoms in PD. *Movement Disorders*, *33*, S263.	Congress abstract on Xie et al. 2018
36	Xie, T., Bloom, L., Padmanaban, M., Takahashi, K., Kang, W., Dachman, A., MacCracken, E., & Warnke, P. (2021). Impact of oral textures on aspiration and changes in swallow dynamics in patients with PD with DBS. Journal of Neurology, Neurosurgery & Psychiatry, 92(4), 447–449. https://doi.org/10.1136/jnnp-2020-324579	Same participants and study as in Xie et al. 2018, different outcomes, but not a full report of the study, "letter to the editor"
37	陈艳红, 孙涛, & 陈敏. (2017). 门德尔松手法联合康复训练治疗帕金森病病人吞咽障碍的疗效观察. Chinese Nursing Research, 31(7), 864–866. ccm. https://doi.org/10.3969/j.issn.1009-6493.2017.07.032	Abstract, full text not available

Appendix 4.6: Excluded outcomes with reason

#	Author, Year	Verbatim	Definition	Measure-ment	Timepoints	Frequency	Reason for exclusion
1	Ayres, 2017	Prior state of secretion in nasopharyngeal structures, oropharynx, laryngopharynx	N/S	FEES	pre, post (4w)	2	Observations that were made, but not measured and not reported
2	Ayres, 2017	Thickening on the posterior laryngeal wall	Presence/absence of thickening on the posterior laryngeal wall	FEES	pre, post (4w)	2	Observations that were made, but not measured and not reported
3	Ayres, 2017	Tremor in structures (BOT, vallecula)	Presence/absence of tremor in structures (BOT, valleculae)	FEES	pre, post (4w)	2	Observations that were made, but not measured and not reported
4	Ayres, 2017	Early escape	Characterized by the presence of food in the hypopharynx or larynx before the swallowing reflex was triggered	FEES	pre, post (4w)	2	Observations that were made, but not measured and not reported
5	Ayres, 2017	Vallecular stasis in glossoepiglottic folds and pyriform sinus	Characterized by accumulation of food after the third swallowing on the mentioned structures	FEES	pre, post (4w)	2	Observations that were made, but not measured and not reported
6	Ayres, 2017	Penetration	Characterized by the presence of food in the laryngeal vestibule	FEES	pre, post (4w)	2	Observations that were made, but not measured and not reported
7	Ayres, 2017	Tracheal aspiration	Characterized by food intake in the region located below the vocal folds, in the subglottic region and in the trachea, at any time of swallowing	FEES	pre, post (4w)	2	Observations that were made, but not measured and not reported
8	Ayres, 2017	Cough reflex	Presence/absence	FEES	pre, post (4w)	2	Observations that were made, but not measured and not reported
9	Ayres, 2017	Ability to manipulate flows	Presence/absence	CSE	pre, post (4w)	2	Observations that were made, but not measured and not reported
10	Ayres, 2017	Postural control	Presence/absence	CSE	pre, post (4w)	2	Observations that were made, but not measured and not reported
11	Ayres, 2017	Fatiguability	Presence/absence	CSE	pre, post (4w)	2	Observations that were made, but not measured and not reported
12	Ayres, 2017	Anatomy and oral, pharyngeal and laryngeal physiology	Presence/absence	CSE	pre, post (4w)	2	Observations that were made, but not measured and not reported
13	Ayres, 2017	Orofacial tones	Presence/absence	CSE	pre, post (4w)	2	Observations that were made, but not measured and not reported
14	Ayres, 2017	Oral apraxia	Presence/absence	CSE	pre, post (4w)	2	Observations that were made, but not measured and not reported
15	Ayres, 2017	Orofacial sensitivity	Presence/absence	CSE	pre, post (4w)	2	Observations that were made, but not measured and not reported

16	Ayres, 2017	Gag pharyngeal contraction	Presence/absence	CSE	pre, post (4w)	2	Observations that were made, but not measured and not reported
17	Ayres, 2017	Saliva swallowing	Presence/absence	CSE	pre, post (4w)	2	Observations that were made, but not measured and not reported
18	Ayres, 2017	Cough and hawk	Presence/absence	CSE	pre, post (4w)	2	Observations that were made, but not measured and not reported
19	Ayres, 2017	Swallowing apraxia	Presence/absence	CSE	pre, post (4w)	2	Observations that were made, but not measured and not reported
20	Ayres, 2017	Oral residue	Presence/absence	CSE	pre, post (4w)	2	Observations that were made, but not measured and not reported
21	Ayres, 2017	Delayed swallowing reflex	Presence/absence	CSE	pre, post (4w)	2	Observations that were made, but not measured and not reported
22	Ayres, 2017	Reduction in laryngeal elevation	Presence/absence	CSE	pre, post (4w)	2	Observations that were made, but not measured and not reported
23	Ayres, 2017	Wet voice	Presence/absence	CSE	pre, post (4w)	2	Observations that were made, but not measured and not reported
24	Ayres, 2017	Multiple swallowing	Presence/absence	CSE	pre, post (4w)	2	Observations that were made, but not measured and not reported
25	Ayres, 2017	History of aspiration pneumonia	Presence/absence	CSE	pre, post (4w)	2	Observations that were made, but not measured and not reported
26	Ayres, 2017	Intake of food	Level of oral food intake	FOIS	pre, post (4w)	2	Observations that were made, but not measured and not reported
27	Ayres, 2017	Interaction attention/ability	Presence/absence	CSE	pre, post (4w)	2	Observations that were made, but not measured and not reported
28	Ayres, 2017	Alert state	presence/absence	CSE	pre, post (4w)	2	Observations that were made, but not measured and not reported
29	Ayres, 2017	Awareness of the swallowing problem	Presence/absence	CSE	pre, post (4w)	2	Observations that were made, but not measured and not reported
30	Ayres, 2017	Awareness of secretion	presence/absence	CSE	pre, post (4w)	2	Observations that were made, but not measured and not reported
31	Feng, 2019	Consciousness	N/S	SSA	pre, post (4w)	2	Observations that were made for overall SSA score
32	Feng, 2019	Head and trunk control	N/S	SSA	pre, post (4w)	2	Observations that were made for overall SSA score
33	Feng, 2019	Lip closure	N/S	SSA	pre, post (4w)	2	Observations that were made for overall SSA score
34	Feng, 2019	Voice strength	N/S	SSA	pre, post (4w)	2	Observations that were made for overall SSA score
35	Feng, 2019	Gag reflex	N/S	SSA	pre, post (4w)	2	Observations that were made for overall SSA score

36	Feng, 2019	Laryngeal movement	Presence/absence by observation of the patient drinking (scale from 18-46)	SSA	pre, post (4w)	2	Observations that were made for overall SSA score
37	Feng, 2019	Repetitve swallowing	Presence/absence by observation of the patient drinking (scale from 18-46)	SSA	pre, post (4w)	2	Observations that were made for overall SSA score
38	Feng, 2019	Laryngeal function after swallowing	Presence/absence by observation of the patient drinking (scale from 18-46)	SSA	pre, post (4w)	2	Observations that were made for overall SSA score
39	Feng, 2019	Time required to swallow	Presence/absence by observation of the patient drinking (scale from 18-46)	SSA	pre, post (4w)	2	Observations that were made for overall SSA score
40	Feng, 2019	Breathing	clinical examination (scale from 18-46)	SSA	pre, post (4w)	2	Observations that were made for overall SSA score
41	Feng, 2019	Wheezing during swallowing	presence/absence by observation of the patient drinking (scale from 18-46)	SSA	pre, post (4w)	2	Observations that were made for overall SSA score
42	Heijnen, 2012	N/S	N/S	FEES	N/S	N/S	No specification of outcomes being measured
43	Heijnen, 2012	N/S	N/S	VFS	N/S	N/S	No specification of outcomes being measured
44	Khedr, 2019	Diagnosis of dysphagia	- N/S (a score on the SDQ of ≥ 11 indicates dysphagia.)	SDQ	pre	1	Is reported as an outcome but was only conducted for assessing dysphagia pre treatment for inclusion
45	Silva-Arone, 2021	N/S	N/S	VFS: Eisenhuber scale	pre, post (3m, 6m)	3	No specification, outcome was only reported in unpublished Master thesis, referenced by Battel et al. (2020)
46	Wei, 2017	Mis-inhalation	N/S	N/S	post (6m)	1	No definition of the outcome, unclear to what this relates and what was measured

CSE: Clinical Swallow Evaluation; FEES: Fibreoptic Evaluation of Swallowing; N/S: not specified; SDQ: Swallowing Disturbance Questionnaire; SSA: Standardized Swallow Assessment; VFS: Videofluorscopy of Swallowing; w: weeks

Appendix 4.7: Definitions of outcomes

Outcome	Definition
Death	N/S
Oropharyngeal dysphagia severity	N/S
Drooling	Severity and frequency of drooling
Salivary pooling	Salivary pooling in vallecula and pyriform sinuses
Orolingual bolus control	3x N/S; Preswallow loss of bolus from lips/into pharynx; Premature spillage: materials spilled over the base of the tongue into the hypopharynx (including the valleculae, the lateral channels, and the piriform sinus) too early during the oral swallowing stage, meaning before the pharyngeal swallow was initiated.
Oral bolus transport	Swallow hesitancy; delayed onset oral transport
Swallowing related lingual movement pattern	Preswallow involuntary repetitive tongue movements
Timing of oropharyngeal swallow components	GPJo (glossopalatal junction opening): N/S
	GPJc (glossopalatal junction closure): N/S
	VPJo (velopharyngeal junction opening): Moment of separation of soft palate and posterior pharyngeal wall with re-entry of air in retrolingual space from nasopharynx (in seconds)
	VPJc (velopharyngeal junction closure): Moment of separation of soft palate and posterior pharyngeal wall with re-entry of air in retrolingual space from nasopharynx (in seconds)
	VPJd (velopharyngeal junction duration): ▲T between VPFc and VPJo (in seconds)
	Lvo (laryngeal vestibule opening): Moment of separation of arytenoid cartilages and underside of epiglottis with re-entry of air in laryngeal vestibule (in seconds)
	LVc (laryngeal vestibule closure): Moment when laryngeal elevation results in making contact between arytenoid cartilages and underside of epiglottis (in seconds)
	LVd (laryngeal vestibule duration): ▲T between LVc and LVo (in seconds)

	UESo (upper esophageal sphincter opening): N/S
	UESc (upper esophageal sphincter closure): Moment of closure of esophagus after bolus transport (in seconds)
	GPJo (glossopalatal junction opening) – LVc (laryngeal vestibule closure): ▲T between GPJo and LVc (in seconds)
	Duration horizontal hyoid motion: Duration between initiation of swallow and moment of maximum horizontal (anterior) motion (in seconds)
	Duration vertical hyoid motion: Duration between initiation of swallow and moment of maximum vertical motion (in seconds)
	Pharyngeal transit time (PTT): From the point where the bolus head moved from the hold position and passed the posterior nasal spine until it fully entered the esophagus after the closure of the upper esophageal sphincter (in seconds)
	Time required for max elevation of the hyoid bone: H2-H1: The time of the first superior-anterior movement of the hyoid bone was assigned as H1, and the time when the hyoid bone reached its maximum elevation was assigned as H2.
	Bolus flow time: Measurement of the time when the bolus is seen in the hypopharynx until it triggers the swallowing reflex (0 = 0-1 s, 1 = 2-4 s, 2= 5-7 s, 3 = 8 + s)
	Oral transit time (OTT): N/S
	Pharyngeal transit time (PTT): N/S
	Pharyngeal response time (PRT): N/S
	Duration of hyoid movement: measurement tags at 1) the initiation of hyoid movement which resulted in the swallow and 2) the point when the hyoid returned to rest following the completion of the swallow. These tags were then used to calculate the duration of hyoid (in seconds) movement.
Laryngeal elevation	N/S
Laryngeal sensation	The response of glottal closure reflex induced by toughing epiglottis with endoscope; Glottal closure and cough reflexes induced by touching epiglottis or arytenoids with endoscope
Piecemeal deglutition	Sequential swallowing of same bolus
Swallowing related hyoid bone movement	Movement patterns of hyoid bone: Anterior/superior corner of hyoid bone (x axis), anterior/inferior corner of third and fifth cervical vertebral bodies (y axis)
	Extent of movement of hyoid bone: Extent of movement in x-y coordinate system over time
	Vertical hyoid motion: Maximum vertical motion during swallowing act (in mm)

	Horizontal displacement of the hyoid bone: The distance (in cm) from the resting position to the maximal excursion position during swallowing; the most supero-anterior point of the hyoid indicates maximum displacement after swallowing
	Vertical displacement of the hyoid bone: The distance (in cm) from the resting position to the maximal excursion position during swallowing; the most supero-anterior point of the hyoid indicates maximum displacement after swallowing
	Hyoid displacement - Onset of bolus transit: Bolus head arrival at posterior edge of ramus of mandible
	Hyoid displacement - UES-opening: Forward displacement of cricoid cartilage from posterior pharyngeal wall
	Hyoid displacement - UES-widset: Widest part of bolus head passing through UES
	Hyoid displacement - UES-closure: Last point when UES is open
	Hyoid displacement - Laryngeal closure: Forward displacement of arytenoid cartilage to epiglottic petiole
	Hyoid displacement - Max laryngeal closure: Maximum contact of arytenoid cartilages with epiglottic petiole
	Hyoid displacement - Laryngeal opening: First separation of arytenoid cartilages from epiglottic petiole
Initiation of pharyngeal swallow	Delayed initiation pharyngeal triggering; the location of the bolus at the time of swallow onset; Initiation of swallowing reflex; Bolus location when the swallowing reflex is triggered; Initiation time of the swallowing reflex
Postswallow oral residue	1x N/S, Pooling in oral cavity after the swallow
Postswallow pharyngeal residue	1x N/S; Material was insufficiently cleared from the hypopharynx during swallowing and remained after swallowing; The extent of pharyngeal clearance after swallowing; Residue location of the food
Postswallow pharyngeal pooling	Postswallow pooling in valleculae; Pooling in valleculae after the swallow; Pooling in pyriform sinuses after the swallow
Piecemeal deglutition	Sequential swallowing of same bolus
Penetration/Aspiration	2x N/S; Definitions of penetration and aspiration according to Rosenbek et al. 1996, Penetration and/or aspiration; Penetration: material entered the laryngeal vestibule (defined by Langmore's epiglottis level 3) but remained at or above the level of the vocal cords; Aspiration: material entered the airway below the vocal cords; Penetration/aspiration of food/liquid before/after swallowing
Cortical reorganization	N/S
Motor evoked potential (MEP)	Amplitude: N/S Latency: the time in milliseconds from the point at which a TMS pulse was delivered to the onset of a MEP

Overall motor symptoms	N/S
Tremor	N/S
Rigidity	N/S
Bradykinesia	N/S
Axial symptoms	N/S
Freezing of gait	Number of Freezing of gait spells, time to complete test
Phonation	N/S
Loudness	N/S
Level of oral intake	N/S
Aspiration pneumonia	Chest radiography: evidence of pneumonia Clinical evaluation: 3 or more of the following: sustained fever (temperature > 100 °F [38 °C]), rales or rhonchi on chest auscultation, sputum Gram stain showing substantial leukocytes, or sputum culture showing a respiratory pathogen
Acitivities of daily living	N/S
Pleasure of oral intake	Patient's degree of enjoyment from food
Swallowing related quality of life	N/S
Self-percepetion of swallowing	4x N/S; Changes in patient subjective dysphagia symptoms as well as swallowing-related quality of life
Self-perception of walking	Patient's perception of walking difficulty
Self-perception of activities of daily living	N/S
Patient's satisfaction with intervention	1x N/S; Preference for different interventions
Patient's adherence to intervention	N/S
Hospitalization	N/S

Adverse events	Any clinically significant event possibly related to the assigned intervention (for example, dehydration), no report of events expected as part of the participant's disease progression or aging process (for example, worsening of Parkinson disease symptoms), all adverse events were rated as mild, moderate, severe, or life-threatening

N/S: not specified; T: time; TMS: Transcranial Magnetic Stimulation; UES: upper esophageal sphincter

Appendix 4.8: Outcome measurement instruments

Outcome	Measurement
Death	N/S
Oropharyngeal dysphagia severity	VFS: DOSS (O'Neil et al., 1999); FDS (Han et al., 1999); VDS-total; -oral; -pharyngeal (Kim et al., 2012) FEES: score 0-108; sum of 5 scores; sum of 4 scores Dysphagia scale (no reference, from Wei et al. 2017); SSA (Perry et al., 2001); CSE
Drooling	UPDRS II: 5-point scale DSFS: 5- and 4-point scale (Thomas-Stonell & Greenberg, 1988)
Salivary pooling	FEES: 4-point scale
Orolingual bolus control	VFS: 5-point scale FEES: 3-point-scale, 5-point scale
Oral bolus transport	VFS: 3-point scale
Swallowing related lingual movement	VFS: 5-point scale
Timing of oropharyngeal swallow components	VFS FEES: 4-point scale
Laryngeal elevation	FEES: 3-point scale
Laryngeal sensation	FEES: touching epiglottis or arytenoids with endoscope, 4-point scale
Swallowing related hyoid bone movement	VFS: in milli- or centimeters
Initiation of pharyngeal swallow	VFS: 3-point scale FEES: endoscopic whiteout, 4-point scale; 3-point scale EMG (submental muscles) in milliseconds
Postswallow oral residue	VFS: 5-point scale
Postswallow pharyngeal residue	VFS: 3-point scale FEES: 4-point scale; 5-point scale
Postswallow pharyngeal pooling	VFS: 3-point scale FEES: 3-point scale
Piecemeal deglutition	VFS: 5-point scale FEES: 5-point scale
Penetration/Aspiration	VFS: PAS, 8-point scale (Rosenbek et al., 1996) FEES: PAS, 8-point scale (Rosenbek et al., 1996); penetration/aspiration before/after swallowing, 4-point scale
Cortical reorganization	Magnetoencephalography: data filtered within theta (4–8 Hz), alpha (8–13 Hz), beta (13–30 Hz), low-gamma, (30–60 Hz), high-gamma (60–80 Hz) frequency bands; in all frequency bands, performance of source localization of each subject's swallowing-associated event-related desynchronization (ERD) of cortical rhythms
Motor evoked potential	Electromyography

Overall motor symptoms	H&Y; UPDRS III
Tremor	UPDRS III
Rigidity	UPDRS III
Bradykinesia	UPDRS III
Axial symptoms	UPDRS III
Freezing of gait	Stand-walk-test (no reference, from Xie et al., 2018)
Phonation	Maximum phonation time on /aa/ in seconds
Loudness	Maximum phonation decibel (dB) on /aa/
Level of oral intake	FOIS (Crary et al., 2005)
Aspiration pneumonia	Chest radiography Clinical evaluation: ≥ 3 of the following: sustained fever (temperature > 100 °F [38 °C]), rales or rhonchi on chest auscultation, sputum Gram stain showing substantial leukocytes, or sputum culture showing a respiratory pathogen
Activities of daily living	IADL (Lawton et al., 1969)
Pleasure of oral intake	POE (Manor et al., 2013)
Swallowing related quality of life	SWAL-QOL (McHorney et al., 2002), MDADI (Chen et al., 2001)
Self-perception of swallowing	SDQ (Manor et al., 2007), DSS (Tohara et al., 2004), A-DHI (Farahat et al., 2014)
Self-perception of walking	FOG questionnaire (Giladi et al., 2009)
Self-perception of activities of daily living	Activities of Daily Living Scale (Brown et al., 1989)
Patient's satisfaction with intervention	Interview: rated as easy/pleasant, average, or difficult/unpleasant SWAL-CARE (McHorney et al., 2002)
Patient's adherence to intervention	Assessed across meals, classified monthly as 0% - 25%, 26% - 50%, 51% - 75%, or 76% - 100%
Hospitalization	N/S
Adverse events	Clinical evaluation

DHI = Arabic Dysphagia Handicap Index, CSE = Clinical Swallow Evaluation, DOSS = Dysphagia Outcome and Severity Scale, DSFS = Drooling Severity and Frequency Scale, DSS = Dysphagia Severity Scale, EMG = Electromyography, FDS = Functional Dysphagia Scale, FEES = Fibreoptic Endoscopic Evaluation of Swallowing, FOG = Freezing of Gait, FOIS = Functional Oral Intake Scale, H&Y = Hoehn & Yahr, IADL = Instrumental Activities of Daily Living, MDADI = MD Anderson Dysphagia Inventory, N/S = not specified, PAS = Penetration-Aspiration-Scale, POE = Pleasure of Eating, SDQ = Swallowing Disturbance Questionnaire, SSA = Standardized Swallowing Assessment, SWAL-CARE = Swallowing Quality of Care Questionnaire, SWAL-QOL = Swallowing Quality of Life Questionnaire, UPDRS = Unified Parkinson's Disease Rating Scale, VDS = Videofluoroscopic Dysphagia Scale, VFS = Videofluorscopic Evaluation of Swallowing

Appendix 4.9: Timepoints and frequency of measurement

Outcome	Timepoints of measurement	Frequency of measurement
Death	continuously during 3m	continously
Oropharyngeal dysphagia severity	pre, post (5min, 30min, 60min, 2w, 1m, 3m, 6m)	1 to 3
Drooling	pre, post (4w)	2
Salivary pooling	pre, post (5min, 30min, 60min)	4
Orolingual bolus control	during, pre, post (5min, 30min, 60min, 15d, 1m, 3m)	1 to 4
Oral bolus transport	during, pre, post (15d)	1 to 2
Swallowing related lingual movement	pre, post (15d)	2
Timing of oropharyngeal swallow components	during, pre, post (0min, 2w, 4w)	1 to 2
Laryngeal elevation	pre, post (5min, 30min, 60min)	4
Laryngeal sensation	pre, post (5min, 30min, 60min)	4
Swallowing related hyoid bone movement	during, pre, post (4w)	1 to 2
Initiation of pharyngeal swallow	during, pre, post (0d, 5min, 30min, 60min, 15d, 2w)	1 to 4
Postswallow oral residue	during, pre, post (15d)	1 to 2
Postswallow pharyngeal residue	pre, post (5min, 30min, 60min, 2w, 1m, 3m)	2 to 4
Postswallow pharyngeal pooling	during, pre, post (15d)	1 to 2
Piecemeal deglutition	during, pre, post (15d)	1 to 2
Penetration/Aspiration	during, pre, post (0min, 15d, 2w, 1m, 3m) V1: during; V2 (at least 6m later): during	1 to 2
Cortical reorganization	pre, post (1m)	2
Motor evoked potential (MEP)	pre, post (0min, 30min)	3
Overall motor symptoms	pre, post (2w, 1m, 2m, 3m) V1: during; V2 (at least 6m later): during	2 to 5
Tremor	V1: during; V2 (at least 6m later): during	2
Rigidity	V1: during; V2 (at least 6m later): during	2
Bradykinesia	V1: during; V2 (at least 6m later): during	2

Axial symptoms	V1: during; V2 (at least 6m later): during	2
Freezing of gait	V1: during; V2 (at least 6m later): during	2
Phonation	pre, post (4w)	2
Loudness	pre, post (4w)	2
Level of oral intake	pre, post (3m, 6m)	3
Aspiration pneumonia	continuously during 3m	continously
Acitivities of daily living	pre, post (2w, 1m, 2m, 3m)	5
Pleasure of oral intake	pre, post (2w, 4w)	3
Swallowing related quality of life	pre, post (1m, 3m, 6m)	2 to 3
Self-percepetion of swallowing	pre, post (2w, 1m, 2m, 3m, 6m) 2 x pre, after each session (13-15), 2 x post V1: post (30 min); V2 (at least 6m later): post (30min)"	2 to 15
Self-perception of walking	V1: during; V2 (at least 6m later): during	2
Self-perception of activities of daily living	pre, post (2w, 1m, 2m, 3m)	5
Patient's satisfaction with intervention	during, post (2w)	1
Patient's adherence to intervention	weekly	36
Hospitalization	continuously during 3m	continously
Adverse events	continuously during 3m	continously

min: minutes; d: days; w: weeks; m: months; V1: visit 1; V2: visit 2

Appendix 5.1: Checklist for Reporting Of Survey Studies (CROSS)

Section/topic	Item	Item description	Reported on page #
Title and abstract			
Title and abstract	1a	State the word "survey" along with a commonly used term in title or abstract to introduce the study's design.	1
	1b	Provide an informative summary in the abstract, covering background, objectives, methods, findings/results, interpretation/discussion, and conclusions.	1
Introduction			
Background	2	Provide a background about the rationale of study, what has been previously done, and why this survey is needed.	2
Purpose/aim	3	Identify specific purposes, aims, goals, or objectives of the study.	2
Methods			
Study design	4	Specify the study design in the methods section with a commonly used term (e.g., cross-sectional or longitudinal).	3
	5a	Describe the questionnaire (e.g., number of sections, number of questions, number and names of instruments used).	3
Data collection methods	5b	Describe all questionnaire instruments that were used in the survey to measure particular concepts. Report target population, reported validity and reliability information, scoring/classification procedure, and reference links (if any).	3
	5c	Provide information on pretesting of the questionnaire, if performed (in the article or in an online supplement). Report the method of pretesting, number of times questionnaire was pre-tested, number and demographics of participants used for pretesting, and the level of similarity of demographics between pre-testing participants and sample population.	3
	5d	Questionnaire if possible, should be fully provided (in the article, or as appendices or as an online supplement).	SI 3
Sample characteristics	6a	Describe the study population (i.e., background, locations, eligibility criteria for participant inclusion in survey, exclusion criteria).	3-4
	6b	Describe the sampling techniques used (e.g., single stage or multistage sampling, simple random sampling, stratified sampling, cluster sampling, convenience sampling). Specify the locations of sample participants whenever clustered sampling was applied.	4
	6c	Provide information on sample size, along with details of sample size calculation.	4
	6d	Describe how representative the sample is of the study population (or target population if possible), particularly for population-based surveys.	4
Survey administration	7a	Provide information on modes of questionnaire administration, including the type and number of contacts, the location where the survey was conducted (e.g., outpatient room or by use of online tools, such as SurveyMonkey).	3

	7b	Provide information of survey's time frame, such as periods of recruitment, exposure, and follow-up days.	3
	7c	Provide information on the entry process:	N/A
		–>For non-web-based surveys, provide approaches to minimize human error in data entry.	
		–>For web-based surveys, provide approaches to prevent "multiple participation" of participants.	
Study preparation	8	Describe any preparation process before conducting the survey (e.g., interviewers' training process, advertising the survey).	3-4
Ethical considerations	9a	Provide information on ethical approval for the survey if obtained, including informed consent, institutional review board [IRB] approval, Helsinki declaration, and good clinical practice [GCP] declaration (as appropriate).	3
	9b	Provide information about survey anonymity and confidentiality and describe what mechanisms were used to protect unauthorized access.	3
Statistical analysis	10a	Describe statistical methods and analytical approach. Report the statistical software that was used for data analysis.	4
	10b	Report any modification of variables used in the analysis, along with reference (if available).	N/A
	10c	Report details about how missing data was handled. Include rate of missing items, missing data mechanism (i.e., missing completely at random [MCAR], missing at random [MAR] or missing not at random [MNAR]) and methods used to deal with missing data (e.g., multiple imputation).	N/A
	10d	State how non-response error was addressed.	N/A
	10e	For longitudinal surveys, state how loss to follow-up was addressed.	N/A
	10f	Indicate whether any methods such as weighting of items or propensity scores have been used to adjust for non-representativeness of the sample.	N/A
	10g	Describe any sensitivity analysis conducted.	N/A
Results			
Respondent characteristics	11a	Report numbers of individuals at each stage of the study. Consider using a flow diagram, if possible.	7
	11b	Provide reasons for non-participation at each stage, if possible.	N/A
	11c	Report response rate, present the definition of response rate or the formula used to calculate response rate.	N/A
	11d	Provide information to define how unique visitors are determined. Report number of unique visitors along with relevant proportions (e.g., view proportion, participation proportion, completion proportion).	N/A
Descriptive results	12	Provide characteristics of study participants, as well as information on potential confounders and assessed outcomes.	7-9
Main findings	13a	Give unadjusted estimates and, if applicable, confounder-adjusted estimates along with 95% confidence intervals and p-values.	N/A

	13b	For multivariable analysis, provide information on the model building process, model fit statistics, and model assumptions (as appropriate).	N/A
	13c	Provide details about any sensitivity analysis performed. If there are considerable amount of missing data, report sensitivity analyses comparing the results of complete cases with that of the imputed dataset (if possible).	N/A
Discussion			
Limitations	14	Discuss the limitations of the study, considering sources of potential biases and imprecisions, such as non-representativeness of sample, study design, important uncontrolled confounders.	22
Interpretations	15	Give a cautious overall interpretation of results, based on potential biases and imprecisions and suggest areas for future research.	21-22
Generalizability	16	Discuss the external validity of the results.	22
Other sections			
Role of funding source	17	State whether any funding organization has had any roles in the survey's design, implementation, and analysis.	Title page
Conflict of interest	18	Declare any potential conflict of interest.	Title page
Acknowledgements	19	Provide names of organizations/persons that are acknowledged along with their contribution to the research.	Title page

Note: indicated page numbers refer to pages on manuscript submitted to journal

Appendix 5.2: Focus groups consent form

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

COS-DIP
Core Outcome Set for Dysphagia Interventions in Parkinson's disease

Development of a Core Outcome Set for Dysphagia Interventions in Parkinson's disease: the COS-DIP Study

Focus Groups Consent Form

We invite you to take part in a focus group interview. Your feedback is important. This interview will take approximately 60 minutes.

Please initial the box if you agree with the statement. Please feel free to ask questions if there is something you do not understand. The end of this form is for the researchers to complete. Thank you for participating.

General	**Tick box**
I confirm I have read and understood the **Participant Information Leaflet** for the above study. The information has been fully explained to me and I have been able to ask questions, all of which have been answered to my satisfaction.	
I understand that this study **is entirely voluntary, and if I decide that I do not want to take part, I can stop taking part in this study at any time without giving a reason**. I understand that deciding not to take part will not affect my future medical care.	
I agree to taking part in an **online video meeting**. I understand that no video recording will be made.	
I agree to the interview being **audio recorded**.	
I agree **to introduce myself at the beginning of the interview** to the researcher and other participants and say my first name, my role (person living with Parkinson's, carer or family member), where I am from and which age range I belong to. This information will not be disclosed to anyone else other than the present interview members and researcher.	
I understand that I **will not be paid for taking part in this study**.	
I understand that there are **no direct benefits** to me from participating in this study, which has been set out in full in the information leaflet which I have been provided with.	
I know how to contact the research team if I need to.	
I agree to being contacted by researchers by email as part of this study.	

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COS-DIP
Core Outcome Set for Dysphagia Interventions in Parkinson's disease

Data Processing	**Tick box**
I understand the study is being conducted by researchers from Trinity College Dublin (TCD) and my personal information will be stored electronically on the TCD server and will be protected in accordance with the General Data Protection Regulation.	
By taking part in this interview, I agree to allow the researchers to anonymously publish the research ideas that I help them to identify. Anonymised means that my personal information will be removed, and it will not be possible to link me as an individual to any of the ideas generated.	
I agree to take part in this focus group interview.	

Participant Name (Block Capitals) Participant Signature Date

To be completed by the Principal Investigator.

I, the undersigned, have taken the time to fully explain to the above participant the nature and purpose of this study in a way that they could understand. I have explained the risks and possible benefits involved. I have invited them to ask questions on any aspect of the study that concerned them.
I have given a copy of the information leaflet and consent form to the participant with contacts of the study team.

Researcher name: Julia Hirschwald
Signature:
Date:

Appendix 5.3: Long list of outcomes with lay terms used in the survey and focus groups

Outcome	Lay term
Oropharyngeal dysphagia severity	Changes in overall swallowing severity scores
Self-perception of swallowing	Changes in overall awareness of swallowing function
Initiation of pharyngeal swallow	Changes in overall speed of swallowing
Level of oral intake	Changes in how food and liquids are taken (e.g., via mouth, feeding tube, pureed diet, thickened liquids)
Dry mouth	Changes in how the mouth feels in terms of dryness
Drooling	Changes in frequency and severity of saliva leaking out of the mouth
Salivary pooling	Changes in amount of saliva accumulated in the mouth or throat
Swallowing related orolingual movement	Changes in tongue "pumping" or "rocking" movements when eating or drinking
Orolingual bolus control	Changes in control over food or liquids in the mouth
Piecemeal deglutition	Changes in need to break down food or liquids in the mouth in order to swallow
Oral bolus transport	Changes in speed of food or liquids passing through the mouth
Nasal regurgitation	Changes in frequency of food or liquids going the wrong way - into the nose
Penetration/Aspiration	Changes in frequency of food or liquids going the wrong way - into the airway/windpipe
Laryngeal sensation	Changes in feeling of food or liquids being stuck in the throat
Choking	Changes in frequency of choking episodes

Post swallow oral residue	Changes in food or liquids left in the mouth after swallowing
Post swallow pharyngeal residue	Changes in food or liquids left in the throat after swallowing
Cough production	Changes in cough strength so the airway/windpipe can be cleared easier
Cough sensitivity	Changes in coughing as a response of feeling food or liquids going down the wrong way
Lingual muscle strength	Changes in strength of tongue muscles
Pharyngeal muscle strength	Changes in strength of throat muscles
Weight loss	Changes in body weight related to swallowing difficulties
Medication intake	Changes in swallowing Parkinson's medication
Pneumonia	Changes in frequency of chest infections
Hospitalization	Changes in number of hospital admissions related to swallowing difficulties
Swallowing related quality of life	Changes in quality of life related to swallowing
Carer burden	Changes in caregiver's quality of life related to swallowing in person with Parkinson's
Fear of eating / drinking	Changes in fear or anxiety related to eating and drinking
Pleasure of oral intake	Changes in pleasure of eating and drinking related to swallowing
Avoidance of food / liquids	Changes in avoidance of food or liquids related to swallowing
Changes in voice, speech	Changes in other areas related to swallowing muscles, e.g., voice or speech
Adverse events	Other unfavorable changes or events that occur due to swallowing treatment
Participant's adherence to intervention	Person with Parkinson's following the intervention as prescribed
Participant's experience with intervention	Opportunity to give feedback on overall swallowing program

Appendix 5.4: Online survey questionnaire

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Coláiste na Tríonóide, Baile Átha Cliath
The University of Dublin

COS-DIP
Core Outcome Set for Dysphagia Interventions in Parkinson's disease

Development of a Core Outcome Set for Dysphagia Interventions in Parkinson's disease: the COS-DIP Study

Online Survey

Dear participant,

We are seeking people living with Parkinson's disease and swallowing difficulties, their caregivers and family members to take part in our study.

Why is this research being done?
Research studies on swallowing treatments need to be able to measure how well the treatment works. Researchers do this by measuring 'outcomes'. Examples of outcomes include 'quality of life' and 'severity of swallowing difficulty'.
However, so far researchers have been using different outcomes when conducting these studies. Some for example measure quality of life and others do not. Furthermore, people like you (living with Parkinson's disease and swallowing difficulties, carers and family members) have not been involved in deciding what to measure in these research studies. Thus, the results of the research might have little importance to you.
We are doing this study to find out what outcomes are important to you when deciding if a swallowing treatment works. If you like you can watch this short video here, where we explain the purpose of this research project and what it involves.

What does this study involve?
This anonymous online survey will take less than 10 minutes to complete. No identifying information is required in this survey. Participation is completely voluntary and even if you agree to participate, you can withdraw from the survey at any time until the point of submission.
The survey will remain open until **10 April 2023**. The results of the survey will inform further stages of the COS-DIP project and will be published and presented fully anonymous at scientific conferences in the future.

Who can take part?
People living with Parkinson's disease and experiencing swallowing difficulties, their carers and family members ≥ 18 years of age.

Who can NOT take part?
People with other conditions than Parkinson's potentially causing swallowing difficulties (for example Progressive Supranuclear Palsy (PSP), Multiple System

Atrophy (MSA), stroke, head and neck cancer, muscular diseases, etc.).

What are the benefits of partaking in this study?
While taking part in this study may not directly benefit you now, the information you share with us will provide valuable data for Parkinson's disease researchers and potentially improve the quality of studies testing swallowing treatments for people with Parkinson's disease in the future. The results of this project will also better inform clinicians on the most appropriate information they should be gathering in treatments with people with Parkinson's disease and swallowing difficulties. This in turn will benefit future people living with Parkinson's disease and swallowing difficulties.

If you have any queries about the study, more information may be obtained from Julia Hirschwald (cosdip.tcd@gmail.com) or Prof. Margaret Walshe (walshema@tcd.ie).

Follow up focus groups will be carried out to explore this topic in more detail. If you would like to take part in this, please email Julia Hirschwald (cosdip.tcd@gmail.com).

Thank you very much for contributing to this research study.

Julia Hirschwald
Dr. Sallyanne Duncan
Prof. Tobias Warnecke
Gary Boyle
Prof. Julie Regan
Prof. Margaret Walshe

By continuing to the next page, you confirm that:

1. You have read the information on the previous page.
2. You understand and have had enough time to consider the information.
3. You understand that your participation is voluntary and that you are free to withdraw at any time by choosing to not complete the survey.

Do you wish to proceed with the survey: yes/no

Q1: Please indicate your current role(s). Check all that apply:
   a. Person living with Parkinson's disease and swallowing difficulties
   b. Caregiver of a person with Parkinson's disease and swallowing difficulties
   c. Family member of a person with Parkinson's disease and swallowing difficulties

2. Which age group do you belong to?
   a. 18-30
   b. 31-45
   c. 46-60
   d. 61-75
   e. 76-90
   f. over 90 years
3. What age were you when you were diagnosed with Parkinson's disease? Please enter in number of years (e.g. 70).
   OR
   What age was the person with Parkinson's disease at the time of diagnosis? Please enter in number of years (e.g. 70).
4. Approximately what year were you diagnosed with Parkinson's disease? Please enter the year of diagnosis (e.g. 2015).
   OR
   Approximately what year did the person with Parkinson's disease receive the diagnosis Parkinson's disease? Please enter the year of diagnosis (e.g. 2015).
5. How do you describe yourself?
   a. Man
   b. Woman
   c. Non-binary
   d. None of the above, I identify as *(text box)*
   e. Prefer not to answer
6. In which country do you primarily reside? *Drop down menu*
7. I complete this survey:
   a. On my own
   b. With help of someone
   c. On behalf of someone

If you receive a swallowing treatment, what is important to you in terms of deciding if the swallowing treatment has worked?

OR

If the person with Parkinson's disease receives a swallowing treatment, what is important to you in terms of deciding if the swallowing treatment has worked?

Please indicate which items in the list below are <u>important</u> and which are <u>not important</u> to you by ticking the according box.

	Important	Not important
Changes in overall swallowing severity scores		
Changes in overall awareness of swallowing function		
Changes in overall speed of swallowing		
Changes in how food and liquids are taken (e.g., via mouth, feeding tube, pureed diet, thickened liquids)		
Changes in how the mouth feels in terms of dryness		
Changes in frequency and severity of saliva leaking out of the mouth		
Changes in amount of saliva accumulated in the mouth or throat		
Changes in tongue "pumping" or "rocking" movements when eating or drinking		
Changes in control over food or liquids in the mouth		
Changes in need to break down food or liquids in the mouth in order to swallow		
Changes in speed of food or liquids passing through the mouth		
Changes in frequency of food or liquids going the wrong way - into the nose		
Changes in frequency of food or liquids going the wrong way - into the airway/windpipe		
Changes in feeling of food or liquids being stuck in the throat		
Changes in frequency of choking episodes		
Changes in food or liquids left in the mouth after swallowing		
Changes in food or liquids left in the throat after swallowing		
Changes in cough strength so the airway/windpipe can be cleared easier		
Changes in coughing as a response of feeling food or liquids going down the wrong way		
Changes in strength of tongue muscles		
Changes in strength of throat muscles		

Changes in body weight related to swallowing difficulties		
Changes in swallowing Parkinson's medication		
Changes in frequency of chest infections		
Changes in number of hospital admissions related to swallowing difficulties		
Changes in quality of life related to swallowing		
Changes in caregiver's quality of life related to swallowing in person with Parkinson's		
Changes in fear or anxiety related to eating and drinking		
Changes in pleasure of eating and drinking related to swallowing		
Changes in avoidance of food or liquids related to swallowing		
Changes in other areas related to swallowing muscles, e.g., voice or speech		
Other unfavourable changes or events that occur due to swallowing treatment		
Person with Parkinson's following the intervention as prescribed		
Opportunity to give feedback on overall swallowing program		

Is there anything that was not listed but is important to you for deciding if a swallowing treatment has worked? *Comment box*

Thank you very much for participating. You completed the survey.

In a next step we will conduct group interviews to better understand the survey results and to learn why these things are important to you. We are currently seeking ethics approval for these interviews. If you are potentially interested in participating, please contact Julia Hirschwald via email cosdip.tcd@gmail.com.

If you have any further questions, please do not hesitate to also contact Julia Hirschwald via email cosdip.tcd@gmail.com.

If you would like to be informed about the results of this survey, please register your interest by clicking here.

Appendix 5.5: Recruitment poster for online survey



Appendix 5.6: Recruitment poster for focus groups



Appendix 5.7: Participant information leaflet for focus groups

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

COS-DIP
Core Outcome Set for Dysphagia Interventions in Parkinson's disease

Participant Information Leaflet

for patients, carers & family members

Study title	**Development of a Core Outcome Set for Dysphagia Interventions in Parkinson's disease: the COS-DIP Study**
Research stage	**Focus group interviews**
Site	**Trinity College Dublin, Ireland**
Principal Investigator Co-Investigators	**Ms. Julia Hirschwald** (cosdip.tcd@gmail.com) **Dr. Sallyanne Duncan** **Prof. Tobias Warnecke** **Mr. Gary Boyle** **Prof. Julie Regan** **Prof. Margaret Walshe** (walshema@tcd.ie)
Data Controllers	**Trinity College Dublin**
Data Protection Officer	**Data Protection Officer, Secretary's Office** **Trinity College Dublin, Dublin 2, Ireland**

You are being invited to take part in a research study. Before you decide whether or not you wish to take part, please read this information sheet carefully. It is important that you understand why the research is being done and what it will involve so that you can make a decision that is right for you. Please do not feel rushed or under pressure to make a quick decision. You can ask Julia Hirschwald any questions. You may wish to discuss it with your family, friends, GP or neurologist.

This leaflet has four parts: (1) The Study, (2) Data Protection, (3) Costs, Funding and Approval, (4) Further Information

Part 1 – The Study

Why is this study being done?

Swallowing difficulties (dyphagia) are common in people with Parkinson's disease (PD) and can cause dehydration and poor nutrition. It may also impact on quality of life. Food and liquids can enter the airway or lungs, leading to choking or possible lung infections. Research into treating swallowing difficulties in people with PD has increased in recent years. Research studies of swallowing treatments

need to be able to measure how well the treatment works, but also what side effects or risks it may have and how they may affect patients. Researchers do this by measuring 'outcomes'. Examples of outcomes include 'quality of life' or 'severity of swallowing difficulty'.

However, so far researchers have been using different outcomes when conducting studies. Some for example measure quality of life and others do not. Furthermore, people like you (living with PD and swallowing difficulties, carers and family members) have not been involved in deciding what to measure in these research studies. Thus, the results of the resarch might have little importance to you.

We are doing this study to find out what the most important outcomes are when deciding if a swallowing treatment works and why these are important to you. Therefore, we are gathering the opinions of people who live with PD and experience swallowing difficulties, their caregivers and family members. We do this to later develeop a minimum standard (a core outcome set) of *what* should be measured and *how* it should be measured in studies on swallowing treatments for people with PD.

Why have I been invited to take part?

You are being invited to participate in this study because you have Parkinson's disease and swallowing difficulties or you provide care for a person with PD and swallowing difficulties or you are a family member of a person with PD and swallowing difficulties. We are intending to involve up to 32 participants.

Do I have to take part? Can I withdraw?

You don't have to take part in this study. **It is entirely voluntary**. If you decide not to take part, it will not affect your current or future medical care. You can change your mind about taking part in the study and opt out at any time, even if the study has started. You don't have to give a reason for not taking part or for opting out. If you wish to opt out, please contact Julia Hirschwald (cosdip.tcd@gmail.com), who will be able to organise this for you.

What happens if I change my mind?

You can change your mind at any time by contacting Julia Hirschwald (cosdip.tcd@gmail.com) and we will be able to organise this for you. If you choose not to continue to take part, this will not affect your medical care in any way. If you wish, you can ask for your data stored to be destroyed. If you request this, we will destroy all data that are still in our possession. We will no longer use or share your data for research from this point onwards. However, it will not be possible to destroy data already used in research studies prior to this time.

How will the study be carried out?

The interview will take part online on June 20th or 22nd 2023. We will conduct up to four group interviews (so called focus groups) with 6-8 participants each. If you wish to participate in one of these group interviews we will get in contact with you to organise a day and time for the online interviews.

What will happen to me if I decide to take part?

If you agree to participate you will take part in **one group interview** together with five to seven people with PD and swallowing difficulties, carers and family members. You will be invited to take part online via a video conference software. The group interview normally takes around 60 minutes. Julia Hirschwald will first ask you to introduce you to the group and say your first name, your role (person with PD, carer or family member), where you are from and which age range you belong to. Thereafter, she will ask you questions about swallowing difficulties your or the person with PD are experiencing, how this may have changed your or the person with PD's life and how you might decide if a swallowing treatment works. You will be able to discuss this within the group and Julia Hirschwald will moderate the discussion. During the interview, we will ask you to turn on your camera, if you feel comfortable with this. The group conversation will only be audio-recorded (one audio file for each participant), not video-recorded. The audio recording helps the researchers as it is difficult to take notes of what people say, listen carefully and think all at the same time. After the interview, the audio-recording is typed up so that we have a full and accurate account of the views that were presented. Thereafter, the audio-recording will be deleted.

What will happen to my data?

All collected data will be coded and pseudonymised. We will note your name, and a participant number only on your consent form. This will be kept separately from all other data. All other data will be coded with a participant number. This way you will not be identifiable. Your identity will not be disclosed to anyone outside of the research team. In the final report of this study no identifiable details will be published or disclosed. All data on electronic devices will be password protected and encrypted.

Are there any benefits to taking part in this research?

Taking part in this study will not directly benefit you. However, this research project will provide valuable information for PD researchers and potentially improve the quality of studies testing swallowing treatments for people with PD in the future. The results of this project will also better inform clinicians on the most appropriate information they should be gathering in treatments with people with PD and swallowing difficulties. This in turn will benefit future patients living with PD and swallowing difficulties.

Are there any disadvantages or risks to me if I take part?

You might find it upsetting to talk about your experiences of living with Parkinson's disease and swallowing difficulties. If you do not wish to answer some questions that is alright. You can stop the conversation at any time you wish.

The only possible risk to taking part in this study is that a connection to your identity could be made as we will have your name and contact details. However, this information will be kept in a separate folder and will be stored securely in a locked filing cabinet. Great care will be taken to ensure the confidentiality of all data. The risk to participants of a breach of confidentiality is considered very low.

What happens if something goes wrong when I'm taking part in the study?

If you have any concerns about any aspects of the study, you can contact Prof. Margaret Walshe (walshema@tcd.ie).

What will happen to the results of the research?

If you wish, we will send you the interview transcripts so that you can check if you agree with it. If you should not agree with the interview transcripts, you can email us with suggestions for change. The results of the study will be reported in medical and scientific journals and presented at (inter-) national medical and scientific conferences. No information revealing your identity will be disclosed. If you wish to be informed about the final results will we will send you a summary of the results, once the focus groups are finished and the results are analysed.

Part 2 – Data Protection

What information about me (personal data) will be used as part of this study? Will my medical records be accessed?

No, your medical records will not be accessed. We will use the following information about you: contact details to arrange an interview, the audio recording of your interview and the final transcript of the audio recordings.

We will use the following information about you (personal data) for this research study: gender, age, nationality and years since diagnosis of PD. Any information will be labelled with a code instead of your name prior to sharing for this research project. This is to protect your identity.

What will happen to my personal data?

Only your personal data essential for the project will be processed in order to achieve the objective of this study. Your personal data will not be processed in a way that can cause damage or distress to you.

Your contact details and audio recordings will be immediately destroyed after the data have been transcribed and have been agreed on with you. The coded transcripts and your personal data will be pseudonymised and the stored pseudonymised and the key code linking personal data with research data will be stored separately from each other for one year after the study is completed and written up.

The collected study information will be kept securely in a password protected and encrypted file on the TCD One Drive. In August 2026, all collected information will be destroyed.

Who will access and use my personal data as part of this study?

Only Julia Hirschwald and Prof. Margaret Walshe will have access to your personal data as part of this study.

Will my personal data be kept confidential? How will my data be kept safe?

Your privacy is important to us. We take many steps to make sure that we protect your confidentiality and keep your data safe. Here are some examples of how we do this:

- Your consent form with your personal data will be stored and locked in a filing cabinet.
- You will not be identifiable in any presentations or publications in relation to this study.
- All principal investigators and co-investigators who have access to your personal data are bound by a professional code of secrecy.
- Training in data protection law and practice has been provided to those individuals involved in carrying out the research.

What is the lawful basis to use my personal data?

By law,[1] we can use your personal information for scientific research[2] (in the public interest[3]). We will also ask for your explicit consent to use your data as a requirement of the Irish Health Research Regulations.

What are my rights?

You are entitled to:

- The right to access to your data and receive a copy of it

[1] The European General Data Protection Regulation (GDPR)
[2] Article 9(2) (j)
[3] Article 6(1)(e)

- The right to restrict or object to processing of your data
- The right to object to any further processing of the information we hold about you (except where it is de-identified)
- The right to have inaccurate information about you corrected or deleted
- The right to receive your data in a portable format and to have it transferred to another data controller
- The right to request deletion of your data

By law you can exercise the following rights in relation to your personal data, unless the request would make it impossible or very difficult to conduct the research. You can exercise these rights by contacting your Julia Hirschwald (hirschwj@tcd.ie) or the Trinity College Data Protection Officer, Secretary's Office, Trinity College Dublin, Dublin 2, Ireland. Email: dataprotection@tcd.ie. Website: www.tcd.ie/privacy.

Part 3 – Costs, Funding and Approval

Has this study been approved by a research ethics committee?

Yes, this study has been approved by the Faculty of Health Sciences Research Ethics Committee of Trinity College Dublin, Dublin 2, Ireland. Approval was granted on 15 May 2023 [HT27]. None of the persons carrying out this study have a link to the committees. Julia Hirschwald is a PhD student and Prof. Margaret Walshe an Associate Professor at Trinity College Dublin.

Who is organising and funding this study? Will the results be used for commercial purposes?

A short version of this study is publicly available at www.comet-initiative.org/Studies/Details/1942. This research is conducted by Julia Hirschwald for the purpose of obtaining the degree of Doctor in Philosophy under the supervision of Prof. Margaret Walshe and Dr. Sallyanne Duncan. Prof. Julie Regan, Mr. Gary Boyle and Prof. Tobias Warnecke are part of the study steering committee and take on an advisory role in this research project. Julia Hirschwald undertakes this research project as part of a Provost's PhD Award funded by Trinity College Dublin, Ireland. The results will not be used for commercial purposes.

Is there any payment for taking part? Will it cost me anything if I agree to take part?

No, neither will we pay you to take part in the study nor will you have to pay to take part.

Part 4 – Further Information

Who should I contact for information or complaints?

If you have any concerns or questions, you can contact:

- Julia Hirschwald: cosdip.tcd@gmail.com, Prof. Margaret Walshe: walshema@tcd.ie
- Data Protection Officer, Trinity College Dublin: Data Protection Officer, Secretary's Office, Trinity College Dublin, Dublin 2, Ireland. Email: dataprotection@tcd.ie. Website: www.tcd.ie/privacy.

Under GDPR, if you are not satisfied with how your data is being processed, you have the right to lodge a complaint with the Office of the Data Protection Commission, 21 Fitzwilliam Square South, Dublin 2, Ireland. Website: www.dataprotection.ie.

Will I be contacted again?

If you would like to take part in this study, you will be asked to sign the consent attached to the email. In case you wish to be informed about further research, please inform Julia Hirschwald (cosdip.tcd@gmail.com).

Appendix 5.8: Semi-structured focus group interview guide

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

COS-DIP
Core Outcome Set for Dysphagia Interventions in Parkinson's disease

Development of a Core Outcome Set for Dysphagia Interventions in Parkinson's disease: the COS-DIP Study

I. Semi structured interview guide

Interview Schedule

1. Introduction
2. Reminder of the purpose of the COS-DIP study
3. Participants will be given the opportunity to ask questions
4. Participants will be asked if they are still willing to participate
5. Participants will be asked if they agree to the interview being audio recorded
6. Introduction
7. Participants will be asked specific interview questions
8. All things considered question
9. Thank you and goodbye

1. Introduction

A warm welcome to our interview session today. We are very grateful that you have taken the time to participate in our focus groups. This session is part of a study we are carrying out to develop a "Core Outcome Set". One of the main delays in the improvement and progression of care for people with Parkinson's and swallowing difficulties is the differences in what things researchers look at to see if a swallowing treatment works or not. These 'things' are called outcomes. If different outcomes are measured in different trials, they can't be compared, and this slows progress in knowing how to improve patient care. Another problem is that often the research carried out does not incorporate the values and opinions of people with Parkinson's and swallowing disorders, their carers and family members. The development of our Core Outcome Set (COS) will:

- Standardise what should be measured in research studies for treating swallowing difficulties in people with Parkinson's disease
- Incorporate the opinions of people living with Parkinson's disease and swallowing difficulties and their carers and family members

Show explainer video

2. Reminder of the purpose of the COS-DIP study

We have previously conducted an online survey on the perspectives of people with Parkinson's disease and swallowing difficulties, carers and family members on what outcomes they think are most important and what should be measured when deciding if a swallowing treatment works. Maybe you participated in this survey as well.

With these focus groups we aim to get a deeper understanding of the findings and would like to clarify some inconclusive results. We also would like to understand why these outcomes are important to you as people living with Parkinson's disease and swallowing difficulties, carers and family members.

After the focus groups, we will compile a list of outcomes that we will then use to develop a questionnaire. This we will then circulate to a wider group also including healthcare professionals and researchers. Therefore, we really want your experiences and perspectives to influence these outcomes.

3. **Participants will be given the opportunity to ask questions**

Before we begin, do you have any questions?

4. **Participants will be asked if they are still willing to participate**

We would like to take this opportunity before we begin to make sure you are still happy to take part in this focus group. If you do not want to participate anymore, don't worry, you can simply let us know now and leave the virtual meeting room. This will not have any medical or any other consequences for you, and we will not use any of your data in our research.

If at any stage you would like a break or the questions asked are too difficult or uncomfortable to answer, please just let us know. There is no problem at all.

5. **Participants will be asked if they agree to the interview being audio recorded**

We will be recording this interview (only the sound) as it is difficult for me to write everything down that you say and moderate the discussion at the same time. We will store the recording safely until we have written it up. Afterwards we will delete the audio recording. We will not mention your names or anything that individually identifies you in anything we do.

6. **Introduction**

I would now kindly like to ask everyone to introduce themselves. Please let us know your name, your role (person living with Parkinson's or a carer or family member), when the PD diagnosis was received, where you are from and what age range you belong to.

Show a slide with the information, so that everyone can remember what to say when introducing themselves

7. **Participants will be asked specific questions (***will be adapted according to survey responses***)**

I. If you think about eating and drinking, what matters most to you?
II. How do the swallowing difficulties manifest in your case or the person with PD?
III. What do you think has changed due to the swallowing difficulties?
    a. How has this impacted on your life or the person's life with PD?
    b. What have you possibly lost due to the swallowing difficulties?
IV. What do you think a swallowing treatment should do for you or the person with PD?

Appendix 5.9: Open text answers by survey participants on what outcomes are considered important but missing in the long list of outcomes

Type of answer	Content	# of participants
General feedback	Not having received swallowing treatment so far	2
	Not knowing that swallowing treatment existed but noticing a decline in swallowing ability, describing only the symptoms	1
	Not assuming that swallowing difficulties are due to PD	1
Outcomes already included in long list	Other unfavorable changes or events that occur due to swallowing treatment	1
	Changes in food or liquids left in the throat after swallowing	1
New outcomes	Changes in fear of choking	4
	Changes of related consequences of fear of choking, e.g., fear of going to sleep	1
	Changes in impact on social functioning	1
	Changes in what and how to eat	1
	Changes in frequency of alternate liquid solid swallows	1
	Changes in ability to communicate during mealtimes	1
	Changes in globus sensation	1
	Method and frequency of outcome measurement, e.g. repeated instrumental assessments	1
	Treatment efficiency	1
	Amount of required commitment to treatment	1
	Context of the delivery of the treatment, e.g., by an experienced and skilled SLP	1
	Impact on others due to the treatment, e.g., due to noise	1

PD Parkinson's disease, *SLP* Speech and Language Pathologist

Appendix 6.1: Checklist for Reporting Of Survey Studies (CROSS)

Section/topic	Item	Item description	page #
Title and abstract			
Title and abstract	1a	State the word "survey" along with a commonly used term in title or abstract to introduce the study's design.	Title page
	1b	Provide an informative summary in the abstract, covering background, objectives, methods, findings/results, interpretation/discussion, and conclusions.	1
Introduction			
Background	2	Provide a background about the rationale of study, what has been previously done, and why this survey is needed.	2
Purpose/aim	3	Identify specific purposes, aims, goals, or objectives of the study.	2-3
Methods			
Study design	4	Specify the study design in the methods section with a commonly used term (e.g., cross-sectional or longitudinal).	3
	5a	Describe the questionnaire (e.g., number of sections, number of questions, number and names of instruments used).	3
Data collection methods	5b	Describe all questionnaire instruments that were used in the survey to measure particular concepts. Report target population, reported validity and reliability information, scoring/classification procedure, and reference links (if any).	3-4
	5c	Provide information on pretesting of the questionnaire, if performed (in the article or in an online supplement). Report the method of pretesting, number of times questionnaire was pre-tested, number and demographics of participants used for pretesting, and the level of similarity of demographics between pre-testing participants and sample population.	3-4
	5d	Questionnaire, if possible, should be fully provided in the article, or as appendices or as an online supplement.	Suppl. Material 2
Sample characteristics	6a	Describe the study population (i.e., background, locations, eligibility criteria for participant inclusion in survey, exclusion criteria).	5
	6b	Describe the sampling techniques used (e.g., single stage or multistage sampling, simple random sampling, stratified sampling, cluster sampling, convenience sampling). Specify the locations of sample participants whenever clustered sampling was applied.	5
	6c	Provide information on sample size, along with details of sample size calculation.	5

	6d	Describe how representative the sample is of the study population (or target population if possible), particularly for population-based surveys.	N/A
Survey administration	7a	Provide information on modes of questionnaire administration, including the type and number of contacts, the location where the survey was conducted (e.g., outpatient room or by use of online tools, such as SurveyMonkey).	4
	7b	Provide information of survey's time frame, such as periods of recruitment, exposure, and follow-up days.	4
	7c	Provide information on the entry process:	
		→ For non-web-based surveys, provide approaches to minimize human error in data entry.	N/A
		→ For web-based surveys, provide approaches to prevent "multiple participation" of participants.	5
Study preparation	8	Describe any preparation process before conducting the survey (e.g., interviewers' training process, advertising the survey).	5
Ethical considerations	9a	Provide information on ethical approval for the survey if obtained, including informed consent, institutional review board [IRB] approval, Helsinki declaration, and good clinical practice [GCP] declaration (as appropriate).	3
	9b	Provide information about survey anonymity and confidentiality and describe what mechanisms were used to protect unauthorized access.	3
Statistical analysis	10a	Describe statistical methods and analytical approach. Report the statistical software that was used for data analysis.	6
	10b	Report any modification of variables used in the analysis, along with reference (if available).	N/A
	10c	Report details about how missing data was handled. Include rate of missing items, missing data mechanism (i.e., missing completely at random [MCAR], missing at random [MAR] or missing not at random [MNAR]) and methods used to deal with missing data (e.g., multiple imputation).	N/A
	10d	State how non-response error was addressed.	N/A
	10e	For longitudinal surveys, state how loss to follow-up was addressed.	N/A
	10f	Indicate whether any methods such as weighting of items or propensity scores have been used to adjust for non-representativeness of the sample.	N/A
	10g	Describe any sensitivity analysis conducted.	N/A
Results			
Respondent characteristics	11a	Report numbers of individuals at each stage of the study. Consider using a flow diagram, if possible.	7
	11b	Provide reasons for non-participation at each stage, if possible.	N/A
	11c	Report response rate, present the definition of response rate or the formula used to calculate response rate.	7

	11d	Provide information to define how unique visitors are determined. Report number of unique visitors along with relevant proportions (e.g., view proportion, participation proportion, completion proportion).	N/A
Descriptive results	12	Provide characteristics of study participants, as well as information on potential confounders and assessed outcomes.	7-16
Main findings	13a	Give unadjusted estimates and, if applicable, confounder-adjusted estimates along with 95% confidence intervals and p-values.	N/A
	13b	For multivariable analysis, provide information on the model building process, model fit statistics, and model assumptions (as appropriate).	N/A
	13c	Provide details about any sensitivity analysis performed. If there are considerable amount of missing data, report sensitivity analyses comparing the results of complete cases with that of the imputed dataset (if possible).	N/A
Discussion			
Limitations	14	Discuss the limitations of the study, considering sources of potential biases and imprecisions, such as non-representativeness of sample, study design, important uncontrolled confounders.	19
Interpretations	15	Give a cautious overall interpretation of results, based on potential biases and imprecisions and suggest areas for future research.	19-20
Generalizability	16	Discuss the external validity of the results.	N/A

N/A = not applicable

Note: indicated page numbers refer to pages on manuscript submitted to journal

Appendix 6.2: Online survey – English version

Q1

Dear Participant,
We are seeking **Dysphagia Clinicians** who work with clients/patients who have **Parkinson's disease** to take part in an **online survey**.

Who can take part?
A **dysphagia clinician** is anyone who has experience in implementing **oropharyngeal dysphagia (OD) interventions** in people with **Parkinson's Disease**, e.g., speech and language pathologists (SLPs) who conduct swallowing interventions.

To take part in this study, the dysphagia clinician must:

- work with clients/patients who have **Idiopathic Parkinson's disease**
- are proficient in one or more of the following languages:
    - English
    - Italian
    - German
    - Mandarin Chinese (simplified)
    - Spanish

Why is this research being carried out?

Research studies on swallowing treatments need to be able to measure how well the treatment works. Researchers do this by measuring 'outcomes'. Examples of outcomes include 'quality of life' and 'severity of swallowing difficulty'. However, so far researchers have been using different outcomes when conducting these studies. Furthermore, key stakeholders such as dysphagia clinicians have not been involved in deciding what to measure in these research studies. Thus, the results of the research may have little importance in clinical practice. We are carrying out this study to find out what outcomes are important to you (as a dysphagia clinician) when deciding if a swallowing treatment works.

What does this study involve?

This anonymous online survey will take less than 15 minutes to complete.
The survey will remain open until 14th August 2023.

What about GDPR (data protection rules)?

No personal data is collected. No identifying information is required in this survey. Participation is completely **voluntary.** As the data subject, you have the option to withdraw consent at any time until the point of submission.

What are the benefits of taking part in this study?

While taking part in this study may not directly benefit you now, having Dysphagia Clinicians' perspectives and opinions considered will help **guide the formation of a Core Outcome Set (COS) for dysphagia interventions in Parkinson's Disease** which will **benefit clinicians and researchers** in this field in the long-run. The formation of a COS will make research more **comparable and clinically useful.**

This could help clinicians in improving communication of outcomes and better records of changes in patients' dysphagia. This in turn will benefit future people living with Parkinson's disease and swallowing difficulties. It could potentially improve the quality of studies testing swallowing treatments for people with Parkinson's disease in the future. The **results** of the survey may be **published and presented at scientific conferences** in the future. **No identifying information will be provided.**

Sharing this Survey

At the end, you will be given the option to share this survey. This is optional.
If you have **any queries** about the study, more information may be obtained by contacting from us.

Thank you very much for taking the time to read this.

By clicking or tapping the "Agree" button, you confirm that:

- You have read the information on the previous page.
- You understand and have had enough time to consider the information.
- You understand that your participation is voluntary and that you are free to withdraw at any time by choosing to not complete the survey. Due to the anonymous nature of this survey, you will not be able to edit or withdraw your responses after submission.

○ Agree

End of Block: Participant Information Leaflet

Start of Block: Perspectives

Q2 What outcome measures do you use when assessing swallowing?
Check all that apply and please feel free to add any that are missing.

- ☐ Penetration aspiration scale (Rosenbek et al., 1996)
- ☐ Functional Oral Intake Scale (Crary, Carnaby Mann & Groher, 2005)
- ☐ ROMP Saliva (Kalf, Borm, de Swart, Bloem, Zwarts & Munneke,2011)
- ☐ SWAL-QOL questionnaire (McHorney et al., 2002)
- ☐ Dysphagia Severity Rating Scale (Gramigna, 2006; Waxman et al., 1990)
- ☐ MD Anderson Dysphagia Inventory (MDADI) (Chen, Frankowski & Bishop-Leone, 2001)
- ☐ Other: _____

Q3 When do you believe these outcomes should be measured? Check all that apply:

- ☐ 4 weeks before start of intervention
- ☐ At start of intervention
- ☐ At end of intervention
- ☐ 4 weeks after intervention
- ☐ 3 months after intervention
- ☐ 6 months after intervention
- ☐ 9 months after intervention
- ☐ 12 months after intervention

- [ ] 15 months after intervention
- [ ] 18 months after intervention
- [ ] 24 months after intervention
- [ ] Other: _____

Q4 In swallowing treatment with a person with Parkinson's Disease, what is important to you in terms of deciding if the swallowing treatment has worked?

Please indicate which items in the list below are:

- Important
- Not important

to you by ticking the according box.

	Important (1)	Not Important (2)
Oropharyngeal dysphagia severity	○	○
Self- perception of Swallow	○	○
Initiation of pharyngeal swallow	○	○
Level of oral intake	○	○
Dry mouth	○	○
Drooling	○	○
Salivary pooling	○	○
Swallow related orolingual movement	○	○

Orolingual bolus control	○	○
Piecemeal deglutition	○	○
Oral bolus transport	○	○
Nasal regurgitation	○	○
Penetration/Aspiration	○	○
Laryngeal Sensation	○	○
Choking	○	○
Post-swallow oral residue	○	○
Post-swallow pharyngeal residue	○	○
Cough production	○	○
Cough sensitivity	○	○
Lingual Muscle strength	○	○
Pharyngeal Muscle Strength	○	○
Weightloss	○	○
Medication Intake	○	○
Pneumonia	○	○

Hospitalization	○	○
Swallowing related quality of life	○	○
Carer Burden	○	○
Fear of eating/ drinking	○	○
Pleasure of oral intake	○	○
Avoidance of food/liquids	○	○
Changes in voice and/or speech	○	○
Adverse events	○	○
Participant's adherence to intervention	○	○
Participant's experience with intervention	○	○

Q5 Are there any other outcomes which you feel are important in evaluating if a swallowing treatment with a person with Parkinson's Disease works?

○ Yes _____

○ No

End of Block: Perspectives

Start of Block: Profile of Caseload

Q6 What is the typical age range of your clients with Parkinson's Disease?

- [ ] Younger than 50 years
- [ ] 50 - 59 years
- [ ] 60 - 69 years
- [ ] 70 - 79 years
- [ ] 80 years or older

Q7 What is the predominant ethnic profile of your clients with Parkinson's Disease?

- ○ African
- ○ Arab
- ○ Asian
- ○ Caribbean
- ○ Mixed or multiple ethnic groups
- ○ White
- ○ Other ethnic group: _____

Q8 What gender are the majority of your clients with Parkinson's Disease?

- ○ Male
- ○ Female
- ○ Non-binary
- ○ Other: _____

Q9 Would most of your clients with Parkinson's Disease have co-occurring cognitive impairment?

- ○ Yes
- ○ No
- ○ Unsure

Q10 Would most of your clients with Parkinson's Disease have co-occurring dysarthria?

- ○ Yes
- ○ No
- ○ Unsure

End of Block: Profile of Caseload

Start of Block: Demographics of Participant

Q11 What is your profession as a dysphagia clinician?

- ○ Speech and Language Therapist/Pathologist
- ○ Occupational Therapist
- ○ Physiotherapist
- ○ Other: _____

Q12 In which country do you primarily work as a dysphagia clinician with people who have Parkinson's Disease?

_____

Q13 Are you currently practicing as a dysphagia clinician?

- ○ Yes
- ○ No, I am on leave
- ○ No, I am currently involved in full time research/teaching
- ○ No, I am retired
- ○ Other: _____

Q14 Please indicate the setting where you work as a dysphagia clinician. Check all that apply:

- ☐ Acute setting
- ☐ Community Setting
- ☐ Residential setting
- ☐ Rehabilitation Setting
- ☐ Other: _____

Q15 For how many years have you worked as a dysphagia clinician?

- ○ <1
- ○ 1
- ○ 2

- ○ 3
- ○ 4
- ○ 5
- ○ 6
- ○ 7
- ○ 8
- ○ 9
- ○ 10
- ○ 11+

Q16 For how many years have you worked with people with Parkinson's Disease and dysphagia?

- ○ <1
- ○ 1
- ○ 2
- ○ 3
- ○ 4
- ○ 5
- ○ 6
- ○ 7
- ○ 8
- ○ 9
- ○ 10
- ○ 11+

Q17 Which age group do you belong to?

- ○ 18-25 years
- ○ 26-33 years
- ○ 34-41 years
- ○ 42-49 years
- ○ 50-57 years
- ○ 58-65 years
- ○ 66+ years

Q18 How do you describe yourself?

- ○ Woman
- ○ Man
- ○ Non-binary
- ○ None of the above, I identify as: _____
- ○ Prefer not to answer

Q19 By tapping/clicking the **arrow (→)** in the bottom right corner, you are **submitting your responses** to this survey.

End of Block: Demographics of Participant



You completed the survey!

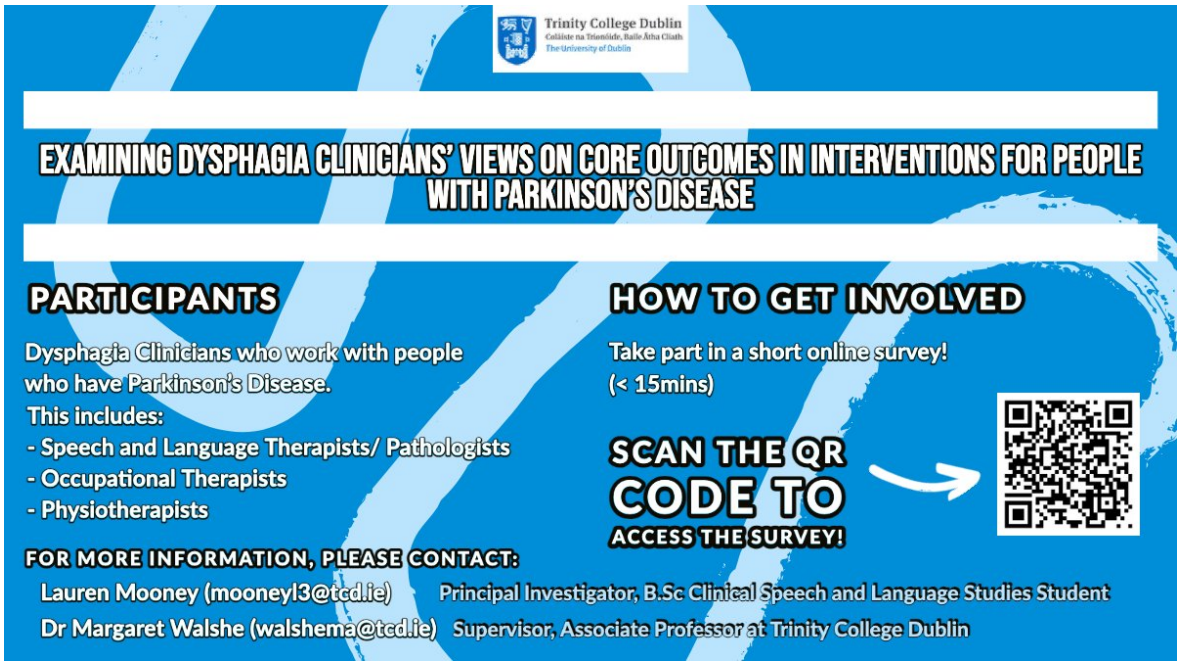
Thank you very much for participating!

If you would like to receive the results of this survey, please register your interest through emailing us.

If you have any further questions, please do not hesitate to also contact **us via email.**
Further information about the research project can be found on our website.

Wish to share this survey? You can do so by copying and sharing the following link.

Appendix 6.3: Recruitment poster for online survey



Appendix 6.4: Outcome measurement instruments suggested by respondents in open text box that could not be classified with reasons

Unable to classify	Reason	n
Clinical predictors of dysphagia and aspiration risk (Daniels et al., 1998)	not an OMI	5
Valid items for screening dysphagia risk (Daniels et al., 2012)	not an OMI	1
Therapy Outcome Measure (TOM) (Enderby & John, 2015)	global OMI, not swallowing specific	1
Functional Independence Measures (FIM) (Keith et al., 1987)	global OMI, not swallowing specific	1
ASHA's National Outcomes Measurement System (ASHA NOMs) (Mullen, 2004)	global OMI, not swallowing specific	2
Handgrip strength (Hathaway et al., 2015)	not swallowing specific	1
Diadochokinetic Rate	not swallowing specific	1
Respiratory Measures	too unspecific	3
Spirometry measures	too unspecific	1
Cough peak flow	outcome, not OMI	1
Peak Voluntary Cough Flow	outcome, not OMI	1
Sequential Voluntary Cough	outcome, not OMI	1
Clinical swallow evaluation	too unspecific	8
Department's own OMI	too unspecific	7
Clinician's own observations	too unspecific	4
DSS (Leonard/Kenall; VFSS)	unclear	1

OMI = Outcome Measurement Instrument

Appendix 6.5: Outcome measurement instruments classified as screening tools

Category	Screening tools that involve swallow trials	n
Overall swallow screening	The Gugging Swallowing Screen (GUSS) (Trapl et al., 2007)	7
Overall swallow screening	Volume-Viscosity Swallow Test (Rofes et al., 2012)	2
Overall swallow screening	Berlin (Bolus) Swallow Test (BST) (Schultheiss et al., 2011)	1
Overall swallow screening	Modified Swallowing Assessment (MSA) (Perry, 2001)	1
Timed swallow screening with one consistency	Timed Water Swallow Test (Hughes & Wiles, 1996)	3
Timed swallow screening with one consistency	3 oz Water Swallow Test (Suiter & Leder, 2008)	2
Timed swallow screening with one consistency	100mL Water Swallow Test (Patterson et al., 2011)	1

Appendix 7.1: Consent form for consensus meetings

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

COS-DIP
Core Outcome Set for Dysphagia Interventions in Parkinson's disease

Development of a Core Outcome Set for Dysphagia Interventions in Parkinson's disease: the COS-DIP Study

Consensus Meeting Consent Form

We invite you to take part in an online consensus meeting. Your feedback is important. This meeting will take approximately 60-90 minutes.

Please initial the box if you agree with the statement. Please feel free to ask questions if there is something you do not understand. The end of this form is for the researchers to complete. Thank you for participating.

General	**Tick box**
I confirm I have read and understood the **Participant Information Leaflet** for the above study. The information has been fully explained to me and I have been able to ask questions, all of which have been answered to my satisfaction.	
I understand that this study **is entirely voluntary, and if I decide that I do not want to take part, I can stop taking part in this study at any time without giving a reason**. I understand that deciding not to take part will not affect my future medical care.	
I agree to taking part in an **online video meeting**. I understand that no video recording will be made.	
I agree to the consensus meeting being **audio recorded**.	
I understand that I **will not be paid for taking part in this study**.	
I understand that there are **no direct benefits** to me from participating in this study, which has been set out in full in the Participant Information Leaflet which I have been provided with.	
I know how to contact the research team if I need to.	
I agree to being contacted by researchers by email as part of this study.	

Data Processing	Tick box
I understand the study is being conducted by researchers from Trinity College Dublin (TCD) and my personal information will be stored electronically on the TCD server and will be protected in accordance with the General Data Protection Regulation.	
By taking part in this consensus meeting, I agree to allow the researchers to anonymously publish the research ideas that I help them to identify. Anonymised means that my personal information will be removed, and it will not be possible to link me as an individual to any of the ideas generated.	
I agree to take part in this consensus meeting.	

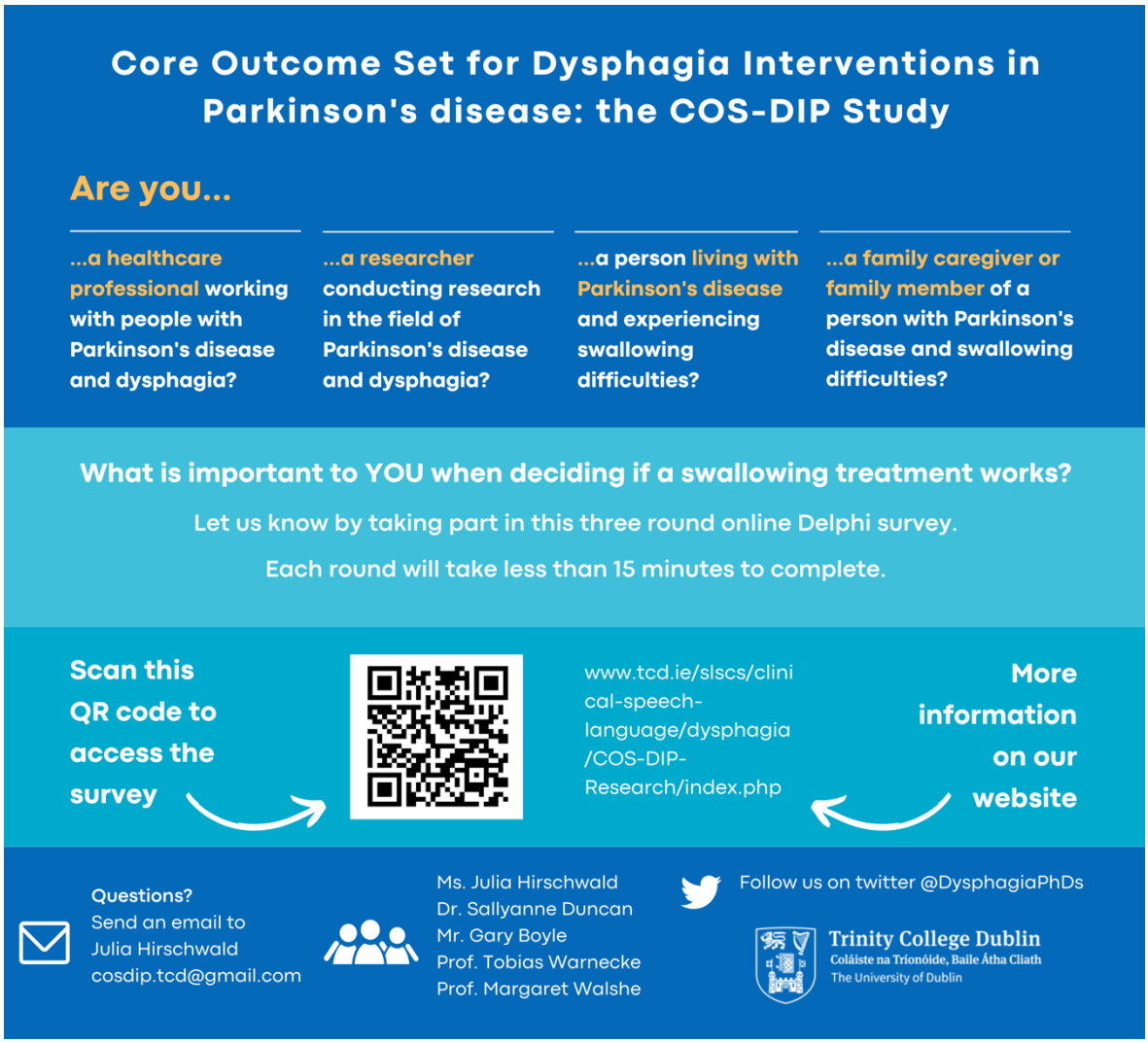
Participant Name (Block Capitals) Participant Signature Date

To be completed by the Principal Investigator.

I, the undersigned, have taken the time to fully explain to the above participant the nature and purpose of this study in a way that they could understand. I have explained the risks and possible benefits involved. I have invited them to ask questions on any aspect of the study that concerned them.
I have given a copy of the participant information leaflet and consent form to the participant with contacts of the study team.

Researcher name: Julia Hirschwald
Signature:
Date:

Appendix 7.2: Recruitment poster for Delphi survey



Appendix 7.3: Participant information leaflet on Delphi survey and consensus meetings for people with OD in PD and family members/caregivers

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

COS-DIP
Core Outcome Set for Dysphagia Interventions in Parkinson's disease

Participant information leaflet

for patients, family caregivers & family members

Study title	**Development of a Core Outcome Set for Dysphagia Interventions in Parkinson's disease: the COS-DIP Study**
Research stage	**Delphi survey and online consensus meeting**
Site	**Trinity College Dublin, Ireland**
Principal Investigator Co-Investigators	**Ms. Julia Hirschwald** (cosdip.tcd@gmail.com) **Dr. Sallyanne Duncan** **Prof. Tobias Warnecke** **Mr. Gary Boyle** **Prof. Margaret Walshe** (walshema@tcd.ie)
Data Controllers	**Trinity College Dublin**
Data Protection Officer	**Data Protection Officer, Secretary's Office** **Trinity College Dublin, Dublin 2, Ireland**

You are being invited to take part in a research study. Before you decide whether or not you wish to take part, please read this information sheet carefully. It is important that you understand why the research is being done and what it will involve so that you can make a decision that is right for you. Don't feel rushed or under pressure to make a quick decision. You can ask Julia Hirschwald any questions. You may wish to discuss it with your family, friends, GP or neurologist.

This leaflet has four parts: (1) The Study, (2) Data Protection, (3) Costs, Funding and Approval, (4) Further Information

Part 1 – The Study

Why is this study being done?

Background: Swallowing difficulties (dysphagia) are common in people with Parkinson's disease (PD) and can cause dehydration and poor nutrition. It may also impact on quality of life. Food and liquids can enter the airway or lungs, leading to choking or possible lung infections. Research studies of swallowing treatments need to be able to measure how well the treatment works, but also what side

effects it may have and how they may affect patients. Researchers do this by measuring 'outcomes'. Examples of outcomes include 'quality of life' or 'severity of swallowing difficulty'.

Current challenge: So far researchers have been using different outcomes when conducting studies. Some for example measure quality of life and others do not. Furthermore, people like you (living with PD and swallowing difficulties, family caregivers and family members) have not been involved in deciding what to measure in these research studies. Thus, the results of the research might have little importance to you.

A solution to this challenge is to establish a core outcome set (COS). A COS is a collection of outcomes that should be measured as a minimum standard in every trial that focuses on a particular area. Establishing a COS is best undertaken using a Delphi Study. A Delphi study is used to get expert consensus on a topic using sequential rounds of questionnaires, whereby anonymised feedback of the responses from the previous round is provided, prompting reflection and gradually consensus evolves.

Purpose of the study: We are doing this study to find out what the most important things to measure are when delivering a swallowing treatment to people with PD. We aim to develop a minimum standard (a COS) of *what* should be measured and *how* it should be measured in studies on swallowing treatments for people with PD.

Study content: We will ask researchers and healthcare professionals working in this field and people living with PD and swallowing difficulties, family caregivers and family members of their opinions on what outcomes are most important to them. To gather the opinions and reach consensus on the COS we will conduct three online surveys (so called Delphi surveys). Thereafter, we will conduct two online consensus meetings to discuss the results and agree on the COS. You do not have to attend a consensus meeting. You can participate in the Delphi survey only, if you wish.

Further information: If you like, you can go to our website and watch this video to get a better understanding of what this project is about and what it involves.

Why have I been invited to take part?

You are being invited to participate in this study because you have PD and swallowing difficulties, you are a family caregiver or family member of a person with PD and swallowing difficulties. Additionally, we are also inviting healthcare professionals who work with people with PD and swallowing difficulties and researchers who perform studies on swallowing interventions in PD. We are intending to involve up to 200 participants for the online Delphi surveys and up tp 40 people for the online consensus meetings.

Do I have to take part? Can I withdraw?

You do not have to take part in this study. **It is entirely voluntary**. If you decide not to take part, it will not affect your current or future medical care. You can change your mind about taking part in the study

and opt out at any time, even if the study has started. You do not have to give a reason for not taking part or for opting out. If you wish to opt out, please contact Julia Hirschwald (cosdip.tcd@gmail.com) who will be able to organise this for you.

What happens if I change my mind?

You can change your mind at any time by contacting Julia Hirschwald (cosdip.tcd@gmail.com) and we will be able to organise this for you. If you choose not to continue to take part, this will not affect your medical care in any way. If you wish, you can ask for your data stored to be destroyed. If you request this, we will destroy all data that are still in our possession. We will no longer use or share your data for research from this point onwards. However, it will not be possible to destroy data already used in research studies prior to this time.

How will the study be carried out?

The study will take part between **January and May 2024** online. We will conduct three online Delphi surveys. Thereafter, we will conduct two online consensus meetings to facilitate attendance of participants from different time zones. If you wish to participate in one of these meetings we will get in contact with you to organise a day and time.

What will happen to me if I decide to take part?

If you agree to participate you will be asked to complete three online questionnaires (Delphi surveys). You will receive a link to each questionnaire via email. One questionnaire will take around 15 minutes to complete. A consent form will be included at the beginning of the online survey and the consent box will need to be ticked by you before commencing the survey. You can contact Julia Hirschwald via email (cosdip.tcd@gmail.com) if you have any queries or difficulties completing any aspect of the online survey. Each online survey will be opened for 3 weeks.

After the three Delphi surveys you will be asked if you would like to participate in one of two online consensus meetings. If you agree to participate, we will contact you via email to inform you whether or not you were randomly selected. Overall, one consensus meeting is estimated to last 60-90 minutes. We will ask you to turn on your camera so that the we can see you. You do not have to turn on your camera. Only audio (one audio file per participant), no video, will be recorded during the consensus meeting. The audio recordings will help us so that we do not have to focus on listening, talking and taking notes at the same time. After the consensus meeting, the audio recordings will be transcribed and the audio recordings will be deleted thereafter. In the following Figure 1 you can see what to expect in each survey round and in the consensus meeting.

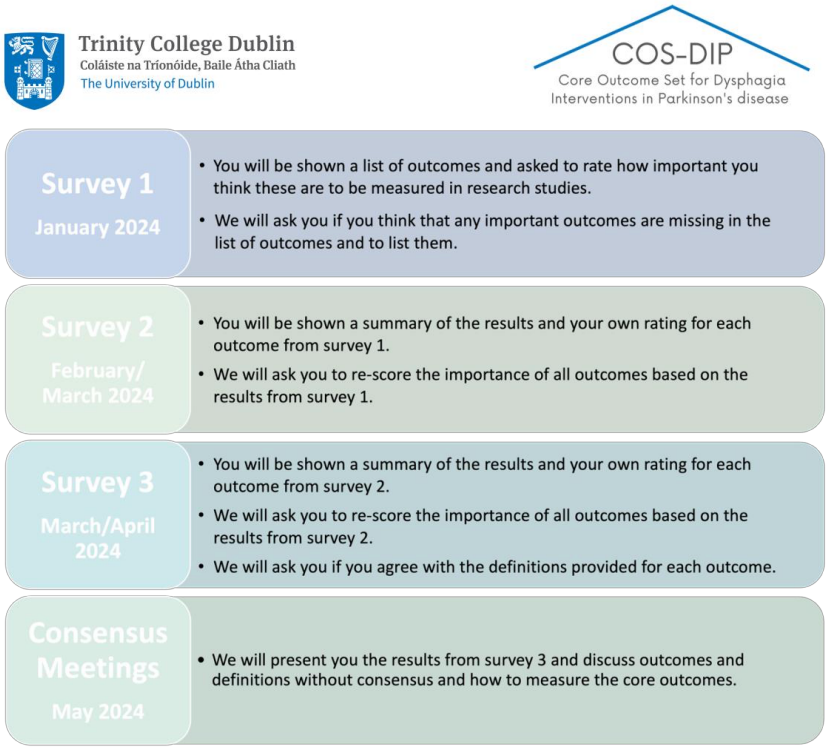


Figure 1: Description of three round online Delphi survey followed by online consensus meetings

What will happen to my Data?

We will give your questionnaire a unique study number. This way you will not be identifiable. Your identity will not be disclosed to anyone outside of the research team. In the final report of this study no identifiable details will be published or disclosed. All data on electronic devices will be password protected and encrypted.

However, we would like to acknowledge your participation in the study in the final publication. After completing the three online surveys, we will send you a thank you letter and ask if you would like to be acknowledged and how your acknowledgement should appear (e.g., name and country). This will appear in an acknowledgement list.

If you participate in one of the consensus meeting, you will keep the same unique study number. This way you will not be identfiable to anyone outside of the consensus meeting. Only other participants during the consensus meeting and the research team will know your identity (name, role). In the final report of this study no identifiable details will be published or disclosed. All data on electronic devices will be password protected and encrypted.

Are there any benefits to taking part in this research?

Taking part in this study will not directly benefit you. However, this research project will provide valuable information for PD researchers and potentially improve the quality of studies testing swallowing treatments for people with PD in the future. The results of this project will also better inform clinicians on the most appropriate information they should be gathering in treatments with people with PD and swallowing difficulties. This in turn will benefit future people living with PD and swallowing difficulties.

Are there any disadvantages or risks to me if I take part?

The only possible risk to taking part in this study is that a connection to your identity could be made as we will have your name and contact details. However, this information will be kept in a separate folder from all research data generated during the online survey and will be stored securely on the TCD server. Great care will be taken to ensure the confidentiality of all data. The risk to participants of a breach of confidentiality is considered very low.

What happens if something goes wrong when I'm taking part in the study?

If you have any concerns about any aspects of the study, you can contact Prof. Margaret Walshe (walshema@tcd.ie).

What will happen to the results of the research?

The results of the study will be reported in medical and scientific journals and presented at (inter-) national medical and scientific conferences. We will also liaise with representatives from national and international funding agencies and policy-makers to ensure the COS decided on in this study will be adopted by future researchers conducting clinical trials in this area. We will provide lay summaries to relevant patient and carer support groups and organisations. No information which reveals your identity will be disclosed.

Part 2 – Data Protection

What information about me (personal data) will be used as part of this study? Will my medical records be accessed?

Before starting the first Delphi questionnaire, we will ask you to provide information about yourself, your name, geographic location, and stakeholder group (healthcare professional/reseracher, person living with PD and swallowing difficulties or family caregiver/family member). Your name will not be

associated with your responses in the Delphi questionnaire feedback and will not be attributed to your responses in any report or publication. Your medical records will not be accessed.

If you agree to participate in an online consensus meeting we will use the following additional information about you for data analysis purposes only: the audio recording of the consensus meeting and the final transcript of the audio recording.

What will happen to my personal data?

Only your personal data essential for the project will be processed in order to achieve the objective of this study. Your personal data will not be processed in a way that can cause damage or distress to you.

Your contact details and audio recordings will be immediately destroyed after the data have been transcribed and the consensus meetings have concluded. The coded transcripts and your personal data will be pseudonymised and the key code linking personal data with research data will be stored separately from each other for one year after the study is completed and written up.

The collected study information will be kept securely in a password protected and encrypted file on the TCD One Drive. In January 2027, all collected information will be destroyed.

Who will access and use my personal data as part of this study?

Only Julia Hirschwald and Prof. Margaret Walshe will have access to your personal data as part of this study.

Will my personal data be kept confidential? How will my data be kept safe?

Your privacy is important to us. We take many steps to make sure that we protect your confidentiality and keep your data safe. Here are some examples of how we do this:

- Your consent form with your personal data will be stored and locked in a filing cabinet.
- You will not be identifiable in any presentations or publications in relation to this study.
- All principal investigators and co-investigators who have access to your personal data are bound by a professional code of secrecy.
- Training in data protection law and practice has been provided to those individuals involved in carrying out the research.

What is the lawful basis to use my personal data?

By law,[1] we can use your personal information for scientific research[2] (in the public interest[3]). We will also ask for your explicit consent to use your data as a requirement of the Irish Health Research Regulations.

What are my rights?

You are entitled to:

- The right to access to your data and receive a copy of it
- The right to restrict or object to processing of your data
- The right to object to any further processing of the information we hold about you (except where it is de-identified)
- The right to have inaccurate information about you corrected or deleted
- The right to receive your data in a portable format and to have it transferred to another data controller
- The right to request deletion of your data

By law you can exercise the following rights in relation to your personal data, unless the request would make it impossible or very difficult to conduct the research. You can exercise these rights by contacting your Julia Hirschwald (cosdip.tcd@gmail.com) or the Trinity College Data Protection Officer, Secretary's Office, Trinity College Dublin, Dublin 2, Ireland. Email: dataprotection@tcd.ie. Website: www.tcd.ie/privacy.

Part 3 – Costs, Funding and Approval

Has this study been approved by a research ethics committee?

Yes, this study has been approved by the Faculty of Health Sciences Research Ethics Committee of Trinity College Dublin, Dublin 2, Ireland. Approval was granted on 15/05/2023. None of the persons carrying out this study have a link to the committees. Julia Hirschwald is a PhD student and Prof. Margaret Walshe an associate professor at Trinity College Dublin.

[1] The European General Data Protection Regulation (GDPR)
[2] Article 9(2) (j)
[3] Article 6(1)(e)

Who is organising and funding this study? Will the results be used for commercial purposes?

A short version of this study is publicly available at www.comet-initiative.org/Studies/Details/1942. This research is conducted by Julia Hirschwald for the purpose of obtaining the degree of Doctor in Philosophy under the supervision of Prof. Margaret Walshe and Dr. Sallyanne Duncan. Mr. Gary Boyle and Prof. Tobias Warnecke are part of the study steering committee and take on an advisory role in this research project. Julia Hirschwald undertakes this research project as part of a Provost's PhD Award funded by Trinity College Dublin. The DelphiManager platform is funded through a TRiSS fellowship awarded by Trinity College Dublin. The results will not be used for commercial purposes.

Is there any payment for taking part? Will it cost me anything if I agree to take part?

No, neither will we pay you to take part in the study nor will you have to pay to take part.

Part 4 – Further Information

Who should I contact for information or complaints?

If you have any concerns or questions, you can contact:

- Julia Hirschwald: cosdip.tcd@gmail.com, Prof. Margaret Walshe: walshema@tcd.ie
- Data Protection Officer, Trinity College Dublin: Data Protection Officer, Secretary's Office, Trinity College Dublin, Dublin 2, Ireland. Email: dataprotection@tcd.ie. Website: www.tcd.ie/privacy.

Under GDPR, if you are not satisfied with how your data is being processed, you have the right to lodge a complaint with the Office of the Data Protection Commission, 21 Fitzwilliam Square South, Dublin 2, Ireland. Website: www.dataprotection.ie.

Will I be contacted again?

If you would like to take part in this study please contact Julia Hirschwald (cosdip.tcd@gmail.com). You will then be contacted before and after each Delphi round. If you wish to participate in one of the consensus meetings, you will be contacted beforehand again.

Appendix 7.4: Participant information leaflet on Delphi survey and consensus meetings for healthcare professionals and researchers

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

COS-DIP
Core Outcome Set for Dysphagia Interventions in Parkinson's disease

Participant information leaflet

for healthcare professionals & researchers

Study title	**Development of a Core Outcome Set for Dysphagia Interventions in Parkinson's disease: the COS-DIP Study**
Research stage	**Delphi survey and online consensus meeting**
Site	**Trinity College Dublin, Ireland**
Principal Investigator Co-Investigator Co-Investigator	**Ms. Julia Hirschwald** (cosdip.tcd@gmail.com) **Dr. Sallyanne Duncan** **Prof. Tobias Warnecke** **Mr. Gary Boyle** **Prof. Margaret Walshe** (walshema@tcd.ie)
Data Controllers	**Trinity College Dublin**
Data Protection Officer	**Data Protection Officer, Secretary's Office** **Trinity College Dublin, Dublin 2, Ireland**

You are being invited to take part in a research study. Before you decide whether or not you wish to take part, please read this information sheet carefully. It is important that you understand why the research is being done and what it will involve so that you can make a decision that is right for you. Don't feel rushed or under pressure to make a quick decision. You can ask Julia Hirschwald any questions.

This leaflet has four parts: (1) The Study, (2) Data Protection, (3) Costs, Funding and Approval, (4) Further Information

Part 1 – The Study

Why is this study being done?

Background: Research studies on oropharyngeal dysphagia interventions for Parkinson's disease (PD) need to be able to measure how well the treatment works (benefits), but also what side effects (adverse events and risks) it may have and how they may affect patients. Researchers do this by measuring outcomes. Examples of outcomes include 'quality of life' or 'severity of swallowing difficulty'.

Current challenge: Clinical trials investigating the effectiveness of dysphagia interventions in PD have collected different outcomes, at different time points, and used different measures. This makes comparison across studies a major challenge. Furthermore, people living with PD and dysphagia, their family caregivers and family members have not been involved in deciding what to measure in these research studies. Thus, the results of the resarch might have little importance to them.

A solution to this challenge is to establish a core outcome set (COS). A COS is a collection of outcomes that should be measured as a minimum standard in every trial that focuses on a particular area. Establishing a COS is best undertaken using a Delphi Study. A Delphi study is used to get expert consensus on a topic using sequential rounds of questionnaires, whereby anonymised feedback of the responses from the previous round is provided, prompting reflection and gradually consensus evolves.

Purpose of the study: We are doing this study to find out what the most important outcomes to measure in clinical trials on dysphagia interventions in people with PD are. We aim to develop a minimum standard (a COS) of *what* should be measured and *how* it should be measured in clinical trials on dysphagia interventions in people with PD.

Study content: We will ask researchers, healthcare professionals, people living with PD and dysphagia, family caregivers and family members of their opinions on what outcomes are most important. To gather the opinions and reach consensus on the COS we will conduct three online surveys (so called Delphi surveys). Thereafter, we will conduct two online consensus meetings to discuss the results and agree on the COS-DIP. You do not have to attend the consensus meeting. You can participate in the Delphi survey only, if you wish.

Further information: If you like, you can go to our website and watch this video to get a better understanding of what this project is about and what it involves.

Why have I been invited to take part?

You are being invited to participate in this study because you work with people with PD and dysphagia or you are conducting research on dysphagia interventions in PD. We also invite people living with PD and dysphagia, family caregivers and family members. We are intending to involve up to 200 participants for the online Delphi survey and 40 people for the two online consensus meetings.

Do I have to take part? Can I withdraw?

You do not have to take part in this study. **It is entirely voluntary**. You can change your mind about taking part in the study and opt out at any time, even if the study has started. You do not have to give a reason for not taking part or for opting out. If you wish to opt out, please contact Julia Hirschwald (cosdip.tcd@gmail.com) who will be able to organise this for you.

What happens if I change my mind?

You can change your mind at any time by contacting Julia Hirschwald (cosdip.tcd@gmail.com) and we will be able to organise this for you. If you wish, you can ask for your data stored to be destroyed. If you request this, we will destroy all data that are still in our possession. We will no longer use or share your data for research from this point onwards. However, it will not be possible to destroy data already used in research studies prior to this time.

How will the study be carried out?

The study will take part between **January and May 2024** online. We will conduct three online Delphi surveys. Thereafter, we will conduct two online consensus meetings to facilitate attendance of participants from different time zones. If you wish to participate in one of these meetings we will get in contact with you to organise a day and time.

What will happen to me if I decide to take part?

If you agree to participate you will be asked to complete three online questionnaires (Delphi surveys). You will receive a link to each questionnaire via email. One questionnaire will take around 15 minutes to complete. A consent form will be included at the beginning of the online survey and the consent box will need to be ticked by you before commencing the survey. You can contact Julia Hirschwald via email (cosdip.tcd@gmail.com) if you have any queries or difficulties completing any aspect of the online survey. Each online survey will be open for 3 weeks.

After the three Delphi surveys you will be asked if you would like to participate in an online consensus meeting. We will invite 20% of participants from the Delphi survey. If you agree to participate, we will contact you via email to inform you whether or not you were randomly selected. Overall, one consensus meeting is estimated to last 60-90 minutes. During the consensus meeting we will ask you to turn on your camera. However, this is voluntary. We will only record audio (one audio file per participant), no videos. After the consensus meeting we will transcribe the audio recordings and delete the audio recordings thereafter. In Figure 1 below you can see what to expect in each survey round.

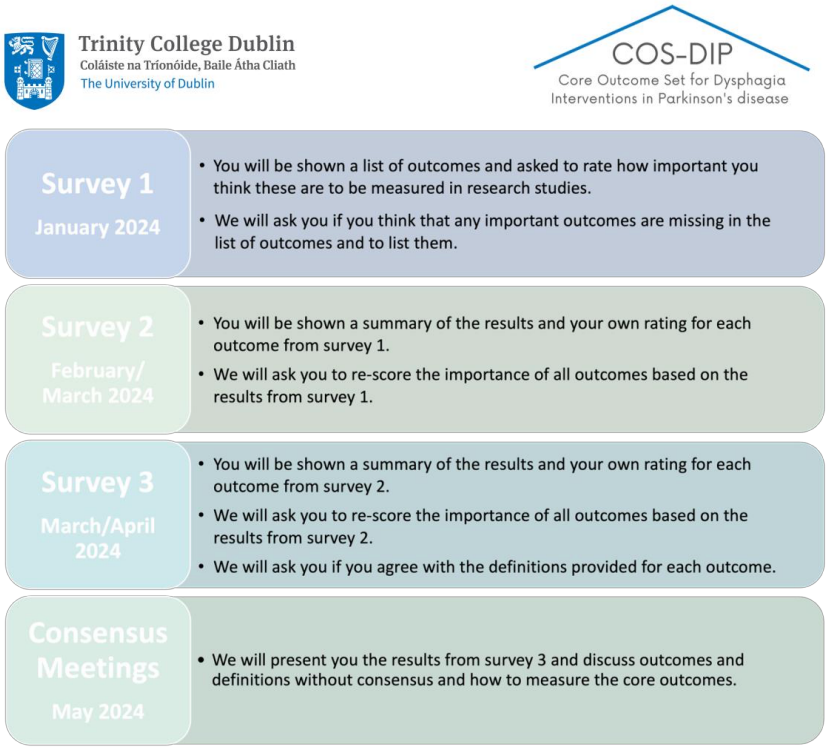


Figure 1: Description of three round online Delphi survey followed by online consensus meetings

What will happen to my Data?

We will give your questionnaire a unique study number. This way you will not be identifiable. Your identity will not be disclosed to anyone outside of the research team. In the final report of this study no identifiable details will be published or disclosed. All data on electronic devices will be password protected and encrypted.

However, we would like to acknowledge your participation in the study in the final publication. After completing the three online surveys, we will send you a thank you letter and ask if you would like to be acknowledged and how your acknowledgement should appear (e.g., name and country, profession/organisation). This will appear in an acknowledgement list.

If you participate in one of the consensus meeting, you will keep the same unique study number. This way you will not be identfiable to anyone outside of the consensus meeting. Only other participants during the consensus meeting and the research team will know your identigy (name, role). In the final report of this study no identifiable details will be published or disclosed. All data on electronic devices will be password protected and encrypted.

Are there any benefits to taking part in this research?

Taking part in this study will not directly benefit you. However, this research project will provide valuable information for PD researchers and potentially improve the quality of studies testing dysphagia interventions for people with PD in the future. The results of this project will also better inform clinicians on the most appropriate information they should be gathering in dysphagia treatments with people with PD and. This in turn will benefit future patients living with PD and dysphagia.

Are there any disadvantages or risks to me if I take part?

The only possible risk to taking part in this study is that a connection to your identity could be made as we will have your name and contact details. However, this information will be kept in a separate folder from all research data generated during the online survey and will be stored securely on the TCD server. Great care will be taken to ensure the confidentiality of all data. The risk to participants of a breach of confidentiality is considered very low.

What happens if something goes wrong when I'm taking part in the study?

If you have any concerns about any aspects of the study, you can contact Prof. Margaret Walshe (walshema@tcd.ie).

What will happen to the results of the research?

The results of the study will be reported in medical and scientific journals and presented at (inter-) national medical and scientific conferences. We will also liaise with representatives from national and international funding agencies and policy-makers to ensure the core set of outcomes decided on in this study will be adopted by future researchers conducting clinical trials in this area. We will provide lay summaries to relevant patient and carer support groups and organisations. No information which reveals your identity will be disclosed.

Part 2 – Data Protection

What information about me (personal data) will be used as part of this study?

Before starting the first Delphi questionnaire, we will ask you to provide information about yourself, your name, geographic location, profession and stakeholder group (healthcare professionals, reserachers, or patients/carers). Your name will not be associated with your responses in the Delphi questionnaire feedback and will not be attributed to your responses in any report or publication.

If you agree to participate in an online consensus meeting we will use the following additional information about you for data analysis purposes only: the audio recording of the consensus meeting and the final transcript of the audio recordings.

What will happen to my personal data?

Only your personal data essential for the project will be processed in order to achieve the objective of this study. Your personal data will not be processed in a way that can cause damage or distress to you.

Your contact details and audio recordings will be immediately destroyed after the data have been transcribed and the consensus meetings have concluded. The coded transcripts and your personal data will be pseudonymised and the key code linking personal data with research data will be stored separately from each other for one year after the study is completed and written up.

The collected study information will be kept securely in a password protected and encrypted file on the TCD OneDrive. In January 2027, all collected information will be destroyed.

Who will access and use my personal data as part of this study?

Only Julia Hirschwald and Prof. Margaret Walshe will have access to your personal data as part of this study.

Will my personal data be kept confidential? How will my data be kept safe?

Your privacy is important to us. We take many steps to make sure that we protect your confidentiality and keep your data safe. Here are some examples of how we do this:

- Your consent form with your personal data will be stored and locked in a filing cabinet.
- You will not be identifiable in any presentations or publications in relation to this study.
- All principal investigators and co-investigators who have access to your personal data are bound by a professional code of secrecy.
- Training in data protection law and practice has been provided to those individuals involved in carrying out the research.

What is the lawful basis to use my personal data?

By law,[1] we can use your personal information for scientific research[2] (in the public interest[3]). We will also ask for your explicit consent to use your data as a requirement of the Irish Health Research Regulations.

[1] The European General Data Protection Regulation (GDPR)
[2] Article 9(2) (j)
[3] Article 6(1)(e)

What are my rights?

You are entitled to:

- The right to access to your data and receive a copy of it
- The right to restrict or object to processing of your data
- The right to object to any further processing of the information we hold about you (except where it is de-identified)
- The right to have inaccurate information about you corrected or deleted
- The right to receive your data in a portable format and to have it transferred to another data controller
- The right to request deletion of your data

By law you can exercise the following rights in relation to your personal data, unless the request would make it impossible or very difficult to conduct the research. You can exercise these rights by contacting your Julia Hirschwald (cosdip.tcd@gmail.com) or the Trinity College Data Protection Officer, Secretary's Office, Trinity College Dublin, Dublin 2, Ireland. Email: dataprotection@tcd.ie. Website: www.tcd.ie/privacy.

Part 3 – Costs, Funding and Approval

Has this study been approved by a research ethics committee?

Yes, this study has been approved by the Faculty of Health Sciences Research Ethics Committee of Trinity College Dublin, Dublin 2, Ireland. Approval was granted on 15/05/2023. None of the persons carrying out this study have a link to the committees. Julia Hirschwald is a PhD student and Prof. Margaret Walshe an associate professor at Trinity College Dublin.

Who is organising and funding this study? Will the results be used for commercial purposes?

A short version of this study is publicly available at www.comet-initiative.org/Studies/Details/1942. This research is conducted by Julia Hirschwald for the purpose of obtaining the degree of Doctor in Philosophy under the supervision of Prof. Margaret Walshe and Dr. Sallyanne Duncan. Mr. Gary Boyle and Prof. Tobias Warnecke are part of the study steering committee and take on an advisory role in this research project. Julia Hirschwald undertakes this research project as part of a Provost's PhD Award funded by Trinity College Dublin. The DelphiManager platform is funded through a TRiSS fellowship awarded by Trinity College Dublin. The results will not be used for commercial purposes.

Is there any payment for taking part? Will it cost me anything if I agree to take part?

No, neither will we pay you to take part in the study nor will you have to pay to take part.

Part 4 – Further Information

Who should I contact for information or complaints?

If you have any concerns or questions, you can contact:

- Julia Hirschwald: cosdip.tcd@gmail.com, Prof. Margaret Walshe: walshema@tcd.ie
- Data Protection Officer, Trinity College Dublin: Data Protection Officer, Secretary's Office, Trinity College Dublin, Dublin 2, Ireland. Email: dataprotection@tcd.ie. Website: www.tcd.ie/privacy.

Under GDPR, if you are not satisfied with how your data is being processed, you have the right to lodge a complaint with the Office of the Data Protection Commission, 21 Fitzwilliam Square South, Dublin 2, Ireland. Website: www.dataprotection.ie.

Will I be contacted again?

If you would like to take part in this study please contact Julia Hirschwald (cosdip.tcd@gmail.com). You will then be contacted before and after each Delphi round. If you wish to participate in one of the consensus meetings, you will be contacted beforehand again.

Appendix 7.5: Development of long list of outcomes for Delphi survey

Outcomes from OD intervention studies in PD with control groups	Additional outcomes from OD intervention studies in PD without control groups	Additional outcomes from survey with people with PD, family members/caregivers	Additional outcomes from survey with dysphagia clinicians	Additional outcomes added by study steering committee
Death	Lingual muscle strength	Fear of choking	Respiratory swallow coordination	Dry mouth
Oropharyngeal dysphagia severity	Swallowing related velopharyngeal movement		Hydration	Nasal regurgitation
Drooling	Swallowing related pharyngeal movement	**> 1 additional outcome**	Mealtime duration	Choking
Salivary pooling	Pharyngeal muscle strength		Independent eating/drinking	Cough sensitivity
Orolingual bolus control	Swallowing capacity		Social aspect of eating/drinking	Avoidance of food / liquids
Oral bolus transport	Time per swallow			Fear of eating / drinking
Swallowing related orolingual movement	Volume per swallow		**> 5 additional outcomes**	Medication intake
Timing of oropharyngeal swallow components	Swallows per bite			Carer burden
Laryngeal elevation	Masticatory cycles per bite			Changes in voice, speech
Laryngeal sensation	Pitch			Weight loss
Swallowing related hyoid bone movement	Speech production			
Initiation of pharyngeal swallow	Respiratory muscle strength			**> 10 additional outcomes**
Postswallow oral residue	Swallowing related respiratory pattern			
Postswallow pharyngeal residue	Swallowing related lung volume			
Postswallow pharyngeal pooling	Cough production			
Piecemeal deglutition	Self-perception of voice			
Penetration/Aspiration	Self-perception of speech			
Cortical reorganization				
Motor evoked potential	**> 3 additional outcomes**			
Overall motor symptoms				
Tremor				
Rigidity				
Bradykinesia				
Axial symptoms				
Freezing of gait				
Phonation				
Loudness				
Level of oral intake				
Aspiration pneumonia				
Activities of daily living				
Pleasure of oral intake				
Swallowing related quality of life				
Self-perception of swallowing				
Self-perception of walking				
Self-perception of activities of daily living				
Participant's experience with intervention				
Participant's adherence to intervention				
Hospitalisation				
Adverse events				
> 26 outcomes				

Note: outcomes in grey font were removed from the final long list of outcomes

Appendix 7.6: Post consensus meeting survey

Dear participant,

thank you for taking part in this last short survey to finalise the Core Outcome Set for Dysphagia Interventions in Parkinson's disease (COS-DIP). Recently, we conducted two online consensus meetings to agree on the core outcomes, definitions, suggested measurements, and timepoints of measurement. While many aspects were agreed upon in both meetings, some definitions and suggested measurements received suggestions for changes and were voted on separately in the two meetings. To ensure a comprehensive consensus, **we are conducting this survey to gather all participants' opinions on the final changes**. We will begin by presenting the outcomes that were agreed upon in both meetings, followed by a section where you can review and vote on the modified definitions and suggested measurements.

Completion of this survey will take approximately 5 minutes.

The following has been agreed on in both consensus meetings by ≥ 70% of participants.

Core outcomes: 82% of participants agreed

- Penetration/aspiration
- Aspiration pneumonia
- Choking
- Oropharyngeal dysphagia severity
- Swallowing related quality of life
- Post swallow pharyngeal residue

Aspiration pneumonia: 71% participants agreed on the definition and suggested measurement

- Definition: An acute respiratory infection likely caused by aspiration of material (e.g., food, fluids or saliva/secretions).
- Suggested measurement: A clinical suspicion of a new or worsening respiratory infection in the context of at least 2 of the following: temperature <35°C or >38°C; abnormal chest x-ray (CT scan); a white cell count of <4x109/l or >11x109/l or purulent (tracheal) secretions.

Swallowing related quality of life: 91% of participants agreed on the definition and suggested measurement

- Definition: An individual's perception of their ability to swallow within the context of their daily life, considering the cultural norms, values, and personal goals that influence their experience.
- Suggested measurement: e.g., SWAL-QOL (McHorney et al., 2002)

For the remaining four outcomes and timepoints of measurement, changes were proposed and voted on in at least one consensus meeting. We have collated the results from both meetings in the following and we would like to ask you to review and vote on the revised definitions, suggested measurements and timepoints.

Do you agree to proceed with the survey?
Yes/No

Core Outcome: Penetration/aspiration

The **definition** of penetration/aspiration was **agreed** on by 91% of participants and is as follows:

- Penetration: Entry of material (e.g., food, fluids or saliva/secretions) into the larynx that does not pass below the vocal folds.
- Aspiration: Entry of material (e.g., food, fluids or saliva/secretions) into the larynx and below the level of the vocal folds.

The **suggested measurements** for penetration/aspiration were **amended** as follows:
(Note: Added items are highlighted in green. Removed items are crossed out and highlighted in red.)

- VFS~~/FEES~~: e.g., Penetration-Aspiration Scale (Rosenbek et al., 1996), Dynamic Imaging Grade of Swallowing Toxicity (DIGEST) safety rating (Hutcheson et al., 2017, 2022)
- FEES: e.g., Penetration-aspiration events (Warnecke et al., 2016), Visual Analysis of Swallowing Efficiency and Safety (VASES) safety rating (Curtis et al., 2022), DIGEST-FEES safety rating (Labeit et al., 2023)

Do you **agree** on the **suggested measurements** for penetration/aspiration?
Yes/No/Unsure (with text box)

Core Outcome: Oropharyngeal dysphagia severity

The **definition** of oropharyngeal dysphagia severity was **agreed** on by 86% of participants and is as follows:

- The overall degree of impairment (i.e., swallowing safety and swallowing efficiency) in swallowing material (e.g., food, liquids or saliva/secretions).

The **suggested measurements** for oropharyngeal dysphagia severity were **amended** as follows:
(Note: Added items are highlighted in green. Removed items are crossed out and highlighted in red.)

- VFS: e.g., overall Dynamic Imaging Grade of Swallowing Toxicity (DIGEST) score (Hutcheson et al., 2017, 2022), Modified Barium Swallow Impairment Profile (MBSImP) (Martin-Harris et al., 2008)
- FEES**:** e.g., overall DIGEST-FEES score (Labeit et al. 2023), ~~Visual Analysis of Swallowing Efficiency and Safety (VASES) (Curtis et al., 2022)~~, Endoscopic PD dysphagia score (Warnecke et al., 2016)

Do you **agree** on the **suggested measurements** for oropharyngeal dysphagia severity?
Yes/No/Unsure (with text box)

Core Outcome: Choking

The **suggested measurement** of Choking was **agreed** on by 70% of participants and is as follows:

- E.g., report the number of choking events over or within a specific time period

The **definition** of Choking was **amended** as follows:
(Note: Amended wording is highlighted in green.)

- A condition that occurs when breathing is perceived to be impeded through the inhalation or ingestion of material (e.g., food, fluids or saliva/secretions) which is felt as an obstruction of the respiratory tract.

Do you **agree** on the **definition** of Choking?
Yes/No/Unsure (with text box)

Core Outcome: Postswallow pharyngeal residue

The **definition** of Postswallow pharyngeal residue was **agreed** on by 94% of participants and is as follows:

- The presence of material (e.g., food, fluids or saliva/secretions) remaining in the pharynx after swallowing.

The **suggested measurements** for Postswallow pharyngeal residue were **amended** as follows:
(Note: Added items are highlighted in green. Removed items are crossed out and highlighted in red.)

- VFS ~~& FEES~~**:** e.g., Yale Pharyngeal Residue Severity Rating Scale (Neubauer et al., 2015)
- VFS: e.g., Bolus Residue Scale (Rommel et al., 2015), ~~Eisenhuber Scale (Eisenhuber et al., 2002)~~
- FEES: e.g., Residue Scale (Warnecke et al., 2016), Visual Analysis of Swallowing Efficiency and Safety (VASES) efficiency rating (Curtis et al., 2022)

Do you **agree** on the **suggested measurements** for Postswallow pharyngeal residue?
Yes/No/Unsure (with text box)

Timepoints of measurement of core outcomes
(Note: Amended items are highlighted in green.)

1 month pre start of intervention	Advisable
Start of intervention	**Minimum**
End of intervention	**Minimum**
3 months post end of intervention	**Minimum**
6 months post end of intervention	Advisable
12 months post end of intervention	Advisable

Do you **agree** on the **timepoints of measurement** for all six core outcomes?
Yes/No/Unsure (with text box)

_____

Overall, how satisfied were you with your participation in the consensus meeting?
Very satisfied/Satisfied/Neither satisfied nor dissatisfied/ Dissatisfied/Very dissatisfied

_____

We would really appreciate learning more about your experience of the consensus meeting. Please feel free to provide feedback in the text box below. (open text box)

_____

You have reached the end of the survey. Thank you very much for taking the time and for sharing your valuable perspective. After analysis, we will send you the final results.

Appendix 7.7: Incomplete participant data

Registered but not completed Round 1: 18/133		
Healthcare professionals/researchers: 5/84 (4,2%)	Family members/caregivers: 3/12 (25%)	People with OD in PD: 10/36 (27,8%)
Completed Round 1 but not Round 2: 10/115		
Healthcare professionals/researchers: 5/74 (6,8%)	Family members/caregivers: 1/8 (12,5%)	People with OD in PD: 4/22 (18,2%)
Completed Round 2 but not Round 3: 14/105		
Healthcare professionals/researchers: 9/65 (13,8%)	Family members/caregivers: 1/7 (14,3%)	People with OD in PD: 4/18 (22%)

Appendix 7.8: Number of participants rating an outcome 10 during the three round Delphi survey

Round 1	# of participants rating 10
Swallowing related hyoid bone movement	15
Laryngeal elevation	12
Adverse events	10
Oral bolus transport	10
Postswallow pharyngeal pooling	10
Hospitalisation	9
Laryngeal sensation	9
Orolingual bolus control	9
Pharyngeal muscle strength	9
Participant's adherence to intervention	8
Piecemeal deglutition	8
Postswallow pharyngeal residue	8
Swallowing related orolingual movements	8
Mortality	7
Nasal regurgitation	7
Participant's experience with intervention	7
Postswallow oral residue	7
Initiation of pharyngeal swallow	6
Penetration/aspiration	6
Timing of oropharyngeal swallow components	6
Carer burden	5
Cough sensitivity	5
Lingual muscle strength	5
Oropharyngeal dysphagia severity	5
Respiratory swallow coordination	5
Aspiration pneumonia	4
Cough production	4
Salivary pooling	4
Fear of eating/drinking	3
Swallowing related quality of life	3
Weight loss	3
Avoidance of food/liquids	2
Choking	2
Drooling	2
Fear of choking	2
Independent eating/drinking	2
Mealtime duration	2
Medication intake	2
Pleasure of oral intake	2
Hydration	1
Level of oral intake	1
Self-perception of swallowing	1
Social aspect of eating/drinking	1
Voice parameters	1
Total	**238**

Round 2	# of participants rating 10
UES opening during swallowing	10
Adverse events	4
Laryngeal sensation	3
Oropharyngeal dysphagia severity	3
Postswallow pharyngeal residue	3
Aspiration pneumonia	2
Cough sensitivity	2
Cough strength	2
Mortality	2
Choking	1
Penetration/aspiration	1
Total	**33**

Round 3	# of participants rating 10
Adverse events	5
Cough sensitivity	2
Laryngeal sensation	2
Mortality	2
Oropharyngeal dysphagia severity	2
Postswallow pharyngeal residue	2
Aspiration pneumonia	1
Cough strength	1
Total	**17**

Appendix 7.9: Ratings of critically importance (scores 7-9) per stakeholder group and Delphi round

Delphi Round 1 - Ratings of critically importance (7-9) per stakeholder group and in total

	Outcome	Total	Family m/c	HCP/Researc	PwPD
1	Oropharyngeal dysphagia severity	**89 (82%)**	**6 (75%)**	**72 (91%)**	11 (50%)
2	Dry mouth	32 (28%)	3 (33%)	21 (27%)	8 (31%)
3	Drooling	56 (50%)	**6 (75%)**	38 (49%)	12 (46%)
4	Salivary pooling	57 (52%)	**6 (75%)**	42 (54%)	9 (38%)
5	Orolingual bolus control	67 (64%)	3 (43%)	**57 (73%)**	7 (35%)
6	Oral bolus transport	69 (66%)	1 (17%)	**61 (78%)**	7 (35%)
7	Lingual muscle strength	52 (48%)	4 (57%)	40 (51%)	8 (33%)
8	Nasal regurgitation	44 (41%)	1 (17%)	32 (41%)	11 (48%)
9	Swallowing related orolingual movements	52 (49%)	3 (50%)	41 (53%)	8 (36%)
10	Timing of oropharyngeal swallow components	65 (60%)	4 (67%)	50 (63%)	11 (48%)
11	Laryngeal elevation	55 (54%)	1 (20%)	47 (60%)	7 (37%)
12	Laryngeal sensation	**80 (76%)**	4 (67%)	**60 (77%)**	**16 (76%)**
13	Swallowing related hyoid bone movement	55 (56%)	**4 (80%)**	45 (58%)	6 (38%)
14	Pharyngeal muscle strength	64 (61%)	3 (50%)	50 (64%)	11 (52%)
15	Initiation of pharyngeal swallow	71 (66%)	3 (50%)	**57 (73%)**	11 (46%)
16	Postswallow oral residue	62 (58%)	3 (50%)	51 (65%)	8 (36%)
17	Postswallow pharyngeal residue	**81 (76%)**	4 (67%)	**67 (86%)**	10 (45%)
18	Postswallow pharyngeal pooling	72 (69%)	4 (67%)	**58 (76%)**	10 (45%)
19	Piecemeal deglutition	50 (47%)	3 (43%)	34 (44%)	13 (62%)
20	Penetration/aspiration	**99 (92%)**	**7 (88%)**	**78 (99%)**	14 (67%)
21	Choking	**87 (78%)**	**8 (89%)**	**66 (84%)**	13 (54%)
22	Medication intake	76 (68%)	4 (44%)	**65 (83%)**	7 (28%)
23	Weight loss	63 (57%)	1 (12%)	**55 (70%)**	7 (29%)
24	Voice parameters	52 (46%)	5 (56%)	34 (44%)	13 (50%)
25	Level of oral intake	77 (68%)	3 (33%)	**64 (81%)**	10 (40%)
26	Hydration	**80 (71%)**	4 (44%)	**66 (84%)**	10 (40%)
27	Avoidance of food/liquids	72 (64%)	6 (67%)	53 (68%)	13 (52%)
28	Respiratory swallow coordination	72 (66%)	6 (67%)	**54 (70%)**	12 (52%)
29	Cough production	**84 (76%)**	4 (50%)	**66 (86%)**	14 (56%)
30	Cough sensitivity	**76 (70%)**	4 (50%)	**59 (78%)**	13 (52%)
31	Aspiration pneumonia	**90 (82%)**	4 (44%)	**72 (92%)**	14 (61%)
32	Mealtime duration	49 (44%)	1 (11%)	42 (54%)	6 (24%)
33	Independent eating/drinking	59 (53%)	4 (44%)	44 (56%)	11 (44%)
34	Social aspect of eating/drinking	65 (58%)	4 (44%)	45 (57%)	16 (64%)
35	Pleasure of oral intake	61 (54%)	4 (44%)	47 (60%)	10 (40%)
36	Fear of eating/drinking	68 (61%)	6 (67%)	50 (64%)	12 (50%)
37	Fear of choking	70 (62%)	**7 (78%)**	50 (64%)	13 (52%)
38	Swallowing related quality of life	**81 (73%)**	**7 (78%)**	**64 (81%)**	10 (43%)
39	Self-perception of swallowing	64 (57%)	3 (33%)	53 (67%)	8 (32%)
40	Carer burden	53 (49%)	4 (44%)	41 (53%)	8 (36%)
41	Participant's experience with intervention	54 (50%)	2 (29%)	42 (54%)	10 (45%)
42	Participant's adherence to intervention	70 (66%)	2 (29%)	**56 (72%)**	12 (57%)
43	Hospitalisation	70 (67%)	1 (11%)	**59 (77%)**	10 (53%)
44	Adverse events	**77 (74%)**	4 (50%)	**60 (79%)**	13 (65%)
45	Mortality	**85 (79%)**	4 (50%)	**68 (86%)**	13 (65%)

Delphi Round 2 - Ratings of critically importance (7-9) per stakeholder group and in total

	Outcome	Total	Family m/c	HCP/Researcher	PwPD
1	Oropharyngeal dysphagia severity	**88 (87%)**	**7 (88%)**	**70 (95%)**	11 (58%)
2	Laryngeal sensation	**81 (80%)**	**5 (71%)**	**62 (85%)**	14 (67%)
3	Postswallow pharyngeal residue	**83 (82%)**	4 (57%)	**68 (93%)**	11 (52%)
4	Penetration/aspiration	**95 (92%)**	**7 (88%)**	**73 (99%)**	**15 (71%)**
5	Choking	**85 (83%)**	**7 (88%)**	**63 (85%)**	**15 (71%)**
6	UES opening during swallowing	55 (59%)	3 (60%)	41 (58%)	11 (61%)
7	Hydration	**84 (81%)**	4 (50%)	**68 (92%)**	12 (55%)
8	Cough strength	**81 (79%)**	4 (57%)	**66 (90%)**	11 (50%)
9	Cough sensitivity	**79 (77%)**	4 (57%)	**63 (86%)**	12 (55%)
10	Aspiration pneumonia	**89 (87%)**	5 (62%)	**70 (96%)**	14 (67%)
11	Swallowing related quality of life	**84 (81%)**	5 (62%)	**68 (92%)**	11 (50%)
12	Adverse events	**80 (80%)**	4 (67%)	**64 (88%)**	12 (57%)
13	Mortality	**84 (82%)**	5 (62%)	**65 (88%)**	**14 (70%)**

Delphi Round 3 - Ratings of critically importance (7-9) per stakeholder group and in total

	Outcome	Total	Family m/c	HCP/Researc	PwPD
1	Oropharyngeal dysphagia severity	**57 (65%)**	3 (43%)	**45 (69%)**	9 (56%)
2	Laryngeal sensation	33 (38%)	1 (14%)	27 (42%)	5 (29%)
3	Post swallow pharyngeal residue	**54 (61%)**	3 (43%)	**45 (70%)**	6 (35%)
4	Penetration/aspiration	**75 (83%)**	**5 (71%)**	**58 (89%)**	**12 (67%)**
5	Choking	**60 (67%)**	**6 (86%)**	**41 (63%)**	**13 (72%)**
6	Hydration	36 (40%)	2 (29%)	28 (43%)	6 (33%)
7	Cough strength	43 (48%)	0 (0%)	38 (59%)	5 (28%)
8	Cough sensitivity	35 (40%)	2 (29%)	26 (41%)	7 (39%)
9	Aspiration pneumonia	**65 (73%)**	4 (57%)	**52 (81%)**	9 (50%)
#	Swallowing related quality of life	**56 (62%)**	2 (29%)	**48 (74%)**	6 (33%)
#	Adverse events	35 (41%)	1 (20%)	28 (45%)	6 (33%)
#	Mortality	46 (52%)	2 (29%)	33 (52%)	**11 (65%)**

Appendix 7.10: Outcomes and definitions uploaded to DelphiManager for each Delphi round

Round 1

Name	HelpText	DomainName
Oropharyngeal dysphagia severity	Changes in overall severity of swallowing difficulty	General swallowing related outcomes
Dry mouth	Changes in how the mouth feels in terms of dryness	General swallowing related outcomes
Drooling	Changes in frequency and severity of saliva leaking out of the mouth	General swallowing related outcomes
Salivary pooling	Changes in amount of saliva accumulated in the mouth or throat	General swallowing related outcomes
Orolingual bolus control	Changes in control over food/liquids in the mouth	General swallowing related outcomes
Oral bolus transport	Changes in the movement of food/liquids through the mouth	General swallowing related outcomes
Lingual muscle strength	Changes in strength of tongue muscles	General swallowing related outcomes
Nasal regurgitation	Changes in frequency of food/liquids going the wrong way - into the nose	General swallowing related outcomes
Swallowing related orolingual movements	Changes in tongue "pumping" or "rocking" movements when eating/drinking	General swallowing related outcomes
Timing of oropharyngeal swallow components	Changes in timing of swallow components in the mouth/throat, e.g. swallow is faster/slower	General swallowing related outcomes
Laryngeal elevation	Changes in upwards movement of the voice box (larynx) when swallowing	General swallowing related outcomes
Laryngeal sensation	Changes in feeling of food/liquids being stuck in the throat	General swallowing related outcomes
Swallowing related hyoid bone movement	Changes in movement of a specific bone in the throat when swallowing	General swallowing related outcomes
Pharyngeal muscle strength	Changes in strength of throat muscles	General swallowing related outcomes
Initiation of pharyngeal swallow	Changes in overall speed of swallowing	General swallowing related outcomes
Postswallow oral residue	Changes in food/liquids left in the mouth after swallowing	General swallowing related outcomes
Postswallow pharyngeal residue	Changes in food/liquids left in the throat after swallowing	General swallowing related outcomes
Postswallow pharyngeal pooling	Changes in food/liquids accumulating in the throat after swallowing	General swallowing related outcomes
Piecemeal deglutition	Changes in need to break down food/liquids in the mouth in order to swallow	General swallowing related outcomes
Penetration/aspiration	Changes in frequency of food/liquids going the wrong way - into the airway/windpipe	General swallowing related outcomes
Choking	Changes in frequency of choking episodes	General swallowing related outcomes
Medication intake	Changes in swallowing Parkinson's medication	General swallowing related outcomes
Weight loss	Changes in body weight related to swallowing difficulties	General swallowing related outcomes
Voice parameters	Changes in voice, e.g. loudness, voice quality	General non-swallowing related outcomes
Level of oral intake	Changes in how food/liquids are taken (e.g., via mouth, feeding tube, pureed diet, thickened liquids)	Nutritional and dietary outcomes
Hydration	Changes in hydration status	Nutritional and dietary outcomes
Avoidance of food/liquids	Changes in avoidance of food/liquids related to swallowing difficulties	Nutritional and dietary outcomes
Respiratory swallow coordination	Changes in the coordination of breathing and swallowing	Respiratory outcomes
Cough production	Changes in cough strength so the airway/windpipe can be cleared easier	Respiratory outcomes
Cough sensitivity	Changes in coughing as a response of feeling food/liquids going down the wrong way	Respiratory outcomes
Aspiration pneumonia	Changes in frequency of chest infections	Respiratory outcomes
Mealtime duration	Changes in the duration of a mealtime related to swallowing difficulties	Physical functioning
Independent eating/drinking	Changes in level of independence while eating/drinking	Physical functioning
Social aspect of eating/drinking	Changes in social activities because of swallowing difficulties	Social functioning
Pleasure of oral intake	Changes in pleasure of eating/drinking related to swallowing	Emotional functioning/wellbeing
Fear of eating/drinking	Changes in fear or anxiety related to eating/drinking	Emotional functioning/wellbeing
Fear of choking	Changes in fear or anxiety of choking related to eating/drinking	Emotional functioning/wellbeing
Swallowing related quality of life	Changes in quality of life related to swallowing difficulties	Global quality of life
Self-perception of swallowing	Changes in overall awareness of swallowing function	Perceived health status
Carer burden	Changes in caregiver's quality of life related to swallowing difficulties in person with Parkinson's	Delivery of care
Participant's experience with intervention	Opportunity to give feedback on overall swallowing program	Delivery of care
Participant's adherence to intervention	Person with Parkinson's following the intervention as prescribed	Delivery of care
Hospitalisation	Changes in number of hospital admissions related to swallowing difficulties	Hospital
Adverse events	Unfavorable or harmful side effects that occur during or after the swallowing treatment	Adverse events/effects
Mortality	The event of death	Mortality/survival

Round 2

Name	HelpText	DomainName
Oropharyngeal dysphagia severity	Changes in overall severity of swallowing difficulty	General swallowing related outcomes
Laryngeal sensation	Changes in feeling of food/liquids/saliva being stuck in the larynx (voice box)	General swallowing related outcomes
Postswallow pharyngeal residue	Changes in food/liquids/saliva left in the throat after swallowing	General swallowing related outcomes
Penetration/aspiration	Changes in frequency of food/liquids/saliva going the wrong way - into the airway/windpipe	General swallowing related outcomes
Choking	Changes in frequency of choking episodes	General swallowing related outcomes
UES opening during swallowing	Changes in opening of the Upper Esophageal Sphincter (short UES). The UES is the junction of the throat and th	General swallowing related outcomes
Hydration	Changes in hydration status	Nutritional and dietary outcomes
Cough strength	Changes in cough strength so the airway/windpipe can be cleared easier	Respiratory outcomes
Cough sensitivity	Changes in coughing as a response of feeling food/liquids/saliva going down the wrong way	Respiratory outcomes
Aspiration pneumonia	Changes in frequency of aspiration pneumonia. Aspiration pneumonia is a lung inflammation that is caused by food/liquids/saliva going into the airway/windpipe.	Respiratory outcomes
Swallowing related quality of life	Changes in quality of life related to swallowing difficulties	Global quality of life
Adverse events	Unfavorable or harmful side effects that occur during or after the swallowing treatment	Adverse events/effects
Mortality	The event of death	Mortality/survival

Note: changes made between rounds are highlighted in red

Round 3

Name	HelpText	DomainName
Oropharyngeal dysphagia severity	Changes in overall severity of swallowing difficulty	General swallowing related outcomes
Laryngeal sensation	Changes in feeling of food/liquids/saliva being stuck in the larynx (voice box)	General swallowing related outcomes
Postswallow pharyngeal residue	Changes in food/liquids/saliva left in the throat after swallowing	General swallowing related outcomes
Penetration/aspiration	Changes in frequency of food/liquids/saliva going the wrong way - into the airway/windpipe	General swallowing related outcomes
Choking	Changes in frequency of choking episodes. Choking is a blockage of the upper airway by food/liquids/saliva, which can disrupt breathing.	General swallowing related outcomes
Hydration	Changes in hydration status	Nutritional and dietary outcomes
Cough strength	Changes in cough strength so the airway/windpipe can be cleared easier	Respiratory outcomes
Cough sensitivity	Changes in coughing as a response of feeling food/liquids/saliva going down the wrong way	Respiratory outcomes
Aspiration pneumonia	Changes in frequency of aspiration pneumonia. Aspiration pneumonia is a lung inflammation that is caused by food/liquids/saliva going into the airway/windpipe.	Respiratory outcomes
Swallowing related quality of life	Changes in quality of life related to swallowing difficulties	Global quality of life
Adverse events	Unfavorable or harmful side effects that occur during or after the swallowing treatment	Adverse events/effects
Mortality	The event of death	Mortality/survival

Note: changes made between rounds are highlighted in red

Appendix 7.11: Suggested outcome definitions and outcome measurement instruments with changes made by consensus meeting participants

Core Outcome		Devised by SSC	Suggested changes in consensus meeting 1	Suggested changes in consensus meeting 2
Penetration/ aspiration	Definition	Penetration: Entry of material (e.g., food, fluids or saliva/secretions) into the larynx that does not pass below the vocal folds. Aspiration: Entry of material (e.g., food, fluids or saliva/secretions) into the larynx and below the level of the vocal folds.		OMI: Add: DIGEST safety rating (Hutcheson et al. 2017, 2022) for VFS, DIGEST-FEES safety rating (Labeit et al., 2023) for FEES, VASES safety rating (Curtis et al., 2022) for FEES Remove: PAS (Rosenbek et al., 1996) for FEES
	OMIs	VFS/FEES: e.g. PAS (Rosenbek et al., 1996) FEES: e.g. Penetration-aspiration events (Warnecke et al., 2016)		
Aspiration pneumonia	Definition	An acute respiratory infection likely caused by aspiration of material (e.g., food, fluids or saliva/secretions).		
	OMI	A clinical suspicion of a new or worsening respiratory infection in the context of at least 2 of the following: temperature <35°C or >38°C; abnormal chest x-ray (CT scan); a white cell count of <4x109/l or >11x109/l or purulent (tracheal) secretions.		
Oropharyngeal dysphagia severity	Definition	The overall degree of impairment (i.e., swallowing safety and swallowing efficiency) in swallowing material (e.g., food, liquids or saliva/secretions).		
	OMIs	VFS: e.g. DIGEST (Hutcheson et al., 2017, 2022) FEES: e.g. DIGEST-FEES (Labeit et al. 2023), VASES (Curtis et al., 2022), Endoscopic PD dysphagia score (Warnecke et al., 2016)	OMI: Remove: VASES (Curtis et al., 2022) for FEES	OMI: Add: MBSImP (Martin-Harris et al., 2008) for VFS Remove: VASES (Curtis et al., 2022) for FEES
Choking	Definition	A condition that occurs when breathing is impeded through the inhalation or ingestion of material (e.g., food, fluids or saliva/secretions), which causes obstruction of the respiratory tract.		Definition: A condition that occurs when breathing is perceived to be impeded through the inhalation or ingestion of material (e.g., food, fluids or saliva/secretions) which is felt as an obstruction of the respiratory tract.
	OMI	e.g. report number of choking events over or within a specific time period		
Swallowing related quality of life	Definition	An individual's perception of their ability to swallow within the context of their daily life, considering the cultural norms, values, and personal goals that influence their experience.		
	OMI	e.g. SWAL-QOL (McHorney et al., 2002)		
Post swallow pharyngeal residue	Definition	The presence of material (e.g., food, fluids or saliva/secretions) remaining in the pharynx after swallowing.		
	OMIs	VFS & FEES: e.g. Yale Pharyngeal Residue Severity Rating Scale (Neubauer et al., 2015) VFS: e.g. Bolus Residue Scale (Rommel et al., 2015), Eisenhuber Scale (Eisenhuber et al., 2002) FEES: e.g. Residue Scale (Warnecke et al., 2016)	OMI: Add: VASES efficiency rating (Curtis et al., 2022) for FEES	OMI: Add: VASES efficiency rating (Curtis et al., 2022) for FEES Remove: Yale Scale (Neubauer et al., 2015) for VFS, Eisenhuber Scale (Eisenhuber et al., 2022) for VFS

DIGEST = Dynamic Imaging Grade of Swallowing Toxicity, FEES = Fibreoptic Endoscopic Evaluation of Swallowing, MBSImP = Modified Barium Swallow Impairment Profile, OMI = outcome measurement instrument, PAS = Penetration-Aspiration Scale, SSC = Study Steering Committee, VASES = Visual Analysis of Swallowing Efficiency and Safety, VFS = Videofluoroscopy