University of Southampton

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Development of a Core Outcome Set to Evaluate Physical Activity Interventions for People Living with Dementia

by

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Thesis for the degree of Doctor of Philosophy

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Abstract

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Research on physical activity interventions for people living with dementia has increased in the last decade. However, literature reviews report limitations in comparing studies due to their heterogeneous selection of outcomes. Therefore, guidance for practice is yet to be established. This thesis aimed to develop a Core Outcome Set – an agreed set of outcomes to be measured, as a minimum, in all effectiveness trials – to evaluate physical activity interventions for people living with dementia, across stages of the disease, intervention settings, in research and practice.

A systematic literature review identified a total of 133 outcomes reported in physical activity interventions for people living with dementia. A qualitative study with key stakeholders (patients, informal carers and professionals) added ten new outcomes to those previously reported. This qualitative study also identified that physical activity was meaningful to patients, not only because of its potential physiological and wellbeing benefits, but also for its impact on identity, perceived roles towards others and connections to the present. A consensus study (modified Delphi survey) was then conducted to reach agreement on the minimum set of core outcomes. Informed by patient and public involvement activities, people living with dementia were included in the consensus process through an innovative card sorting strategy. Consensus was reached on seven outcomes: “preventing falls”; “doing what you can do”; “staying healthy and fit”; “walking better, being able to stand up and climb stairs”; “feeling brighter”; “enjoying the moment”; and, “feeling useful and having a purpose”. However, physical activity may incur side effects and does not happen in isolation from informal carers. A prioritisation exercise was, therefore, completed with professionals and carers, which identified “becoming agitated and confused”, “falling over” and “feeling discomfort and pain” as the most undesirable negative side effects of physical activity for people living with dementia; and, “carer feeling positive and satisfied”; “carer improving wellbeing” and “making lives of carers easier”, as priority outcomes for carers.

This PhD contributes to methodological knowledge of Core Outcome Set development (through the use of mixed methods and an innovative card sorting strategy to engage people living with dementia) and informs our understanding of meaningful outcomes of physical activity for people living with dementia. These outcomes should be measured, as a minimum, in all future trials of physical activity in this population.

Key words: Core Outcome Set; Physical activity; Dementia; Consensus; Patient and public involvement.

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Research Thesis: Declaration of Authorship

Print name:	Ana Carolina Vieira Gonçalves
Title of thesis:	Development of a Core Outcome Set to Evaluate Physical Activity Interventions for People Living with Dementia.

I declare that this thesis and the work presented in it are my own and has been generated by me as the result of my own original research.

I confirm that:

1. This work was done wholly or mainly while in candidature for a research degree at this University;
2. Where any part of this thesis has previously been submitted for a degree or any other qualification at this University or any other institution, this has been clearly stated;
3. Where I have consulted the published work of others, this is always clearly attributed;
4. Where I have quoted from the work of others, the source is always given. With the exception of such quotations, this thesis is entirely my own work;
5. I have acknowledged all main sources of help;
6. Where the thesis is based on work done by myself jointly with others, I have made clear exactly what was done by others and what I have contributed myself;
7. Parts of this work have been published as:

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Abbreviations

COMET: Core Outcome Measures in Effectiveness Trials

COS-STAD: Core Outcome Set – STAndards for Development

COS-STAR: Core Outcome Set – STAndards for Reporting

ICHOM: International Consortium for Health Outcomes Measurement

OMERACT: Outcome Measures in Rheumatology

Chapter 1 Introduction

1.1 Thesis introduction and overview

Dementia is a syndrome characterised by progressive degeneration of the structure and function of the brain. Dementia symptoms include memory loss, language difficulties and functional impairment (All-Party Parliamentary Group on Dementia, 2016;OECD, 2015). It can be caused by a variety of underlying diseases, Alzheimer’s disease being the most common, affecting approximately two thirds of those living with dementia (Alzheimer’s Disease International, 2018). Other common types of dementia are Vascular dementia, Mixed dementia, dementia with Lewy bodies and Frontotemporal dementia (van der Flier and Scheltens, 2005). Dementia is a chronic condition with no current cure available and with an incidence that increases with age. Therefore, the number of people living with dementia is predicted to continue to increase, as life expectancy continues to rise (Alzheimer's Disease International, 2015).

Dementia impacts not only on the cognitive and physical health on those living with the condition, but also has important impact on the person’s financial and social health domains (Alzheimer's Disease International, 2015). As the disease progresses, people living with dementia become progressively more dependent on their families as well as on social and health care services, leading to a high burden of disease (Alzheimer’s Disease International, 2018). This fact, together with its growing prevalence, has meant that tackling dementia has been identified as a priority in several developed countries (Department of Health, 2015;Chow et al., 2018). While receiving more attention from governments and funders, research in the field of dementia has also expanded in recent years.

Physical activity is one of the growing areas of research within the field of dementia care. Physical activity is broadly defined as any body movement that involves activation of skeletal muscle and energy expenditure (Caspersen et al., 1985). Examples of physical activity include formal exercise, activities of daily living, walking or housekeeping. Physical activity is associated with healthy ageing. Healthy ageing is defined by the World Health Organisation as a status where functional abilities and wellbeing are maintained in older age, regardless of the presence of disease (World health Organization, 2015c). Older adults who take part in physical activity, at sufficient intensity to improve cardiorespiratory fitness, are known to have better cognitive function than their peers (Bherer et al., 2013). Those taking part in resistance training have been shown to have better gait speed and therefore, reduced mortality (Bherer et al., 2013). Further, in a general population of community dwelling older adults, those who take part in medium to high levels of physical activity are known to have a reduced risk of disability in basic activities of daily living (Tak et al., 2013). It has, therefore, often been hypothesised that physical activity could be associated with similar health benefits for people living with dementia (Lautenschlager et al., 2012). The potential benefits of physical activity are numerous (e.g., improvements in functional independence, cognition and wellbeing; and reductions in behaviour disturbances, burden of care and number of falls), and researchers have spread their attention across many of these possible outcomes.

As researchers look into numerous possible outcomes, the number of publications, including systematic reviews with meta-analyses, in this area of research has increased. However, an heterogeneity of reported outcomes means that these efforts have not resulted in clearer recommendations for clinical practice, and no guidance is yet available on the recommended dose(s) and type(s) of activity for people living with dementia. To address this heterogeneity of reported outcomes, and with the view to fast-tracking recommendations to practice, this thesis aimed to develop a Core Outcome Set to evaluate physical activity interventions for people living with dementia. Core Outcome Sets are agreed sets of outcomes and measurement tools, to be reported as a minimum in all clinical trials of a given health condition and/or intervention (Williamson et al., 2017). Core Outcome Sets should represent outcomes that are most relevant to all key stakeholder groups, in this case, people living with dementia, their family carers and professionals involved in their care. By doing so, Core Outcome Sets guide researchers on “what to measure” (outcomes) and “how to measure” (measurement tools). If successfully implemented, Core Outcome Sets can facilitate comparisons between clinical trials, enabling all trials to contribute meaningful data whilst minimising publication bias (Williamson et al., 2012).

The development of this Core Outcome Set is presented in seven chapters, which include five original publications[1] and address the first stage of development of this Core Outcome Set – “What to measure”. The “How to measure” stage (identification of measurement tools) is beyond the scope of this thesis and is described as planned future work. Chapter I introduces the research problem and presents an overview of the thesis and its specific research questions. Chapter 2 provides background information on the ontological and epistemological positions assumed in the development of this Core outcome Set and this thesis. It also provides background for the current knowledge on dementia prevalence and burden of disease, possible outcomes of physical activity for people living with dementia and Core Outcome Sets. Chapter 3 is the methods chapter and includes Paper I: a protocol for the development of this Core Outcome Set. This paper provides important insights into the methodological approaches in the development of this Core

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[1] The first author of all publications was responsible for the study design, data collection, data analysis and drafting of the manuscripts. The remaining authors were consulted for advice on study design, data collection and data analysis and critically reviewed the manuscripts prior to submission for publication.

Outcome Set and it describes the adaptations undertaken to fully include people living with dementia. Chapter 3 also includes a description of the changes made to the protocol since its publication, and provides a rationale for those changes. Chapter 4 aimed at identifying a comprehensive list of possible outcomes of physical activity for people living with dementia, often named by Core Outcome Set developers as the “long list of outcomes”. This is accomplished through two publications. Paper II reports on a systematic literature review identifying all outcomes measured or described in the last decade of research in the field of physical activity for people living with dementia. Paper III is a qualitative study with key stakeholders (patients, informal carers and professionals) that adds to the list of outcomes reported in the previous literature review, by identifying any relevant outcomes not previously investigated, while gaining deeper understanding on why those outcomes are considered important. Chapter 5 aims to narrow the long list down to those outcomes considered core by all stakeholders involved. This was achieved through two original publications. Paper IV reports on a modified Delphi survey, using an innovative approach to include people living with dementia to reach consensus on what positive outcomes should be included in the final Core Outcome Set. Paper V describes a prioritisation exercise of negative outcomes of physical activity; and outcomes that physical activity for the person living with dementia may have on their carers. Although not part of the final consensus, these outcomes can provide important guidance for the implementation physical activity interventions in research and practice. An integrated discussion of all publications and recommendations for future research is presented in Chapter 6. Lastly, Chapter 7 outlines the conclusion of this thesis. Figure 1, below, presents a visual summary of the structure of this thesis, including its aims, research questions and publications.

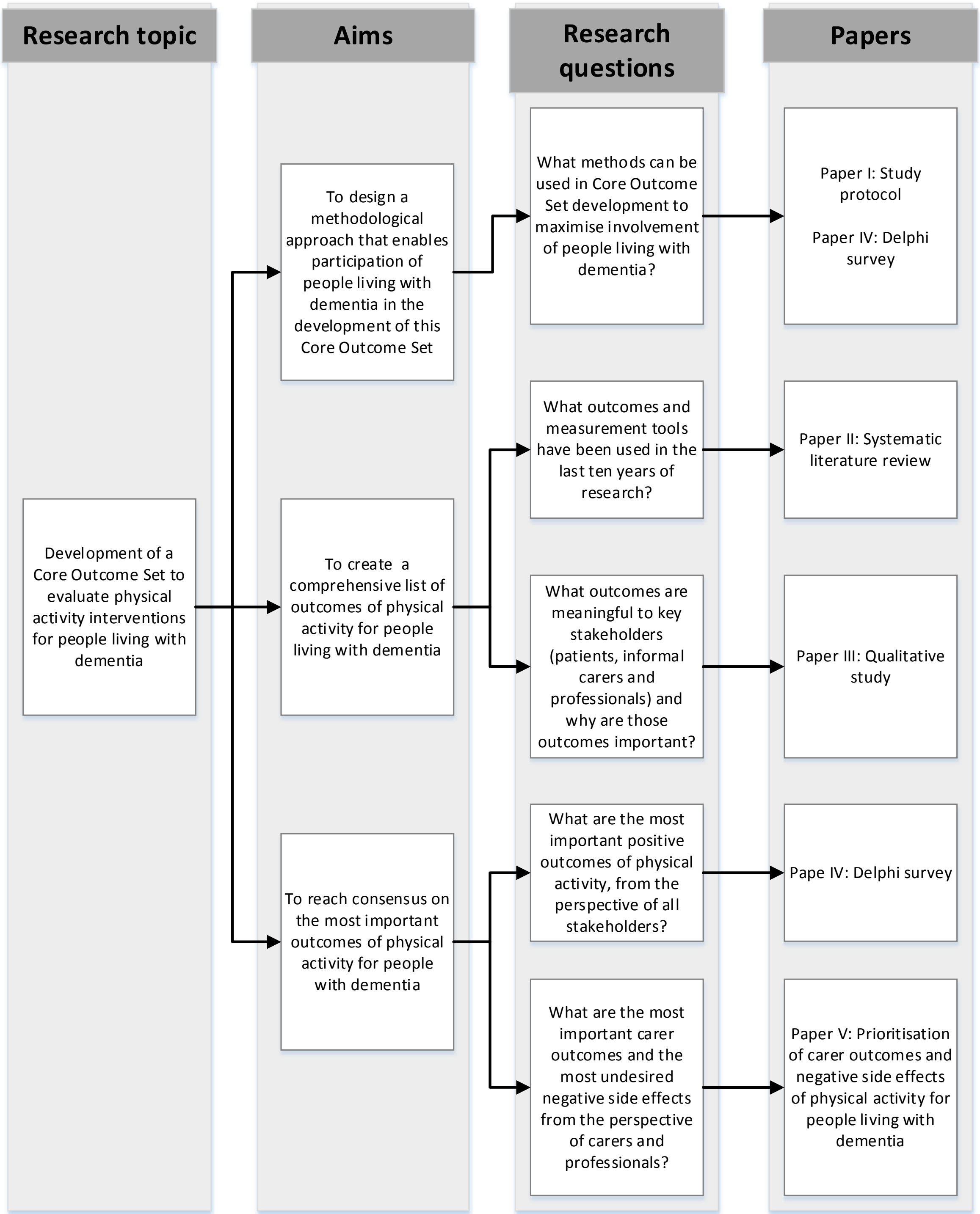

Figure 1 - Outline of the five papers included in this thesis.

Chapter 2 Background

2.1 Ontology and epistemology

All research questions, and all methods used to answer research questions, are always underpinned by the researchers' stance on what is perceived as "real" (ontology) and what counts as "valid knowledge" (epistemology). The research questions that build the development of the Core Outcome Set outlined in this thesis (listed in Chapter 1, Figure 1) are no exception. It is therefore important to provide a background to the ontological and epistemological positions that informed the development of this Core Outcome Set. Ontology and epistemology are interdependent as one's view of what is real will determine what counts as valid knowledge. The variety of ontological and epistemological positions that can be assumed by researchers form a spectrum. The two ends of the spectrum are briefly outlined below, as well as the middle ground that was assumed in the development of this Core Outcome Set.

On the one end of the spectrum, realism is the ontological position that sees reality as logical, factual and objective; a single reality, based on one single truth, which is not influenced by context or interpretation (Braun and Clarke, 2013;Tebes, 2005). In response to realism, positivism is the epistemological position that assumes that truth can be deducted from experiments and that valid knowledge is accurate, replicable, generalizable, based on hypotheses and tested using empirical measurements (Tebes, 2005). These frameworks are typically assumed in quantitative studies (Braun and Clarke, 2013).

In contrast, at the other end of the spectrum, relativism is the ontological position that assumes a reality that cannot be found, discovered or tested: it is only produced by human interpretation. Therefore, relativists assume not one, but multiple realities and that knowledge can never reach beyond the filters of discourse, social and time constructs. Assuming a "relative" reality, constructionism is the epistemological position that sees knowledge as a creation of discourse, culture, society, time and context. For constructionists, knowledge is a product of how we understand and make sense of the word (Braun and Clarke, 2013;Nightingale and Cromby, 2002). These positions tend to be adopted in qualitative studies.

Critical realism is an ontological position that "sits on the fence" between realism and relativism, and it is the ontological position that underpins the research presented in this thesis. Critical realists accept that a reality exists behind social constructs, but that reality can only be partially reached. In line with critical realism, the epistemological position of contextualism accepts that knowledge depends on context and, therefore, pursues valid knowledge, of that reality, through a plurality of methods. Multiple methods can all offer valid scientific knowledge of that same truth, which is legitimate, but situated and contextual (Braun and Clarke, 2013;Tebes, 2005;Nightingale and Cromby, 2002). Critical realism and contextualism are a natural fit for mixed methods research, such as the development of this Core Outcome Set. A possible example of a contextualist approach to the research question: “What is the minimum core of outcomes that should be measured in physical activity interventions for people with dementia?” can be presented as follows. If participants describe that “muscle strength” can be improved through physical activity, “muscle strength” is accepted as a reality, that exists beyond human interpretation, and which can be objectively measured (with a dynamometer for instance). But it is the context around the participants and the value they give to “being stronger” that makes “improving strength” a meaningful outcome of taking part in physical activity. Therefore, both methods of pursuing knowledge (quantitative via the dynamometer, and qualitative, through interviews) will allow a closer look at the “truth”, and help address the research questions included in the development of this Core Outcome Set.

This first section of the background chapter sets the philosophical grounds of the development of this Core Outcome Set to evaluate physical activity interventions for people living with dementia. The following sections will provide background information on the prevalence and disease burden of dementia, the current knowledge regarding outcomes of physical activity in this population, and methodological considerations for Core Outcome Set development.

2.2 Dementia: Prevalence and burden of disease

In 2018, 50 million people lived with dementia worldwide, and 152 million are predicted to be living with the condition by 2050 (Alzheimer’s Disease International, 2018). The estimated incidence of 7.7 million new cases worldwide each year, corresponds to a new case every three seconds (Alzheimer’s Disease International, 2018). The high incidence and prevalence of dementia has substantial implications for health and social care costs - currently 1 trillion US$ per year, a figure predicted to double by 2030 - making dementia one of the greatest challenges of the century (van der Flier and Scheltens, 2005;Alzheimer’s Disease International, 2018;Livingston et al., 2017). It is estimated that 85% of those costs are attributed to care provided by family and social services (Livingston et al., 2017).

Dementia is the condition with single greatest impact on disability, and need for support with basic care, amongst older people (Alzheimer's Disease International, 2013b). Its impact on the quality of life of patients and families can be devastating (Alzheimer's Disease International, 2013a;Wimo et al., 2007). With no cure currently available and remaining uncertainty about the pathophysiology of dementia, international organisations have indicated the need for research that aims to address not only curative treatments, but also opportunities for prevention, disease modifiable interventions and provision of care, throughout the course of the condition (OECD, 2015;Livingston et al., 2017;Alzheimer’s Disease International, 2018).

Physical activity is amongst the interventions investigated as both a risk reduction strategy (Livingston et al., 2017;Hamer and Chida, 2009;Aarsland et al., 2010) and a component of care for people living with dementia (Forbes et al., 2015). The role of physical activity in preventing or delaying the onset of dementia is beyond the scope of this thesis. Instead, the next section will focus on the current knowledge about the possible effects of physical activity on those already diagnosed with the condition and the current recommendations regarding physical activity for people living with dementia.

2.3 Physical activity: Current knowledge of clinical outcomes and recommendations for people living with dementia

The sections below present the available knowledge on clinical outcomes of physical activity interventions for people living with dementia. For purposes of clarity they have been organised into: “cognition, behaviour, mood and mental health”, “physical health”, “use of health services”, “carer outcomes” and “adverse events”. Whilst this section will focus on the current available knowledge on clinical outcomes of physical activity for people living with dementia, Chapter 4 – Paper II provides detail on how frequently each of the outcome has been reported in recent literature.

2.3.1 Cognition, behaviour, mood and mental health

Conflicting results have been reported by literature reviews on the impact of physical activity on cognition for people living with dementia, with four reviews reporting improvements in cognition (Heyn et al., 2004;Groot et al., 2016;Learner and Williams, 2016;Lee et al., 2016) and three reviews describing no effects in this domain (Ohman et al., 2014;Zeng et al., 2016;Park and Cohen, 2018). Of the four literature reviews advocating positive effects of physical activity on cognition, only two completed a meta-analysis regarding this outcome (Heyn et al., 2004;Groot et al., 2016), which showed small to medium effects sizes: 0.42 with 95% confidence interval of 0.23 to 0.62 (Groot et al., 2016); 0.46 with 95% confidence interval of 0.26 to 0.66 and 0.57 with 95% confidence interval of 0.43 to 1.17 (Heyn et al., 2004). However, all these meta-analyses were limited by the heterogeneity of populations, interventions and use of measurement tools. One literature review included original studies with participants living with mild cognitive impairment as well as with dementia (heterogeneity of population) (Heyn et al., 2004). Three literature reviews highlighted that the included studies reported effects on cognition using multiple different measurement tools, which may have affected the accuracy of their analyses (Groot et al., 2016;Heyn et al., 2004;Lee et al., 2016). All four literature reviews reported on heterogeneous physical activity interventions, with duration varying from two to 112 weeks, one to seven times per week, with sessions ranging from 20 to 150 minutes (Heyn et al., 2004;Groot et al., 2016;Lee et al., 2016;Learner and Williams, 2016). None of the literature reviews concluding that physical activity had no effect, was able to produce a meta-analysis for this outcome due to heterogeneity of measurement tools used to assess cognition in the included studies (Ohman et al., 2014;Zeng et al., 2016;Park and Cohen, 2018). Moreover, a large variety of study designs was also reported (Ohman et al., 2014;Zeng et al., 2016;Park and Cohen, 2018), similarly to the reviews describing positive effects on cognition. Even less clarity exists regarding long-term effects. Recent research has found no evidence of the maintenance of positive effects of physical activity on cognition in the long term, either because the included studies in literature reviews did not include long term follow ups (Learner and Williams, 2016), or because the follow up times were very disparate, ranging from three to 36 months, which impaired reaching definitive conclusions (Park and Cohen, 2018).

With regards to other aspects of mental health and wellbeing, evidence from clinical trials remains inconclusive. It has been suggested that any activity that meets the interests and abilities of people with dementia has the potential to reduce behavioural symptoms (Trahan et al., 2014). However, no statistical evidence of improvement in overall levels of challenging behaviour in people with dementia has been found in recent systematic literature reviews exploring the effectiveness of physical activity in this domain (Barreto et al., 2015;Ginis et al., 2017;Lamb et al., 2018;Park and Cohen, 2018). Small, mixed or no effects have also been described with regard to depression in this population (Barreto et al., 2015;Ginis et al., 2017;Lee et al., 2016;Zeng et al., 2016;Morris et al., 2017). Another possible outcome of physical activity for people living with dementia is a reduction in use of anti-psychotic medication, but data on this outcome is often not collected and thus, literature reviews remain inclusive on this outcome (Barreto et al., 2015). A recent pilot randomised controlled trial (Maltais et al., 2019) noted a reduction of the total number of medications taken by participants in the exercise group compared to control (social intervention), however this difference was no longer noticeable when considering antipsychotic medication alone (Maltais et al., 2019). Data from larger randomised controlled trials on this outcome are still needed. Mood is another possible wellbeing outcome of physical activity for people living with dementia with scarce data available. Mood has been reported to improve with physical activity interventions for people living with dementia (Rolland et al., 2008), however, this literature review included one single primary study measuring mood as an outcome. In qualitative studies, people with dementia have reported positive effects on wellbeing, levels of alertness, expressions of creativity, humour and sense of purpose (Wright, 2016).

2.3.2 Physical health

Physical health outcomes, including mobility, number and risk of falls, balance and pain have also been explored as possible positive outcomes of physical activity interventions for people living with dementia. However, there is still little certainty about the effects of physical activity on these outcomes. Mobility, for instance, has been the focus of a systematic literature review (Pitkala et al., 2013a), however, a meta-analysis was not possible due to the poor quality of the included studies. A description of the included studies considered of high quality (two in the community and two in institutional settings) suggests that physical activity may have a positive effect on mobility (Pitkala et al., 2013a). Number and risk of falls is also an outcome frequently considered in physical activity interventions for people living with dementia. A consensus process where a recommendation is made towards physical activity for people living with dementia for its “promising evidence” on falls risk reduction has been reported (Ginis et al., 2017). However, the empirical literature used to support this recommendation cannot, in fact, provide robust evidence that physical activity for people living with dementia can indeed reduce falls, as it includes: one pilot randomised controlled trial where there was no significant difference in the number of falls between groups (Mackintosh and Sheppard, 2005); three clinical trials that measured balance and mobility, but not falls (Santana-Sosa et al., 2008;Christofoletti et al., 2008;Ries et al., 2010) and one case study (Mirolsky-Scala and Kraemer, 2009). Therefore, despite the available recommendations, it is yet to be clarified if physical activity for people living with dementia can or cannot reduce the incidence of falls in this population.

Scarce evidence also exists for the effect of physical activity on pain levels of people living with dementia, as pain management is not a frequently reported outcome (see Chapter 4, Paper II). A recent pilot clustered randomised controlled trial including people living with dementia in nursing homes, measured pain as secondary outcome but found no statistical difference between an exercise group and a social group with regards to pain levels or use of analgesic medication (Maltais et al., 2019).

Reduction in mortality has also been explored as a possible outcome of physical activity interventions for people living with dementia, but recent literature reviews concluded that there is no evidence of reduction in mortality as an outcome of physical activity (Barreto et al., 2015). Nevertheless, it has been highlighted that many of the included studies measuring mortality do not have follow up times long enough to capture deaths in either intervention or control groups (Barreto et al., 2015).

Conversely, physical activity has consistently shown positive benefits on functional independence for those living with dementia in literature reviews and meta-analyses (Forbes et al., 2015;Rao et al., 2014;Groot et al., 2016;Lee et al., 2016;Pitkala et al., 2013a;Park and Cohen, 2018). Given the strong evidence that physical activity can slow down the loss of independence, and as adherence to physical activity interventions can predict improvements in function (Rao et al., 2014), researchers have already started exploring strategies to better engage people living with dementia in physical activity (Trahan et al., 2014;van Alphen et al., 2016a).

2.3.3 Use of health services

Physical activity has also been explored as a possible option to manage the health economic burden of dementia (Alzheimer's Disease International, 2013b;Olazaran et al., 2010). As a non-pharmacological intervention, physical activity tends to incur lower costs when compared to medication. Thus, it has been considered an attractive option for health systems, particularly in developing countries (Olazaran et al., 2010) where low resources and an increased incidence of dementia are combined (Alzheimer's Disease International, 2015). Moreover, the possible effect sizes of physical activity on cognition, for instance, have been comparable to those of medication (Groot et al., 2016) . Therefore, physical activity and other non-pharmacological interventions have been suggested to be a possible low cost “add-on intervention” to the use alongside pharmacology (Groot et al., 2016).

Non-pharmacological interventions for people with Alzheimer’s disease have been considered as a possible intervention to delay institutionalisation (Olazaran et al., 2010) and therefore, reduce costs to health services. However, lack of sufficient duration of interventions in physical activity studies, has limited any conclusions regarding this outcome (Olazaran et al., 2010). A few published randomised controlled trials have reported on the cost-effectiveness of physical activity interventions in the community (Pitkala et al., 2013b), but data on possible savings from implementing physical activity in the care of people with dementia have not yet been established.

2.3.4 Carer outcomes

Dementia is characterised by a gradual increase in functional dependence (OECD, 2015). Informal carers of people living with dementia are known to experience higher levels of burden than carers of people living with other health conditions (Karg et al., 2018). Physical activity interventions delivered to "carers only" are beyond the scope of this thesis, however, as described above, physical activity can improve functional independence of people living with dementia, which may indirectly reduce burden of care. Recent literature reviews have aimed to show the impact that physical activity for people living with dementia may have on carers (Zeng et al., 2016;Forbes et al., 2015). However, their conclusions remain limited by the small number of trials measuring carer outcomes and by the diversity of measures used (Zeng et al., 2016;Forbes et al., 2015). For instance, a literature review by Zeng et al. (2016) pooled data from four randomised controlled trials measuring carer burden and found a statistically significant mean difference of -2.33 in the Neuro-psychiatric inventory caregiver score, with a 95% confidence interval ranging from -3.65 to -1.01 (p=0.0005). However, in a Cochrane review, two trials were identified using two different tools, and only one provided data to be included in the analysis (with a positive effect on carer burden, using the Zarit Burden Interview scale, mean difference score of -15.30 with 95% confidence interval of -24.73 to -5.87, p=0.001) leading the authors to rate this as low quality evidence for the effectiveness of physical activity for people living with dementia on burden of care (Forbes et al., 2015). Forbes et al. (2015) also attempted to conduct a meta-analysis on the outcomes "caregiver quality of life" and "caregiver mortality" but found no studies that met the inclusion criteria to report on these outcomes. Thus, the limited amount of data available appears to report possible improvements in carer burden, but further research is needed to strengthen these findings.

2.3.5 Adverse events

Physical activity is not always free of adverse events. Robust knowledge of the possible negative outcomes (or side effects) of physical activity would enable researchers, clinicians, as well as patients and their families to accurately judge the overall benefit of physical activity. Recent literature reviews report that only few of their included empirical studies report adverse events (Forbes et al., 2015;Heyn et al., 2004), and when adverse events are reported, they are either not linked to the intervention, or considered not serious, such as dissatisfaction of the patient's relatives with the intervention (Rolland et al., 2007) or joint pain (Steinberg et al., 2009). This may indicate that physical activity is generally a very safe intervention for people living with dementia, or that possible adverse effects may be under-reported or not always linked to the intervention. A comprehensive list of the currently reported adverse effects in clinical trials of physical activity for people living with dementia is identified in Chapter 4, Paper II.

It is also possible that side effects of physical activity may vary, not only with the type of interventions, but also with the type of dementia. A recent randomized controlled trial, for instance, noted an increase in the number of falls during the intervention period in the exercise group compared to control, but only in the subgroup of participants living with Alzheimer's disease, and not in the participants with other types of dementia (Toots et al., 2018). Therefore, more research is warranted to clarify the dimension of the possible adverse effects of physical activity for people with dementia as well as the population groups where adverse effects are most likely.

2.3.6 Current recommendations

The previous sections have highlighted that the effectiveness of physical activity for people living with dementia is still equivocal to the large majority of its suggested clinical outcomes. Despite this uncertainty, recently published literature reviews appear to have shifted their research question from "Does physical activity work?" (Heyn et al., 2004;Forbes et al., 2008) to "Confirming that it works, and finding the most effective exercise protocol" (Learner and Williams, 2016;Lee et al., 2016). A group of experts in the field of physical activity in dementia care, has recently argued that the lack of current guidelines is a missed opportunity, which is holding back the implementation of physical activity interventions with potential benefits for people living with dementia. After reviewing the available literature, a recommendation statement has been published indicating that, by being active, people with Alzheimer's disease can benefit from improved functional independence, balance and cognition (Ginis et al., 2017). Similarly to other international statements (Ardern, 2012), this recommendation statement only focuses on Alzheimer's disease. Yet, there are national and international policies supporting physical activity, not only in Alzheimer's disease, but expand across dementia types, stages of disease progression and intervention settings (National Institute for Health and Care Excellence, 2006;Alzheimer's Society, 2015;Department of Health, 2009;Dyer et al., 2016). It is possible, however, that the effects of physical activity may vary from types of dementia, stage of disease progression and intervention setting. Examples of how the effects of physical activity may differ in across types of dementia, stages of disease progression and intervention types and settings are presented in section 2.2.7.

Across all these characteristics (types of dementia, stages of disease, types of physical activity and intervention setting), the reported benefits of keeping active presented in policy documents and in the grey literature (Alzheimer's Society, 2015;Alzheimer's Society, 2013a) are numerous: improved functional independence (Ardern, 2012;Department of Health, 2009;Dyer et al., 2016;National Institute for Health and Care Excellence, 2006), cognition (Physical Activity Guidelines Advisory Committee, 2008), mood (Ardern, 2012), behaviour (Department of Health, 2009), confidence (Department of Health, 2009), quality of life (Department of Health, 2009), reductions in depression (Ardern, 2012;Department of Health, 2009), comorbidities, falls (Miskovski, 2014), mortality (Department of Health, 2009) and opportunities for meaningful activity (Department of Health, 2009), healthy routine and social interaction, relaxation and enjoyment (Miskovski, 2014;Department of Health, 2009). However, as illustrated in the previous sections, little evidence can be found in the available literature to support most of the claims made in these documents. These health promotion messages are aimed at enhancing the overall health of the person living with dementia, but have been made public ahead of clear knowledge of the benefits of physical activity, and ahead of the development of robust guidelines for the implementation of physical activity in this population. Providing guidance through policy documents aimed at care providers, people living with dementia and their families, without research evidence to support it, may be detrimental. It may lead to inappropriate provision of care, and incorrect information being given to patients and families with regards to the outcomes they should expect from being physically active, potentially increasing burden of care with unknown real benefit to those living with dementia.

The next section will therefore look at possible reasons for the absence of evidence-based guidance on physical activity interventions for people living with dementia.

2.3.7 Limitations to the development of guidelines on physical activity for people with dementia.

The previous sections highlighted that, despite recent increase in research investigating the effectiveness of physical activity in dementia, few conclusive answers have emerged. It was noted that international agencies and expert panels in this field are promoting physical activity for the care of those with dementia, even though robust guidance on the recommended dose and types of physical activity have not yet been published. This section will consider the possible causes for the delay in the development of these guidelines. It is likely that absence of evidence-based guidance on physical activity for people living with dementia is multi-factorial. Literature reviews and meta-analyses are vital to the development of guidelines. As noted in the previous sections, many such literature reviews exist but have highlighted important methodological limitations in relation to the included studies. These limitations include small sample sizes or poor methodological quality (Olazaran et al., 2010); selection of measurement tools with inadequate validity and reliability (Heyn et al., 2004), interventions with insufficient duration or follow up (Park and Cohen, 2018;Groot et al., 2016), high dropout rates and/ or lack of intention to treat analyses (Learner and Williams, 2016) and, absence of clear randomisation and blinding methods (Learner and Williams, 2016). All of these methodological constraints are valid contributors to the lack of evidence-based guidelines in this field. Arguably, all these factors can be improved by conducting well designed randomised controlled trials. The argument proposed in this thesis is that even if large, well designed randomised controlled trials are implemented in the future, robust conclusions may still be limited by the heterogeneity in this field of research. People living with dementia are a heterogeneous population (different stages of disease progression, different types of dementia, different levels of comorbidity etc.). Physical activity is also a broad term including a wide range of possible interventions (different types of activity, different intensities, frequencies, durations and settings) and numerous outcomes and measurement tools can be used in this field of research. Aspects related to heterogeneity of population, interventions as well as outcomes and measurement tools are presented in greater detail in the following sections, and explored further in the systematic literature review presented in Chapter 4, Paper II.

2.3.7.1 Heterogeneity of the population

People living with dementia are a heterogeneous group. Multiple dementia subtypes have different symptom profiles and patterns of progression (Alzheimer's Association, 2016), which means that those with different types of dementia may have a different physiological response to physical activity. This is corroborated by the example on adverse effects presented earlier in this chapter (Section 2.2.5 - Adverse events), where people living with Alzheimer's disease had more falls than the control group, but not people living with other types of dementia (Toots et al., 2018). Even within the same dementia type, a paucity of studies including patients living with moderate or severe stages of dementia has been highlighted (Park and Cohen, 2018). This was also found in the systematic literature review in Chapter 4 (Paper II), which identified 72 primary studies including people living with mild to moderate dementia, but only five including people with severe dementia. Therefore, despite the growing evidence on physical activity in early stages of dementia, little is known about the effects of physical activity on those living in the later stages of the condition (Gonçalves et al., 2018a).

Dementia is also more prevalent in older age, which means that “dementia rarely travels alone” and people with dementia tend to live with other health conditions (All-Party Parliamentary Group on Dementia, 2016). The presence of comorbidities and disabilities may alter the effectiveness of physical activity interventions (Ohman et al., 2014) and may lead to disparity in the effects of physical activity, even when comparing similar interventions. One of many possible examples is a study describing a walking intervention which included people living with different types of dementia living in a long-term care facility, but excluded participants living with dementia who needed assistance to mobilise (Harris and Johnson, 2017). This hinders comparisons with another walking intervention in a nursing home, which only included people living with Alzheimer's disease, dependent for at least two basic activities of daily living listed in the Barthel Index (Venturelli et al., 2011). These are examples of very similar interventions, in the same setting, both including people living with dementia, but clearly describing very different populations (one including independently mobile people with a variety of dementia types; and one including people with Alzheimer's disease and more functionally dependent). Both studies also included small sample sizes which precludes any type of sub-group analysis (for dementia type or level of independence). Therefore it remains unclear, whether or not walking is effective for people with Alzheimer's disease versus people with other types of dementia; or whether physical activity can effective for participants more versus less functionally dependent.

2.3.7.2 Heterogeneity of interventions

The concept of physical activity is broad, including any activity involving activation of skeletal muscles (i.e., any exercise, activities of daily living, walking, dancing, gardening etc.). Therefore, physical activity interventions tested in people with dementia have varied greatly with regards to type, intensity, duration and frequency, which has limited the comparison between trials in literature reviews (Ohman et al., 2014;Rao et al., 2014;Pitkala et al., 2013a;Park and Cohen, 2018). All literature reviews on the topic of physical activity (including the literature review in Chapter 4, Paper II) report a large range of activity types. For instance, a literature review focussing on physical activity interventions to improve cognition in people living with dementia noted at least eight activity types: tai chi, dance, walking, strengthening, aerobic, flexibility, physiotherapy and occupational therapy (Ohman et al., 2014). The duration of interventions can be equally varied. In another literature review, studies were included lasting between two weeks to more than two years (Heyn et al., 2004) and frequencies ranging between one to seven days per week (Lee et al., 2016). A sub-group analysis identified that both high and low frequency interventions led to improvements in cognition for people living with dementia but only if the intervention included a component of aerobic activity (Groot et al., 2016). This highlights the complexity of determining the ideal intervention dose to maximise benefits to people living with dementia. Two other factors can add to this complexity: intensity and adherence. However, very few empirical studies report data on these parameters (Groot et al., 2016;Park and Cohen, 2018), and therefore, it remains unclear what intensity of activity should be targeted and what are the expected levels of adherence.

The heterogeneity of interventions with regards to activity type, duration and frequency may be a natural result of the current search for an ideal dose of physical activity for people with dementia. Furthermore, the very nature of dementia implies that physical activity interventions need to be individually tailored to the participant. On the one hand this may promote adherence to the intervention – a possibility that is yet to be confirmed as data on adherence is so scarce. On the other hand, heterogeneous types and frequencies of activity may also represent an obstacle for the standardisation of physical activity protocols (Pitkala et al., 2013a).

2.3.7.3 Heterogeneity of outcomes and measurement tools

Multiple literature reviews of physical activity interventions for people with dementia have reported limitations to their analysis due to a heterogeneous selection of outcomes and use of measurement tools. Many identified this as a reason for caution in the interpretation of any possible positive results (Heyn et al., 2004;Ohman et al., 2014;Forbes et al., 2015). For instance, two literature reviews measuring the effectiveness of physical activity on cognition have found a total of seven (Lee et al., 2016) and 11 (Ohman et al., 2014) different measurement tools. The findings from the literature review presented in Chapter 4, Paper II, highlight this problem by identifying a total of 133 different outcomes in 130 studies reporting on physical activity for people living with dementia, measured by 267 different measurement tools (Gonçalves et al., 2018a).

To add to these concerns regarding the selection and reporting of outcomes and measurement tools, it is also important to note a variety of terminologies used to report on the similar outcomes, or an overlap between outcomes, which impairs the ability of literature reviews to find all studies measuring the same outcomes, hinders comparisons between studies and limits meta-analyses. A good example of this, is the Cochrane review, which aimed to assess non-pharmacological interventions measuring “wandering” as an outcome for people living with dementia at home, but found no randomised controlled trials measuring “wandering” as an outcome, and could make no conclusions on this topic (Hermans et al., 2007). The authors noted that “wandering” is often reported among other aspects of “behavioural and psychological symptoms of dementia” (Hermans et al., 2007) and this may explain the lack of studies reporting on “wandering” as an outcome. Therefore, heterogeneous reporting of outcomes and selection of measurement tools has been considered a research waste (Clarke and Williamson, 2015) and recent literature reviews advocate better standardisation of study designs and more accurate reporting of methods in future research (Groot et al., 2016;Lautenschlager et al., 2012).

The problem of heterogeneous reporting of outcomes and measurement tools is also known in clinical practice, and has limited the ability of services to compare their performance and learn from each other. This has been addressed by the International Consortium for Health Outcomes Measurement (ICHOM), which provides a standard set of outcomes to be measured in clinical practice for dementia care (International Consortium for Health Outcomes Measurment, 2017). However, the standard set recommended by ICHOM is not specific to physical activity interventions which means that it may include outcomes that are not relevant to physical activity interventions (this is discussed in greater detail in Chapter 6, section 6.2).

This section highlighted that: i) an ideal dose of physical activity is yet to be established; ii) different types of dementia may respond differently to physical activity; and iii) physical activity interventions may need to be adapted to different settings and different stages of disease progression. Thus, measuring the same outcomes in all interventions will allow a clear understanding of what type and dose of physical activity works, in each setting, in each type of dementia and each stage of disease progression, by enabling conclusive meta-analyses in this field of research. Further, if professionals in clinical practice measure the same outcomes, with the same tools, they will be able to benchmark their intervention outcomes against those described by other health care providers. With standardised sets of outcomes, professionals will also be able to audit their service delivery against research findings.

These agreed minimum sets of outcomes are called “Core Outcome Sets”. The present thesis aims to reach consensus on a Core Outcome Set to be applicable in physical activity interventions, across different types of dementia, stages of disease progression and intervention settings, in research and in practice. The next section will explain in greater detail what Core Outcome Sets are and how they can be fundamental in minimising heterogeneity in reported outcomes and use of measurement tools.

2.4 Core outcome Sets: definition and methodological considerations

A Core Outcome Set is an agreed minimum set of outcomes and measurement tools to be used as standard, across clinical trials, involving people with a particular health condition and/or type of intervention (Williamson et al., 2012;Clarke and Williamson, 2015;Kirkham et al., 2016). Approximately 12% of all currently developed Core Outcome Sets are designed to be applicable in both research and in practice (Gargon et al., 2018). Researchers, and clinicians, are not restricted to using only those outcomes in the Core Outcome Set but can continue to explore and report further outcomes, in addition to the minimum set defined by the Core Outcome Set (Clarke and

Williamson, 2015). The concept of Core Outcome Sets was first introduced by the World Health Organisation in 1979 with the view to standardising results of cancer interventions (World Health Organization, 1979), and further developed in the field of Rheumatology by the Outcome Measures in Rheumatology initiative (OMERACT) in 1992 (Boers et al., 2014b). More recently, the Core Outcome Measures in Effectiveness Trials initiative (COMET) has expanded Core Outcome Set methodology and uptake across clinical areas and the number of Core Outcome Sets being developed has increased in recent years (Gargon et al., 2018).

If well developed, Core Outcome Sets represent the outcomes of relevance to all key stakeholders and consequently, can guide the design of relevant interventions (Clarke and Williamson, 2015) as well as guide literature reviews to focus on outcomes that matter to all stakeholder groups (Bastian et al., 2010). The implementation of a Core Outcome Set allows more effective comparisons between trials, aiding meta-analyses and thus, fast tracking the development of clinical guidelines (Tugwell et al., 2007); minimises publication bias and ultimately reduces research waste (Williamson et al., 2017;Clarke and Williamson, 2015). Based on these methodological and ethical benefits, the use of Core Outcome Sets is recommended by the National Institute of Health Research (Clarke and Williamson, 2015) and considered beneficial by the majority of Cochrane coordinating editors (Kirkham et al., 2013).

Although guidance is available from the COMET and OMERACT initiatives on multiple methodological approaches used to develop Core Outcome Sets (Williamson et al., 2017;Boers et al., 2014b), a gold standard is yet to be established, and the methodological approaches to Core Outcome Set development are still evolving. While a gold standard is not yet available, the COMET initiative has recently published minimum standards for the development (COS-STAD – (Kirkham et al., 2017)) and reporting (COS-STAR - (Kirkham et al., 2016)) of Core Outcome Sets. These documents report on minimum standards with regards to determining the scope, stakeholder involvement and consensus process. A variety of methodologies can and have been used by Core Outcome Set developers (Gargon et al., 2018). The development of a Core Outcome Set often starts by ascertaining “what to measure”. This process begins by listing all possible outcomes of a given intervention to the population of interest; and then reaching consensus on outcomes that should be included in the final Core Outcome Set. The use of mixed methods has been reported as an innovation (Keeley et al., 2016) in the process of defining “what to measure”.

Mixed methods can be defined as an approach or methodology that employs both rigorous quantitative and qualitative research. This allows an assessment of both the magnitude and frequency of constructs (quantitative, mainly deductive) and a deeper understanding of those constructs (qualitative, mainly inductive) (Creswell et al., 2011). In mixed methods, data from both qualitative and quantitative paradigms are integrated in three possible ways: 1) merging data, where qualitative and quantitative results are presented together and have the same weight in the analysis (e.g., quantitative numeric data, explained by quotes from qualitative interviews); 2) embedding data, where a primary or larger data set includes a secondary smaller set of data embedded in it (e.g., a large randomized controlled trial includes a small qualitative exploration from the participants' experiences in taking part in the trial) and 3) connecting data, where data is collected in sequence and the findings of one type of data collection will inform the next stage of data collection (e.g., qualitative interviews to inform the development of a measurement instrument, which will subsequently be piloted using quantitative methods) (Creswell et al., 2011). In the context of a Core Outcome Set, the latter approach (connecting data) has recently been described. Qualitative studies have been found potentially useful to identify important outcomes, from the perspectives of different stakeholders, which may have not been reported in previous literature (Keeley et al., 2016). Outcomes identified using qualitative methods are then added to those described in the literature - a process often referred to as "creating a long list of outcomes". This "long list" of outcomes is then used in the consensus process (quantitative) to determine which outcomes gain consensus to be part of the final Core Outcome Set.

The use of qualitative methods is still an innovation in the development of Core Outcome Sets and yet to be considered standard practice. Some Core Outcome Set developers report that their qualitative study did not add any new outcomes to their "long list" generated from the literature (Harman et al., 2015), raising questions regarding its utility. Core Outcome Set developers are aware that qualitative studies are resource intensive and have considered carefully whether they are necessary. One of the contexts in which mixed methods may be particularly valuable is when views of patients are not well addressed in previous literature (Keeley et al., 2016). This is definitely the case with regards to physical activity for people living with dementia, where qualitative work is scarce and often presented as description of patients' experiences of taking part in a particular clinical trial and not their in-depth views of what are valued outcomes of physical activity (see the background section in Chapter 4, Paper III for further detail). It is argued, therefore, that in the context of physical activity for people living with dementia, a qualitative study would most certainly add value by highlighting outcomes valued by patients and by gaining in-depth understanding of why certain outcomes are important. The knowledge of what outcomes are important and why may also be key to inform the design of physical activity interventions and promote adherence to physical activity in this population group.

Including patients in the development of Core Outcome Sets (through qualitative and/or quantitative methods) has been undoubtedly recognised as vital, as patients may value different outcomes to those identified by clinicians and researchers (Williamson et al., 2017). More recently, the involvement of patients has gained weight beyond their role as research participants, and the involvement of patients and the public in planning, design and delivery of research, commonly known as "patient and public involvement", is now welcomed in multiple areas of heath research (Staley, 2009). "Patient and public involvement" is defined by the National Institute of Health Research as "research being carried out with or by members of the public, rather than to, about and for them". It aims to make research more relevant to the needs of patients (National Institute for Health Research, 2014) and it has been reported to make research more effective by improving recruitment, assisting with analysis of qualitative data and ensuring the use of relevant outcome measures in clinical trials (Staley, 2009). All these reported benefits of patient and public involvement are equally relevant to the development of Core Outcome Sets. As the methodological principles of Core Outcome Set development evolve, so do the strategies to involve patients as collaborators. A recent publication from the COMET initiative reflects on specific challenges of including patients in the design and development of Core Outcome Sets (Young and Bagley, 2016). Some of these challenges include ensuring that the methods used are suitable for patients to take part, and that patients can contribute in a meaningful way to the selection of outcomes included in the final Core Outcome Set (Young and Bagley, 2016). The authors also highlight the use of patient and public involvement in the design of Delphi surveys (a commonly used consensus method by Core Outcome Set developers) as some patients may find a long list of outcomes intimidating; whereas others tend to value the fact that they can participate anonymously (Young and Bagley, 2016). These two key aspects of including patients in Delphi surveys were also important findings of the patient and public activities conducted as part of the Core Outcome Set presented in this thesis. Chapter 5, Paper IV presents in greater detail how patient and public involvement activities informed a modified Delphi survey that enabled the participation of people living with dementia in the consensus process of this Core Outcome Set.

2.4.1 Lessons learnt from previous Core Outcome Sets including people living with cognitive impairment

The fact that the present Core Outcome Set is to be applicable to people living with dementia poses some challenges, related to the inclusion of people living with cognitive impairment in the selection and prioritisation of outcomes, to be included in the final Core Outcome Set. As described above, patient and public involvement is key to the development of any Core Outcome Set, and arguably particularly important when developing a Core Outcome Set for people living with a cognitive impairment. Additionally to patient and public involvement, it is relevant to review other Core Outcome Sets in the field of dementia, and in other populations with a cognitive impairment. Previous work in these areas may allow an understanding of possible strategies to successfully include people living with a cognitive impairment in the selection of core outcomes.

A total of five other Core Outcome Sets have been previously reported in the field of dementia. The first Core Outcome Sets developed in this field were published in the early 2000s (Katona et al., 2007;Moniz-Cook et al., 2008) and did not actually include people living with dementia in their development. This highlights how our understanding of the importance of patient involvement has progressed in recent years. More recent Core Outcome Sets have included views of people living with dementia through face to face interactions, either through workshops or focus groups (JPND, 2015;International Consortium for Health Outcomes Measurment, 2017;Webster et al., 2017). However, little detail is offered in these publications about any adaptations made to accommodate to the needs of people living with dementia during data collection. This may suggest that face to face interaction may be an important factor when involving people living with dementia in Core Outcome Set development, but practical questions on how to present outcome related information to people living with dementia and how to gain their views on particular outcomes still remain.

In order to gain further insight into the process of reaching consensus on core outcomes, including people living with a cognitive impairment, two other key publications can be drawn upon: the work by Morbey et al. (2019) and by Morris et al. (2015). The first is a methodological paper, which describes the adaptations made to a traditional Delphi survey, in the context of a Core Outcome Set to evaluate dementia interventions in the community (Morbey et al., 2019). The second is the final paper of a Core Outcome Set for children with neurodisability, which reports the use of Q-sorting strategy to gain the views of children in its consensus phase (Morris et al., 2015). Three design modification features have been suggested to a traditional Delphi survey, in order to include people living with dementia: (1) the use of three point rating scale for each outcome, rather than the traditional nine point Likert scale, in which each outcome is presented individually on an A4 page; (2) the use of accessible language to describe each of the outcomes being presented; and (3) data collection on a one-to-one face to face interaction, where the person living with dementia could express their choices verbally (Morbey et al., 2019). In line with previous Core Outcome Sets, the use of face to face interaction appears to be valued (Morbey et al., 2019;JPND, 2015;International Consortium for Health Outcomes Measurment, 2017;Webster et al., 2017). The use of simplified rating scales, use of accessible language and the consideration of one outcome at a time are also important lessons learnt from the study by Morbey et al. (2019). Instead of a Delphi study, Morris et al. (2015) used a Q-sorting strategy. In this method, young children, their parents and health professionals were presented with 33 outcomes. Each of the outcomes were presented in individual laminated cards with a pictorial and written description. All stakeholders worked together in groups to organise the 33 cards into a pyramid (forced choice frequency distribution grid), from “less” to “more important”. Whilst this was a very successful strategy to include children with cognitive impairment in the prioritisation of outcomes, the authors recognised as challenging the large number of outcomes to be considered and ranked at one time. They therefore recommended preliminary activities to reduce the number of outcomes being considered in the final sorting tasks. The lessons learnt from this study included, once more, that face to face interaction was used successfully; the use of cards with pictures was well understood by participants with a neurodisability, but care must be taken to avoid a large number of cards/outcomes being considered at one time. The large number of cards (outcomes) being considered at one time in the Q-sorting strategy might, in fact, limit its full applicability when including people living with dementia. Patient and public involvement activities, carried out prior to the consensus stage of the present Core Outcome Set, clearly indicated that considering one outcome at a time would be a key feature to include people living with dementia in the consensus process. This is also in line with the study by Morbey et al. (2019), conducted with people living with dementia, which suggested that it was helpful to present one outcome at a time when involving people living with dementia.

In summary, previous research in the field of Core Outcome Set development including people living with cognitive impairment suggests that: (i) the inclusion of people living with dementia in Core Outcome Sets has not always been a reality and adaptations may be necessary to enable their participation; (ii) face to face interactions have been valued, as well as the use of simple rating scales and accessible language; (iii) a Q-sorting strategy offers some of these features and also suggests the use of cards with pictorial representations, but is limited by the fact that a large number of outcomes must be considered at any one time. Therefore, a novel approach may be needed to incorporate the lessons learnt from previous research described above, in order to maximise participation of people living with dementia in the consensus process. This new approach will be described as a “card sorting strategy”. It was developed and implemented in this thesis (Chapter 5, paper IV), and it was shaped by previous literature, and by input from patients and the public.

The present chapter provided background information regarding the development of a Core Outcome Set to evaluate physical activity interventions for people living with dementia, across different types of dementia, intervention settings and stages of disease progression, to be applicable in research and in clinical practice. The present research followed a mixed methods approach and utilised innovative strategies to include people living with dementia in the development of this Core Outcome Set. It also benefited from patient and public involvement initiatives, including collaborative work with a former carer of a person living with dementia throughout the research design, recruitment and data analysis; and consultation with a wider group of patients and carers, for some of the methodological decisions involved in its development. The following chapters present a series of publications that led to the development of this Core Outcome Set.

Chapter 3 Methods

3.1 Paper I - Development of a core outcome set to evaluate physical activity interventions for people living with dementia: study protocol

Gonçalves, A.C., Marques, A; Demain, S and Samuel, D. 2018. International Journal of Therapy and Rehabilitation, 25(7), 346-52.

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ABSTRACT

Background: Evidence on the benefits of physical activity for people with dementia remains disparate, mainly due to the selection of heterogeneous outcomes and measurement tools. This delays clear and specific recommendations for research and clinical practice. The development of Core Outcome Sets can contribute to overcoming this heterogeneity.

Content: This is a study protocol for the development of a Core Outcome Set applicable to physical activity interventions, in any setting, for people with dementia, across stages of disease progression. This is a mixed methods study divided in four phases: i) literature review to identify outcomes used in previous literature; ii) a qualitative study to explore valued outcomes in the perspective of different stakeholders; iii) a Delphi survey and consensus meeting to reach a minimum set of outcomes and iv) a literature review to link the agreed core outcomes to the most appropriate measurement tools.

Conclusions: A Core Outcome Set in this field has the potential to allow fast-tracking recommendations to research and clinical practice. However, dissemination activities are required to encourage researchers to implement the Core Outcome Set.

Key words: Core Outcome Set; Dementia; Physical activity; Methodology

BACKGROUND

It is estimated that currently 47 million people live with dementia worldwide (World Health Organization, 2015b), a number that may reach to 76 million by 2030 (Alzheimer's Disease International, 2013a). Enormous costs are being predicted, informal care being a significant component of these (Wimo et al., 2007). Higher levels of functional dependence are linked to an increased carer burden and consequently an increased risk of institutionalisation (Stephan et al., 2014). Evidence suggests that physical activity interventions may have a positive impact on the levels of independence of people living with dementia (Forbes et al., 2015), potentially reducing care needs. Physical activity is also recommended for the general older population as it is known to have a positive impact on levels of mobility, risk of depression and mental wellbeing (National Institute for Health and Care Excellence, 2008). In line with these potential benefits, the National Institute for Health and Care Excellence (2006) guides health professionals to recommend appropriate physical activity for people with dementia. Yet, caution is needed. Despite the large body of research, systematic reviews report limitations in their results due to the use of heterogeneous outcomes and measurement tools (Forbes et al., 2015;Rao et al., 2014). This heterogeneity hinders the effective synthesis of evidence (Macefield et al., 2014) and delays the development of clear recommendations for research and clinical practice.

The use of Core Outcome Sets has emerged as a solution for the heterogeneity of reported outcomes in clinical trials (Williamson et al., 2012;Idzerda et al., 2014). Core Outcome Sets are an agreed minimum set of outcomes that are recommended to be measured and reported as a minimum standard across clinical trials of a particular health condition or trial population (Williamson et al., 2012). The adherence to Core Outcome Sets ensures that clinical trials measure meaningful outcomes for different stakeholders (Clarke and Williamson, 2015); reduces reporting bias; and allows a direct comparison between trials in meta-analysis, which will subsequently lead to clearer recommendations for clinical practice (Williamson et al., 2012;Waters et al., 2014). A Core Outcome Set to evaluate the effectiveness of physical activity interventions for people with dementia may also inform health professionals delivering these interventions. Health professionals can use this Core Outcome Set to select meaningful outcomes for patients and monitor the effects of their interventions against the results reported in the literature.

Study aims and overview

No “gold standard” methodology currently exists for Core Outcome Set development. The COS-STAR statement therefore recommends that Core Outcome Set protocols are made publically available to increase the transparency of the Core Outcome Set development and minimise any biases (Kirkham et al., 2016). The present protocol represents the proposed methodology to develop a Core Outcome Set to evaluate physical activity interventions for people with dementia.

Specific objectives for each of the four phases that form the development of this Core Outcome Set are to: phase I) comprehensively list the outcomes and measurement tools used in previous literature; phase II) explore what outcomes are meaningful for professionals delivering physical activity, people with dementia and their friends, relatives or informal carers, adding to the list of outcomes identified in the literature; phase III) reach consensus, across stakeholders, of what outcomes should be prioritised into the Core Outcome Set; and phase IV) link each of the agreed outcomes to the most appropriate measurement tool.

METHODS

Registration and Ethical approval

This project has been registered with the COMET initiative and its registration is available from:

http://www.comet-initiative.org/studies/details/708?result=true.

Informed consent will be obtained from all participants of each of the empirical phases of this study. This protocol has received ethical approval from the Ethics Committee of the Faculty of Health Sciences of the University of Southampton, United Kingdom. The design and implementation of this project was informed and supported by the involvement of patient representatives.

Scope

The present Core Outcome Set will be applicable to any physical activity intervention, as per the World Health Organisation definition: “Any body movement produced by skeletal muscles that requires energy expenditure”, for people with dementia, at any stage of the condition, in any setting. This excludes interventions for people diagnosed with a mild cognitive impairment or people with a cognitive impairment as a result of any other health conditions but dementia. It is anticipated that the final Core Outcome Set will be subdivided into “mild to moderate” and “severe stages”, as different outcomes might have more or less relevance in different stages of the disease. The final Core Outcome Set will be recommended for use in clinical trials. Additionally, guidance on the assessment of effectiveness of physical activity interventions for people with dementia in clinical practice will be drawn.

Stakeholders

The selection of participants for the development of this Core Outcome Set aims to reflect the variety of stakeholders involved in physical activity interventions for people with dementia. Two stakeholder groups will be included. A professional group including health and social care professionals, researchers and members of volunteering organisations; and a second group including people with dementia, their relatives, friends and informal carers.

Phase I: systematic literature review

One systematic mixed studies literature review will be conducted with the aim of comprehensively listing the outcomes and measurement tools used in previous literature.

Information sources and search strategy

The search strategy will begin with a key word search on Delphis, a single interface that allows a key word search in providers such as Medline, PsycINFO, Cinahl, Scopus and ScienceDirect. The search strategy below has been developed in collaboration with an experienced librarian in health sciences:

S1. “Physical activity” OR exercis*

S2. dement* OR Alzheimer

S3. S1 AND S2

S4. S3 AND source type: academic journals OR reviews OR thesis/dissertations (excluded books, magazines, news, conference materials, electronic resources and reports).

S5. S4 AND studies written in English, Portuguese or Spanish.

S6: studies published from the 1st of January 2005.

A subheading search will be performed using the database identified as the most important source of studies (based on the Delphis results) to ensure literature saturation.

Participants

Studies including people with dementia at any stage of disease progression will be included.

Types of studies and interventions

Experimental designs (with or without comparators), qualitative studies and study protocols investigating the impact of any physical activity intervention, will be included. No restrictions will be made regarding intervention setting.

Exclusion criteria

Studies will be excluded if they are not written in English, Portuguese or Spanish; or relate to physical activity interventions for relatives or carers only. All searches will be limited to studies published from January 2005 *to April 2015*[2]. Although this decision is recognised as a limitation, it is anticipated that any important outcomes not captured by this review will emerge during the interviews with different stakeholders (phase II).

Data extraction and analysis

The screening and eligibility of papers generated by the searches will be conducted by one author. A random sample of 10% of the studies will be independently screened by a second author to ensure accurate application of inclusion and exclusion criteria. Standardised data extraction will ensure the identification of all outcomes (positive or negative) and measurement tools reported by the included papers in their methods, results and discussion sections.

A content analysis methodology (Macefield et al., 2014) will be used to synthesise the diversity of the outcomes used in physical activity interventions for people with dementia. Verbatim outcomes, from qualitative, quantitative and mixed methods studies will be extracted and analysed using the same content analysis approach. Verbatim outcomes will be grouped in outcome domains (outcomes with different taxonomies but the same perceived meaning). The outcome domains will subsequently be organised into broader themes by the research team. An analysis of the outcome domains per stage of disease progression, study paradigm and identified by each stakeholder group, will be performed. With regards to the stakeholder groups, outcomes reported in clinical trials will be considered as outcomes selected by professionals, unless described otherwise in the papers. Outcomes reported by qualitative studies will be linked to the participants in these studies.

Risk of bias

The methodological quality of included papers will be assessed using the Mixed Methods Assessment tool – version 2011 (Pluye et al., 2009), a tool designed for the purpose of complex reviews, including studies from different paradigms. The quality of the included studies will be used purely to inform the readers of the quality of research in this field. It will not be used as an exclusion criterion and will not influence data analysis.

Phase II: qualitative Interviews

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[2] *Information added post publication to enhance content.*

This qualitative study will aim to complement the results from the literature review in the previous phase and allow a deeper understanding of what outcomes are valued to each stakeholder group. Phases I and II will be conducted in a sequence because the findings from the qualitative study will complement the results from literature review. However, it is possible that a temporal overlap may occur between data analysis for the literature review and data collection for this qualitative study.

Participants

The “professional” stakeholder group includes any health and social care professional or member of a volunteering organisation who has been involved in the design, implementation or support of physical activity for people with dementia, in any setting. To be included, “professionals” have to live or work in the United Kingdom and have sufficient English language skills.

People with dementia, at any stage of the disease progression, who have been involved in any type of physical activity since diagnosis, will be eligible regardless of age or accommodation setting. Capacity to consent to take part in research is required. Sufficient verbal communication skills in English language are also required to undertake the interview. Relatives, friends or informal carers of people with dementia, who have been in contact with the patient during their involvement in physical activity, and have sufficient English skills, will be interviewed either independently or in a joint interview with the patient.

Factors such as age, gender, accommodation setting, levels of physical activity and stage of disease progression will be used for purposive sampling (Coyne, 1997). Both stakeholder groups will be recruited from charities, community centres, privately run care and nursing homes, support and professional groups. The sample size will follow the principles of data saturation (Guest, 2006), to a maximum of 30 participants (8 to 10 professionals, 4 to 10 people with dementia and 4 to 10 relatives).

Interview format

A Semi-structured interview format will be followed. The interviews will be conducted through the use of open-ended questions which will not be influenced by the results of the literature review. People living with dementia will be interviewed face-to-face, to allow for ongoing capacity assessment throughout the interview. Telephone interviews will be a possibility for other participants. Topic guides will address the valued outcomes of physical activity for all stakeholders. It is anticipated that the concept of “outcomes” may be unfamiliar for many participants. Thus, this terminology will be replaced by “effects” or “results” of physical activity, for purposes of clarity during the interviews. In addition to outcomes, participants will be asked about barriers and facilitators for the application of a Core Outcome Set for this population, in research and clinical practice. These data will inform final recommendations for the applicability and dissemination of the Core Outcome Set.

People with dementia will be encouraged to have a relative or friend with them at all times, for their own comfort. The interview will be conducted in a familiar venue (i.e., their home) to reduce possibilities of distress caused by being in an unfamiliar location. The researcher conducting the interview will have experience in communicating with people with dementia.

Interview analysis

All interview data will be audio-recorded, transcribed verbatim. NVivo software (NVivo10 software, QSR International, Burlington, Massachusetts, United States) will be used to aid data management. A framework methodology will be followed (Ritchie and Spencer, 1994), coding the interview data against a framework of outcomes generated by the literature review in Phase I. This methodology also enables novel outcomes, emergent from the interviews, to be added to the initial framework.

At the end of Phase II, a comprehensive list of potential outcomes will be generated and used in the Delphi survey, described in Phase III.

Phase III: Delphi and consensus meeting – what to measure

The OMERACT initiative recommends that the number of outcomes in a Core Outcome Set is limited to a maximum of nine, in order to promote its applicability (Boers et al., 2014a). A Delphi survey will be used as a method to reach consensus regarding what outcomes should be prioritised for inclusion in the Core Outcome Set. A Delphi technique utilises several rounds where participants receive feedback from previous rounds and have opportunity to review their choices. The main advantages of this method are the anonymous participation of experts, minimising possible role pressures from fellow participants; and expenses and logistical challenges of face-to-face meetings (Boers et al., 2014a;Prinsen et al., 2014;Sinha et al., 2012), making it a commonly used consensus method in the development of Core Outcome Sets. A two round modified Delphi survey, including both stakeholder groups is planned for the development of this Core Outcome Set. Modifications to the Delphi survey, detailed below, were made to enable the participation of people with dementia in this phase of the study. Each item in the Delphi survey will consist of one outcome identified in the literature review and/or qualitative interviews (Phases I and II) and reviewed by patient representatives, to guarantee content clarity of the items.

Participants and sampling

Participants from both professional and lay stakeholder groups will be invited to participate in the Delphi survey. Equivalent inclusion/exclusion criteria and recruitment strategies will be used. People with dementia will require face-to-face contact; but *carers will be recruited from anywhere in the UK and professionals from anywhere in the world*[3]. The first page of the Delphi survey will list the inclusion criteria and all participants will be asked to confirm these criteria before completing the survey.

The optimum number of participants in a Delphi survey is yet to be established, however previous studies have reported sample ranging from 46 (Sinha et al., 2012) to 218 (Devane et al., 2007). MacLennan et al. (2015) suggested a sample size of up to 150 participants. Therefore, we aim to recruit between 80 (40 for each stakeholder group) and 150 participants. Participants from the qualitative interviews will also be invited for the Delphi study. Additionally, a snowball sampling strategy will be implemented for the online surveys, where participants will be asked to invite peers who may wish to participate (Kottner et al., 2016).

Methodological adaptation to enable the participation of People with Dementia

A card-sorting alternative, in a face-to-face interaction, will be offered to people with dementia, aiming to reduce the cognitive demand of the task. Participants will be shown a set of cards, each with a simple description of the outcome and pictorial representation. The participant will be asked to choose the cards that represent their valued outcomes of physical activity. Card sorting strategies are used with people with dementia as a form of assessment, for instance through the use of the Nelson's Modified card sorting test (Chao et al., 2013). This indicates that using cards to facilitate the selection of information, according to an established criteria, might be appropriate for this population. People with dementia and their carers, from local support groups will be asked to contribute to the development of the survey and pilot its first version before the beginning of the Delphi survey. Participants other than people with dementia will receive an on-line or paper survey, via post, according to their preference.

Round one

Based on what is already known regarding the heterogeneity of the literature on this topic, it is expected that the round one survey will consist of over 100 outcomes (survey items). This is a large amount of information, potentially too challenging for people with dementia, even when using the card sorting strategy described above. Therefore, in this first round, people with dementia will not be included, and the stakeholder group one will be represented only by their

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[3] *Correction made post publication to enhance clarity (previously read: "but all other participants can be recruited from any part of the globe").*

relatives, friends or informal carers. All participants of round one will be asked to choose responding to the survey designed for mild to moderate or severe stages of dementia (or both) according to their own experience or choice. To each of the surveys (mild to moderate and severe stages) each participant will be ask to choose (without rating) up to nine outcomes from the list.

Data analysis and definition of consensus

Descriptive statistics will be used to describe participants' characteristics and ascertain consensus as follows. At the end of the first round, outcomes will be excluded if: selected by 15% of the participants or less (Waters et al., 2014), and had not been identified by a person living with dementia in the interview stage. All other outcomes will be taken through round two. At the end of round two, any outcomes selected by 70% or more of the participants in both stakeholder groups or by 80% in one stakeholder group will be included in the Core Outcome Set (Waters et al., 2014;Boers et al., 2014a;Potter et al., 2015). It is anticipated that the Delphi survey will be divided into "mild to moderate" and "severe" stages and in that case, this definition of consensus will apply individually to each of these stages of the condition.

Round two

All participants of round one will be asked to review their answers based on the feedback from round one. The feedback will consist of the percentage of all participants; and percentage of participants from each stakeholder group, who selected each of the outcomes.

People with dementia will be included in this round, when the number of outcomes remaining on the survey are likely to be substantially lower. People with dementia will complete a face-to-face survey, using a card sorting approach as previously described, regarding their own stage of the disease only (mild to moderate or severe). The interaction between the researcher and the participant will be audio recorded.

At the end of this round, all outcomes that remained in the Delphi (selected by 16% or more of the participants) will be taken to a consensus meeting for validation and discussion of possible disagreements.

Consensus meeting – final decision on what to measure

A final consensus meeting aims to present and validate the agreed outcomes from the Delphi survey (as per definition of consensus), resolve any disagreements and to seek consensus for the outcomes in which an agreement has not yet been achieved (MacLennan et al., 2015). Results from the Delphi survey will be presented and discussed by a group consisting of at least one representative of each stakeholder group. An open group discussion methodology will be followed. Consensus will be defined as 90% of agreement to include one more outcome to the Core Outcome Set. If consensus cannot be achieved, a smaller Core Outcome Set, including only the fully agreed outcomes will be defined (Williamson et al., 2012). A separate meeting per stakeholder group may be arranged according to the preference of people with dementia and theirs carers, relatives or friends. This option will also be used if a marked disparity in opinions per stakeholder group, would have been noted in the Delphi results (Waters et al., 2014).

Phase IV: literature review – how to measure

This final literature review aims to link each of the outcomes agreed at the end of the previous phase to the most appropriate measurement tool. Practical guidance on how to select measurement tools for outcomes in a Core Outcome Set has recently been published a result of a collaboration between COMET and the Consensus-based Standards for the selection of health Measurement Instruments (COSMIN) initiatives (Prinsen et al., 2016). This guidance suggests four steps, which will be followed as described below.

Conceptual considerations

A clear definition of the concepts behind each of the outcomes will be decided upon by the research team.

Finding existing measurement tools

The selection of the measurement tools will consider the stage of disease progression (mild to moderate and severe stages separately) and the intervention settings. The process of finding existing measurement tools would have started in Phase I (literature review). Additional literature searches will be undertaken to update the literature review on phase I, and to identify the psychometric properties of each of the measurement tools identified. The search strategy will follow the guidance of Prinsen et al. (2016) and the advice from a librarian.

Quality assessment of the instrument tools

This step will follow the criteria indicated by COSMIN: internal consistency, reliability, measurement error, content validity, structural validity, hypothesis testing, cross cultural validity, criterion validity, responsiveness and interpretability (Mokkink et al., 2010) to assess the quality of the evidence available on the measurement properties of measurement tools linked to each of the outcomes, according to this population group and per intervention setting.

Generic recommendations

Only tools with high quality evidence for good content validity, good internal consistency and that are considered feasible (on the grounds of application time, availability and costs) will be recommended in the final Core Outcome Set. To encourage consistency in clinical trials, each outcome should be linked to one measurement tool only. If multiple tools fit quality and feasibility criteria, an expert panel will be arranged with the stakeholder groups to reach a consensus on which measurement tool will be recommended in the final Core Outcome Set (Coulter et al., 2016).

Measurement tools and their characteristics identified in the literature will be shown to the panel members. Lay terms and examples will be used to explain psychometric properties to non-scientific members of the panel. Patients, friend and relatives stakeholder group may be represented by friends or relatives only. However, if people with dementia are recruited to take part in this stage, quiet environments and shorter sessions will be arranged to accommodate their needs and facilitate their participation. Advice from patient representatives will also be sought in the planning of this expert panel. Each panel member would then rate each measurement tool individually. The results of the voting will be revealed and followed by a group discussion, which would then lead to another round of voting, until a consensus of 70% of more votes in favour of a particular measurement tool can be reached.

CONCLUSION

Adherence to a Core Outcome Set applicable to all physical activity interventions for people with dementia in any setting and able to cover all stages of the disease progression will increase the consistency of clinical trials in this field; allow a direct comparison between interventions and consequently lead to more clear guidance for research and clinical practice.

Of the methodology presented above, the Delphi study is the phase requiring particular attention, and careful adaptations to enable people with dementia to take part. A card sorting strategy and the absence of a *rating*[4] system, typically used in Delphi surveys, is suggested. These adaptations will be trialled by carers before implementation. The possible expert panel at the end of phase IV, to vote on one of multiple possible tools to measure one outcome, is also an adaptation to previously described methods, with the view to include people with dementia. The use of consensus and prioritisation methods involving people with dementia and other cognitive impairments requires further methodological research.

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[4] *Correction made post publication to enhance clarity (previously read: ranking)*

Despite the scope for important benefits of the use of a Core Outcome Set in this field, these will be dependent on the adherence of the trialists to the outcomes and measurement tools set by the Core Outcome Set. Therefore dissemination work should not be overlooked once the final Core Outcome Set is achieved and published.

CONFLICTS OF INTEREST

The authors have no conflicts of interest to declare.

3.2 Changes to the protocol since publication

3.2.1 Changes to the qualitative study

The method of analysing qualitative data was changed from the planned framework analysis to thematic analysis. This decision was made after the interviews had been conducted, and the literature review had been completed. At this point, it was found that the framework of outcomes generated by the literature review was extensive (133 different outcome domains - see Chapter 4 for further details). This would mean starting the analysis with an already complex framework. In addition, data collected during the interviews were surprisingly rich and had the potential to be analysed in-depth. It was therefore important to go beyond a “validation exercise” and verify which outcomes had been reported in the qualitative study that had or had not yet been explored in the literature. Instead, the aim of this qualitative study became also focused on “why” and “how” certain outcomes were important to the participants. Thematic analysis would allow an in-depth analysis of the outcomes participants had reported (Braun and Clarke, 2013), rather than spread the data “thin” through a wide framework of outcomes.

The maximum sample size of the qualitative study remained unchanged (n=30), however the proportion of carers and patients was altered, from 10 to 15 for carers and from 10 to five for patients. Two factors contributed to this decision. Firstly, the 10 carers initially recruited were all female. The views of male carers on the impact of physical activity as a health intervention, within their caring role, might differ from female carers (Sharma et al., 2016). Therefore, it was important to recruit up to five more male carers. Lastly, recruiting people living with dementia was found particularly challenging, possibly due to the inclusion criteria used: mental capacity to consent to research and ability to communicate verbally to take part in an interview. Thus, all patients included were in the earlier stages of the disease. Therefore, it was felt that the views of people with moderate to severe stages of dementia would need to be represented by their carers, hence increasing the sample size of carers to 15 and limiting the sample of patients to five. A higher proportion of carers and small proportion of people living with dementia made the recruitment strategy more feasible within the available time frame. These changes were submitted and approved by the ethics committee of the Faculty of Health Sciences (now named: Faculty of Environmental and Life Sciences).

3.2.2 Changes to Delphi survey

At the end of the qualitative study (Chapter 4, Paper III), it was ascertained that there were no differences between valued outcomes in different stages of dementia. Whilst the tools to measure those outcomes may need to be different in different stages of the condition, the valued outcomes were common across stages. Thus, the protocol for the Delphi study was changed and a single survey was used with outcomes to be considered across all stages of dementia (see Chapter 5, Paper IV for further details on the Delphi study).

3.2.3 Selection of measurement tools

The last study described in this protocol was a literature review of measurement tools to be linked to the core of outcomes agreed upon in the consensus study (Delphi survey). It is, however, important to note that the research process of linking the agreed core of outcomes to the most appropriate measurement tools goes beyond the development of a single literature review. Following the guidance by Prinsen et al. (2016), the first step in this process is a consensus exercise to determine the exact constructs within each outcome that should be measured; the second step is indeed a literature review to find all available tools to measure the agreed constructs; the third step is an analysis of the quality of the identified measurement tools (through their psychometric properties) and an analysis of the quality of studies conducted to validate the measurement tools; lastly, a further consensus study may be necessary to ensure that only one tool is recommended per outcome, and that all recommended tools have high quality evidence for at least good content validity and internal consistency. Thus, it was understood early in the development of this Core Outcome Set that time restrictions would prevent the link between outcomes and measurement tools to be completed with sufficient rigor during the Clinical Doctoral Fellowship. The scope of this thesis was therefore limited to establishing a minimum set of outcomes. The process of linking the agreed core outcomes to the most appropriate measurement tools is planned as future work and highlighted in Chapter 6, section 6.4 – Recommendations for future research.

Chapter 4 Creating a “long list” of outcomes

4.1 Paper II – Evaluating physical activity in dementia: a systematic review of outcomes to inform the development of a core outcome set

“This is a pre-copyedited, author-produced version of an article accepted for publication in Age and Aging, following peer review. The version of record *Gonçalves, A.C.; Cruz, J.; Marques, A.; Demain, S. and Samuel, D. 2018. Age and Ageing, 47(1), 34-41,* is available online at: https://academic.oup.com/ageing/article/47/1/34/4079771”

ABSTRACT

Background: Physical activity is recommended for people living with dementia, but evidence for the positive effects of physical activity is limited by the use of heterogeneous outcomes and measurement tools. This systematic literature review aimed to summarise previously reported outcomes and identify the measurement tools used most frequently in physical activity interventions for people with dementia.

Methods: Literature searches were conducted in April 2015, on Delphis and Medline. Qualitative, quantitative and mixed methods studies reporting on any type of physical activity, in any setting, across types of dementia, stages of disease progression and published from 2005 onwards were included. A content analysis approach was used to report on the frequency of reported outcomes and measurement tools.

Results: The 130 included studies reported on 133 different outcome domains and 267 different measurement tools. “Functional abilities and independence” (n=69), “Global cognitive function” (n=65), “Balance” (n=43), “Global behavioural symptoms of dementia” (n=42) and “Health related quality of life” (n=40) were the most frequently reported outcome domains. “Enjoyment” was the outcome most frequently sought by patients and carers.

Conclusion: The need for the development and implementation of a Core Outcome Set has been reinforced. Ahead of the completion of the Core Outcome Set, researchers and clinicians are advised to measure the impact of physical activity interventions on these frequently reported outcome domains.

Keywords: Physical activity; Dementia; Outcomes; Measurement tools; Core Outcome Set; Systematic review; Older people.

Key Points:

Research into physical activity for people with dementia is heterogeneous, limiting guidance to practice;

This literature review summarises the outcomes and measurement tools used in the past 10 years of research;

Clinicians and researchers are encouraged to continue to assess the impact of physical activity on Functional independence;

Researchers should prefer measurement tools frequently used in previous research – these tools can be found in this review;

The effects of physical activity in people with severe dementia are under-researched.

BACKGROUND

Dementia is predicted to affect 42.3 million people worldwide by 2020 (Hill et al., 2009). Physical activity is defined by The World Health Organisation as “any body movement produced by skeletal muscles that requires energy expenditure”, including therefore not only formal exercise but also any activity involving body movement. Physical activity is recommended for people with dementia by several international agencies (National Institute for Health and Care Excellence, 2006;Forbes et al., 2015;Miskovski, 2014;Physical Activity Guidelines Advisory Committee, 2008), as there is promising evidence of its benefits. However, the use of heterogeneous outcomes leads to a need for caution in the interpretation of these results (Forbes et al., 2015). Two recent literature reviews (Forbes et al., 2015;Rao et al., 2014) reported important limitations in their statistical analyses due to a lack of consistent use of measurement tools. This heterogeneity adds to an already diverse area of research, as physical activity is a broad concept, and interventions tend to vary in type of activity, intensity, setting, type of dementia and stage of disease progression. The use of inconsistent outcomes and measurement tools hinders the effective synthesis of evidence (Macefield et al., 2014) making it difficult to compare interventions and therefore delaying the development of clear clinical guidelines.

The development of Core Outcome Sets has been suggested as a solution to this heterogeneity. A Core Outcome Set is an agreed minimum collection of outcomes to be used as standard in a particular pathological condition or type of intervention (Hopkins et al., 2015a;Williamson et al., 2012;Clarke and Williamson, 2015;Idzerda et al., 2014;Sinha, 2011). The use of Core Outcome Sets allows direct comparison of the effects of different interventions, minimising the risk of outcome reporting bias and increasing the power of meta-analysis (Williamson et al., 2012;Sinha, 2011;MacLennan et al., 2015), from which clearer clinical guidance can be generated (Tugwell et al., 2007).

The present systematic literature review is the first phase of the development of a Core Outcome Set, to evaluate physical activity interventions for people with any type of dementia, across stages of disease progression and in different settings. This review aims to i) list all the outcomes reported in physical activity research in the last 10 years ; and ii) identify the most frequently reported measurement tools for each of the identified outcomes.

METHODS

A systematic literature review was conducted. The review protocol is available on request.

Registration:

The overall Core Outcome Set is registered with the COMET initiative, available at: http://www.comet-initiative.org/studies/details/708?result=true.

Eligibility criteria

Design: Any study design describing a physical activity intervention, or exploring patients', relatives', carers' or professionals' views on physical activity was included. Study protocols, pilot or feasibility studies were also included. Literature reviews were not included, but their reference lists were screened and considered according to the eligibility criteria. Single experimental case studies, surveys, cross-sectional studies or studies not published in a peer-reviewed journal were excluded.

Study participants: Studies including people with any type of dementia, at any stage of disease progression, formal and informal carers and/or health care professionals working with people with dementia were included. Interventions including only carers of people with dementia were excluded.

Intervention: Interventions with at least one component of physical activity, in any setting, were included. Qualitative studies exploring participants' views on physical activity were also included.

Comparators: Having a comparator or control group was not a requirement for inclusion; when available, outcomes used as comparators were considered for data analysis.

Language and date of publication: Studies written in English, Portuguese and Spanish published from 1st of January 2005 onwards were included.

Information sources

The search strategy was developed with the advice of a health sciences librarian. The searches were initially conducted in Delphis, a single interface integrating the providers: Medline, Psycinfo, CINAHL, Web of Science, Scopus and ScienceDirect. To ensure literature saturation, a subheading search was then performed on Medline - the database that had generated the greatest number or records in the Delphis search.

Search strategy

A full detail of the search strategy can be found in appendix 1[5] (tab 1), available from the journal website.

Selection criteria

The selection process (screening and eligibility) was completed by one researcher; other authors were consulted in cases of uncertainty. To guarantee consistency, a random sample of 10% of abstracts were independently screened by a second author.

Risk of bias

The quality of included papers was assessed using the Mixed Methods Assessment tool – version 2011 (Pluye et al., 2009). This tool was designed for the appraisal of studies included in complex, mixed studies reviews. For the purpose of this review, the quality of the included studies was used purely as an indicator of the quality of the evidence in the field, not as an exclusion criterion and it did not influence data analysis.

Data collection process

A pre-developed standardised form was used, to extract the following data: paradigm and study design; country; stage of disease progression; intervention outcomes and measurement tools. Outcomes were defined as any effect (positive or negative) of physical activity, which had been measured or described as a result of a physical activity intervention. Outcomes were identified from the methods, results and discussion sections of the included papers. To avoid double counting of outcomes, multiple publications of the same study were analysed as one (i.e., protocol and pilot of the same study).

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[5] This is an interactive excel file, found with this publication but not possible to attach to the thesis document. Please refer to the journal website access appendix 1.

Data analysis

NVivo (NVivo qualitative data analysis Software; QSR International Pty Ltd. Version 10, 2014) was used for data management.

From verbatim outcomes to outcome domains: Content analysis (Elo and Kyngas, 2008) was used to quantify the number of studies referencing each outcome domain. Firstly, outcomes were extracted verbatim. One author, with clinical experience in promoting physical activity in dementia care, grouped verbatim outcomes with the same semantic meaning, into outcome domains. For instance, the verbatim outcomes “*Functional independence*”, “*Ability to develop basic activities of daily living*” and “*Functional performance*” were grouped into the outcome domain “Functional abilities and independence”. The total number of studies allocated to each outcome domain was then counted.

Post publication note: In a recent paper, published since the paper presented in this thesis, Young et al. (2019) used the following definition of outcome domain: “A trial outcome is one that has original meaning and context”. This aligns with the definitions used in this thesis where outcomes with the same meaning but different wording were described as one single outcome. Additionally, the same outcome measured in different time points was also counted as one outcome/outcome domain[6]*.*

From outcome domains to themes: Four authors individually organised the outcome domains into broader themes. Any discrepancies were resolved through group discussion until full consensus was achieved. For instance, the outcome domains “*Keep fit and active*”, “*Levels of physical activity*” and “*Levels of restricted physical activity*” were listed under the theme: “Levels of physical activity”.

Appendix1 (tab 3), aids transparency by presenting all verbatim outcomes, their groupings into outcome domains and organisation into themes. The analysis and presentation of results followed the guidance provided by the PRISMA statement (Moher et al., 2009), with the necessary adaptations for a mixed studies review.

RESULTS

The searches were conducted in April 2015. The PRISMA flowchart is presented in Figure 1. A total of 4868 records were identified, from which 130 studies were included in the final analysis.

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[6] *Added post publication to enhance content.*

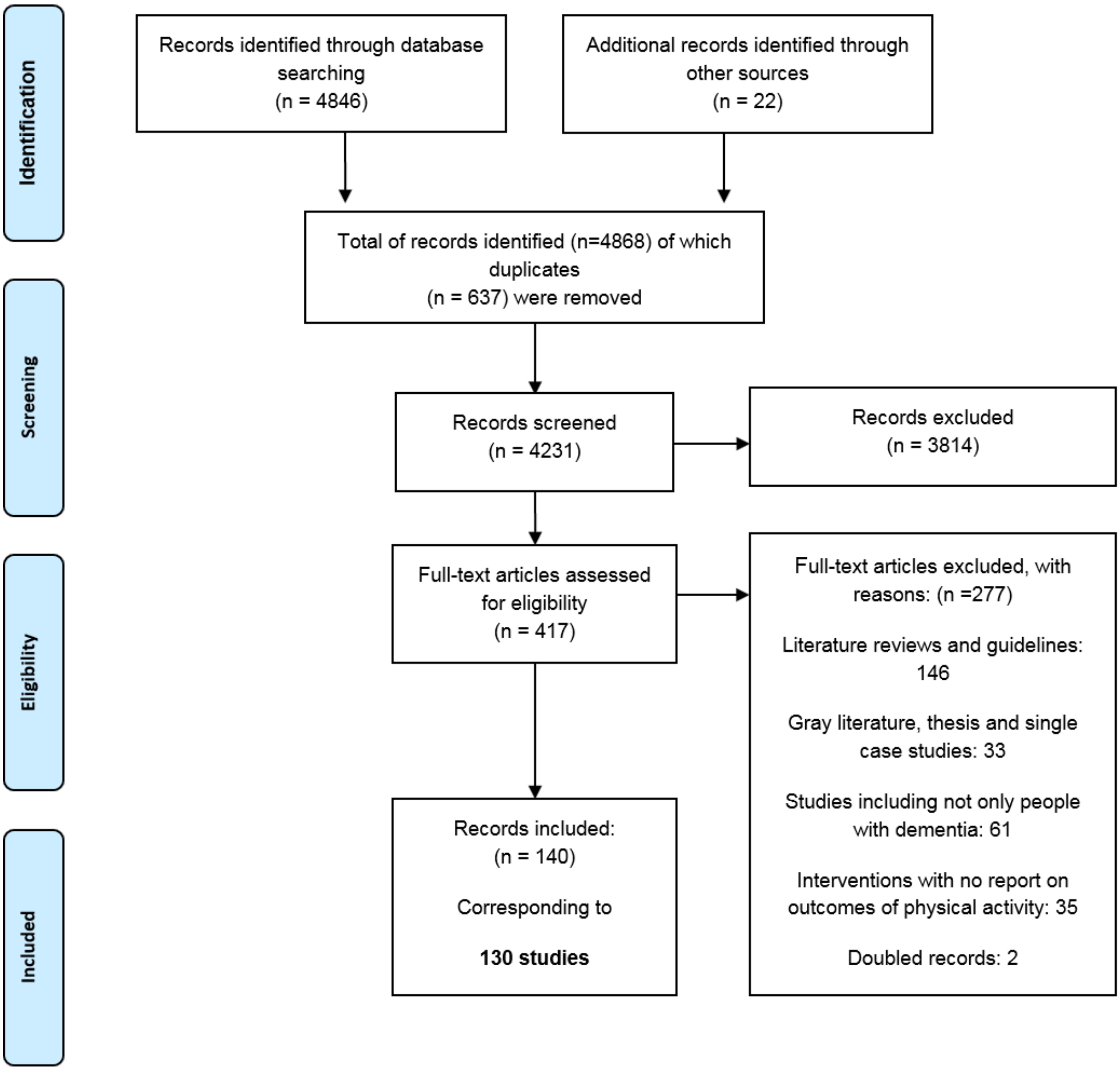

Figure 1- PRISMA flow chart: number of records identified, screened, excluded (with reasons) and included in the present literature review.

Study selection

The screening of 10% of the abstract by a second author revealed good consistency in the use of the eligibility criteria. Of the 500 abstracts screened by both authors, 468 were screened equally by both authors. Of the remaining 32 abstracts, 15 were not considered clear enough by one of the authors and the full texts were considered to make a decision; 17 were screened contradictorily by both authors – nine would have been only included by the first author and eight only by the second author. Measurement of agreement using Cohen’s Kappa was 0.69. Disagreements were resolved through consensus and the eligibility criteria were reviewed to ensure clarity.

Study characteristics

The 130 included studies were conducted in 22 countries; 113 studies used quantitative methods, nine qualitative and eight used mixed methods. The overall quality of the studies was considered good (Mixed Methods Assessment tool average score: 65%). Appendix 1 (tab 10), presents the quality assessment of each of the included papers. Most interventions took place in the community (n=70) or in institutional settings (n=43). Studies including only people with severe dementia (n=5) were all performed in nursing homes. A large number of interventions were multimodal or complex interventions (n=60) and were delivered in groups (n=62). Table 1 presents further details on the description of the interventions, settings and participants from the included studies.

Study characteristics	Reference
Stage of Disease progression	
All stages (n=26)	(Ferrer and del Valle, 2014;Lowery et al., 2014;Rodriguez-Sanchez et al., 2014;Guzman-Garcia et al., 2013;Harmer and Orrell, 2008;Ferrer et al., 2013;Garuffi et al., 2013;Yao et al., 2013;Rogers and Jarrott, 2012;Litchke et al., 2012;Hooghiemstra et al., 2012;Thurm et al., 2011;Volkers and Scherder, 2011;McCurry et al., 2011;McCurry et al., 2010;Kemoun et al., 2010;Yao et al., 2008;Lee and Kim, 2008;Jarrott et al., 2008;Netz et al., 2007;Connell et al., 2007;Rolland et al., 2007;Woodhead et al., 2005;Mackintosh and Sheppard, 2005;Kolanowski et al., 2011;Roach et al., 2011;Williams and Tappen, 2007).
Mild to moderate (n=72)	(Hill et al., 2009;Holthoff et al., 2015;Padala et al., 2012;Brown et al., 2015;Bossers et al., 2015;Li et al., 2014;Frederiksen et al., 2014;Miu et al., 2008;Soderhamn et al., 2014;Yu et al., 2014;Khoo et al., 2014;Cedervall et al., 2015;Schwenk et al., 2010;Aguiar et al., 2014;Malthouse and Fox, 2014;Bossers et al., 2014;Cadore et al., 2014;Cheng et al., 2014b;Nascimento et al., 2014;Schwenk et al., 2014a;Arcoverde et al., 2014;Coelho et al., 2014;Cheng et al., 2014a;Yoon et al., 2013;Slaughter and Estabrooks, 2013;de Andrade et al., 2013;Hoffmann et al., 2013;Tanaka et al., 2013;Wesson et al., 2013;Coelho et al., 2013;Yu et al., 2013a;Groppo et al., 2012;Vidoni et al., 2012;Suttanon et al., 2012a;Vital et al., 2012;Canonici et al., 2012;Yu and Swartwood, 2012;Nascimento et al., 2012;Yerokhin et al., 2012;Maci et al., 2012;Pedroso et al., 2012;Pitkala et al., 2011;Quintero et al., 2011;Prick et al., 2011;Yaguez et al., 2011;Gil et al., 2006;Lam et al., 2010;Yu et al., 2011;Stella et al., 2011;Cyarto et al., 2010;Kang et al., 2010;Ramstrom, 2010;Schwenk et al., 2014b;Cedervall and Aberg, 2010;Callahan et al., 2006;Ries et al., 2010;Steinberg et al., 2009;Eggermont et al., 2009b;Eggermont et al., 2009a;Santana-Sosa et al., 2008;Fajersztajn et al., 2008;Christofoletti et al., 2008;Kwak et al., 2008;Littbrand et al., 2006;Stevens and Killeen, 2006;Hernandez et al., 2010;Manckoundia et al., 2014;Suttanon et al., 2012b;Hauer et al., 2012;Burgener et al., 2008;Barnes et al., 2015;Pitkala et al., 2013b;Ferretti et al., 2014;Zieschang et al., 2013;Huger et al., 2009;Yu et al., 2013b;Pitkala et al., 2010;Burgener et al., 2011;Wu et al., 2015)
Moderate to Severe (n=19)	(Hwang and Choi, 2010;Henwood et al., 2015;Hutson et al., 2014;McCaffrey et al., 2014;Neville et al., 2014;Duignan et al., 2009;Cruz et al., 2013;Venturelli et al., 2012;Fan and Chen, 2011;Venturelli et al., 2011;Kader, 2010;Edwards et al., 2008;Thelander et al., 2008;Williams and Tappen, 2008;Volicer et al., 2006;Logsdon et al., 2005;McCurry et al., 2005;Ries et al., 2015;Cruz et al., 2011;Marques et al., 2013)
Severe (n=5)	(Galik et al., 2014;Aman and Thomas, 2009;Dayanim, 2009;Galik et al., 2008;Schreiner et al., 2005)

Stage not specified (n=8)	(Dal Bello-Haas et al., 2014;Johnson et al., 2012;Mapes, 2012;Chang et al., 2011;Parkinson and Milligan, 2011;Richeson and Croteau, 2010;Cerga-Pashoja et al., 2010;Ootani et al., 2005)
Setting	
Community (n=70)	(Litchke et al., 2012;McCurry et al., 2011;McCurry et al., 2010;Yao et al., 2008;Jarrott et al., 2008;Netz et al., 2007;Woodhead et al., 2005;Mackintosh and Sheppard, 2005;Lowery et al., 2014;Rodriguez-Sanchez et al., 2014;Garuffi et al., 2013;Yao et al., 2013;Rogers and Jarrott, 2012;Holthoff et al., 2015;Padala et al., 2012;Brown et al., 2015;Frederiksen et al., 2014;Soderhamn et al., 2014;Yu et al., 2014;Khoo et al., 2014;Cedervall et al., 2015;Malthouse and Fox, 2014;Wesson et al., 2013;Coelho et al., 2013;Yu et al., 2013a;Vidoni et al., 2012;Suttanon et al., 2012a;Vital et al., 2012;Yu and Swartwood, 2012;Nascimento et al., 2012;Yerokhin et al., 2012;Vreugdenhil et al., 2012;Maci et al., 2012;Pitkala et al., 2011;Quintero et al., 2011;Prick et al., 2011;Yaguez et al., 2011;Gil et al., 2006;Lam et al., 2010;Stella et al., 2011;Cyarto et al., 2010;Kang et al., 2010;Ramstrom, 2010;Schwenk et al., 2014b;Cedervall and Aberg, 2010;Callahan et al., 2006;Ries et al., 2010;Steinberg et al., 2009;Fajersztajn et al., 2008;Kwak et al., 2008;Hernandez et al., 2010;Manckoundia et al., 2014;Suttanon et al., 2012b;Hauer et al., 2012;Yu et al., 2011;Burgener et al., 2008;Barnes et al., 2015;Pitkala et al., 2013b;Ferretti et al., 2014;Zieschang et al., 2013;Huger et al., 2009;Yu et al., 2013b;Pitkala et al., 2010;Burgener et al., 2011;Wu et al., 2015;Henwood et al., 2015;McCaffrey et al., 2014;Neville et al., 2014;Logsdon et al., 2005;McCurry et al., 2005;Ries et al., 2015;Dal Bello-Haas et al., 2014;Johnson et al., 2012;Mapes, 2012;Chang et al., 2011;Parkinson and Milligan, 2011;Cerga-Pashoja et al., 2010;Ootani et al., 2005)
Institution (n=43)	(Hooghiemstra et al., 2012;Thurm et al., 2011;Volkers and Scherder, 2011;Kemoun et al., 2010;Lee and Kim, 2008;Connell et al., 2007;Rolland et al., 2007;Kolanowski et al., 2011;Roach et al., 2011;Williams and Tappen, 2007;Guzman-Garcia et al., 2013;Harmer and Orrell, 2008;Bossers et al., 2015;Bossers et al., 2014;Cadore et al., 2014;Cheng et al., 2014b;Cheng et al., 2014a;Yoon et al., 2013;Slaughter and Estabrooks, 2013;Tanaka et al., 2013;Eggermont et al., 2009b;Eggermont et al., 2009a;Santana-Sosa et al., 2008;Christofoletti et al., 2008;Littbrand et al., 2006;Stevens and Killeen, 2006;Hwang and Choi, 2010;Hutson et al., 2014;Duignan et al., 2009;Cruz et al., 2013;Venturelli et al., 2012;Fan and Chen, 2011;Venturelli et al., 2011;Edwards et al., 2008;Thelander et al., 2008;Williams and Tappen, 2008;Volicer et al., 2006;Cruz et al., 2011;Marques et al., 2013;Galik et al., 2014;Aman and Thomas, 2009;Dayanim, 2009;Galik et al., 2008;Schreiner et al., 2005;Richeson and Croteau, 2010)
Hospital (n=7)	(Ferrer et al., 2013;Ferrer and del Valle, 2014;Li et al., 2014;Miu et al., 2008;Schwenk et al., 2010;Aguiar et al., 2014;Schwenk et al., 2014a)

Not specified (n=10)	(Nascimento et al., 2014;Arcoverde et al., 2014;Coelho et al., 2014;de Andrade et al., 2013;Hoffmann et al., 2013;Groppo et al., 2012;Canonici et al., 2012;Pedroso et al., 2012;Kader, 2010;Soderhamn et al., 2014)
Delivery	
Group intervention (n=62)	(Jarrott et al., 2008;Nascimento et al., 2014;Schwenk et al., 2014a;Cheng et al., 2014a;Hoffmann et al., 2013;Groppo et al., 2012;Vital et al., 2012;Canonici et al., 2012;Nascimento et al., 2012;Maci et al., 2012;Quintero et al., 2011;Yaguez et al., 2011;Lam et al., 2010;Stella et al., 2011;Kang et al., 2010;Ramstrom, 2010;Schwenk et al., 2014b;Callahan et al., 2006;Ries et al., 2010;Eggermont et al., 2009a;Santana-Sosa et al., 2008;Fajersztajn et al., 2008;Littbrand et al., 2006;Stevens and Killeen, 2006;Hernandez et al., 2010;Hauer et al., 2012;Yu et al., 2011;Barnes et al., 2015;Zieschang et al., 2013;Huger et al., 2009;Yu et al., 2013b;Burgener et al., 2011;Wu et al., 2015;Hwang and Choi, 2010;Henwood et al., 2015;Hutson et al., 2014;McCaffrey et al., 2014;Neville et al., 2014;Duignan et al., 2009;Cruz et al., 2013;Venturelli et al., 2012;Fan and Chen, 2011;Edwards et al., 2008;Thelander et al., 2008;Volicer et al., 2006;Ries et al., 2015;Marques et al., 2013;Dayanim, 2009;Johnson et al., 2012;Mapes, 2012;Parkinson and Milligan, 2011;Soderhamn et al., 2014;Ferrer and del Valle, 2014;Guzman-Garcia et al., 2013;Garuffi et al., 2013;Rogers and Jarrott, 2012;Litchke et al., 2012;Netz et al., 2007;Connell et al., 2007;Rolland et al., 2007;Woodhead et al., 2005;Padala et al., 2012;Brown et al., 2015;Li et al., 2014;Frederiksen et al., 2014;Yu et al., 2014;Khoo et al., 2014;Schwenk et al., 2010;Cheng et al., 2014b)
Individual intervention (n= 27)	(Arcoverde et al., 2014;Coelho et al., 2014;Slaughter and Estabrooks, 2013;Tanaka et al., 2013;Yu et al., 2013a;Vidoni et al., 2012;Suttanon et al., 2012a;Yu and Swartwood, 2012;Cyarto et al., 2010;Eggermont et al., 2009b;Manckoundia et al., 2014;Ferretti et al., 2014;Williams and Tappen, 2008;Cruz et al., 2011;Galik et al., 2014;Aman and Thomas, 2009;Galik et al., 2008;Richeson and Croteau, 2010;Ootani et al., 2005;Cedervall et al., 2015;Hill et al., 2009;Roach et al., 2011;Williams and Tappen, 2007;Holthoff et al., 2015;Bossers et al., 2015;Miu et al., 2008;Aguiar et al., 2014;Bossers et al., 2014)
Intervention including dyads (n=14)	(Wesson et al., 2013;Vreugdenhil et al., 2012;Prick et al., 2011;Steinberg et al., 2009;Kwak et al., 2008;Suttanon et al., 2012b;Venturelli et al., 2011;Logsdon et al., 2005;McCurry et al., 2005;Cerga-Pashoja et al., 2010;Lowery et al., 2014;Rodriguez-Sanchez et al., 2014;McCurry et al., 2011;McCurry et al., 2010)

A combination of delivery options (n= 14)	(Pitkala et al., 2011;Gil et al., 2006;Cedervall and Aberg, 2010;Christofoletti et al., 2008;Burgener et al., 2008;Pitkala et al., 2013b;Pitkala et al., 2010;Schreiner et al., 2005;Dal Bello-Haas et al., 2014;Yao et al., 2013;Yao et al., 2008;Hooghiemstra et al., 2012;Mackintosh and Sheppard, 2005;Kolanowski et al., 2011;Malthouse and Fox, 2014;Cadore et al., 2014)
Not specified (n= 13)	(Yoon et al., 2013;de Andrade et al., 2013;Coelho et al., 2013;Yerokhin et al., 2012;Pedroso et al., 2012;Kader, 2010;Chang et al., 2011;Harmer and Orrell, 2008;Ferrer et al., 2013;Thurm et al., 2011;Volkers and Scherder, 2011;Kemoun et al., 2010;Lee and Kim, 2008)
Type of physical activity	
Aerobic (e.g. walking, dancing, cycling, ball games) (n= 33)	(Arcoverde et al., 2014;Coelho et al., 2014;Yoon et al., 2013;Hoffmann et al., 2013;Yu et al., 2013a;Vidoni et al., 2012;Yu and Swartwood, 2012;Cyarto et al., 2010;Cedervall and Aberg, 2010;Eggermont et al., 2009b;Stevens and Killeen, 2006;Yu et al., 2011;Yu et al., 2013b;Hwang and Choi, 2010;Duignan et al., 2009;Venturelli et al., 2012;Venturelli et al., 2011;Kader, 2010;McCurry et al., 2005;Dal Bello-Haas et al., 2014;Mapes, 2012;Richeson and Croteau, 2010;Cerga-Pashoja et al., 2010;Ootani et al., 2005;Guzman-Garcia et al., 2013;Frederiksen et al., 2014;Yu et al., 2014;Bossers et al., 2015;Miu et al., 2008;Lowery et al., 2014;McCurry et al., 2011;McCurry et al., 2010;Hooghiemstra et al., 2012;Volkers and Scherder, 2011)
Strength and balance (e.g. yoga, tai chi, resistance and balance training) (n=27)	(Cheng et al., 2014a;Tanaka et al., 2013;Wesson et al., 2013;Vital et al., 2012;Yerokhin et al., 2012;Maci et al., 2012;Quintero et al., 2011;Callahan et al., 2006;Ries et al., 2010;Manckoundia et al., 2014;Ferretti et al., 2014;Zieschang et al., 2013;McCaffrey et al., 2014;Fan and Chen, 2011;Edwards et al., 2008;Williams and Tappen, 2008;Ries et al., 2015;Johnson et al., 2012;Parkinson and Milligan, 2011;Garuffi et al., 2013;Rogers and Jarrott, 2012;Litchke et al., 2012;Cheng et al., 2014b;Hill et al., 2009;Yao et al., 2013;Yao et al., 2008;Arcoverde et al., 2014).
Functional activities (e.g. gardening, activities of daily living) (n= 9)	(Slaughter and Estabrooks, 2013;Lam et al., 2010;Fajersztajn et al., 2008;Thelander et al., 2008;Galik et al., 2014;Connell et al., 2007;Li et al., 2014;Kolanowski et al., 2011;Lee and Kim, 2008)

Multimodal interventions (n=60)	(Nascimento et al., 2014;Schwenk et al., 2014a;de Andrade et al., 2013;Coelho et al., 2013;Suttanon et al., 2012a;Canonici et al., 2012;Nascimento et al., 2012;Vreugdenhil et al., 2012;Pedroso et al., 2012;Pitkala et al., 2011;Prick et al., 2011;Yaguez et al., 2011;Gil et al., 2006;Stella et al., 2011;Kang et al., 2010;Ramstrom, 2010;Schwenk et al., 2014b;Steinberg et al., 2009;Santana-Sosa et al., 2008;Christofoletti et al., 2008;Kwak et al., 2008;Littbrand et al., 2006;Hernandez et al., 2010;Suttanon et al., 2012b;Hauer et al., 2012;Burgener et al., 2008;Barnes et al., 2015;Pitkala et al., 2013b;Huger et al., 2009;Pitkala et al., 2010;Burgener et al., 2011;Wu et al., 2015;Henwood et al., 2015;Hutson et al., 2014;Neville et al., 2014;Cruz et al., 2013;Volicer et al., 2006;Logsdon et al., 2005;Cruz et al., 2011;Marques et al., 2013;Aman and Thomas, 2009;Dayanim, 2009;Galik et al., 2008;Schreiner et al., 2005;Chang et al., 2011;Soderhamn et al., 2014;Ferrer and del Valle, 2014;Netz et al., 2007;Rolland et al., 2007;Padala et al., 2012;Brown et al., 2015;Khoo et al., 2014;Cedervall et al., 2015;Schwenk et al., 2010;Roach et al., 2011;Williams and Tappen, 2007;Holthoff et al., 2015;Bossers et al., 2015;Aguiar et al., 2014;Bossers et al., 2014;Rodriguez-Sanchez et al., 2014;Mackintosh and Sheppard, 2005;Malthouse and Fox, 2014;Cadore et al., 2014;Harmer and Orrell, 2008;Ferrer et al., 2013;Thurm et al., 2011;Kemoun et al., 2010;Jarrott et al., 2008;Groppo et al., 2012)
Hand motor activity (n= 1)	(Eggermont et al., 2009a)
Not specified (n= 1)	(Woodhead et al., 2005)

Table 1 - Characteristics of the included studies. Legend: n – number of studies (please note that some studies were described in multiple publications and therefore multiple references can be linked to the same study).

Outcomes reporting positive effects of physical activity for people with dementia

A total of 1552 verbatim outcomes were identified. Those verbatim outcomes were grouped[7] into 133 positive outcome domains, organised into 17 themes, and measured by 267 different measurement tools. A considerable number of outcome domains (36 out of 133) were reported only once. Each study incorporated an average 9.49 different outcome domains. Table 2 lists the themes, outcome domains and the most frequently used measurement tool per domain. Due to space restrictions, only the two most frequently reported outcome domains per theme are listed in Table 2. The complete table, including the all the 133 positive outcome domains and the three most frequently used measurement tools per outcome domain, can be found in appendix 1 (tab 2).

The most frequently reported outcome domains were “Functional abilities and independence” (n=69), “Global cognitive function” (n=65), “Balance” (n=43), “Global behavioural symptoms of dementia” (n=42) and “Health related quality of life (n=40)”. In qualitative studies, “Enjoyment” was the outcome most frequently reported by patients and carers (n=22). Outcomes such as “Mobility”, “Health related quality of life”, all outcome domains under the themes “Social interaction” and “Carer outcomes” were not reported in patients with severe dementia. Outcomes such as “Fatigue management” and “Sense of normality” have been identified as important by patients in qualitative studies, but never objectively measured. Other outcomes, such as “Balance”, although frequently measured (n=43) were identified by only one carer in qualitative studies.

Most frequently reported measurement tools

The most frequently reported measurement tools in the included studies were Mini Mental State Examination as a measure of “Global cognitive function” (used in 43 of the 65 studies reporting on this domain); the Berg Balance Scale, as a measure of “Balance” (15 of the 43 studies) and the Timed Up And Go as an assessment of “Mobility” (14 out of 23 studies). Conversely, some measurement tools, such as the Timed Up And Go, were used to measure multiple outcome domains (in this instance “Ability to conduct movements rightly”, “Motor skills”, “Functional gait”, “Mobility” and “Falls risk”). See Table 2 and appendix 1 (tab 2) for further details. Appendix 1 (tabs 4 to 6) also offers an interactive overview of themes, outcomes and measurement tools per setting and stage of disease progression and across the last 10 years of research.

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[7] *Added post publication to enhance content.*

Table 2 - Short list of outcome domains and measurement tools organised by themes.

PLEASE NOTE: Due to space restrictions only the two most frequently reported outcome domains per theme, and the most frequently reported measurement tool per outcome domain are presented in this table. Please refer to appendix 1 (tab 2), available from the journal website, to access the full table. Legend: P&C: patients and carers; HP: health professionals; R: researchers; NA: Not applicable, used when the outcome domain was not objectively measured; "Study Specific Questionnaire/Survey", refers to data collection tools designed specifically for a particular study.

Theme: Levels of physical activity	**Frequency of use per stakeholder**			**Most frequently used measurement tool (frequency of use)**	***Primary reference for the measurement tool***[8]
Outcome domain (number of studies)	**P&C**	**HP**	**R**		
Levels of physical activity (23)	5	0	18	Physical Activity Scale For The Elderly (PASE) (4/23);	*(Washburn et al., 1993)*
Keep fit and active (5)	5	0	0	Study Specific Questionnaire/Survey (1/5)	*NA*

Theme: Health related physical fitness	**Frequency of use per stakeholder**			**Most frequently used measurement tool (frequency of use)**	
Outcome domain (number of studies)	**P&C**	**HP**	**R**		
Balance (43)	1	2	41	Berg Balance Scale (BBS) (15/43);	*(Berg, 1989)*
Physical performance and fitness (33)	3	0	30	6 Minute Walk Test (6/33);	*(Enright, 2003)*

Theme: Movement quality and control	**Frequency of use per stakeholder**			**Most frequently used measurement tool (frequency of use)**	***Primary reference for the measurement tool***
Outcome domain (number of studies)	**P&C**	**HP**	**R**		

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[8] *Content added post publication to enhance clarity.*

Ability to conduct movements rightly (5)	1	2	3	Timed Up And Go (TUG) (1/5)	*(Podsiadlo and Richardson, 1991)*
Agility (5)	0	0	5	*All Different*	*NA*
Theme: Walking ability and functional independence	**Frequency of use per stakeholder**			**Most frequently used measurement tool (frequency of use)**	***Primary reference for the measurement tool***
Outcome domain (number of studies)	**P&C**	**HP**	**R**		
Functional abilities and independence (69)	3	2	66	Barthel Index (12/69); Timed Up And Go (12/69)	*(Mahoney and Barthel, 1965;Podsiadlo and Richardson, 1991)*
Mobility (23)	5	0	18	Timed Up And Go (TUG) (14/23);	*(Podsiadlo and Richardson, 1991)*
Theme: cardiorespiratory function	**Frequency of use per stakeholder**			**Most frequently used measurement tool (frequency of use)**	***Primary reference for the measurement tool***
Outcome domain (number of studies)	**P&C**	**HP**	**R**		
Heart rate (5)	0	0	5	Blood Pressure Monitor (2/5)	*NA*
Blood pressure (4)	0	0	4	Blood Pressure Monitor (3/4)	*NA*
Theme: Reducing falls	**Frequency of use per stakeholder**			**Most frequently used measurement tool (frequency of use)**	***Primary reference for the measurement tool***
Outcome domain (number of studies)	**P&C**	**HP**	**R**		
Number of falls (17)	1	0	16	Study Specific Questionnaire/Survey (12/17)	*NA*
Falls risk (13)	0	0	13	Timed Up And Go (TUG) (6/13);	*(Podsiadlo and Richardson, 1991)*
Theme: General Health	**Frequency of use per stakeholder**			**Most frequently used measurement tool (frequency of use)**	***Primary reference for the measurement tool***
Outcome domain (number of studies)	**P&C**	**HP**	**R**		
Medication (10)	1	1	8	Study Specific Questionnaire/Survey (2/10)	*NA*
Health and social care service use (9)	0	0	9	Study Specific Questionnaire/Survey (2/9)	*NA*
Theme: Dementia physiological indicators	**Frequency of use per stakeholder**			**Most frequently used measurement tool (frequency of use)**	***Primary reference for the measurement tool***
Outcome domain (number of studies)	**P&C**	**HP**	**R**		

Disease progression (9)	1	1	7	Clinical Dementia Rating (CDR) (3/9);	*(Hughes et al., 1982)*
Brain volumes, physiology and imaging (5)	0	0	5	Magnetic Resonance Imaging (MRI) (3/5);	*NA*
Theme: Cognitive function	**Frequency of use per stakeholder**			**Most frequently used measurement tool (frequency of use)**	***Primary reference for the measurement tool***
Outcome domain (number of studies)	**P&C**	**HP**	**R**		
Global cognitive function (65)	3	0	62	Mini-Mental State Examination (MMSE) (43/65);	*(Folstein et al., 1975)*
Executive function (18)	0	0	18	Stroop Test (5/18);	*(Stroop, 1935)*
Theme: Verbal and non-verbal communication	**Frequency of use per stakeholder**			**Most frequently used measurement tool (frequency of use)**	***Primary reference for the measurement tool***
Outcome domain (number of studies)	**P&C**	**HP**	**R**		
Expression and communication (6)	2	3	1	Study Specific Questionnaire/Survey (1/6)	*NA*
Language ability (6)	0	0	6	*Verbal* Fluency Test (VF) (2/6)	*No development study found*
Theme: Behavioural and psychological symptoms of dementia	**Frequency of use per stakeholder**			**Most frequently used measurement tool (frequency of use)**	***Primary reference for the measurement tool***
Outcome domain (number of studies)	**P&C**	**HP**	**R**		
Global Behavioural symptoms of dementia (42)	0	1	41	Neuropsychiatric Inventory (NPI) (14/42);	*(Cummings et al., 1994)*
Depression (33)	1	0	32	Cornell Scale For Depression In Dementia (CSDD) (12/33);	*(Alexopoulos et al., 1988)*
Theme: Emotional status	**Frequency of use per stakeholder**			**Most frequently used measurement tool (frequency of use)**	***Primary reference for the measurement tool***
Outcome domain (number of studies)	**P&C**	**HP**	**R**		
Enjoyment (22)	12	2	10	NA	*NA*
Affective function (9)	0	1	8	Apparent Affect Rating Scale (AARS) (3/9)	*(Lawton et al., 1996)*
Theme: Quality of life and personhood	**Frequency of use per stakeholder**			**Most frequently used measurement tool (frequency of use)**	***Primary reference for the measurement tool***
Outcome domain (number of studies)	**P&C**	**HP**	**R**		

Health related quality of life (40)	1	0	39	Quality Of Life In Alzheimer’s Disease (QoL-AD) Scale (8/40);	*(Logsdon et al., 1999)*
Sense of wellbeing (11)	2	1	9	*All Different*	*NA*
Theme: Social interaction	**Frequency of use per stakeholder**			**Most frequently used measurement tool (frequency of use)**	***Primary reference for the measurement tool***
Outcome domain (number of studies)	**P&C**	**HP**	**R**		
Global social interaction (16)	7	3	7	Holden Communication Scale (1/16)	*No development study found*
Opportunity to leave the house (3)	2	0	1	NA	*NA*
Theme: Setting	**Frequency of use per stakeholder**			**Most frequently used measurement tool (frequency of use)**	***Primary reference for the measurement tool***
Outcome domain (number of studies)	**P&C**	**HP**	**R**		
Institutionalisation (7)	1	0	6	Study Specific Questionnaire/Survey (1/7)	*NA*
Stay at home for as long as possible (2)	0	0	2	NA	*NA*
Theme: Adherence	**Frequency of use per stakeholder**			**Most frequently used measurement tool (frequency of use)**	***Primary reference for the measurement tool***
Outcome domain (number of studies)	**P&C**	**HP**	**R**		
Adherence (30)	2	0	28	Study Specific Questionnaire/Survey (24/30);	*NA*
Theme: Carer outcomes	**Frequency of use per stakeholder**			**Most frequently used measurement tool (frequency of use)**	***Primary reference for the measurement tool***
Outcome domain (number of studies)	**P&C**	**HP**	**R**		
Carer burden (33)	3	0	30	Zarit Caregiver Burden Inventory (ZBI) (7/33);	*(Zarit et al., 1980)*
Carer quality of life (6)	0	0	6	Quality Of Life In Alzheimer’s Disease (QoL-AD) Scale (2/6)	*(Logsdon et al., 1999)*

Outcomes reporting negative effects (or adverse/side effects) of physical activity for people with dementia

A total of 31 adverse effects were reported. “Falls during the activity” was the most common (n=15). Most studies (n=82) did not mention the occurrence of adverse effects. Negative outcomes, or adverse effects are also detailed in the appendix 1 (tab 9).

DISCUSSION

The present systematic literature review is the first phase in the development of a Core Outcome Set to assess physical activity interventions for people living with dementia. It identified 133 outcome domains, measured by 267 different measurement tools. Ahead of the establishment of the Core Outcome Set, this review provides valuable information for researchers designing studies investigating physical activity interventions for people with dementia. Researchers may choose to focus either on the most frequently reported outcomes and measurement tools, enabling the comparison of novel interventions against the current evidence, or explore outcomes where research is still lacking.

Functional abilities and independence (n=69), was frequently measured by the Barthel Index (n=12) and the Timed Up And Go (n=12). This outcome domain was reported in all stages of dementia and by all stakeholder groups (patients and carers, health professionals and researchers). A recent Cochrane review highlighted the positive influence of physical activity on this outcome domain (Forbes et al., 2015). The available international guidance also directs professionals to promote independence in people with dementia through physical activity (National Institute for Health and Care Excellence, 2006;Forbes et al., 2015;Miskovski, 2014).

Global cognitive function (n=65), was most commonly measured by the Mini-Mental State Examination (n=43), but with inconclusive results. Researchers have hypothesised that physical activity improves cognition, through physiological mechanisms, such as an increase in blood supply to the brain, synaptogenesis and reduced inflammation (Lautenschlager et al., 2012), nevertheless some uncertainty remains about the impact of physical activity on cognition. A recent Cochrane review was inconclusive in supporting this hypothesis, due to the important heterogeneity of the included studies (Forbes et al., 2015). Qualitative studies reporting the perspectives of patients also reflect uncertainly about the impact on cognition, with some patients reporting they have been able to "think more deeply" (Duignan et al., 2009) as a result of physical activity, whilst others reported no changes in this domain (Yu et al., 2014).

Balance (n=43), measured using Berg Balance Scale in 15 studies, was linked to a reduction in the risk of falling in 12 studies (Yao et al., 2013;Thurm et al., 2011;Yao et al., 2008;Padala et al., 2012;Cadore et al., 2014;Arcoverde et al., 2014;de Andrade et al., 2013;Pedroso et al., 2012;Santana-Sosa et al., 2008;Hernandez et al., 2010;Suttanon et al., 2012b;Ferretti et al., 2014). This is important due to the well-known health and economic burden of falls in older people (National Institute for Health and Care Excellence, 2013). Nevertheless, "Balance" and "Falls risk" are often "researcher led" outcome domains; only one qualitative study (with carers) (Yao et al., 2008) identified "Balance", and none reported a reduction in "Falls risk" as desirable outcomes. Examples of outcomes reported by participants as meaningful include "Sense of self", "Social interaction", "Fatigue management" and "Enjoyment". These outcomes are yet to be objectively measured in quantitative studies.

Global behavioural symptoms of dementia (n=42) was measured using the Neuropsychiatric Inventory in 13 studies. Recent literature has correlated the presence of behavioural symptoms with quality of life in people with dementia (Beerens et al., 2013;Mjorud et al., 2014), carer burden (Reed et al., 2014) and perceived reasons for institutionalisation (Stephan et al., 2014). The presence of these symptoms was also linked to an increased risk of psychotropic medication misuse (Cerejeira et al., 2012), hence the importance of managing behavioural symptoms through non-pharmacological interventions, such as physical activity (Thune-Boyle et al., 2012). However, a recent Cochrane review found limited evidence for the effectiveness of physical activity in this domain (Forbes et al., 2015), with some of the included studies showing conflicting results or positive results not maintained post-intervention. Future studies using this outcome should report the duration of the impact on these symptoms.

Health related quality of life (n=40), most frequently measured using the Quality Of Life In Alzheimer's Disease Questionnaire (n=19), concurs with the guidance given by the National Institute for Health and Care Excellence on promoting activities that people living with dementia enjoy (National Institute for Health and Care Excellence, 2006). Better quality of life can also be linked to a decrease in depression, also prevalent among people living with dementia (Curran and Loi, 2012). The availability of measures of quality of life suitable for completion by a proxy may also account for its common use in research.

Outcomes where research is lacking

Very little is known regarding effects of physical activity for people with severe dementia. Research is lacking in relation to multiple settings and numerous outcome domains, namely “Mobility”, “Health related quality of life”, “Social interaction” and all the outcome domains related to carers. Carer related outcomes deserve particular attention in future research due to the known correlation between severity of cognitive impairment and carer burden (Reed et al., 2014).

Heterogeneous measurement tools

In line with previous literature reviews (Forbes et al., 2015;Rao et al., 2014), substantial heterogeneity was identified in the use of measurement tools. It is important to emphasise that whilst “the three most commonly used measurement tools” per outcome domain were identified, the psychometric properties of the instruments were not considered. The most appropriate measurement tools for each of the outcomes included in the Core Outcome Set will be determined in the final phase of its development.

Limitations

Several limitations need to be acknowledged. Due to limited time and resources, language filters (English, Portuguese and Spanish) were added to the initial searches. The inclusion of papers written in three different languages implies that a large proportion of the available literature would have been screened. Yet, the number of papers missed due to the use of the language filter is unknown. The data analysis process was conducted by a single researcher. Whilst it is possible that other researchers would have defined the outcome domains differently, this risk was minimised by presenting appendix 1, to ensure transparency of the process.

CONCLUSION

Research into physical activity for people living with dementia is common and necessary. However the considerable heterogeneity in the outcomes sought and the tools used hinders the development of definitive clinical guidance, reinforcing the need for a Core Outcome Set. The present systematic literature review not only forms the first phase of development of a Core Outcome Set, but also offers useful information for interventions being currently designed. “Functional abilities and independence” is the outcome domain i) most frequently reported across stages of dementia; ii) most frequently shown to improve; and iii) reported as important by patients and carers as well as health professionals and researchers. Thus, clinicians and researchers are encouraged to continue to evaluate this outcome domain when developing Physical activity interventions for people with dementia. Other frequently reported outcomes are “Global cognitive function”, “Balance”, “Global behavioural symptoms of dementia” and “Health related quality of life”, but the evidence of benefit is less conclusive in these domains.

CONFLICTS OF INTEREST

The authors have no conflicts of interest to declare.

4.2 Paper III - Outcomes of Physical Activity for People Living with Dementia: Qualitative Study to Inform a Core Outcome Set

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ABSTRACT

Background: The need for a Core Outcome Set to evaluate physical activity interventions for people living with dementia, across stages of disease and intervention settings has been established. This qualitative study precedes the consensus phase of developing this Core Outcome Set and aims to: (i) compare the outcomes identified by patients, carers and professionals to those previously reported in the literature; (ii) and understand "why" certain outcomes are considered important.

Design and Methods: In-depth semi-structured interviews were conducted with people living with dementia, family carers and professionals (n=29). The outcomes identified in the interviews were mapped to a list of outcomes reported in a recent literature review. An in-depth thematic analysis was conducted to understand the importance of physical activity in dementia care.

Results: A comprehensive, inductively derived list of 77 outcomes, common across stages of dementia and intervention setting, was put together for the consensus phase of this Core Outcome Set: ten of these were new outcomes generated by this qualitative study. Five themes explained why stakeholders perceived physical activity outcomes as important for people living with dementia: "being well and staying well", "having a role towards others", "maintaining identity", "being connected to the present" and "delivering good quality care".

Conclusion: Ten new outcomes of physical activity, not previously reported in recent literature, were identified. Physical activity is considered important to people living with dementia due to its positive impact on multiple health outcomes for both patients and carers.

Contribution of the paper:

- Qualitative methods ensure relevant outcomes are not missed in Core Outcome Sets.
- Physical activity can enable a constant adaptation of self-identity to patients and carers as the dementia progresses and may bring positive outcomes to those living with severe dementia and/or comorbidities.
- Physical activity for people living with dementia is linked to good quality care, overall wellness, living in the present and being connected with others.

Key Words: Physical activity; Dementia; Core Outcome Set; Qualitative study.

INTRODUCTION

Dementia is a major cause of dependency and disability among older people, resulting in increased burden on individuals, carers and health care services (World Health Organization, 2015a). The need for research into interventions to improve care of people living with dementia is evident (Alzheimer's Society, 2013b). Physical activity, defined by the World Health Organization as “any bodily movement produced by skeletal muscles that requires energy expenditure”, may be one such intervention. Despite the large number of available studies, guidance on the ideal dose of physical activity for people living with dementia is lacking. A recent literature review (Gonçalves et al., 2018a) identified substantial heterogeneity of outcomes reported in physical activity research for people living with dementia. This is one factor limiting meta-analyses and the establishment of robust conclusions in this field. The development of a Core Outcome Set – a minimum set of outcomes to be measured across trials of a particular intervention and/or health condition (Williamson et al., 2017) – has therefore been recommended for physical activity interventions for people with dementia (Gonçalves et al., 2018a). This Core Outcome Set has been designed to be applicable across stages of disease progression and intervention settings (Gonçalves et al., 2018a) to maximise homogeneity in this field of research, knowing that specific outcomes for different stages of disease, types of dementia or activity settings can still be measured by researchers, in addition to those recommended by the Core Outcome Set (Williamson et al., 2017).

A gold standard for Core Outcome Set development is yet to be defined. The inclusion of qualitative methods preceding the consensus phase of Core Outcome Set development, represents a methodological innovation considered particularly necessary when research in the field of the Core Outcome Set lacks sufficient qualitative work (Keeley et al., 2016). Most qualitative studies about physical activity for people with dementia are linked to a specific intervention and are aimed at exploring feasibility, barriers and facilitators to that intervention (Khoo et al., 2014;Malthouse and Fox, 2014;Suttanon et al., 2012a;Yu and Swartwood, 2012;McCaffrey et al., 2014). The few available qualitative papers, not linked to a specific intervention, include only care home settings (Guzman-Garcia et al., 2013;Harmer and Orrell, 2008) or people living with early stages of dementia (Soderhamn et al., 2014;Cedervall et al., 2015) and do not meet the needs of a Core Outcome Set applicable across intervention settings, types of dementia and stages of the condition. Furthermore, these qualitative studies are not centred on intervention outcomes, and therefore many outcomes, relevant to participants, may have been missed during data collection.

Views of multiple stakeholders - patients, carers and professionals - may facilitate the selection of meaningful outcomes for this Core Outcome Set, leading to the implementation of tailored interventions, in research and clinical practice, as recommended in dementia research and policy (National Institute for Health and Care Excellence, 2006).

The present qualitative study aimed to inform the next stage (a consensus study) of a Core Outcome Set to evaluate physical activity interventions for people with dementia by (i) comparing the outcomes identified by professionals, carers and patients against those previously described in the literature and (ii) gaining a deeper understanding of “why” certain outcomes are considered important in dementia care.

DESIGN AND METHODS

The present study was reported according to the Consolidated Criteria for Reporting Qualitative Research (Tong et al., 2007).

Registration and Ethical Approval

This study is part of the development of a Core Outcome Set, registered with the COMET initiative. Ethical approval was obtained from the ethics committee at the Faculty Health Sciences, University of Southampton (ethics number: 19524). All participants provided informed consent prior to taking part in this study.

Recruitment and Patient and Public Involvement

Recruitment visits were made to support groups, exercise groups and dementia related activities and events. Posters were made available in public spaces (e.g. libraries, churches, social centres) and flyers distributed via gatekeepers in charities and support groups. The study poster and a short description of this research was made available through websites and newsletters of professional organisations. Participants interested in taking part contacted the research team using the contact details provided on posters and flyers. Information about the study was provided to potential participants (verbally and in a writing via the participant information sheet). A date for interview was only arranged with those who wanted to participate. A phone call was made the day before the interview to offer any further clarifications and confirm the interview date. Formal consent was recorded on the day of the interview.

This study benefited from patient and public involvement during recruitment and data analysis. A former carer of a person living with dementia, who also had a role as a leader in a carer support group, assisted the team to identify local dementia services and advised on strategies to approach gatekeepers in those organisations. This strategy had a very positive impact on recruitment for this study. She also contributed to the data analysis, by discussing findings with the authors as analysis progressed.

Inclusion criteria

The following participants were included:

- People with a diagnosis of dementia, capacity to consent to take part in research at the time of data collection and able to communicate verbally in English;
- Family members who were currently involved or had previously been involved in the care of a relative living with dementia;
- Professionals who have been involved in promoting, designing or supporting physical activity for people with dementia.

Exclusion criteria

People living with dementia were excluded if they had been bedridden since before the diagnosis. Family members were excluded if they were not in contact with the person with dementia since their diagnosis. Professionals were excluded if their professional role in dementia care was undertaken exclusively outside of the United Kingdom. Exclusion criteria were discussed with those who expressed an interest in taking part.

Sampling

Purposive sampling was used, aiming for a maximum variation strategy (Palinkas et al., 2013) across stakeholder groups, using the following criteria:

- Stage of disease progression, determined by the Mini-Mental State Examination score, completed by the first author (ACG - interviewer) immediately prior to the interview: "Mild to moderate" – 17 to 26; "Moderate to severe" - 10 to 16 and "Severe" – less than 10 (Folsteins et al., 1975);
- Type of dementia;
- Activity setting (i.e. community, care or nursing homes, assisted living; hospital);
- Levels of physical activity, determined by the score on the Physical Activity Scale for the Elderly (Washburn et al., 1993), completed immediately before the interview, with input from the carer; Scores range from zero (no physical activity completed in the previous seven days) to 693.9 (maximum physical activity level score);
- Socioeconomic deprivation, determined by the Index of Multiple Deprivation (Department of Communities and Local Government, 2015), using the participants' post code, with scores ranging from 1 (most deprived) to 10 (least deprived).

Recruitment aimed to reach data saturation, which was defined as the point where new data being collected made little or no difference to the codes being generated from the data analysis (Guest, 2006).

Data collection

In-depth semi-structured interviews were conducted and audio-recorded by the first author [ACG], a female clinical doctoral research fellow and physiotherapist with previous experience of conducting qualitative studies and, at the time of data collection, was a senior clinician in an Older Person's Mental Health in-patient service, including a dementia ward. Participants were aware of the professional background of the interviewer. Reflective notes were written after each interview.

The interview topic guide[9] was designed to enable participants to discuss outcomes of physical activity and explore the importance of such outcomes. Professionals and carers were interviewed face-to-face or remotely (via telephone or video call), according to their preference. People living with dementia were interviewed face-to-face, in a private venue of their choice, and encouraged to have a relative or friend with them at all times. During the interviews with people living with dementia, the following strategies were used, based on previous research (Quinn, 2017), and the interviewer's [ACG] experience of communicating with people with living with dementia: asking simple questions, giving time for the person to answer, rephrasing the question with help from the carer, maintaining good eye contact, reading body language for signs of fatigue or distress, offering breaks and choosing the time and venue for the interview according to the person's needs. These strategies also facilitated the capacity assessment. Capacity to consent to research was assessed based on the British Psychological Society guidance (Dobson, 2008). When present, the carer helped the researcher to explain the study to the person with dementia, providing all possible opportunities to make an informed decision. Carers were also encouraged to note any signs of discomfort or distress, which may indicate the need to stop the interview.

Data analysis

To address the aims of this study, two separate types of analyses were completed. Both analysis' steps were completed predominantly by the first author, with regular guidance and in consultation with other authors.

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[9] Available in Appendix A of this thesis document.

Aim 1: Interviews were transcribed verbatim and analysed thematically (Braun and Clarke, 2013). The transcripts were not returned to the participants for comments or corrections. Initial inductive (data driven) and semantic codes (close to the participants' own words) were used across the data set, to identify potential outcomes of physical activity. An outcome was defined as "a perceived consequence or impact, positive or negative, for people living with dementia, their carers or health services, whether intended or incidental, arising from undertaking physical activity". Codes with equivalent semantic meanings were merged and grouped into higher level codes (outcome domains). At this stage, a deductive approach was used, and the outcome domains were compared, by two independent researchers [ACG and a researcher independent from this research team], against the outcome domains previously reported in the literature (Gonçalves et al., 2018a). A merged final list of outcome domains was created and the scope of each outcome domain defined.

Aim 2: Outcome domains were thematically organised, by identifying patterns and connections between outcomes that could be described by a higher level theme (Braun and Clarke, 2013). Each theme represented a meaningful concept to answer the question: "why is physical activity important to people living with dementia?" The final data analysis was presented back to four participants for member checking (Birt et al., 2016).

RESULTS

Participant Characteristics

Interviews were conducted with 29 participants (5 people living with dementia; 14 carers and 10 professionals). The average interview duration was 67 minutes, ranging from 35 to 101 minutes. Table 1 describes the sample characteristics. Previous research suggests that interviews involving people in severe stages of dementia can be triangulated with interviews of their carers (Beuscher and Grando, 2009). When it was not possible to gather the views of the person living with dementia, the carer was interviewed instead, aiming to get as close as possible to the views of those at the later stages of dementia. In this study, nine of the carers interviewed, were carers of people living with severe dementia and/or people with dementia who were unable to communicate verbally or provide consent.

Table 1- Sample characterisation.

Stakeholder group 1: people with dementia and their carers (n=19)	**Mean (SD) or n (%) People living with dementia**	**Mean (SD) or n (%) Carers**
Role		
People living with dementia	5 (26%)	-
Spouses	-	6 (32%)
Adult children or children in law	-	8 (42%)
Demographics		
Age (years)	75 (11.8)	62 (12.4)
Gender (female)	3 (60%)	10 (71%)
Levels of physical activity	58.5 (74.0)	-
Mini Mental State Examination	19.2 (3.9)	-
Index of Multiple Deprivation	7.8 (1.9)	6 (2.7)
Experience of doing or supporting physical activity in which of the following setting(s)		
Community	3 (60%)	9 (64%)
Care or nursing home	2 (40%)	2 (14%)
Assisted living	0 (0%)	4 (29%)
Hospital	0 (0%)	2 (14%)
Living with the following stage of disease progression or having experience of supporting physical activity for a relative through the following stage(s) of disease progression		
Mild to moderate	4 (80%)	6 (43%)
Moderate to severe	1 (20%)	0 (0%)
All stages	-	7 (50%)
Not known	-	1 (7%)
Type of dementia		
Alzheimer's disease	2 (40%)	9 (64%)
Mixed dementia	1 (20%)	1 (7%)
Vascular dementia	1 (20%)	1 (7%)
Lewy body dementia	0 (0%)	2 (14%)
Young onset Alzheimer's disease	1 (20%)	0 (0%)
Not Known	0 (0%)	1 (7%)
Stakeholder group 2: professionals (n=10)	**Mean (SD) or n (%)**	
Role		
Physiotherapists	4 (40%)	
Occupational Therapists	2 (20%)	
Activity coordinators	1 (10%)	
Charities	2 (20%)	
Private companies	1 (10%)	
Involved in delivering research	1 (10%)	
Demographics		
Age (years)	43 (10.1)	
Gender (female)	5 (50%)	
Professional setting		
Community	7 (70%)	
Care or nursing home	7 (70%)	
Hospital	4 (40%)	
Experience in care for people living with dementia per stage of disease progression		
Mild to moderate dementia	1 (10%)	
All stages of dementia	9 (90%)	

Aim 1: Comparing outcome domains generated by this study, against those found in the literature.

A final list of 77 outcome domains (60 positive/beneficial) was generated. Figure 1 illustrates the process of defining this list of outcome domains.

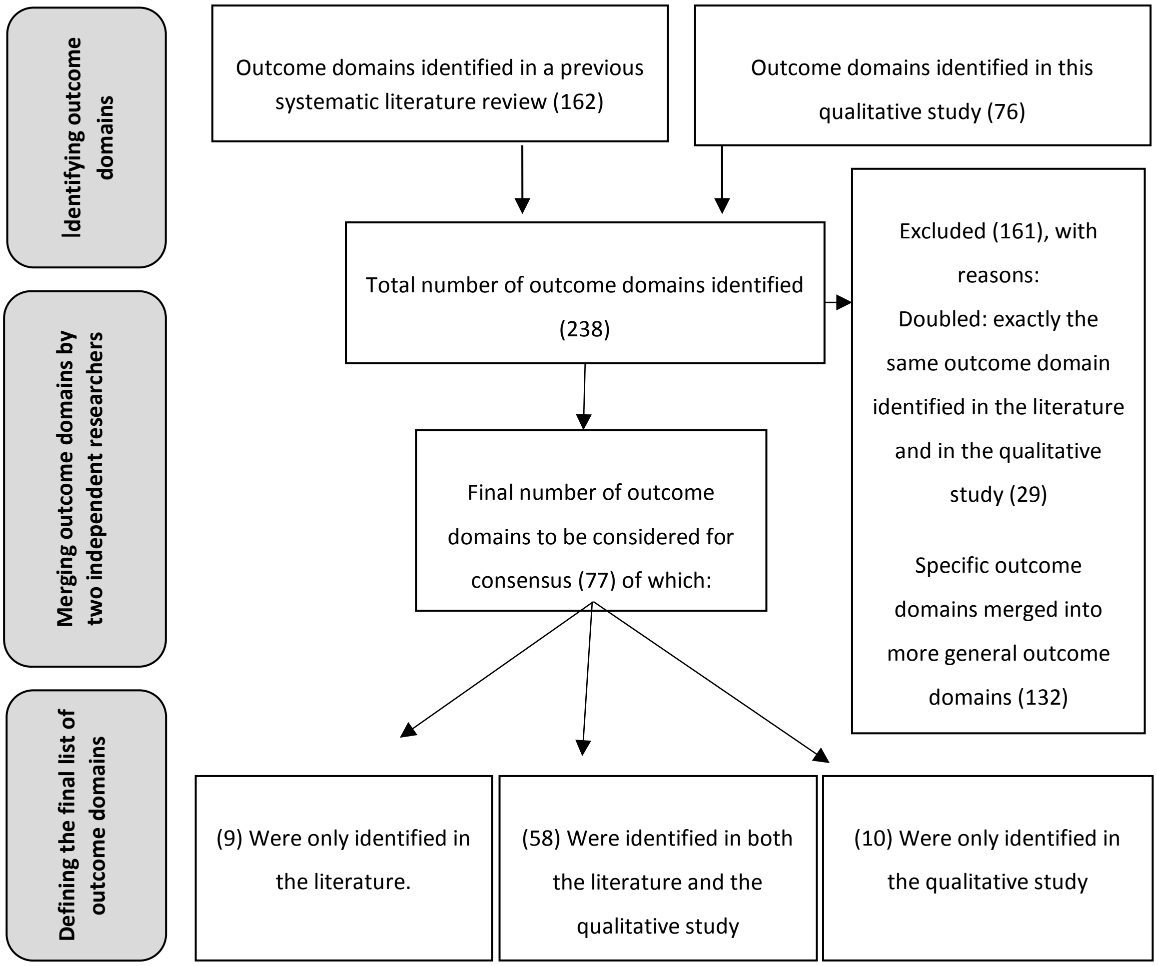

Figure 1- Flowchart of the development of final list of outcome domains to be considered in the next phase: consensus study.

Of the final 77 outcome domains, 10 (8 positive/beneficial) were new outcomes generated by this qualitative study and not reported in the literature; nine (4 positive/beneficial) had been identified previously but were not mentioned by participants in this study. Tables 2a and 2b highlight these outcome domains presented per stakeholder group, stage of disease progression and activity setting. An extended version of these tables including all 77 outcome domains, which will be considered in the consensus phase, can be found in supplementary material[10].

Most outcome domains (69 out of 77) were reported by both stakeholder groups, in multiple settings and dementia stages (Tables 2a, 2b and supplementary material). Data saturation was reached in both stakeholder groups, with no new outcome domains being generated by the last professional interview and only two new outcomes generated in the last patient/carer interview.

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[10] This supplementary file available from the journal website, and can be found in Appendix B of this thesis document.

Table 2a – NEW Outcome domains identified in this qualitative study, but not previously identified in the literature (n=10). Legend: *“prof. and researchers” - professionals and researchers; “patients and carers” – people living with dementia and their informal carers or relatives. Boxes identified with "x" indicate that the outcome domain has been identified in that context. Boxes identified with “-“ indicate that an outcome domain has not been identified in that particular context.*

Outcome domain identified:	**Scope**	**Stakeholder**		**Stage**			**Setting**		
		Prof. and researchers	**Patients and carers**	**Mild to moderate**	**Moderate to severe**	**Severe**	**Community**	**Institution**	**Hospital**
Positive outcome domains (n=8):									
Improving posture	Includes sitting and standing posture.	x	x	x	x	x	x	-	x
Managing lower limb oedema	Fluid retention in the lower limbs.	-	x	-	-	x	x	-	x
Managing pressure ulcers	Preventing pressure ulcers and recovering skin integrity.	x	x	x	x	x	x	-	x
Preventing incontinence	Regularly using the toilet, by mobilising to the toilet or commode instead of using a pad.	-	x	-	-	x	x	-	x
Carer developing friendships and peer support	Family carers meeting other relatives of people living with dementia and developing a network of friendship and peer support.	x	x	x	x	x	x	-	x
Meeting individual needs	Meeting individual needs of the person living with dementia. These includes identity, physical, spiritual,	x	x	x	x	x	x	x	x

Outcome domain identified:	Scope	Stakeholder		Stage			Setting		
		Prof. and researchers	Patients and carers	Mild to moderate	Moderate to severe	Severe	Community	Institution	Hospital
of patient and family	emotional and social needs. Meeting tailored needs of families.								
Becoming interested and gaining interest of others	Gaining the interest of the person living with dementia and the interest of staff, formal carers and relatives. Getting relatives and formal carers to know the person with dementia and becoming interested in what they can do.	x	x	x	x	x	x	x	x
Reframing and revisiting the person with dementia	Recovering one's "old self" even if for brief moments. Relatives having moments of seeing the "old" person with dementia by seeing them active as they were before the diagnosis. Relatives and staff or formal carers being surprised by the ability of the person living with dementia. Using activity to highlight what the person with dementia can still do.	x	x	x	x	x	x	x	-
Negative outcome domains or risks of physical activity (n=2):									
Eating or drinking something harmful	Eating or drinking something that should not be ingested, by confusing it with real food or drink.	x	x	x	x	x	x	-	-
Not being able to sleep after an activity	This was linked to possible changes of environment or routine caused by the activity (e.g. coming back to care after having been on holiday).	-	x	-	x	x	-	x	-

Table 2b - Outcome domains measured and referred to in the literature, but not identified in this qualitative study (n=9). Legend: *"prof. and researchers" - professionals and researchers; "patients and carers" – people living with dementia and their informal carers or relatives. Boxes identified with "x" indicate that the outcome domain has been identified in that context. Boxes identified with "-" indicate that an outcome domain has not been identified in that particular context.*

Outcome domain identified:	**Scope**	**Stakeholder**		**Stage**			**Setting**		
		Prof. and researchers	**Patients and carers**	**Mild to moderate**	**Moderate to severe**	**Severe**	**Community**	**Institution**	**Hospital**
Positive outcome domains (n=4)									
Carer mobility	Carer mobility and balance, ability to walk with less joint pain.	x	x	x	x	x	x	-	-
Carer mood	Simply described as "carer mood".	x	-	-	x	-	x	-	-
Carer quality of life	Carer wellbeing and quality of life. At times linked to carer burden.	x	-	x	x	x	x	-	x
Carer sleep	Improved carer sleep quality.	x	-	-	x	-	x	-	-
Negative outcome domains or risks of physical activity (n=5)									
Being admitted to hospital or experiencing a deterioration in health	Being hospitalized or admitted to an emergency department. Experiencing a deterioration of overall health or becoming too unwell to continue activity.	x	-	x	x	x	x	x	-
Discovering cysts	Ganglion cyst.	x	-	x	-	-	x	-	-

Outcome domain identified:	Scope	Stakeholder		Stage			Setting		
		Prof. and researchers	Patients and carers	Mild to moderate	Moderate to severe	Severe	Community	Institution	Hospital
Feeling dizzy, nauseous or fainting	Feeling dizzy, nauseous or light-headed. Having a syncopal episode.	x	-	x	x	x	x	-	-
Increasing mortality	Number of deaths during activity or within the time period while the person with dementia was a participant in an activity intervention.	x	-	x	x	x	x	x	-
Moving into care	Being placed into an institution for permanent full-time care.	x	-	x	x	x	x	-	-

Aim 2: Understanding why physical activity is important

Five core themes explained the importance of physical activity. All themes were described across stages of disease, intervention settings, and incorporated the views of both stakeholder groups (professionals and patients/carers). There were no opposing views between stakeholders. The generated themes are described below.

Being well and staying well

Participants across all stakeholder groups reported using physical activity to improve or maintain multiple aspects of health and wellbeing of the person living with dementia, keeping an overall state of wellness - physical, cognitive and emotional - despite the diagnosis of dementia.

> *"Mental and physical health. I mean as a starting, as a baseline point. You are better, you are in better health, the more you get to do it. (…) Well, I mean physical stamina, strength, these things support everything. Body and mind are connected. (…) It is promoting health. It promotes health across the board." Occupational therapist (All stages - Care homes).*

> *"It gets me out of my routine, because (…) if I was at home, and things like that, it would drive me up the wall." Person living with dementia (mild to moderate stages – home).*

The data demonstrated that potential positive health benefits of physical activity for people living with dementia are often influenced by other factors: participants weighed up the benefits against the burden that physical activity may generate for people living with dementia; and the extent to which supporting physical activity was beneficial or detrimental to carers. All stakeholder groups also expressed concerns about the potential for increased physical activity increasing the risk of falls:

> *"ACG: would anything stop you from walking? Participant: NO! No. (…) Unless you fall flat on your face." Person living with dementia (Moderate to severe stages – home)*

The negative effects or burden of physical activity were not only related to accidents and injuries but also to a range of emotional and behavioural negative effects, such as becoming agitated or distressed, feeling exhausted or not being able to sleep after the activity.

> *"ACG: You said you stopped the ball game because (…) she was aggressive…" Participant: "It wasn't an aggression as in it didn't cause harm to anyone, but the way she was kicking. It was a bit forceful, with quite a grimaced face so that would demonstrate to me that she wasn't particularly happy about this task." Occupational Therapist (All stages – Hospital).*

Carers also reported on the burden of supporting activity: increased workload from keeping the person living with dementia active, while managing their own health; and perceived negative emotions associated with the activity, such as responsibility, boredom, guilt and worry.

> *"To be honest I think it was like a tedious kind of thing to do [walking with her mother up and down the corridor] because it was very slow and… I mean but it did feel like it was a good thing to do. I did feel it was like a good thing to do. But I did find it a little bit tedious. And I would feel quite anxious that mum might fall and she was anxious! And I would be very tired as well by this stage…" Daughter (Severe stages – Community).*

Conversely, it was suggested that carers also had positive experiences of keeping the person with dementia active, such as bonding with the person living with dementia during the activity, having quality time together, developing support networks with peers and improving their own health.

> *"I think that it is quite bonding because I think they are learning more about their family." Activity coordinator (All stages - Nursing home).*

In addition, carers could see their burden of care being alleviated by witnessing an improved behaviour from the person with dementia and ensuring their functional independence.

> *"I think we hadn't really thought ahead, to when mum would lose her mobility… what would we do then? Or how would we cope? So it seemed very important [to keep her mobile]" Daughter (Severe stages – Community).*

Maintaining independence was not only considered important for managing carer burden, but also key for people living with dementia:

> *"Well, you need exercise, don't you? Otherwise you just would just tighten up and wouldn't be able to do things, surely". Person living with dementia (Mild to moderate stages – Care home).*

When supported by others, activity was also an opportunity to give family carers a break.

"In respect to the fact that I know John is somewhere doing something he enjoys, that is independent of me, he is not reliant on me for that at all, I don't have a huge part in it, he has his own friends down there, he has something that is entirely his. And it is a form of activity. It gives me free time…" Wife (Mild to moderate stages – Community).

Maintaining identity

This theme related to a continuity of identity that was achieved through activity, by enabling people living with dementia to do what they are passionate about. Activity was described by patients, carers and professionals, as a means by which the person with dementia could stay in control, keep life as "normal as possible" and minimise the disruption caused by the diagnosis of dementia.

"I suppose it is important… it sort of, as far as you can, just carry on! You know, it might not be as normal but, just do what you can! As long as you can!" Person living with dementia (Mild to moderate stages – Community).

Having a role towards others

Activity was also reported to be used as a source of social

benefits; fighting loneliness and isolation.

"Because I imagine it can be a very lonely existence [to live with dementia], and if, if through sport in general, or whatever, that can be improved, that can only be fantastic." Private company (All stages – Community).

Activity generated benefits such as feeling included, gaining a sense of belonging to groups, teams and being active members of society. It also allowed people with dementia to maintain or assume new roles, giving meaning to a new stage of life. It offered people with dementia the opportunity of being interested in a cause and feeling valued, by gaining interest from others.

"That's why I came here [became a volunteer in a gardening centre]. Just to be able to help them (…), because I know what it is like (…) and now I understand how the team gets together and do it.

> *You know what I mean? It is great!" Person living with dementia (Mild to moderate stages – Community).*

Being connected to the present

While the diagnosis of dementia was described by participants as taking the person away to a difficult-to-access "dementia world", activity was described as an anchor to the present. It is a way of retrieving positive memories and emotions to the present and "living in the moment". Whilst active, people with dementia aimed to have a fulfilling time, filled with opportunities and new experiences. "Being active" was seen as having a positive, proactive attitude of wanting to be alive and wanting to fight the hopelessness of the diagnosis.

> *"So, so yeah if you've got, if you have just been diagnosed I would say: try not to be put off by the bigger picture. (…) And try and still do things. (…) Do things that they are used to do. But differently. So sort of fight the dementia if you like. Not fight against it, because you can't. Hopefully we will get a cure one day, but actually still do things in spite of it if you like." Daughter (All stages – Community and Care home).*

> *"Well, because it [gardening] gets me to stay out and being alive." Person living with dementia (Mild to moderate stages – home).*

Delivering good quality care

For those providing formal or informal care, delivering activity that people living with dementia could adhere to was considered part of delivering "good care". It was considered "good" because it was cost-effective and specialised for dementia. Participants also reported that providing activity promotes a positive reputation for institutional settings.

> *"From the perspective of the retirement community, (…) I think they liked the fact that it reflected well on them, that they were doing this, so they (…) had it permanently on their website and things like that." Private company (All stages - Community and residential homes).*

Providing activity also promoted staff satisfaction and retention. From the perspective of family carers, the provision of physical activity helped them feel that they had chosen the right care for their loved one.

> *For the carers [live-in carers, privately funded by relatives], you know, our job is to keep the carers content. If we can keep them happy we can keep them. And changing a live-in carer is a challenge as you can imagine. Son (Stage not known – Community).*

DISCUSSION

This qualitative study precedes the consensus phase of developing a Core Outcome Set to be used to evaluate physical activity interventions for people with dementia, and represents an innovative methodological approach in Core Outcome Set development. To our knowledge this is also the first qualitative study exploring outcomes of physical activity across dementia types and stages of disease progression, and first qualitative study being published as a pre-consensus phase of a Core Outcome Set. A list of 77 outcome domains, including 10 novel outcome domains, was developed. Most outcome domains were relevant across stakeholders, settings and stages of disease progression, and were organised under five themes: “Being well and staying well”, “Maintaining identity”, “Having a role towards with others”, “Being connected to the present” and “Delivering good quality care”. This study makes two distinct contributions to knowledge: it informs the development of Core Outcome Set methodology; provides new in-depth knowledge about the importance of physical activity in dementia care.

Implications for Core Outcome Set methodology

A total of 10 outcome domains would have been missed in the consensus phase, had this qualitative study not been undertaken. This is a surprisingly high number of outcome domains, as it is often reported that qualitative studies do not add any outcomes to those already in the literature (Harman et al., 2015).

In line with other Core Outcome Sets for progressive conditions, it was anticipated that the consensus study would be subdivided according to the different stages of disease progression (Gonçalves et al., 2018b). However, our findings indicate that outcomes of physical activity overlap greatly across settings and stages of dementia. The initial protocol (Gonçalves et al., 2018b) was therefore changed to a single consensus survey. The potential for qualitative findings to inform the structure and design of a Core Outcome Set had not been previously identified (Keeley et al., 2016), and it should be taken into account by future Core Outcome Set developers.

Contributions to knowledge about relevant effects of physical activity

The new outcomes “carer developing friends and getting support”; “meeting individual needs of patient and family”; “becoming interested and gaining interest from others”; “reframing and revisiting the person with dementia” (Table 2a) indicate the important role that physical activity has in supporting people living with dementia and their carers in both adapting to the diagnosis and as dementia progresses. The use of physical activity to maintain the identity as that of “someone active” despite the diagnosis is explained in this study, supporting findings from previous qualitative work (Cedervall et al., 2015). However, the use of activity to embrace new roles (as shown, by the new outcome domains “reframing the person with dementia” and “carer making friends and getting support”) is a new perspective, indicating that physical activity can be used as a tool, not only to maintain, but also to adapt a sense of self-identity, as dementia progresses. The concept of exercise identity was shown to be key in a recent model of exercise participation for people with chronic conditions, highlighting how patients choose to exercise according to their age and gender identity, social and cultural norms (Pentecost and Taket, 2011). Physical activity can therefore be used by people living with dementia, as in other chronic conditions, to continuously (re)frame their identity, while living with a progressive condition.

The remaining new outcomes generated by this study reflect potential physical needs of people, either at later stages of dementia, or of those with multiple comorbidities: “improving posture”, “preventing incontinence”, “managing lower limb oedema” and “managing pressure ulcers”. The fact that these are new outcomes, demonstrates the lack of research into physical activity for people with severe dementia (Gonçalves et al., 2018a) and provides new insights into the potential that physical activity may have across stages of disease and for addressing the multi-morbidity that often accompanies the progression of dementia (All-Party Parliamentary Group on Dementia, 2016).

The present study highlights how physical activity interventions can support families living with dementia from early diagnosis and throughout their dementia journeys; a key message in dementia care guidelines and policies (Department of Health, 2015).

Strengths and Limitations

In-depth interviews were a challenging method for patients at later stages of dementia or those with limited verbal communication, leading to a sample with more carers than people living with dementia, and a relatively small sample of people living with dementia. This is a potential limitation because this Core Outcome Set aims to be applicable to physical activity interventions including people with dementia in all stages of the condition, and with no restrictions in terms of communication abilities. Including a higher proportion of carers was a successful strategy to gather the views of those closest to people living with late stage dementia. The present study also informs the next stage of the development of this Core Outcome Set (a consensus study), where a larger sample of people living with dementia will be recruited.

The identification of outcomes from the interviews was conducted by one researcher only. To minimise this potential limitation, the analysis benefited from patient and public involvement through regular reflections with a former carer of someone living with dementia as the data analysis progressed; the final analysis findings were also ‘member checked’ by four participants (Birt et al., 2016). The process of merging the list of outcomes from this qualitative study with the outcomes in the literature was conducted independently by two researchers, increasing the rigor of this stage of data analysis.

CONCLUSION

This qualitative study makes an important contribution to Core Outcome Set methodology. By adding 10 outcome domains to the list previously generated from published literature it has demonstrated the important role qualitative methods can play in ensuring outcomes meaningful to patients, carers and professionals are not missed. This study also highlights the value of physical activity across the stages of dementia progression. Whilst confirming findings from previous studies that physical activity can contribute to “Being well and staying well” and “Delivering good quality care” it has also identified an important new finding that physical activity is important in the process of adapting to the diagnosis through “Maintaining identity”, “Having a role towards and with others” and “Being connected to the present”.

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CONFLICTS OF INTEREST

None to declare.

4.2.1 Reflection on the concept of saturation, post publication of paper III

Paper III reports on saturation being reached, as an indicator of the completion of data collection and a justification for the sample size utilised in this qualitative study. However, the concept of saturation in qualitative research can be complex and controversial (Saunders et al., 2017) and therefore, it deserves further reflection on how it was used in this paper, and what role it may have in the design of qualitative studies that are part of Core outcome Sets.

Saturation was first described in grounded theory and consisted of a point in which no new data was being found to further describe or understand a category within the theory (Glaser and Strauss, 1967). Reaching to this stage would give the researchers, developing a theory, enough confidence that a category within a theory is rich, in-depth and well explained and thus, further data collection may not be needed to understand that category, and the analysis can proceed to the next stages (Glaser and Strauss, 1967). The concept of saturation has since been defined and used in a variety of other ways, by different methods within qualitative research, and arguably, misused as a stamp of quality within qualitative work. Recently, a small body of researchers has critically analysed the concept of saturation and made recommendations for its use (Nelson, 2017;Saunders et al., 2017;Morse, 2015). Four models of saturation have been described: i) theoretical saturation, the closest to the original definition, in which saturation means that no new data is being found to further develop a category within a theory; ii) inductive thematic saturation, in which new interviews do not generate new codes or themes; iii) *a priori* thematic saturation, which is used in deductive studies and it is demonstrated when all themes from a pre-defined theory have been covered by the collected data; and lastly iv) data saturation, which is used at the point of data collection (rather than analysis, unlike the other models) and is based on the researcher’s perception of redundant or repeated remarks being made by participants during interviews or focus groups (Saunders et al., 2017). Of these four models, the inductive thematic saturation is the most suitable to describe the use of the concept of saturation in paper III. It is therefore worth looking into this model in greater detail.

Inductive thematic saturation relates to the non-generation of new themes or codes by new moments of data collection. This model of saturation has been criticised for focusing on the generation of new codes and themes, rather than the depth of the analysis within each of those themes. This critique is particularly relevant when saturation is considered in relation to codes, more than themes, as it means saturation can be claimed at an early stage of the analysis (based on initial codes) and therefore, losing meaning as the analysis progresses (Hennink et al., 2017). It may be seen as controversial that paper III used saturation at a code level to determine sample size. As reported in the results section of paper III, saturation was considered “achieved” as “no new outcome domains [were] generated by the last professional interview and only two new outcomes [were] generated in the last patient/carer interview”. In response to arguments critiquing definitions of saturation at a code level, it is relevant to relate back to the primary aim of the study. Paper III, and other qualitative studies that precede Delphi processes in Core Outcome Set development aim to generate a list of outcomes, which are identified at a code level. If the aim of the qualitative study is to complement a “long list of outcomes” the method used to determine the sample size, and when to stop data collection, should reflect the aim of the study. It was later demonstrated in Chapter 5 (paper IV) that the initial list of outcomes generated at the end of paper III was in fact comprehensive, as no new positive outcomes were suggested by participants in the first round of the Delphi survey. This may indicate that saturation had been approached (rather than “reached”, as explained in the next paragraph) in paper III, and the sample size had been appropriately determined.

The Inductive thematic saturation model described here has its limitations and complexities, and care is advised to Core Outcome Set developers with regards to claims made about saturation. Firstly, saturation is recommended to be used in light of the aim of the study. If the aim of the qualitative study also includes understanding why certain outcomes are important (as it was the case in paper III), the use of inductive thematic saturation at a theme level (rather than at a code level) could arguably be equally appropriate – it is up to the developers’ judgement as to which is the main goal of the study and that should guide saturation: the list of codes (outcomes); or the depth of understanding about each of the outcomes (themes). Secondly, as argued by Saunders et al. (2017), saturation is a process, not a single point in time. Authors are recommended to refer to degrees of saturation, rather than saturation being reached, or not reached. These aspects of saturation (saturation at theme versus code level; and saturation as a process rather than a single point in time) should have been made clearer in paper III and it is suggested that they are taken into account by other core outcome set developers. Lately, saturation may be used in parallel to other indictors to inform sample size (Saunders et al., 2017). In Paper III, a maximum variation sampling strategy was used alongside saturation, to ensure representation from a variety of stakeholders in the generation of the long list of outcomes. This approach was considered appropriate in the context of Core Outcome Set development, and it may be used by other developers looking to use qualitative methods in the pre-consensus phase of Core Outcome Sets.

Chapter 5 Reaching consensus: what to measure

5.1 Paper IV- A Core Outcome Set to evaluate physical activity interventions for people living with dementia

"This is a pre-copyedited, author-produced version of an article accepted for publication in The Gerontologist, following peer review. The version of record Gonçalves, A.C..; Samuel, D.; Ramsay, M.; Demain, S.; and Marques, A. 2019. The Gerontologist, 100, is available online at: https://doi.org/10.1093/geront/gnz100

ABSTRACT

Background and objectives: A Core Outcome Set to evaluate physical activity interventions for people living with dementia is needed, as the development of guidelines is currently limited by important heterogeneity in this field of research. Development of Core Outcome Sets often includes Delphi surveys, but people living with dementia are often excluded. This study aimed to reach consensus on this Core Outcome Set using a modified Delphi survey to enable the participation of people living with dementia.

Research Design and Methods: Two stakeholders groups took part in a Delphi survey (group 1: people living with dementia and family caregivers; group 2: professionals from different backgrounds, including physiotherapists, occupational therapists and researchers). Caregivers and professionals completed the survey remotely. Participants living with dementia took part face-to-face, using a card sorting strategy. The consensus process was finalised with a consensus meeting.

Results: 95 participants of both groups completed the modified Delphi. Of those, 11 attended the consensus meeting. The card sorting strategy was successful at including people living with dementia. Seven outcomes reached consensus: preventing falls; doing what you can do; staying healthy and fit; walking better, being able to stand up and climb stairs; feeling brighter; enjoying the moment; and, feeling useful and having a purpose.

Discussion and implications: Robust and innovative methodological strategies were used to reach a consensus on a Core Outcome Set (what to measure) to evaluate physical activity for people living with dementia. Future work will focus on the selection of the most appropriate tools to measure these outcomes (how to measure).

Key words: Exercise; Alzheimer’s disease; What to measure; Delphi

INTRODUCTION

Dementia is estimated to affect 46.8 million people worldwide (Alzheimer's Disease International, 2015). A progressive and debilitating disease, dementia impacts those living with the condition, their carers and health care providers (Alzheimer's Disease International, 2015;Pinzon et al., 2013;Reed et al., 2014). Physical activity, defined by the World Health Organization as “any body movement produced by skeletal muscles that requires energy expenditure” is often recommended for people living with dementia (Miskovski, 2014;National Institute for Health and Care Excellence, 2006). Potential benefits include improvements in independence for functional activities of daily living, physical performance and executive function (Forbes et al., 2015;Ohman et al., 2016;Rao et al., 2014). However, the precise impact of physical activity, including the ideal dose to maximise benefits for people living with dementia, is not yet known. Recent systematic reviews have reached limited conclusions due to heterogeneity in the selection and reporting of outcomes (Rao et al., 2014;Forbes et al., 2015). Consequently, no guidelines are currently available for physical activity in this population, and health professionals are limited in the advice they can provide. Physical activity interventions are therefore not used to maximum benefit due to the lack of concrete guidance on ideal dose and expected outcomes. The development of a Core Outcome Set for use in research evaluating physical activity interventions for people living with dementia has therefore been recommended (Gonçalves et al., 2018a).

A Core Outcome Set is an agreed minimum set of outcomes to be measured and reported in clinical trials of a particular health condition and/or intervention (Kirkham et al., 2016), with a view to allow studies to be comparable and fast-track guidance to practice. Core Outcome Set development has increased substantially in recent years across a wide range of conditions (Gargon et al., 2018). Core Outcome Set development includes a consensus process to agree which outcomes should be included (i.e., what to measure) (Kirkham et al., 2016). Delphi surveys are commonly used by Core Outcome Set developers as a method to achieve consensus (Gargon et al., 2014), as they allow remote and anonymous participation from stakeholders (Keeney et al., 2011). While multiple variations of the Delphi method have been reported in the literature, a Delphi always involves several survey rounds where participants are asked to score or rank a list of items, while receiving feedback on the scores of peers in previous rounds (Keeney et al., 2011;McMillan et al., 2016). The inclusion of patients, in Core Outcome Set development - in this case, people living with dementia - is considered vital, as they bring a unique perspective about living with the condition. If they are not included, the Core Outcome Set risks missing important outcomes relevant to patient care (Sinha et al., 2011;Young and Bagley, 2016). People living with dementia experience a decline in cognitive function, affecting their ability to learn and retain new information as well as their ability to maintain attention levels, reason, judgment and make decisions (Baudic et al., 2006;Capucho and Brucki, 2011;Lafleche and Albert, 1995;Storandt, 2008). They may therefore find it difficult to fully engage in a traditional Delphi survey, which may be why several previous Delphi surveys about aspects of dementia care did not include people living with dementia in their expert panels (Annear et al., 2015;Page et al., 2015;Patterson et al., 2016;Verkade et al., 2010).

The present study aimed to first reach consensus on a Core Outcome Set applicable to effectiveness trials and clinical practice to evaluate physical activity interventions in people living with dementia, across different stages of disease progression and activity settings. Second, it aimed to develop an innovative adaptation to the Delphi method, using a card sorting strategy, to ensure full participation of people living with dementia in the consensus process.

DESIGN AND METHODS

The development of this Core Outcome Set followed the standards for reporting Core Outcome Sets provided by the COS-STAR statement (Kirkham et al., 2016) and this consensus study meets the agreed reporting guidelines for Delphi studies (Diamond et al., 2014;Sinha et al., 2011).

Registration and ethical approval

This Core Outcome Set was registered in the COMET initiative database (http://www.comet-initiative.org/studies/details/708?result=true) and was approved by the ethics committee at the Faculty of Environmental and Life Sciences, University of Southampton (ethics number: 19542). A protocol for the development of this Core Outcome Set, including this consensus phase, has been published elsewhere (Gonçalves et al., 2018b). All included participants had capacity to consent to taking part in research at the time of data collection, and provided formal written consent.

Participants and recruitment

Two stakeholder groups were considered key to the development of this Core Outcome Set.

Group 1: People living with dementia and their informal carers (relatives or friends) were recruited in the United Kingdom. Adverts and research information activities were conducted through gatekeepers of charities providing support to people living with dementia and their carers, and via the "Join Dementia Research" database. Potential participants were considered for inclusion if they contacted the research team expressing an interest in taking part.

Group 2: Professionals (e.g., physiotherapists, occupational therapists, nurses, formal carers etc.) involved in the design or delivery of physical activity interventions for people living with dementia, in research and in practice, were also recruited. Professional organisations in different countries (e.g. universities, physiotherapy associations, dementia and ageing related charities) disseminated information about this study and potential participants made contact with the research team, if interested in taking part.

The ideal sample size for a Delphi study has not been identified, with studies using sample sizes ranging from five to over 1000. However, Akins et al. (2005) compared the response characteristics of an homogenous group of 23 participants against computer generated samples of 1000 and 2000 participants and found all responses to be stable, concluding that small and homogenous groups of Delphi participants (e.g., n=23) can generate reliable responses (Akins et al., 2005). The present study aimed to include 40 participants in each stakeholder group. This was considered an appropriate number as whilst the group was homogeneous (all participants were required to have experience of physical activity in dementia, as a person living with dementia or carer; or a professional) some intra-group variation was sought, to allow representation of all stages of dementia, a range of dementia types, professional backgrounds and activity settings. A snowball recruitment strategy (Valerio et al., 2016) was used for both groups. In this recruitment strategy, participants were asked to share the link to the survey, and/or the contact details of the research team, with peers who may be interested in taking part. The use of this strategy meant that some participants may have been aware of the identity of some of their peers who were also part of the Delphi, making it a quasi-anonymised survey.

Inclusion and exclusion criteria

Group 1: Carers were included if they had current or previous experience of supporting someone living with dementia doing any form of physical activity, at any stage of their disease progression and in any setting. Carers included spouses, friends or adult children. Those unable to understand written English were excluded.

People living with dementia were included if they had a known diagnosis of dementia (self-reported), capacity to consent to take part in research (assessed immediately prior to data collection and following guidance from the British Psychological Society (Dobson, 2008)) and were able to communicate verbally in English. People living with dementia provided written consent prior to data collection, and verbal consent throughout data collection. They were included from three counties in the United Kingdom: Dorset, Hampshire and Sussex. Those who had been bedridden since before the diagnosis or unable to complete the survey, due to their cognitive impairment, were excluded.

Group 2: Professionals were included if they had any experience in the design or delivery of any type of physical activity interventions for people living with dementia, in any stage or setting, and in any country in the world. Professionals were excluded if they were unable to understand written English.

Delphi survey: Round one

The first round of a Delphi survey traditionally consists of open ended questions, asking participants to generate an initial list of topics (in this case, outcome domains) to be agreed upon in subsequent survey rounds (Keeney et al., 2011). The present study used a modified Delphi approach: the 50 outcome domains (referred to as “outcomes”) presented to participants in round one of this Delphi survey were identified through a systematic literature review (Gonçalves et al., 2018a) and a qualitative study previously conducted with both stakeholder groups (Gonçalves et al., 2019a). Patient and public involvement consultations were critical to the design of this study. These identified that prioritisation from such a long list (n=50) would be too cognitively demanding for people with dementia, regardless of the method used to allow their participation. A decision was therefore taken to further modify the original Delphi method, i.e., not include people living with dementia until round two, when the number of outcomes to be prioritised was reduced. This compromise was considered acceptable because people living with dementia had been pivot to the long list development through their participation in the qualitative study (Gonçalves et al., 2019a).

In round one, participants (carers and professionals) completed the survey remotely, either online using SurveyGizmo software, or using a paper format, with pre-paid envelopes distributed with the surveys. Consent was provided with the return of the surveys. The online and paper surveys were identical and prompted participants to select up to nine outcomes they considered most important, from the initial list of 50 outcomes. Participants were also asked to suggest any additional outcomes. The selection of up to nine outcomes was based on Core Outcome Set development recommendations to include no more than nine outcomes in the final Core Outcome Set, in order to promote its applicability (Boers et al., 2014a). To minimise the length of the survey, participants were not asked to provide a justification for their choices. A glossary, with definitions for each of the outcomes was available with all surveys.

Delphi survey: Round two

In round two, the list of outcomes was presented in order from the most to the least frequently selected during round one. The percentage of all participants selecting each individual outcome was also shown. Each participant was reminded of the outcomes ($n \leq 9$) they had personally selected in round one. Participants from round one were sent the survey for round two, in the same format they had used to complete round one (either paper or online). A booklet providing more detailed results from round one, such as percentages of agreement per stakeholder group, and an updated glossary document (both available on request), were also made available with the round two survey. Participants were asked to consider the results from round one before re-selecting the outcomes (up to nine) from this list they considered most important.

People living with dementia took part in round two, using a face-to-face card sorting strategy. The same principles described above for the round two survey were followed. People living with dementia were shown the outcomes in order, from the most to the least frequently selected in round one, but instead of presenting the outcomes in a list format, the outcomes were presented as a pile of cards. Each card had a written and pictorial representation of one outcome. Participants living with dementia were asked to consider one card at a time, and decide if the outcome was “very important” or “not so important”. After going through the entire pile, the cards in the “not so important” pile were excluded and those in the “very important” pile were counted. If they were nine or less, the survey ended. If they were more than nine the same process was repeated, until a pile with nine or less outcomes was achieved. Flowcharts illustrating this process, and photographic examples of cards, can be found in “supplementary material A”[11]. All face-to-face sessions were audio recorded and participants were asked to “think aloud” as they completed the task. In the same session, people living with dementia completed the Mini-

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[11] This supplementary material can be found in Appendix C of this thesis document.

Mental State Examination (Folsteins et al., 1975) and the Physical Activity Scale for the Elderly (Washburn et al., 1993), with support from a carer, for purposes of sample characterization only.

Selection of outcomes and definition of consensus

Consensus was defined a priori, as previously published in the study protocol (Gonçalves et al., 2018b). At the end of round one, outcomes selected by ≤15% of all participants were eliminated except for those that had been mentioned by a person living with dementia during the qualitative study that informed the list of outcomes in round one (Gonçalves et al., 2019a). This exception ensured that outcomes potentially relevant to those living with dementia were not inadvertently excluded during round one, in which only professionals and carers participated.

After round two, outcomes selected as one of the top nine priorities by ≥70% **of all participants** or ≥80% of participants **of one stakeholder group** were included, without further discussion, in the final Core Outcome Set. Outcomes selected by ≤15% of all participants were permanently excluded. Any remaining outcomes (voted between 16 and 69% of all participants) were taken for further discussion at a consensus meeting. At the consensus meeting, only outcomes with a minimum agreement of 90% were include in the final Core Outcome Set (Gonçalves et al., 2018b).

Consensus meeting

Patient and public representatives advised on a sample size of up to 15, so that the meeting remained dementia friendly. In order to minimize bias in the selection of participants attending the consensus meeting whilst also controlling the sample size, all round two survey participants living within a 60 mile radius of the meeting venue, and that were not housebound, were invited to attend. Potential participants were sent the results of the Delphi round two alongside their invitation to the consensus meeting. Invitations were sent out three weeks before the meeting and a reminder sent to non-respondents one week prior to the meeting. Purposive sampling was used to ensure both groups were represented, as well as more than one activity setting, and more than one stage of disease progression.

The meeting was facilitated by the first author, who prompted both groups to present arguments towards the inclusion or exclusion of each of the outcomes. The discussion on each outcome was followed by a vote. In the voting of each outcome, participants were asked to raise their hand if they wanted to include the outcome that had just been discussed. The meeting was audio recorded.

Patient and public involvement

This study benefited from close advice of a carer research partner and co-author [MR]. She was involved in the recruitment, design of participant documentation, revision of the Delphi survey before it was distributed and planning of the consensus meeting, to ensure full participation from people living with dementia and their carers. Additional patient and public involvement was used in the following activities:

- Use of lay terminology during the Delphi:

A carer, a professional who is also a family carer, and a member of a dementia related charity were involved in the process of renaming the outcomes into lay terms, before they were used in the Delphi round one. They also advised on the glossary that accompanied the Delphi surveys.

- Design of the cards for the card sorting strategy:

A patient support group and two carers' groups were consulted during the design of the cards used by people living with dementia in the Delphi round two. Design advice consisted of: the size of the cards (15x10cm), the use of pale yellow as a background colour for the cards, the use of photographs of people (rather than icons), and not using photographs with background (e.g. one person shopping with a shopping trolley, but without a busy shop as a background). Advice was also received on showing only one card at a time to participants living with dementia.

RESULTS

Delphi Participants' characteristics

In round one, a total of 44 carers (14 male) and 47 professionals (10 male) completed the survey. Of those, 82% of the carers (n=36; 12 male) and 83% of professionals (n=39; 10 male) also completed round two. A total of 20 people living with dementia (10 male; with Mini Mental State Examination scores ranging from 9 to 28; mean score: 21.1 ±5.3) joined the round two survey. See table 1 for detailed demographic characterization of all participants in round two.

Table 1 - Characteristics of participants who completed the Delphi round two.[12]

Group 1: People living with dementia and family carers (n=56)	**Mean (SD) or n (%)**	**Group 2: Professionals (n=39)**	**Mean (SD) or n (%)**
Role		*Role*	
People living with dementia	20 (35.7%)	Physiotherapists	14 (35.9%)
Family carers	36 (64.3%)	Occupational Therapists	6 (15.4%)
		Rehabilitation Assistants	4 (10.3%)
		Nurses	1 (2.6%)
		Members of charities or volunteer organisations	7 (17.9%)
		Social Workers	1 (2.6%)
		Health Care Support Workers	1 (2.6%)
		Academics	8 (20.5%)
Demographics			
Age (Years)		Age (Years)	
18-29	1 (1.8%)	18-29	8 (20.5%)
30-39	0 (0%)	30-39	9 (23.1%)
40-49	2 (3.6%)	40-49	6 (15.4%)
50-59	12 (21.4%)	50-59	12 (30.8%)
60-69	10 (17.9%)	60-69	3 (7.7%)
70-79	18 (32.1%)	70-79	1 (2.6%)
80-89	7 (12.5%)	80-89	
90-99	6 (10.7%)	90-99	
Gender (Female)	34 (60.7%)	Gender (Female)	29 (74.4%)
Years post diagnosis	6.1 (±4.1)	Years of experience in dementia care	9.1 (±6.3)
Physical Activity Scale for the elderly (people living with dementia only)	52.0 (±45.1)		
Living with or supporting people living the following dementia types (self-reported)			
Alzheimer's Disease	23 (41.1%)		
Vascular Dementia	8 (14.3%)		
Mixed Dementia	16 (28.6%)		
Dementia with Lewy Bodies	2 (3.6%)		
Frontotemporal Dementia	1 (1.8%)		
Not known	6 (10.7%)		
Experience of doing or supporting physical activity per stage of disease progression (self-reported by carers and according to Mini-Mental stage examination score for people living with dementia)		*Experience of supporting physical activity for people living with dementia per stage of disease progression*	

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[12] A table with the sample characteristics of those who dropped out after the round one Delphi is available as supplementary material to this publication, and can be found in Appendix C, in this thesis document.

Group 1: People living with dementia and family carers (n=56)	**Mean (SD) or n (%)**	**Group 2: Professionals (n=39)**	**Mean (SD) or n (%)**
Mild to Moderate	27 (48.2%)	Mild to Moderate	8 (20.5%)
Moderate to Severe	16 (28.5%)	Moderate to Severe	7 (17.9%)
Severe	1 (1.8%)	Severe	2 (5.1%)
All stages	11 (19.6%)	All stages	26 (66.7%)
Not known	2 (3.6%)	Not known	0 (0%)
Mini Mental state examination scores (people living with dementia only)	21.1 (±5.3)		
Experience of doing or supporting physical activity per setting		Experience in supporting physical activity per setting	
Home or other community settings	52 (92.9%)	Home or other community settings	35 (89.7%)
Sheltered accommodation	6 (10.7%)	Sheltered accommodation	8 (20.5%)
Care or nursing home or assisted living	18 (32.1%)	Care or nursing home or assisted living	22 (56.4%)
Hospital	2 (3.6%)	Hospital	24 (61.5%)

Legend: Please note that one participant may have supported people living with dementia in multiple settings and stages of disease progression. A total of 44 carers and 47 professionals had completed the previous round of the Delphi survey.

Usability of the card sorting strategy

The card sorting strategy was feasible for enabling people living with dementia to prioritise outcomes. Only one person living with dementia (Mini Mental State Examination score: 19) was excluded for not being able to complete the card sorting task. The session audio-recordings showed that participants were making thoughtful choices and fully understood the process of sorting the cards, as illustrated in the quotes below.

Participant number 8, (male, living with dementia at home, Mini Mental State Examination score: 27) mentioned how he found the process easy to understand. He was also always aware that the goal was to reduce the pile of cards to up to nine:

> *ACG: “We are going to organise them [cards] into “very important” and “not so important””. P8 - “So we will make a pile”. ACG: “yes, that’s right”. P8: I see, this is very high tech research [laughing]. Well, I like it. I can understand it.”*

> *P8: "Well, I am a one vote man [referring to be putting all first five cards on the "very important pile"]. But I understand this is not a practical situation because at some point I will have to choose".*

Participant 11, (male, living in a care home, Mini Mental State Examination score: 11), found it difficult to read the cards accurately, but when the cards were read to him, he would carefully weigh his decision of whether to include or exclude an outcome.

> *ACG, showing and reading the card: "feeling less depressed and avoiding depression". P11: "That is definitely on my top nine". ACG: "Ok. What about this one "slowing down the dementia"".P11: can we put that on the side before I decide on that one?" ACG: "That's a good plan."*

Participants also often referred to their own experiences and preferences while selecting outcomes, demonstrating that their choices were meaningful and well thought through.

> *"Feeling useful and having a purpose. Well, I like to feel useful, so that is very important [to me]." (Participant 17, female, living in assisted living accommodation,* Mini Mental State Examination *score: 22).*

All participants were able to complete the task without help from their carers (although in some cases participants felt reassured by the presence of a relative or friend during the session). On average, each face-to-face card sorting session took 30 minutes, ranging from 17 to 43 minutes.

Consensus meeting: characteristics of the expert panel

Of those who completed round two, 69 participants (37 carers, 18 people living with dementia and 14 professionals) met the inclusion criteria and were invited to attend the consensus meeting. A total of 11 participants were available to take part on the selected date. Among those who attended the meeting, seven were carers (two male, five had experience of supporting someone living with dementia at home, or in sheltered accommodation, and four in a care home setting); one person living with dementia, supported by her husband (living in their own home) and three health care professionals (all female, one nurse, one occupational therapist and one rehabilitation assistant, with experience of supporting people living with dementia in community and in hospital settings). The 11 participants self-declared having experience of living with, or supporting people living with all stages of dementia progression and a variety of dementia types, including Alzheimer's disease (n=4), Dementia with Lewy Bodies (n=2), Mixed Dementia (n=2) and vascular dementia (n=1).

Selection of outcomes and recommendations from the expert panel at the consensus meeting

The process of selection of outcomes from Delphi round one through to the consensus meeting is illustrated in figure 1.

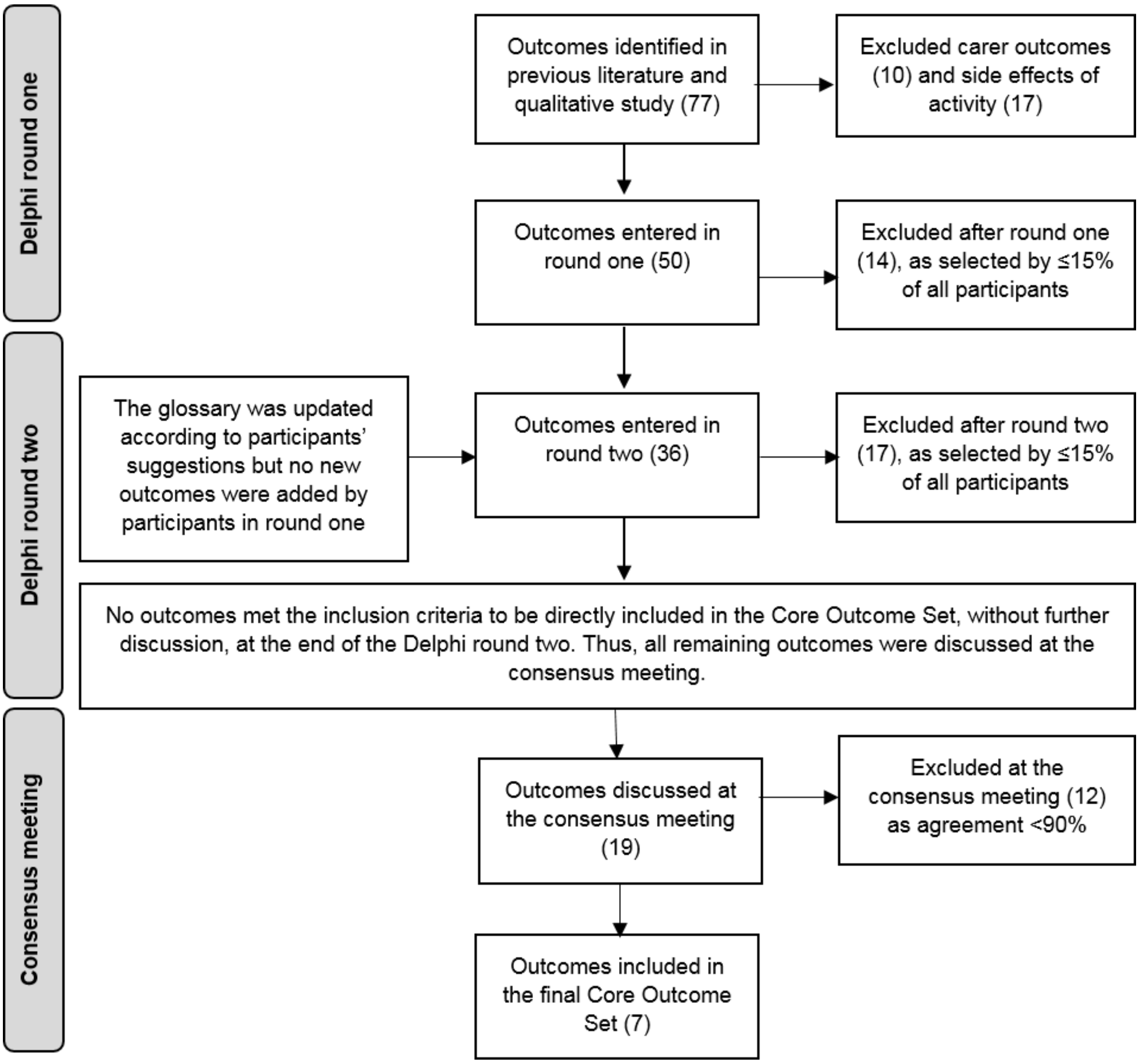

Figure 1- Flowchart of the development of final Core Outcome Set to evaluate physical activity in people living with dementia.

The list of all outcomes considered in both Delphi rounds and the consensus meeting is available in “Supplementary material B”[13]. At the end of round two of the Delphi survey, 31 of the 50 outcomes had been selected by less than 15% of all participants and were therefore excluded. However, none of the outcomes met the criteria to be directly included in the Core Outcome Set without further discussion (selected by ≥70% of all participants or ≥80% of participants of one group). Thus, all remaining outcomes (n=19) were discussed at the consensus meeting. Consultation with members of patient and public group revealed that a consensus meeting including the discussion of 19 outcomes would be feasible. At the consensus meeting, a total of seven outcomes met the definition of consensus and were included in the final Core Outcome Set: preventing falls; doing what you can do; staying healthy and fit; walking better, being able to stand up and climb stairs; feeling brighter; enjoying the moment; and feeling useful and having a purpose. Table 2 presents further details on these seven outcomes, including definitions and the breakdown of the percentages of agreement per participant group and per stakeholder within each group. In addition to these seven outcomes, participants at the consensus meeting also felt the need to make two specific recommendations for intervention delivery. See table 2 for further details.

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[13] This supplementary material can be found in Appendix C of this thesis document.

Table 2 – Core Outcome Set to evaluate physical activity interventions for people living with dementia. At the consensus meeting, the percentage of agreement was 100% to all the outcomes included in this final Core Outcome Set.

Outcome in lay terms included in the final Core Outcome Set	**Definition, as in the glossary made available to the Delphi participants, and with corrections made by the expert panel at the consensus meeting.**	**Percentage of agreement: Delphi round two**
Preventing falls	Number of fallers; number of falls; number of falls sustained by one person living with dementia; health care costs associated with managing fall related injuries (e.g. fractures); falls risk, including balance and fear of falling.	All participants: 67.4% Group 1: 62.5% Carers: 63.9% People with dementia: 60% Group 2 (professionals): 74.4%
Walking better, being able to stand up and climb stairs	Keeping mobile or regaining mobility; keeping moving; being more mobile indoors and outdoors; standing up and walking up and down. Climbing stairs. Includes all aspects of gait, using less supportive walking aids and keeping the ability to change position. Keeping movement in the joints.	All participants: 56.8% Group 1: 51.8% Carers: 63.9% People with dementia: 30% Group 2 (professionals): 61.4%
Staying healthy and fit	Overall health, general ability or performance. Being fit, strong and ready for activity. Staying well and having less medical diagnoses.	All participants: 25.3% Group 1: 19.4% Carers: 39.3% People with dementia: 75% Group 2 (professionals): 5.1%
Doing what you can do	Using one’s physical being. Using all available skills and abilities. Expressing a physical self. Seeking movement as a routine part of life.	All participants: 17.9% Group 1: 23.2% Carers: 16.7% People with dementia: 35% Group 2 (professionals): 10.3%

Outcome in lay terms included in the final Core Outcome Set	Definition, as in the glossary made available to the Delphi participants, and with corrections made by the expert panel at the consensus meeting.	Percentage of agreement: Delphi round two
Feeling useful and having a purpose	Feeling useful and having a role or purpose. Sense of belonging and of being included. Being part of a family, team or a group. Being a volunteer. Helping family, neighbours and peers. Being occupied with meaningful activity. Doing what one is passionate about and having something to look forward to.	All participants: 51.6% Group 1: 44.6% Carers: 44.4% People with dementia: 45% Group 2 (professionals): 61.5%
Feeling brighter	Lifting mood or being in good mood; also referred to as "improving mood levels", feeling content, bright, and happy or having fun. Having a positive attitude. Showing fewer negative emotions. Note: To be measured immediately post activity.	All participants: 18.9% Group 1: 21.4% Carers: 22.2% People with dementia: 20% Group 2 (professionals): 15.4%
Enjoying the moment	Being anchored to the present by activity. Learning or doing something new. Living in the moment. Having an adventure. Not having to think about anything else. *Having moments of joy.*[14] Note: To be measured during activity.	All participants: 38.9% Group 1: 33.9% Carers: 50% People with dementia: 5% Group 2 (professionals): 46.2%
Additional recommendations by the expert panel at the consensus meeting:		
Seeing the person, not the dementia	Although not voted as a core outcome of physical activity, the panel recommended that all activities should be designed and delivered based on the principle that people living with dementia are seen as people and not a diagnosis, and interventions should be tailored to the individual, who should be always seen as person.	
Reducing anxiety	Reducing anxiety was not voted as a potential benefit of physical activity, but the panel recommended it should be measured as a potential side effect. Activities should focus on improving the mood of the person living with dementia ("feeling brighter") but "anxiety" should be monitored as a counterweight. It is accepted that an activity will not necessarily make "everyone feel brighter" and therefore it is important to monitor and report on possible increases in anxiety to weigh up the benefit of the activity on mood.	

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[14] *Overlooked in the original publication. Added post publication to enhance content.*

DISCUSSION

This is the first study to identify a Core Outcome Set to evaluate physical activity interventions for people living with dementia, across activity settings and different stages of disease progression. A novel card sorting strategy was found to be feasible to include people living with dementia in the consensus process. Through the use of this innovative approach, people living with dementia, joined carers and professionals to reach consensus on seven outcomes: preventing falls; doing what you can do; staying healthy and fit; walking better, being able to stand up and climb stairs; feeling brighter; enjoying the moment; and feeling useful and having a purpose (see definitions in table 2).

“Staying healthy and fit” is a good example of the positive impact of the inclusion of patients in this Core Outcome Set. During the Delphi, this outcome was selected by 5% of the professionals and a contrasting 75% of people living with dementia. At the consensus meeting, the views of those with dementia were honoured and participants were unanimous on the inclusion of this outcome in the final Core Outcome Set. This is a key example, to add to others provided in the literature (Hewlett et al., 2005;Mease et al., 2008) of the importance of including patients in the development of Core Outcome Sets.

Other Core Outcome Sets, relating to a wide range of pathologies, often reach consensus on the outcome: “quality of life” (Allin et al., 2017;Haywood et al., 2014;Potter et al., 2015;Sinha et al., 2012). Quality of life is also known to be an important outcome for people living with dementia. However, previous literature has identified that people living with dementia particularly value specific constructs within the broader category of quality of life, such as the concept of “feeling useful” (de Boer et al., 2007). In line with these findings, the present Core Outcome Set includes three specific and non-overlapping outcomes, related to quality of life, but not “quality of life” as whole: “Feeling useful and having a purpose”, “Enjoying the moment” and “Feeling brighter”. The selection of these specific outcomes shows how the present Core Outcome Set represents what truly matters to patients, as well as to those who care for them.

Clinicians may also be aware of the ICHOM, which has produced a Standard Set of outcomes for dementia care (International Consortium for Health Outcomes Measurment, 2017). Standard Sets by ICHOM are sets of outcomes, encouraged to be reported in routine health care provision worldwide, allowing clinicians to collect data on the same outcomes and benchmark the services they provide. The ICHOM Standard Set for dementia includes the outcome “falls”, which is also an outcome included in this Core Outcome Set to evaluate physical activity. Therefore, if this Core Outcome Set is implemented in both research and clinical practice, as planned, professionals delivering physical activity interventions in clinical practice will be able to benchmark their intervention outcomes against not only research outcomes, but also other healthcare providers internationally.

The standard set provided by ICHOM also includes the outcome “Functional abilities and independence”. This outcome was also identified as the most frequently reported in research published in the last decade about physical activity for people living with dementia (Gonçalves et al., 2018a). However, “functional activities and independence” did not reach consensus in the present study. “Functional activities and independence” is a very broad outcome, including independence for basic activities of daily living such as personal hygiene, dressing, completing transfers and mobility, but also instrumental and more complex tasks such as managing finances (Mlinac and Feng, 2016). It is possible that within the specific context of physical activity, functional independence aspects specifically related to mobility (included in the present Core Outcome Set as “walking better, being able to stand up and climb stairs”) are more relevant. For instance, exercise is a well-established intervention for rehabilitation post hip fracture (Beaupre et al., 2013) and the Core Outcome Set for hip fracture trials, includes “mobility” as an outcome (Haywood et al., 2014). Thus, “mobility” may be a specific outcome of functional independence, possibly more relevant with regard to physical activity interventions.

Strengths and limitations

The development of this Core Outcome Set used innovative and robust methods of consensus and followed a pre-defined level of consensus. It also benefited from patient and public involvement and meaningful participation of people living with dementia in the consensus process, which is considered a priority in Core Outcome Set development (Sinha et al., 2011;Young and Bagley, 2016). This consensus study included a Delphi survey. Delphi surveys are known to face challenges with regards to low response rates (Keeney et al., 2011). Other published Core Outcome Sets report Delphi surveys with variable attrition rates between rounds one and two, ranging from two (McGrattan et al., 2018) to 50% (Chiarotto et al., 2015). Thus, the present study describes a two-round Delphi survey with low attrition rate between rounds, followed by a consensus meeting with meaningful and in-depth participation of both groups of stakeholders. The card sorting strategy described here as a Delphi adaptation provides future researchers in this field with a possible tool to enable prioritisation and decision making by people living with dementia. By enabling full participation of people living with dementia in defining their own priorities, this card sorting strategy is also in line with the current national and international agenda to give people living with dementia the opportunity to take part in research and actively influence their care (Curry, 2017;Department of Health, 2015).

Some limitations need to be acknowledged. The card sort-sorting strategy was developed to be as similar as possible to the remote survey completed by professionals and carers. However, the two approaches were not exactly the same. The card sorting strategy involves considering one outcome at a time (in contrast to seeing the whole list of outcomes). It also involved one-to-one interaction with the researcher. Future methodological research is recommended to compare results obtained from a card-sorting strategy with the remote version of the survey with carers and professionals. Only one person living with dementia was at the consensus meeting. All efforts were made to enable the participation of more people living with dementia, however reasons such as caring responsibilities, transport, decline in cognitive abilities since the Delphi round two and stigma of talking about living with dementia in public meant that all other participants living with dementia declined the invitation to take part. To overcome this limitation, some of the carers discussed the results booklet that had been sent with the invitation with their relative/friend with dementia before attending the consensus meeting, and used this information to inform their voting at the meeting.

This Core Outcome Set also had only limited representation from international stakeholders. While this may be seen as a limitation, it was an active choice to prioritise resources on the meaningful inclusion of people living with dementia and their carers, rather than the inclusion of an extensive international group of experts. Evidence is available for the need to include patients in Core Outcome Sets (Hewlett et al., 2005), yet to our knowledge, no previous studies have ascertained if a Core Outcome Set would be different or have a stronger uptake if developed internationally. Therefore, further work may be necessary to determine if this Core Outcome Set is applicable outside the United Kingdom.

Implications for policy, research and practice

The seven outcomes agreed to be core in physical activity interventions for people living with dementia can be used to guide the design and promotion of physical activity in this population, by clinicians, researchers and policy makers. For instance, the inclusion of elements of physical activity that can improve walking, standing and stairs and reduce falls are worth including.

The card sorting strategy presented in the current study can be used in future research, policy and practice, to help gather views and priorities of people living with dementia. Examples of the use of this strategy include consulting people living with dementia to determine the primary outcome of a research intervention, involving people living with dementia in setting research priorities in dementia care, and designing individual and person-centred care plans in clinical practice.

Conclusion

This innovative, robust and inclusive methodological approach has identified a Core Outcome Set of the outcomes that should be measured to determine the effectiveness of all physical activity interventions in people living with dementia, in research and clinical practice. This Core Outcome Set is designed for all types of dementia, at any stage of the disease and in hospital, community and care home settings. The seven outcomes identified by people with dementia, their informal carers and professionals to be included as a minimum were: preventing falls; doing what you can do; staying healthy and fit; walking better, being able to stand up and climb stairs; feeling brighter; enjoying the moment; feeling useful and having a purpose. Future work will include the identification of measurement tools for this set of outcomes (i.e., how to measure). A card sorting method, designed with robust patient and public consultation, successfully enabled the full participation of people living with dementia in the selection and prioritisation of meaningful outcomes.

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CONFLICTS OF INTEREST

We have no conflict of interest to declare.

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5.2 Paper V - Physical activity for people living with dementia: a prioritisation of carer outcomes and side effects from the perspectives of professionals and family carers

Gonçalves, A.C..; Demain, S.; Samuel, D.; and Marques, A. 2019. Submission being currently planned.

ABSTRACT

Background: Adherence of people living with dementia to physical activity is challenging, and largely dependent on carers' involvement. Carers are likely to support physical activity based on a balance of benefits to patients, carers and potential side effects. The present study aimed to identify the priorities of carers and professionals regarding 1) outcomes of physical activity for people living with dementia on carers 2) side effects on patients and carers.

Material and methods: This was a 2-round prioritisation exercise. In round one, participants were asked to rank, from most to least important, two lists of outcomes generated in a previous systematic review and qualitative study: i) 10 outcomes on carers; ii) 17 side effects on patients and carers. In round two, participants were asked to consider their own ranking in round one against the overall group ranking and re-rank both lists.

Results: 36 carers and 39 professionals completed both rounds. The carer outcomes ranked as highest priority were "carer feeling positive and satisfied"; "carer improving wellbeing" and "making lives of carers easier". The most undesirable side effects were "becoming agitated and confused", "falling over" and "feeling discomfort and pain".

Conclusion: Carers and professionals value the potential impact that physical activity for the person living with dementia may have in reducing carer burden. Behaviour and psychological symptoms, falls and pain are the most undesired side effects of physical activity. Future research should aim to address, and consistently report on these outcomes.

Key words: Physical activity, dementia, caregivers, adherence, side effects

INTRODUCTION

By 2050, dementia is predicted to affect 131.5 million people worldwide (Alzheimer's Disease International, 2015). In the absence of a cure, there is a need for interventions aimed at improving the care of those living with the condition (World Health Organization, 2015b). Physical activity is one such intervention, which has received increased research attention in the last decade (Gonçalves et al., 2018a), due to its promising benefits, including potential improvements in independence in activities of daily living (Forbes et al., 2015;Rao et al., 2014), balance (Zeng et al., 2016), physical performance (Rao et al., 2014) and carer burden (Zeng et al., 2016). However, due to impairments in cognition and possible loss of motivation (van Alphen et al., 2016a), professionals may find it challenging to promote and maintain adherence to physical activity in this patient group. People living with dementia are known to be more sedentary than their cognitively healthy peers (van Alphen et al., 2016b), and largely depend on their carers to engage in physical activity (van Alphen et al., 2016a). However, carers of people living with dementia experience higher carer burden than carers of people with other health conditions (Karg et al., 2018) and expecting them to organise and promote physical activity for their loved one may be unrealistic (van Alphen et al., 2016a). Previous research suggests that professionals need to focus on carer satisfaction whilst designing physical activity, in order to optimise adherence (Chung et al., 2008). It has also been noticed that carers' attitudes feelings and perceptions about physical activity are associated with patients' levels of physical activity (Kim et al., 2018). Therefore, from the perspective of carers, the perceived benefit and subsequent adherence to physical activity is likely to be a trade-off between potential benefits to patients, carers themselves and the possibility of negative side effects caused by physical activity.

A core set of positive outcomes of physical activity for people living with dementia has recently been established (Gonçalves et al., 2019b), including outcomes such as: "preventing falls", "enjoying the moment" and "staying healthy and fit". This core outcome set aims to increase consistency in the reporting of positive outcomes of physical activity for people living with dementia and thus fast-track intervention guidelines. However, the most important outcomes of physical activity interventions for people living with dementia on carers (e.g. carer improving wellbeing or making friends and getting support through the participation of the person living with dementia in physical activity) and the most undesired negative side effects of physical activity (e.g. becoming agitated or experiencing a fall while being active) have not yet been identified. This leaves negative side effects and carer outcomes at risk of being overlooked or inconsistently reported, limiting the inclusion of these important outcomes in literature reviews, meta-analyses and guidance to practice. The present study is a prioritisation exercise, aiming to supplement the already established core outcome set, by defining the priorities of carers and professionals regarding: 1) possible positive outcomes that physical activity for people living with dementia may have on carers and 2) negative side effects of physical activity on people living with dementia and/or their carers.

MATERIAL AND METHODS

Study design

This prioritisation exercise was nested in a Delphi survey, which aimed to determine a core set of positive patient outcomes to be measured in physical activity interventions for people living with dementia, across settings and stages of disease progression, and which findings have been published previously (Gonçalves et al., 2019b). A more detailed methodological description of this Delphi survey is available elsewhere (Gonçalves et al., 2018b;Gonçalves et al., 2019b). Including negative side effects and carer outcomes in the Delphi survey was not possible as this would increase the length of the survey and limit participation of people living with dementia. Therefore, alongside the Delphi consensus process, informal carers and professionals were presented with a list of possible negative side effects of physical activity for people living with dementia and their carers; and a list of possible positive outcomes that physical activity for the person living with dementia may have on their carers. These lists were generated from a systematic literature review (Gonçalves et al., 2018a) and a qualitative study (Gonçalves et al., 2019a). Carers and professionals were asked to rank both lists (from most to least important) in a 2-round iterative survey, which is described under “Survey design and data collection”.

Ethics approval

This study received ethical approval from the Ethics Committee of the Faculty of Health Sciences of the University of Southampton (ethics number: 19542). Informed consent was ascertained by the completion and return of the surveys.

Participants and recruitment

Two stakeholder groups were recruited: informal carers of people living with dementia (relatives or friends), referred to as “carers”; and professionals involved in the design, delivery and support of physical activity interventions for people living with dementia, in research and/or clinical practice. Carers were recruited from any location in the United Kingdom and they self-declared their role as informal carers, as well as the stage of disease progression of the person living with dementia they cared for, and the setting where physical activity took place. Professionals who were able to understand written English were recruited from any country in the world and also self-declared their role and experience in dementia care. Recruitment sought volunteers through dementia and carer related charities and support groups (e.g. Alzheimer’s Society; Carers in

Southampton), and through professional organisations (e.g. Chartered Society of Physiotherapy). Additionally, a snowball recruitment strategy was adopted (Valerio et al., 2016), where participants were asked to share the survey link or the contact details for the research team with peers who may also be interested in taking part.

The ideal sample size for prioritisation of health outcomes has not been defined. Previous prioritisation studies report sample sizes ranging from 26 (Sanderson et al., 2012) to 3000 (Duarte et al., 2007). In the current study, it was considered feasible to aim for a sample of 40 participants in each stakeholder group to complete the first survey round.

Survey design and data collection

The two lists of outcomes included: i) 10 positive outcomes that supporting physical activity for the person living with dementia may have on carers; ii) 17 negative side effects of physical activity for the person living with dementia and/or their carers. Patient and public representatives assisted the authors to write these outcomes in lay terms.

The survey was made available in paper (using pre-paid envelops for the return of the surveys) and electronic formats (using the SurveyGizmo software). The surveys in both formats were very similar. In the paper format, the participants were asked to use numbers to rank the outcomes (“1” being the most important). In the electronic format, participants were instructed to click and drag the outcomes in order (with the most important on top, equivalent to ranking position “1”). This was a forced ranking exercise and therefore two outcomes could not be ranked with the same number, or dragged and dropped in the same position.

Round one:

The first round of the survey included demographic questions for sample characterisation purposes, as well as the two lists of outcomes described above. The order in which outcomes were presented was randomised, as the order in which survey items are presented is known to influence participants’ choices (Brookes et al., 2018). There were 15 randomly ordered versions of the paper surveys that were distributed during recruitment, and the electronic version of the survey automatically randomised the order of the outcomes every time a new participant opened the survey link. In round one, participants were also asked to add any missing outcomes to either list. A glossary with the definitions of each of the outcomes in the survey was available to all participants.

Round two:

Participants who completed round one, were sent the round two survey in the format they had used to complete round one (either electronic or paper). In round two, outcomes were presented in ranked order from round one. Each participant was reminded of their own round one ranking order choices. New outcomes suggested in round one were added to the list and the glossary was updated accordingly. A detailed booklet showing how outcomes had been ranked per stakeholder group was also made available. The glossary, booklet and copy of the surveys are available on request. Participants were asked to consider the results from the previous round and re-rank the outcomes. This allowed participants to make an informed judgement regarding their final ranking, as the second round included new outcomes introduced by participants in round one, and each individual participant could compare their own priorities against those of other carers and professionals.

Data analysis

Descriptive statistics were used for demographic data. Ranking data was only considered from round two and therefore only data from participants who completed both survey rounds were included. For each outcome, all ranking positions were summed, and the outcome with the lowest overall ranking number was considered the most important. Non-ranked outcomes (left blank by the participants) were scored as: “number of outcomes plus one”.

RESULTS

Participant characteristics

A total of 75 participants (36 carers and 39 professionals) completed both survey rounds, out of 91 (44 carers and 47 professionals) who had completed round one. Of the participants who completed both rounds, all professionals completed their surveys electronically, whereas 15 carers chose to take part using the paper format. Carers (12 male) included 14 spouses, 17 adult children, four children in law, one grandchild and one friend of a person living with dementia. Professionals (10 male) had a variety of professional backgrounds, including 14 physiotherapists, eight academics, seven members of volunteering organisations, six occupational therapists, four rehabilitation assistants, one social worker, one nurse and one health care support worker. Professionals from six countries (being the large majority from England) and three different continents completed the survey. Both stakeholder groups included participants with experience of supporting physical activity across stages of dementia and activity settings (table 1).

Table 1 - Characteristics of participants in the round two survey

	Carer stakeholder group (n=36)	Professional Stakeholder groups (n=39)
	Mean (SD) or n (%)	Mean (SD) or n (%)
Gender (male)	12 (33.3%)	10 (25.6%)
Age (Years):		
18-29	1 (2.8%)	8 (20.5%)
30-39	0 (0%)	9 (23.1%)
40-49	2 (5.6%)	6 (15.4%)
50-59	12 (33.3%)	12 (30.8%)
60-69	9 (25%)	3 (7.7%)
70-79	10 (27.8%)	1 (2.6%)
Supporting people in the following stages of dementia progression:		
Mild to Moderate	12 (33.3%)	8 (20.5%)
Moderate to Severe	13 (36.1%)	7 (17.9%)
Severe	0 (0%)	2 (5.1%)
All stages	11 (30.6)	26 (66.7%)
Not known	2 (5.6%)	0 (0%)
Supporting activity in the following settings:		
Community	34 (94.4%)	35 (89.7%)
Sheltered accommodation	3 (8.3%)	8 (20.5%)
Care or nursing home	12 (33.3%)	22 (56.4%)
Hospital	2 (5.6%)	24 (61.5%)
Country:		
England	36 (100%)	35 (89.7%)
Wales	0 (0%)	1 (2.6%)
France	0 (0%)	1 (2.6%)
Portugal	0 (0%)	1 (2.6%)
Brazil	0 (0%)	1 (2.6%)
Singapore	0 (0%)	1 (2.6%)

Ranking results

Round one participants added two new carer outcomes: “Carer feeling less worried” and “carer living longer”. A total of four new negative side effects were also added: “becoming more disabled”, “forgetting the activity”, “carer feeling heartbroken” and “creating a conflict between the carer and the person living with dementia”. Therefore, 12 carer outcomes and 21 negative side effects of physical activity were considered in round two. In this last round, the top three carer outcomes identified by both stakeholder groups were: “carer feeling positive and satisfied”; “carer improving wellbeing” and “making the lives of carers easier”. The three most undesirable side effects across all participants were: “becoming agitated and confused”, “falling over” and "feeling discomfort and pain". When considering the rankings by stakeholder groups, carers ranked the same three negative side effects as the overall group but professionals put "falling over" in the ranking position number four, and "having a bad experience" in third place (table 2). Refer to supplementary material[15] for a full list of all outcomes ranked by stakeholder group.

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[15] This supplementary material can be found in Appendix D of this thesis document.

Table 2 - Top three carer outcomes and top three side effects of physical activity from the perspective of carers and professionals.

Top three carer outcomes in lay terminology	**Sum of ranking positions from all participants (n=75)**	**Final overall ranking position**	**Definition, as in the glossary made available to participants**
Carer feeling positive and satisfied	228	1	Carers feeling positive about the person living with dementia being active, improving and having a fulfilling time. Carers feeling proud of the person living with dementia and seeing them doing activities they used to do in the past. Carer having better self-esteem. In the literature this was linked to confidence in their care abilities and carers' satisfaction with the intervention.
Carer improving wellbeing	230	2	Carer wellbeing and quality of life. Carer having fun.
Making the lives of carers easier	319	3	Physical activity may reduce the burden of care in the long-term by: maintaining functional independence of the person with dementia and finding the person living with dementia more agreeable to tasks, lightening the workload that need to be done by the carer; carer accessing support from professionals; and carer experiencing less challenging behaviour, including less sun downing from the person living with dementia. In the short-term, carers' lives can be made easier by: giving the carer a break; time and space to themselves or some respite, while the person with dementia is involved in physical activity and needing less input from the carer.
Top three side effects of activity	**Sum of ranking positions from all participants (n=75)**	**Final overall ranking position**	**Definition, as in the glossary made available to participants**
Becoming agitated and confused	319	1	Becoming challenging, frustrated, rude or overstimulated during physical activity. Refusing to go back into a care setting after an activity in a different environment. In some cases, activities with these effects were considered not appropriate for the person living with dementia and are often interrupted.
Falling over	400	2	Sustaining falls or increasing falls risk by being active. Sustaining injuries after a fall (e.g. fractures) and having to attend emergency care because of falls. Being about to fall, but being able to save oneself. Increasing fear of falling and reduced confidence in walking due to fear of a fall.
Feeling discomfort and pain	419	3	Includes joint pain, muscle soreness or stiffness after exercising. Complaining of pain or experiencing physical discomfort during physical activity. Not being able to be as active as usual in the day(s) after the activity.

DISCUSSION

Physical activity interventions for people living with dementia require effort, carer involvement and are not free from potential negative side effects. The present study successfully identified the top positive carer outcomes of supporting physical activity for the person living with dementia (i.e., “carer feeling positive and satisfied”; “carer improving wellbeing” and “making lives of carers easier”); as well as the most undesired side effects of physical activity (i.e., “becoming agitated and confused”, “feeling discomfort and pain” and “falling over”), from the perspectives of carers and professionals.

The three carer outcomes ranked as most important by the carers and professionals, relate to key aspects of carer burden, which are well described in the literature about dementia caregiving (van der Lee et al., 2017). Reduced burden of care (“making lives of carers easier”) has been linked to carer sense of competence or self-efficacy (“carer feeling positive and satisfied”) and carer quality of life (“carer improving wellbeing”) (van der Lee et al., 2017). Interventions to reduce carer burden do not always consider physical activity for the person living with dementia as a possible solution (Acton and Kang, 2001). Yet this could be a possibility, as a recent systematic literature review found physical activity for the person living with dementia to be effective at reducing carer burden (Zeng et al., 2016). However, the relationship between physical activity for the person living with dementia and carer burden may be complex. Relying on carers to organise and support physical activity is likely to increase their workload and perceived burden (Kang et al., 2018;van Alphen et al., 2016a); however, physical activity for the person living with dementia, without carer involvement, may give carers a break, reducing their burden of care (Watts and Teitelman, 2005). Moreover, perceived burden of care, may be linked to more than just the number of tasks carers are required to undertake to make the physical activity happen, but also the way a particular physical activity intervention is designed and delivered. Current carer policy notes that carers value being involved and listened to, in the design of interventions for the person living with dementia (Newbronner et al., 2013) and professionals are encouraged to establish a proactive and respectful collaboration with carers (van Alphen et al., 2016a), which may increase their self-efficacy, decrease the perceived burden of care, ultimately promoting adherence to physical activity.

The key role of carers in engaging the person living with dementia in physical activity was also reflected in how the participants in this study ranked possible negative side effects of physical activity. The three most undesirable side effects of physical activity for the person living with dementia can all be related back to carer burden.

Firstly, behavioural and physiological symptoms of dementia (“increased confusion and agitation”) have been associated with carer burden in previous research (van der Lee et al., 2017;Regier and Gitlin, 2018). These are also commonly reported negative side effects of physical activity (Gonçalves et al., 2018a) and could be a direct barrier to participation in physical activity (Yu and Kolanowski, 2009).

Secondly, pain (“feeling discomfort and pain”) is a known possible cause of behavioural and psychological symptoms of dementia and also a cause of functional dependence for the person living with dementia (Flo et al., 2014). Symptoms of pain in a person living with dementia can therefore be indirectly linked to carer burden. Although important, pain has not often been reported as a negative effect of physical activity for people living with dementia (Gonçalves et al., 2018a). It is unclear if this is because pain has actually been linked to inactivity (rather than activity) (Plooij et al., 2012), or if pain has simply been missed as a side effect, since expressing themselves is challenging and poses difficulties to recognise pain levels in people living with dementia (Agit et al., 2018). Further research is needed to understand this further.

Lastly, falls also known to have a major impact on carer burden and carer perceived ability to care (McIntyre and Reynolds, 2012). People living with dementia are more likely to fall, be injured and admitted to hospital after a fall than people without dementia (Chen et al., 2018;Allan et al., 2009). In previous research, falls prevention has been agreed to be amongst the most important positive outcome of physical activity for people living with dementia, and recommended to be measured in all future research in this area (Gonçalves et al., 2019b). The present study shows that it is important to record falls both as a positive outcome (falls prevention) and a potential side effect (increased falls during activity) in future physical activity intervention for people living with dementia.

Strengths and limitations

To our knowledge, this was the first study to provide insight into the priorities of carers and professionals regarding both carer outcomes from supporting physical activity for the person living with dementia, and the possible negative side effects of physical activity. This prioritisation exercise was embedded in a Delphi survey, which allowed some anonymous interaction between participants, who were able to reflect on their own ranking based on feedback from other participants. This method also allowed participants to take part remotely, nationally and internationally.

However, this study is not without limitations, including most importantly, the lack of involvement of people living with dementia in this exercise. People living with dementia took part in the primary Delphi survey in which this prioritisation exercise was embedded (Gonçalves et al., 2019b) using novel card sorting strategies to enable them to prioritise outcomes. This was however a lengthy process (on average 30 minutes per session) and they were therefore not asked to complete these prioritisation tasks, with the aim of minimising fatigue. Future work should include such methods to gather the views of people living with dementia, particularly on the negative side effects of physical activity.

CONCLUSION

Physical activity for people living with dementia is valued by carers and professionals, not only for its benefits for patients, but also for its potential to reduce carer burden. Negative effects, such as behavioural and psychological symptoms of dementia, pain and falls are potentially the most undesirable side effects of physical activity and can also influence perceived burden of care. These outcomes should be consistently reported in future research in this field, to allow professionals and carers to make informed decisions on the safety of the intervention, according to outcomes meaningful to them. Designing interventions that take into account these outcomes on carers and possible negative side effects may influence adherence to physical activity.

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CONFLICT OF INTEREST STATEMENT

The authors have no conflicts of interest to declare.

Chapter 6 General Discussion

The main aim of the present thesis was to develop a Core Outcome Set to evaluate physical activity interventions for people living with dementia, across different stages of disease progression, settings and applicable in research and clinical practice. This research was presented in a series of papers. Paper I established the protocol for this Core Outcome set. Paper II was a systematic literature review of outcomes and measurement tools reported in the last 10 years. This literature review initiated the process of creating a “long list of outcomes”. Paper III was a qualitative study with key stakeholders (people living with dementia, their informal carers and the professionals involved in their care) that aimed at adding any missing outcomes to this “long list”, as well as gaining deeper understanding of what makes physical activity meaningful for people living with dementia. This “long list of outcomes” formed the base of a consensus exercise (a modified Delphi survey), reported in Paper IV, which led to the final seven outcomes being included in the present Core Outcome Set: “walking better, being able to stand up and climb stairs”; “Doing what you can do”; “preventing falls”; “staying healthy and fit”; “feeling useful and having a purpose”; “feeling brighter” and “enjoying the moment”. To maximise engagement of people living with dementia in the consensus exercises, the length of data collection sessions was reduced to a minimum and only positive outcomes were considered in the final Core Outcome Set. To complement this, Paper V reports on a prioritisation exercise, completed alongside the Delphi survey, where carers and professionals ranked the most important outcomes of physical activity for the person living with dementia on carers and the most undesired negative side effects of physical activity to both patients and carers.

Findings of Papers I to V have already been discussed in the individual publications. This general discussion will integrate the findings of all papers and will debate the main contributions to knowledge made in this thesis in two main areas: i) methodological contributions to Core Outcome Set development and the involvement of people living with dementia in research; ii) contributions to knowledge about meaningful outcomes of physical activity for people living with dementia, their carers and health professionals involved in their care.

6.1 Contributions to knowledge: core outcome set methodology and involvement of people living with dementia in research

This thesis presents five key contributions to knowledge regarding the methodology of Core Outcome Set development and involvement of people living with dementia in research.

The first methodological contribution to Core Outcome Set development was the publication of Paper I. Although written before the publication of the COS-STAD (Kirkham et al., 2017), this study protocol meets all the 11 minimum standards in the COS-STAD document (see COS-STAD checklist in appendix E), indicating this is a robust protocol. It is also in line with other published protocols with regards to sample size (Harding et al., 2018;Sherratt et al., 2017) and completion of data collection mainly in the United Kingdom (Sherratt et al., 2017;Harding et al., 2018). Further, this protocol provides guidance on an innovative approach to the consensus process that can now be used to include patients with cognitive impairment in consensus processes. In the absence of a gold standard for Core Outcome Set development (Kirkham et al., 2017), this protocol gives an example of a mixed methods Core Outcome Set and guides other developers to use patients and the public to adapt their methodologies and continue to fully include patients in Core Outcome Set development.

The second contribution to methodology was made through the systematic literature review of outcomes (Paper II). Despite being a very common first step in core outcome set development (Gargon et al., 2018), there are no specific methodological recommendations on how to conduct these reviews of outcomes. In fact, they vary greatly amongst Core Outcome Set developers. Some developers conduct more than one review of the literature: one of quantitative (Hopkins et al., 2015b) and one of qualitative (Coulman et al., 2017) studies, before the consensus phase of a Core Outcome Set. Others have made recommendations on the use of outcome measures from a single literature review (Ball et al., 2013). Some reviews focused on patient reported outcomes (Coulman et al., 2013) while others focus on side effects of interventions (Blazeby et al., 2015). Some include only peer reviewed studies (MacLennan et al., 2015); or only randomised controlled trials (Wilkinson et al., 2016) whilst others also include grey literature (Blazeby et al., 2015). Paper II describes a systematic literature review of qualitative and qualitative studies, which listed all outcomes reported in the last 10 years in peer reviewed literature, including: clinical outcomes, patient reported outcomes, both positive and negative outcomes as well as carer outcomes. This approach aimed to cover the potential complex impact of physical activity on those living with dementia and their families and it may be used by other developers looking at complex interventions. This literature review also contributes to the knowledge on outcomes of physical activity for people living with dementia by making available with the publication an interactive file where all outcomes and measurement tools can be filtered based on intervention setting and stage of disease progression. This allows researchers and clinicians to interact with the data from the literature review in a way that is useful to their own setting and population of interest. This file has received multiple expressions of interest from researchers and clinicians looking at “what to measure” in physical activity interventions for people living with dementia, ahead of the completion of this Core Outcome Set. Future Core Outcome Set developers may choose to use similar interactive ways of presenting literature review data, to expand the usability of their literature reviews, from a simple “long list of outcomes” to an informative data set that can be used by researchers and clinicians, from the early stages of Core Outcome Set development.

The third contribution to knowledge about Core Outcome Set methodology is linked to the findings of Paper III (qualitative study). To our knowledge, this was the first qualitative study to be published in the pre-consensus stage of a Core Outcome Set, and it may therefore be used as a guide by the growing number of developers using mixed methods to generate Core Outcome Sets (Gargon et al., 2018). The possible value of qualitative studies in Core Outcome Set development had already began to be explored when Paper III was written (Keeley et al., 2016). However, paper III showed additional benefits as its findings. It suggested the majority of meaningful outcomes of physical activity were common across stages of disease progression, and therefore, the structure of this Core Outcome set was changed (from a planned Core Outcome Set subdivided based on the stages of disease progression, to a single Core Outcome Set across all stages of dementia). This may prompt other developers of Core Outcome Sets of progressive diseases to use qualitative methods in order to determine if outcomes that are meaningful to stakeholders vary with the progression of disease.

The fourth contribution to knowledge is a re-enforcement of the argument that patients (in this case, people living with dementia) and their carers can and should be involved in the design of the Core Outcome Set methodology as well taking part as participants (Young and Bagley, 2016). Paper IV described how patient and public involvement activities informed the consensus process of this Core Outcome Set. With the number of Core Outcome Sets including patients (even “only” as participants) gradually increasing overtime (Gargon et al., 2018), and the inclusion of patients as research partners recognised as necessary (Young and Bagley, 2016), more Core Outcome Set developers will look into strategies to include patients and the public to design their core outcome sets, in which case, Paper IV can serve as guidance.

The last contribution to knowledge, regarding core outcome set methodology, was the use of a card sorting strategy to enable participation of people living with dementia in the Delphi survey (Paper IV). Delphi surveys are known to be a challenging method to implement with people living with dementia (Wood, 2014), which was also in agreement with the feedback received during the patient and public activities conducted during the planning stage of Paper IV. In order to address this challenge, an innovative card sorting alternative was developed through several patient and public involvement activities, and was also inspired by other Core Outcome Set developers who successfully adapted their methodological approach to include patients in the consensus process

(Morris et al., 2015). As described in the background chapter of this thesis (Chapter 2), Morris et al. (2015) used a Q-sorting task, where children with neuro-disabilities, their parents and professionals used laminated cards to “force” outcomes in order of importance, with some similarities to the method implemented in Paper IV. Both in Paper IV and in the study by Morris et al. (2015), these adaptations were successful at engaging patients with a possible cognitive impairment to prioritise health outcomes meaningful to them, and might now guide others wishing to involve these populations in research studies.

Other adaptations to a Delphi survey to enable participation of people living with dementia have recently been reported (Morbey et al., 2019), and highlighted in Chapter 2. Researchers working on a Core Outcome Set to evaluate non-pharmacological interventions for people living with dementia in the community (Morbey et al., 2019) described smaller but equally successful adaptations to a more traditional Delphi survey (i.e., Likert scale with three items only; use of face-to-face surveys; verbal feedback in round two; and acceptance of partially completed surveys). Whilst an ideal method to include people living with dementia in Delphi surveys is yet to be established, the card sorting strategy described in Papers I (protocol) and IV (consensus study), presents a possible solution to ensure that people with dementia are fully involved in the selection of outcomes that matter to them. It is also a more interactive way to engage people living with dementia, with the possibility of being used across stages of the disease progression. Therefore, this innovative card sorting strategy contributes to knowledge in two important areas of research and practice: i) it provides core outcome set developers with an alternative to the traditional Delphi survey, when including patients cognitive impairment and ii) provides a suggestion to health and social care professionals on how to engage people living with dementia in prioritisation of health outcomes.

6.2 Contributions to knowledge: meaningful outcomes of physical activity for people living with dementia

The present thesis has made valuable contributions to knowledge regarding meaningful outcomes of physical activity for people living with dementia.

Firstly, Paper II (systematic literature review) identified the most frequently reported outcomes of physical activity interventions in the last 10 years of dementia research. As noted in research involving other populations and interventions, this review highlighted important heterogeneity in the reporting of outcomes. Reviews of outcomes from other Core Outcome Sets identified similar or even higher numbers of outcomes (164 outcomes identified by Whitehead et al. (2015) in a literature review of outcomes in cardiac arrest clinical trials; 1088 outcomes in a literature review of outcomes in bariatric surgery by Hopkins et al. (2015b)). Other reviews of outcomes also reported on heterogeneity in the use of measurement tools and definition of outcomes, highlighting how this impairs the possibility of comparisons being made between studies in multiple health conditions (Hopkins et al., 2015b;Rodgers et al., 2014;Blencowe et al., 2012). The heterogeneity in the use of instrument tools has raised concerns regarding research waste as a global issue, which has recently prompted the COMET initiative to highlight the disease with greater global prevalence that still lack Core Outcome Sets (Gorst et al., 2016).

Whilst findings of heterogeneity were expected, it was surprising that the most frequently reported outcome domains identified by this literature review did not coincide with those that reached consensus as the final Core Outcome Set (Paper IV). This adds new knowledge to the field of physical activity for people living with dementia, by highlighting that the outcomes most frequently measured by researchers are not necessarily the outcomes most valued by professionals, patients and carers. It also emphasises the importance of consulting key stakeholders in the selection of research outcomes, in order to minimise research waste.

This disparity between the most reported outcomes in the literature, and those agreed to be core, was not found in other literature reviews of (now completed) Core Outcome Sets. For instance, the most reported outcome domain in the literature review of outcomes in the Core Outcome Set for cardiac arrest was “survival” (Whitehead et al., 2015); and “survival” also gained consensus to be included in the final core of outcomes (Haywood et al., 2018). Similarly, the most frequently reported outcome in the literature on bariatric surgeries was “weight loss” (Hopkins et al., 2015a) and “weight” was also included in the final core outcome set to evaluate bariatric surgery (Coulman et al., 2016). It can be hypothesised that this did not happen in the Core Outcome Set presented in this thesis, due to its scope involving physical activity interventions for people living with dementia. It may be clear that the main outcome for a patient in cardiac arrest is “to survive” and the main outcome for a patient going through bariatric surgery is to “lose weight”. But the link between physical activity for people living with dementia and its main outcome(s) is less clear. People living with dementia may be active for many reasons and professionals may attempt to achieve an array of possible outcomes when promoting physical activity for the person living with dementia (e.g., increased functional independence, improved cognition, reduced falls etc). This complexity of possible outcomes of physical activity, in the context of dementia care, reinforced the need for a qualitative study, aiming to gain deeper understanding of “why” physical activity is important for those living with dementia. Paper III (qualitative study), therefore, had a dual aim of adding to the list of outcomes created at the end of Phase II (discussed under methodological contributions to knowledge), and provide in-depth understanding on why physical activity is meaningful for all key stakeholders.

The second contribution to knowledge about meaningful physical activity outcomes for people living with dementia was therefore accomplished in Paper III. This qualitative study clarified that people living with dementia, their families and professionals involved in their care, promote physical activity with the view to support “being well and staying well”, “having a role towards others”, “maintaining identity”, “being connected to the present” and “delivering good quality care”. Paper III concluded that physical activity can be important for the person living with dementia and their carers to continuously adapt and reframe their identity throughout the progression of the disease. It could be questioned whether these themes are only valued in physical activity interventions, or if they represent valued outcomes in any intervention in dementia care. The argument presented here is that the themes identified in Paper III, are in line with the overall values of many different interventions in dementia care. However, within those themes, the outcomes agreed in the consensus study (Paper IV) are particularly relevant in physical activity interventions, and so will be the measurement tools to measure these outcomes. For instance, a qualitative study about the experiences of people living with dementia and their carers attending a singing group reported outcomes such as “sense of belonging”, “enjoyment”, “having something to talk about” and “improving relationships” (Osman et al., 2016), which are all outcomes included in the themes identified in Paper III. When considering the literature about the overall experiences and expectations of people living with dementia and their carers, once more, similar themes can be noted: the importance of maintaining function and independence, staying connected with their pre-diagnosis identity and maintaining dignity (Read et al., 2017;van Gennip et al., 2016). These commonly valued themes of outcomes indicate that physical activity can be a key intervention to allow people with dementia to achieve what is meaningful to them. Of those common themes, the valued specific outcomes of physical activity were made clearer when consensus was achieved in Paper IV. For instance, from the theme “being well and staying well”, outcomes related to falls prevention, mobility and fitness reached consensus. These more specific outcomes are commonly measured in physical activity interventions for multiple health conditions and populations (Oliveira et al., 2019;Morales et al., 2018;Dean et al., 2012). This revalidated the need for a core outcome set in dementia care, specific to physical activity interventions; and reinforces the role of physical activity in achieving outcomes meaningful in dementia care.

The third contribution to knowledge about meaningful outcomes of physical activity for people living with dementia were the seven outcomes that reached consensus in Paper IV: “walking better, being able to stand up and climb stairs”; “doing what you can do”; “preventing falls”; “staying healthy and fit”; “feeling useful and having a purpose”; “feeling brighter” and “enjoying the moment”. These findings clarify that people with dementia value physical activity for its physiological benefits - related to mobility, falls, independence and fitness. But also value physical activity in its own right, as it brings enjoyment and a sense of purpose. This concept of physical activity being important, regardless of its potential benefits to the body, was in fact a topic of in-depth discussion in the consensus meeting in Paper IV, when participants voted to exclude the outcome “slowing down the dementia” from this Core Outcome Set. Participants agreed that it would be very positive if physical activity could delay the progression of the disease, but felt strongly that perceived benefit of physical activity should not be centred only on physiological factors. Instead, its value in terms of bringing enjoyment and a sense of purpose should be emphasised. These findings are similar to those reported in qualitative studies including patients living with other (currently) incurable pathologies. For instance, a qualitative study about the perceptions of teenagers with cystic fibrosis on physical activity concluded that alongside the physiological benefits of activity, patients valued outcomes such as “enjoyment”, “personal satisfaction” and “sense of accomplishment” (Swisher and Erickson, 2008). The perspectives of patients receiving exercise interventions in the context of palliative care are also in line with these findings, suggesting that the psychological and psychosocial benefits of physical activity (such as a sense of “feeling normal”, “feeling joyful”, “increasing confidence, self-esteem and self-worth” are very important outcomes, alongside its physiological benefits, which may or may not be achieved (Malcolm et al., 2016). This thesis therefore contributes to knowledge on meaningful outcomes of physical activity for people living with dementia, by clarifying that physiological outcomes related to mobility, independence, fitness and falls prevention are important but also that the participation in physical activity in itself, may bring overall benefits such as enjoyment and sense of purpose.

The last contribution to knowledge regarding meaningful outcomes of physical activity for people living with dementia was the identification of the most important outcomes for carers (“carer feeling positive and satisfied”; “carer improving wellbeing” and “making lives of carers easier”) and the most undesirable negative side effects of physical activity for people living with dementia (“becoming agitated and confused”, “falling over” and “feeling discomfort and pain”). Side effects such as falls, fractures (and consequently pain) are known possible side effects of antipsychotic medication (Steinberg and Lyketsos, 2012), prescribed to people living with dementia who experience neuro-psychiatric symptoms (such as delusions and agitation). Carers and professionals may therefore be very aware of the high risk of falls in people living with dementia (particularly those on such medication) and be very vigilant to avoid additional risks that physical activity may bring, with regards to falls, injuries and also agitation and distress, which may lead to the need for antipsychotic medication, and potentially feed into a spiral of side effects. This study contributes to knowledge as it was the first study to provide insight into the priorities of carers and professionals about potential negative side effects of physical activity, in the context of dementia care. It also adds to knowledge about the possible impact of physical activity for the person living with dementia on their carers. These findings may inform clinicians and researchers on how to promote adherence to physical activity, by focusing not only on patients' but also on carers' priorities; and by concentrating on addressing the most undesired potential negative side effects of physical activity on patients. These findings can also be key in the process of informed consent to take part in physical activity, as professionals will be able to provide information on the positive outcomes of the intervention on both patients and carers; and inform them about potential risks (meaningful to them) and how they will be addressed.

6.3 Strengths and limitations

The present thesis contributes to knowledge on meaningful outcomes of physical activity for people living with dementia, through innovative methods that will inform Core Outcome Set methodology and the inclusion of people living with dementia in prioritisation of health outcomes. The main strength of this research, was having the needs of people living with dementia at its very core, which was possible due to two important factors. Firstly, this research was part of a Clinical Academic Doctoral Programme. Having a clinical role as a physiotherapist in a dementia ward allowed the experience and confidence in communicating with people living with dementia, recognised in the literature as a key factor to allow inclusion and involvement of people living with dementia in research (Morbey et al., 2019); it also provided the experience of communicating with carers and with multiple health and social care professionals, all key stakeholders in the development of this thesis. Secondly, this research benefited from patient and public involvement in all empirical studies. Patient and public involvement was conducted using two different approaches: collaboration and consultation. Collaboration from a former carer of someone living with dementia who advised on recruitment, design of data collection and debated research findings. Consultation activities were also conducted for the development of the card sorting strategy reported in Paper IV (consensus study) with more than 40 people living with dementia and their carers. This resulted in the development of a Core Outcome Set, which not only followed robust methods, but was also innovative and successful at maximising inclusion of people living with dementia.

Specific limitations to each of the papers have been detailed in the individual publications (Papers II to V discussion sections). Overall, the extent to which the Core Outcome Set presented in this thesis can be generalised internationally is debatable. A small proportion of international participants was included in the Delphi survey, yet more international work is needed to involve more participants from different countries and promote adherence to this Core Outcome Set internationally. Recommendations on how to validate and support the uptake of this Core Outcome Set internationally are also made in the next section: "Recommendations for future research".

Even within a national context, it is important to highlight that the card sorting strategy (developed for the purpose of study IV) was created based on consultation activities involving patients and members of the public, locally. It is possible that, gender, age, ethnicity, culture and levels of health literacy of those involved in the consultation processes, may have influenced the final outlook of the cards. This may imply limitations on the usability and acceptability of the card sorting strategy in other cultural contexts. For instance, all participants in the patient and public consultation processes were English and Caucasian, as were all participants living with dementia who used the card sorting strategy during data collection. All photos printed on the cards, representing each of the outcomes, were photos of Caucasian people. It is possible that adaptations may be needed if this card sorting strategy was to be used in different cultural contexts. Additionally, the selection of the photos in each of the cards were limited by the resources available, as only images that were free of copyright could be used. Care was taken to ensure that all pictures had an implicit positive emotion (e.g. people smiling). This aimed to avoid outcomes being selected by participants living with dementia because "the person in the card looked happy" in detriment to those in which the "person in the card might have looked sad". However, it is still unclear how much the photos in the cards (rather than the outcome itself) influenced participants' choices. Whilst this uncertainty was balanced by asking participants to think aloud and to provide examples that justify their selection of outcomes, future work is necessary to further understand the influence of the photos in the cards, on outcome selection. Suggestions on how to pursue such work are outlined in the next section: "Recommendations for future research".

The present Core Outcome Set also lacked direct involvement of people living with dementia who lacked capacity to consent to taking part in research at the time of data collection. People living with dementia who lack capacity to take part in research will, in many cases, be involved in physical activity. Their views are therefore very important. In this Core Outcome Set, they were included through their carers. It is possible that people who lack capacity to consent to taking part in research could complete the card sorting strategy in a meaningful way, as organising cards can be cognitively less demanding than understanding concepts around research, data protection etc. However, the card sorting strategy was a new method that had not been used before and therefore, it was necessary to try it first with people with capacity to consent. It may now inform future studies looking at actively engaging people without capacity to consent in research studies.

6.4 Recommendations for future research

The next step of this this Core Outcome Set is the identification of measurement tools to measure the outcomes agreed to be core at the end of Paper IV (consensus study). This process will follow four steps: a consensus exercise on the construct to measure; a literature review of all existing measurement tools to measure the agreed construct; an assessment of the quality of the tools (measurement properties and feasibility); and a consensus process to agree on measurement tools to include in the Core Outcome Set (Prinsen et al., 2016). This process of selection of measurement tools will be an opportunity to include more international stakeholders, ahead of the implementation of this Core Outcome Set. The Core Outcome Set presented in this thesis is aimed to be applicable in research and clinical practice. Whilst having seven core outcomes may be feasible in a research setting, this it is likely not be the case in clinical practice, due to time restrictions. What is a feasible number of measurement instruments, and which measurement instruments can be recommended to be used in practice will also be part of the consensus process at this stage.

Once consensus has been reached on the measurement tools, work on promoting the uptake of this Core Outcome Set should not be overlooked, as the benefits of developing a Core Outcome Set are only achieved if it is consistently used by researchers and clinicians. The COMET and the OMERACT initiatives have done extraordinary work promoting the use of Core Outcome Sets as a concept. In addition, participants in Paper III (Qualitative study) were asked about strategies for implementation of this particular Core Outcome Set. They recommended dissemination at conferences related to the topic of physical activity in dementia care and through professional organisations. They also recommended the development of a booklet, with all measurement tools included in the Core Outcome Set and clear instructions on how to use them. These specific recommendations will inform the promotion and dissemination of the present Core Outcome Set. Further work is also welcomed to replicate the methods used in the development of this Core Outcome Set, in other countries. Such work would determine whether or not the outcomes identified in the present core outcome set would be valid across languages and cultural contexts. If so, this validation process may also support the uptake of this Core Outcome set, at an international level.

Some recommendations for future research can also be drawn from the specific publications completed as part of the development of this Core Outcome Set. The qualitative study (Paper III) had a dual aim of adding to the list of outcomes and gain in-depth understanding of the importance of physical activity for people living with dementia. For the purpose of this Core Outcome Set, it was important that this study had a greater focus on the “list of outcomes”. However, the data collected during the interviews includes greater detail on multiple aspects of physical activity in dementia care, including, to name a few, factors that promote adherence, required skill sets for family carers and professionals to deliver physical activity for people living with dementia, and impact of physical activity on relationships with others as the dementia progresses. These aspects of physical activity have received very little attention in previous literature and could be key to understanding and promoting physical activity in this population. Therefore, the next steps in this research will include a more comprehensive and in-depth analysis of the qualitative data, already collected, to better understand the complexity of physical activity for people living with dementia beyond health outcomes.

The card sorting strategy presented in Paper IV (Delphi survey) was successful at including people living with dementia in the consensus process, and therefore further work is recommended to validate this method and maximise its applicability. No gold standard has yet been established for the use of the Delphi method, or for the use of questionnaires for people living with dementia, to examine the validity of the card sorting strategy described in this thesis. In the absence of such gold standards, examples can be drawn from other areas of research, where different questionnaire types are compared. For instance, a market research study asked consumers to complete the same questionnaire in two different formats within one week – one questionnaire used a six-point Likert scale; the second questionnaire included the same questions but with binary answer options: “applies” or “does not apply” (Dolnicar et al., 2011). The authors found that both answer formats were equally reliable, but the binary answer questionnaire took less time to be completed and was considered easier to fill out by the consumers (Dolnicar et al., 2011). Similar strategies could be used to compare the selection of outcomes from a list (traditional Delphi format) versus the card sorting strategy reported in paper IV, where participants are asked to complete both formats of the same Delphi questionnaire. The answers can then be compared to ensure reliability, and participants can be asked to provide feedback on the usability of both formats. A similar process is recommended for the use of pictorial representation of outcomes versus written words only. This recommendation for future research should be extended to participants with and without cognitive impairment and it could guide future questionnaire research in health care.

Focusing particularly on the Delphi method, which is a commonly used consensus method by Core Outcome Set developers (Gargon et al., 2018), it is important to note that the card sorting strategy described in the thesis was used in one single Delphi round. Further work should explore how this card sorting strategy could be used to incorporate feedback and include people living with dementia in multiple Delphi rounds. Future work is also needed to explore if people without capacity to consent to research can engage with this method of data collection in a meaningful way, promoting their inclusion in research and enabling their views to be included.

The prioritisation of carer outcomes and negative side effects described in Paper V did not include people with dementia. Future work should aim to include people with dementia in this prioritisation exercise (possibly using the same card sorting strategy used to prioritise positive outcomes during the Delphi). Once people living with dementia have been included in this prioritisation exercise, consensus on these outcomes could be reached and side effects and carer outcomes could be included in the final Core Outcome Set.

More research is also needed to demonstrate the impact of the development of Core Outcome Sets on the homogeneity of the intervention studies published after the publication of Core Outcome Sets. A literature review of this nature has been completed to assess the impact of a Core Outcome Set in ankylosing spondylitis. Clinical trials of ankylosing spondylitis were compared for their selection of outcomes up to two year post publication of the Core Outcome Set; against those published more than two years post Core Outcome Set (Bautista-Molano et al., 2014). The authors showed an increase of 20% in studies reporting on all core outcomes, two years after the publication of the Core Outcome Set. Objective evidence like this is useful to strengthen the argument for the importance of Core Outcome Sets and monitor its applicability, so that Core Outcome Sets themselves do not add to research waste. Thus, future research should include a literature review to monitor the implementation of this Core Outcome Set to measure its impact on the homogeneity of reporting outcomes in physical activity research for people living with dementia.

Chapter 7 Conclusion

This thesis contributes to knowledge through the development of a Core Outcome Set to evaluate physical activity interventions for people living with dementia, across dementia types, stages of disease progression, intervention settings and applicable to research and clinical practice. It was found that physical activity research for people living with dementia has grown in recent years, but has not been translated into clear guidance to practice due to considerable heterogeneity in the reporting of outcomes and use of measurement tools. Moreover, recent literature had overlooked ten outcomes considered meaningful to patients, families and professionals. The use of qualitative studies within core outcome set development is therefore, encouraged to ensure all meaningful outcomes are taken into consideration in the consensus process. Core Outcome Set developers are also encouraged to adapt their consensus methods, through patient and public activities to enable meaningful inclusion of patients. An innovative card sorting strategy has been successful at including people living with dementia in the prioritisation of health outcomes.

This thesis also contributes to knowledge regarding meaningful outcomes of physical activity for people living with dementia. It was revealed that physical activity is not only considered important because of its physiological and wellbeing benefits, but also for the benefits that participation can bring to the person living with dementia, in terms of identity and perceived roles towards others. This knowledge can tailor delivery of physical activity interventions for this population group. While reporting on the effectiveness of physical activity interventions, researchers and clinicians are encouraged to report on the core outcomes identified in this thesis: “walking better, being able to stand up and climb stairs”; “doing what you can do”; “preventing falls”; “staying healthy and fit”; “feeling useful and having a purpose”; “feeling brighter” and “enjoying the moment”. Researchers and clinicians are also encouraged to monitor any increased incidence of falls, pain and behavioural symptoms as the most undesired negative effects of physical activity; and aim to address carer satisfaction and reduced burden of care, alongside the effect of physical activity on the person living with dementia.

The next steps for the completion of this Core Outcome Set are the identification of the most appropriate measurement tools to measure the seven core outcomes; and further consensus, including people living with dementia, on the core negative side effects of physical activity and the effects of physical activity for the person living with dementia on their carers. Additional research is also needed to further develop the card sorting strategy here presented, to enable its applicability in multiple rounds of a Delphi survey, and in the prioritisation of health outcomes in clinical practice.

Appendix A Interview topic guide – used in data collection for Paper III

Qualitative interview: topic guide (Stakeholder group one- People with dementia, their friends, relatives and informal carers)

Valued outcomes of physical activity interventions

Aims and objectives:

The overall aim of this study is to understand what is meaningful in the experience of doing physical activity as a person with dementia, from the perspective of both the patient, their relatives, friends and informal carers, who have been in touch with the person with dementia during their participation in these activities.

Introduction (aim: to introduce the research and set the context for the interview)

- Introduce self and role at University of Southampton
- Introduce the study and what it is about (give examples of physical activity)
- Purpose and length of the interview (45 to 90 min)
- Procedures of the interview if dyad will be interviewed simultaneously
- Voluntary participation, right to stop to have a break and/or withdraw
- Confidentiality
- Reasons to record the interview
- Any questions
- Verbal consent

Background (aim: to gain understanding of background information that may influence data regarding experiences of physical activity)

- Household circumstances (lives where and with whom).
- Main daytime activity (i.e. routine).
- Hobbies and interests.

Mapping participation in physical activity (PROBE FULLY)

- Types of physical activity has been involved with / supported since diagnosis (or perception of memory loss).

- Description of these activities.

Characterising activities

- Where did they took place?
- Who was involved?

Decision making

- Prompts to start doing these activities **(PROBE FULLY)**
- Reasons to choose these activities (rather than other activities available).
- What factors were taken in consideration to start these activities?
- What needed to be planned or in place to allow participation in activities?
- Any other reasons why this activity way chosen?

Experience of the activity (PROBE FULLY)

- How would they describe the experience of being part of these activities?
- What were the positive results?
- What were the negative results?
- Reasons to stop doing the activity.

Prioritising effect

- Select the most important results of the activity.
- How to recommend these activities to others memory loss/dementia.

Suggestions

- What could be done better to promote physical activity for people with dementia?
- Anything else to add?

Conclusion

- Thank you.
- Reassure confidentiality.
- Ask if would like to see the results of the study.

Qualitative interview: topic guide (Stakeholder group two – professionals and volunteers)

Valued outcomes of physical activity interventions

Aims and objectives:

The overall aim of this study is to understand what outcomes of physical activity interventions have been observed by those responsible for its design, support or implementation. Additionally it would be important to know how can the Core Outcome Set can be promoted and implemented.

Introduction (aim: to introduce the research and set the context for the interview)

- Introduce self and role at University of Southampton
- Introduce the study and what it is about (give examples of physical activity)
- Purpose and length of the interview (30 to 60 min)
- Voluntary participation, right to stop to have a break and/or withdraw
- Confidentiality
- Reasons to record the interview
- Any questions
- Verbal consent

Background (aim: to gain understanding of background information that may influence data regarding experiences of physical activity)

- Main daytime activity (i.e. routine).
- Hobbies and interests.

Mapping participation in physical activity (PROBE FULLY)

- Types of physical activity for people living with that the participant has been involved with
- Describe these activities.
- Describe professional role.

Characterising activities

- Where did they took place?
- Who else was involved?

Decision making

- Motivators to be involved these activities (motivators).
- Reasons to choose this activity (rather than other activities available).
- Factors taken into consideration to start these activities.
- Plans to promote adherence to the activities.

Experience of the activity (PROBE FULLY)

- How would they describe the experience of being part of these activities?
- Positive results for the participants.
- Negative results for the participants.
- Positive results to other people (i.e. relatives, carers, staff).
- Negative results to other people (i.e. relatives, carers, staff).
- Reasons to stop doing the activity.

Prioritising effect

- Select the most important results of the activity.
- How to promote this activity to people with memory loss/dementia?

Suggestions

- What could be done better to promote physical activity for people with dementia?
- How could a Core Outcome Set for physical activity be promoted **(PROBE FULLY)**
- What challenges are behind this implementation?
- Anything else to add.

Conclusion

- Thank you.
- Reassure confidentiality.
- Ask if would like to see the results of the study.

Appendix B Supplementary material of Paper III (Qualitative study)

Outcomes of Physical Activity for People Living with Dementia: Qualitative Study to Inform a Core Outcome Set.

GONÇALVES, A. C., MARQUES, A., SAMUEL, D. & DEMAIN, S. 2019a. Outcomes of Physical Activity for People Living with Dementia: Qualitative Study to Inform a Core Outcome Set. Physiotherapy, published online (in press). https://doi.org/10.1016/j.physio.2019.05.003

Supplementary Material – Outcome domains (n=77) with the scope and context in which each outcome domain has been identified[16].

Legend: "prof. and researchers" - professionals and researchers; "patients and carers" – people living with dementia and their informal carers or relatives. *Boxes identified with "x" indicate that the outcome domain has been identified in that context. Boxes identified with "-" indicate that an outcome domain has not been identified in that particular context. New outcome domains generated by this qualitative study are highlighted; outcomes measured or referred to in previous literature, but not mentioned in this qualitative study are underlined.*

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[16] Please note that in Appendix C (supplementary material for paper IV – Delphi study) these same outcomes are described again, but presented in lay terminology, as they were used in the Delphi survey.

Outcome domain identified:	**Scope**	**Stakeholder**		**Stage**			**Setting**		
		Prof. and researchers	**Patients and carers**	**Mild to moderate**	**Moderate to severe**	**Severe**	**Community**	**Institution**	**Hospital**
THEME: being well and staying well									
Achieving or maintaining functional abilities and independence	Maximising abilities using multiple body movements, being able to complete transfers, avoiding being bedridden, being independent in basic and instrumental activities of daily living, including handling cutlery and maintaining coordination to feed oneself. Feeling independent.	x	x	x	x	x	x	x	x
Attenuating disease progression	Maintaining abilities for as long as possible. Preventing decline and delaying onset of health complications. Includes physical and cognitive decline as well as perceived/expected decline in wellbeing.	x	x	x	x	x	x	x	x
Avoiding being admitted to hospital or being discharged quicker	Avoiding hospital admissions and reducing length of stay following a hospital admission, reducing social and health care use.	x	x	x	x	x	x	x	x

Outcome domain identified:	Scope	Stakeholder		Stage			Setting		
		Prof. and researchers	Patients and carers	Mild to moderate	Moderate to severe	Severe	Community	Institution	Hospital
Being more sociable and talkative	Initiating and keeping conversations, sharing stories and having something to talk about.	x	x	x	x	x	x	x	x
Enjoying activity and getting a sense of success and pleasure out of it	Having a sense of satisfaction, achievement, completion, success and reward or getting pleasure and joy as a result of activity. Finding activity appealing and feeling good about doing it.	x	X	x	x	x	x	x	x
Feeling more relaxed, settled and peaceful	Feeling calmer. Getting a sense of peace and relaxation. Being released from anxiety or confusion and feeling better in oneself post activity. Absence of physical and mental distress.	x	x	x	x	x	x	x	x
Having a healthy routine and a balanced life	Having a sense of order in the day, Keeping the momentum, getting the recommended hours of sleep, having good sleep hygiene and sleep patter. Having a healthy number of meals per day. Having a balance of leisure, rest and activities of daily living. Having	x	x	x	x	x	x	x	x

Outcome domain identified:	Scope	Stakeholder		Stage			Setting		
		Prof. and researchers	Patients and carers	Mild to moderate	Moderate to severe	Severe	Community	Institution	Hospital
	structure to the day. Knowing and keeping one's own routine.								
Improving ability to complete movements and tasks without difficulty	Incorporates fine motor skills, dexterity, hand-eye coordination, praxis and use of quick-reactions. The literature also refers to reaction time, agility, motor skills and ability to conduct movements rightly.	x	x	x	x	x	x	x	-
Improving cardiorespiratory and cardiovascular health	Maintaining a healthy weight, maintaining or improving endurance, circulation and lung function. The literature also included the following components: heart rate, blood pressure, maximum oxygen consumption, breath holding duration, oxygen saturation, respiratory exchange rate, respiratory rate and vital capacity.	x	x	x	x	x	x	x	x
Improving depressive symptoms and preventing depression	Feeling less depressed. Impacting on how the person sees her/himself; wanting to get out of bed in the morning and gaining motivation to do things.	x	x	x	x	x	x	x	x

Outcome domain identified:	Scope	Stakeholder		Stage			Setting		
		Prof. and researchers	Patients and carers	Mild to moderate	Moderate to severe	Severe	Community	Institution	Hospital
Improving dietary intake	Being hungry or having good appetite; eating and drinking more than usual; eating well; eating normally as opposed to not eating enough.	x	x	x	x	x	x	x	-
Improving mood and affective function	Mood levels; lifting mood or being in better mood; being content, brighter, happier or having fun. Having a positive attitude. Showing less negative emotions.	x	x	x	x	x	x	x	x
Improving or maintaining balance	Balancing oneself while moving and shifting weight. Challenging balance. Linked to falls risk.	x	x	x	x	x	x	x	x
Improving or maintaining mobility	Keeping mobile or regaining mobility; keeping moving; being more mobile indoors and outdoors; standing up and walking up and down. Climbing stairs. Includes all aspects of gait, using less supportive walking aids and keeping the ability to change position.	x	x	x	x	x	x	x	x
Improving or maintaining structure and	Improving, cognition, concentration, alertness, attention, memory and reducing confusion. Seeing changes in	x	x	x	x	x	x	x	x

Outcome domain identified:	Scope	Stakeholder		Stage			Setting		
		Prof. and researchers	Patients and carers	Mild to moderate	Moderate to severe	Severe	Community	Institution	Hospital
function of the brain	brain volumes and physiology of the brain. Being able to calculate, read and recognise places. Keeping the brain active. The literature includes additional specific aspects of cognition, such as: executive function, processing speed, different types of memory, Perception and visual-spatial awareness, body awareness, agnosia, reasoning and ability to follow instructions. Fluid biomarkers are also included in the literature.								
Improving posture	Includes sitting and standing posture.	x	x	x	x	x	x	-	x
Improving quality of life and wellbeing	Health related quality of life, in opposition to living longer with poor quality. Also referred to as “raising standards of living”. Includes physical, mental, emotional and social wellbeing. Generally feeling better, comfortable (physically and emotionally), improving stamina and managing fatigue. Feeling joy and satisfaction about being alive.	x	x	x	x	x	x	x	x

Outcome domain identified:	Scope	Stakeholder		Stage			Setting		
		Prof. and researchers	Patients and carers	Mild to moderate	Moderate to severe	Severe	Community	Institution	Hospital
Increasing levels of physical activity	Amount of physical activity performed by the person with dementia within a defined period of time. Includes measures of time, distance, repetitions and intensity (e.g. weights). The literature also includes the levels of restricted activity (e.g. number of days with no activity).	x	x	x	x	x	x	x	x
Keeping healthy, fit and active	Overall health, general ability or performance. Being fit, strong and ready for activity. Staying well and having less medical diagnosis.	x	x	x	x	x	x	x	x
Living longer	Reducing mortality, preventing deaths, lasting longer.	x	x	x	x	x	x	x	-
Maintaining current care arrangements	Living at home for as long as possible and avoid institutionalisation. Avoiding increasing need for higher level of care within the same setting.	x	x	x	x	x	x	x	x
Maintaining functional range of movement	Maintaining joint health and flexibility from upper and lower limbs. Avoiding contractures and maintaining	x	x	x	x	x	x	x	x

Outcome domain identified:	Scope	Stakeholder		Stage			Setting		
		Prof. and researchers	Patients and carers	Mild to moderate	Moderate to severe	Severe	Community	Institution	Hospital
	movement quality. Maintaining or increasing amplitude of movement. Includes achieving the necessary range of movement to complete functional talks (e.g. putting a coat on).								
Managing behaviours	Overcoming challenging behaviours or behavioural issues, including agitation and aggression, apathy, passivity or lack of eye contact, distress, shouting behaviours, restlessness, pacing, wandering, feeling angry and frustrated. Using inappropriate language or handling objects inappropriately and "sun downing". Reducing need for restraint.	x	x	x	x	x	x	x	x
Managing lower limb oedema	Fluid retention in the lower limbs.	-	x	-	-	x	x	-	x
Managing pain	Lower limb pain; arthritic and muscular pain; pain due to constipation.	x	x	x	x	x	x	-	x
Managing pressure ulcers	Preventing pressure ulcers and recovering skin integrity.	x	x	x	x	x	x	-	x

Outcome domain identified:	**Scope**	**Stakeholder**		**Stage**			**Setting**		
		Prof. and researchers	**Patients and carers**	**Mild to moderate**	**Moderate to severe**	**Severe**	**Community**	**Institution**	**Hospital**
Opening bowels	Regular bowel movements. Linked to a reduction in the use of laxative medication.	x	x	x	x	x	-	x	x
Preventing boredom	Relieving and preventing boredom by changing scenery, experiencing sensory or cognitive stimulation. Avoiding excessive sleep due to lack of interest or stimulation.	x	x	x	x	x	x	x	x
Preventing falls	Number of fallers, overall number of falls, number of falls sustained by one person with dementia. Link to health care costs associated with managing fall related injuries (e.g. fractures). The literature also includes concepts of falls risk and fear of falling.	x	x	x	x	x	x	x	x
Preventing incontinence	Regularly using the toilet, by mobilising to the toilet or commode instead of using a pad.	-	x	-	-	x	x	-	x
Reducing anxiety	Distracting from a state of anxiety and reducing levels of anxiety.	x	x	x	x	x	x	-	-

Outcome domain identified:	Scope	Stakeholder		Stage			Setting		
		Prof. and researchers	Patients and carers	Mild to moderate	Moderate to severe	Severe	Community	Institution	Hospital
Reducing use of medication	Taking a reduced number of different drugs and and/or the overall amount of medication. This includes medication for physical and mental health purposes and was linked to a reduction in health related costs.	x	x	x	x	x	x	x	x
Subtheme: Impacting the lives of carers									
Carer developing friendships and peer support	Family carers meeting other relatives of people living with dementia and developing a network of friendship and peer support.	x	x	x	x	x	x	-	x
Carer mobility	Carer mobility and balance, ability to walk with less joint pain.	x	x	x	x	x	x	-	-
Carer mood	Simply described as “carer mood”.	x	-	-	x	-	x	-	-
Carer quality of life	Carer wellbeing and quality of life. At times linked to carer burden.	x	-	x	x	x	x	-	x
Carer regaining sense of satisfaction and achievement	Carers feeling positive about the person with dementia being active, improving and having a fulfilling time. In the literature this was linked to confidence	x	x	x	x	x	x	x	x

Outcome domain identified:	**Scope**	**Stakeholder**		**Stage**			**Setting**		
		Prof. and researchers	**Patients and carers**	**Mild to moderate**	**Moderate to severe**	**Severe**	**Community**	**Institution**	**Hospital**
	in their care abilities and carers' satisfaction with the intervention.								
Carer sleep	Improved carer sleep quality.	x	-	-	x	-	x	-	-
Decreasing carer burden or distress	Activity may reduce the burden of care in the long term by maintaining functional independence of the person with dementia, accessing support from professionals and experiencing less challenging behaviour; In the short term by giving the carer a break while the person with dementia is involved in activity and needing less input from the carer.	x	x	x	x	x	x	x	x
Improving carer general health	Includes weight management, and overall wellbeing and health. In the literature this was measured as the carer's use of health and social care services and therefore linked to costs.	x	x	x	x	x	x	-	-
Increasing carer levels of physical activity	Joining in with the person with dementia, adding to their habitual levels of activity.	x	x	x	x	x	x	x	x

Outcome domain identified:	Scope	Stakeholder		Stage			Setting		
		Prof. and researchers	**Patients and carers**	**Mild to moderate**	**Moderate to severe**	**Severe**	**Community**	**Institution**	**Hospital**
Reducing carer depression	Lack of activity and engagement for the person with dementia was linked to carer depression.	x	x	x	x	-	x	-	-
Subtheme: negative effects or risks of physical activity for patients and carers									
Becoming aggressive, agitated, more confused, anxious, depressed or distressed during activity	Becoming challenging, frustrated, rude or overstimulated during the activity. In some cases, activities with these effects were considered not appropriate for the person with dementia and are often interrupted. Refusing to go back into a care setting after an activity in a different environment.	x	x	x	x	x	x	x	x
Becoming short of breath during activity	Shortness of breath, or needing to “catch one’s breath” and interrupting the activity because of it.	x	x	x	-	-	x	x	-
Being admitted to hospital or experiencing a deterioration in health	Being hospitalised or admitted to an emergency department. Experiencing a deterioration of overall health or becoming too unwell to continue activity.	x	-	x	x	x	x	x	-

Outcome domain identified:	Scope	Stakeholder		Stage			Setting		
		Prof. and researchers	Patients and carers	Mild to moderate	Moderate to severe	Severe	Community	Institution	Hospital
Being in pain or experiencing discomfort during or after the activity	Includes muscle soreness or stiffness after exercising. Complaining of pain or experiencing physical discomfort during activity. Not being able to be as active as usual in the day(s) after the activity.	x	x	x	x	x	x	x	x
Carer outcome: Increasing carer burden or distress	Carers may face an increase in workload: being/feeling responsible to offer activity and sometimes persuade them to join in activities. For carers, activity can imply effort, fatigue and negative emotions of guilt, frustration and worry about the patient safety. It may increase risks of negative health outcomes to the carer.	x	x	x	x	x	x	x	x
Discovering cysts	Ganglion cyst.	x	-	x	-	-	x	-	-
Eating or drinking something harmful	Eating or drinking something that should not be ingested, by confusing it with real food or drink.	x	x	x	x	x	x	-	-
Feeling dizzy, nauseous or fainting	Feeling dizzy, nauseous or light-headed. Having a syncopal episode.	x	-	x	x	x	x	-	-

Outcome domain identified:	**Scope**	**Stakeholder**		**Stage**			**Setting**		
		Prof. and researchers	**Patients and carers**	**Mild to moderate**	**Moderate to severe**	**Severe**	**Community**	**Institution**	**Hospital**
Feeling tired or exhausted	Feeling tired, drained, exhausted. Described as something that would stop the activity and that should be used to monitor the intensity of the activity.	x	x	x	x	x	x	x	x
Getting lost	Not being able to find the way back after an activity. Having to use technology to find the person with dementia.	x	x	x	x	x	x	x	-
Having a negative experience	Triggering negative emotions or feelings (e.g. embarrassment or unhappiness). Having an unsatisfying experience. Activity reinforcing illness and loss of roles (e.g. Letting the team down) and therefore having a negative impact on perceptions of wellbeing.	x	x	x	x	x	x	x	x
Increasing cardiovascular risk	Suffering a transient ischemic attack or developing cardiac pathology. This was also a reason for caution when involving a person with dementia in activity.	x	-	x	x	x	x	-	x
Increasing falls, falls risk, fall	Sustaining falls or increasing falls risk by being active. Sustaining injuries after a	x	x	x	x	x	x	x	x

Outcome domain identified:	Scope	Stakeholder		Stage			Setting		
		Prof. and researchers	Patients and carers	Mild to moderate	Moderate to severe	Severe	Community	Institution	Hospital
related injuries, near falls and fear of falling	fall (e.g. fractures) and having to attend emergency care because of falls. Being about to fall, but being able to save oneself. Increasing fear of falling and reducing confidence in walking due to fear of a fall.								
Increasing mortality	Number of deaths during activity or within the time period while the person with dementia was a participant in a physical activity intervention.	x	-	x	x	x	x	x	-
Moving into care	Being placed into an institution for permanent full-time care.	x	-	x	x	x	x	-	-
Not being able to sleep after an activity	This was linked to possible changes of environment or routine caused by the activity (e.g. coming back to care after having been on holiday).	-	x	-	x	x	-	x	-
Sustaining injuries (not fall related) or hurting others during activity.	Includes injuries to muscles and joints. May result from doing the wrong exercise or overdoing it; or from “bumping into” objects or fellow participants.	x	-	x	x	x	x	x	x

Outcome domain identified:	**Scope**	**Stakeholder**		**Stage**			**Setting**		
		Prof. and researchers	**Patients and carers**	**Mild to moderate**	**Moderate to severe**	**Severe**	**Community**	**Institution**	**Hospital**
THEME: Maintaining identity									
Feeling free and in control of one's life	Doing something independent from the carer, having rights, choices and interests and doing what one wants to do. Being in control of one's own life and feeling free to ask for activity.	x	x	x	x	x	x	x	x
Improving self-esteem, self-worth and self-respect	Sense of self, feeling good about (still) being active and strong and (still) being good at a certain activity. Feeling listened to, valued and respected.	x	x	x	x	x	x	x	x
Making the best of ones' physical abilities	Using one's physical being. Using all available skills and abilities. Expressing a physical self. Seeking movement as a routine part of life.	x	x	x	x	x	x	x	x
Meeting individual needs of patient and family	Meeting individual needs of the person living with dementia. These includes identity, physical, spiritual, emotional and social needs. Meeting tailored needs of families.	x	x	x	x	x	x	x	x

Outcome domain identified:	**Scope**	**Stakeholder**		**Stage**			**Setting**		
		Prof. and researchers	**Patients and carers**	**Mild to moderate**	**Moderate to severe**	**Severe**	**Community**	**Institution**	**Hospital**
Seeing the person before the disease	Being able to connect with the person rather than focusing on the disease through activity. Activity as a constant reminder that the person with dementia is a person and not a diagnosis.	x	x	x	x	x	x	x	x
Wanting life to be as normal as possible and continue doing what they have always done.	Feeling normal by keeping moving and keeping activities that are part of a routine, define identity and outline what "normal life" looks like for an individual. Keeping going despite the diagnosis.	x	x	x	x	x	x	x	x
THEME: Having a role towards others									
Becoming interested and gaining interest of others	Gaining the interest of the person living with dementia and the interest of staff, formal carers and relatives. Getting relatives and formal carers to know the person with dementia and becoming interested in what they can do.	x	x	x	x	x	x	x	x
Experiencing relationships	Doing activities together with staff or relatives and rebuilding relationships.	x	x	x	x	x	x	x	x

Outcome domain identified:	Scope	Stakeholder		Stage			Setting		
		Prof. and researchers	Patients and carers	Mild to moderate	Moderate to severe	Severe	Community	Institution	Hospital
	Seeking interaction with others; fighting isolation and loneliness.								
Getting involved, engaged, joining in and feeling included	Taking part and being part of an activity. Being involved and included.	x	x	x	x	x	x	x	x
Having a role, a purpose towards others and sense of belonging	Feeling useful. Being part of a family, team or a group. Being a volunteer. Helping family, neighbours and peers. Being occupied with meaningful activity. Doing what one is passionate about and having something to look forward to.	x	x	x	x	x	x	x	x
THEME: Being connected to the present									
Bringing back memories and emotions – reminiscence	Tapping into past experiences. Doing activity that relates to and can be recognised by the person with dementia. Triggering memories.	x	x	x	x	x	x	x	x
Communicating and expressing	Using movement as means of communicating information and emotions. Moving spontaneously and	x	x	x	x	x	x	x	x

Outcome domain identified:	Scope	Stakeholder		Stage			Setting		
		Prof. and researchers	Patients and carers	Mild to moderate	Moderate to severe	Severe	Community	Institution	Hospital
oneself through movement	naturally. The literature includes the using facial expressions, and improving language ability and verbal fluency.								
Having increased life opportunities	Getting out of the house, accessing daylight and the outdoors and accessing public spaces and services (including rehabilitation). Being allowed and having opportunity to experience activity. Not being forgotten about.	x	x	x	x	x	x	x	x
Living in the moment	Being anchored to the present by activity. Learning or doing something new. Having an adventure. Not having to think about anything else.	x	x	x	x	x	x	x	x
Living a fulfilled life rather than just existing	Dementia is not the end. Not be willing to "just sit around waiting to die". Being lively, or coming back to life when active. Staying engaged with life, enjoying every small aspect of life and having a fulfilling time while having dementia.	x	x	x	x	x	x	x	x
Reframing and revisiting the	Recovering one's "old self" even if for brief moments. Relatives having	x	x	x	x	x	x	x	-

Outcome domain identified:	Scope	Stakeholder		Stage			Setting		
		Prof. and researchers	Patients and carers	Mild to moderate	Moderate to severe	Severe	Community	Institution	Hospital
person with dementia	moments of seeing the "old" person with dementia by seeing them active as they were before the diagnosis. Relatives and staff or formal carers being surprised by the ability of the person living with dementia. Using activity to highlight what the person with dementia can still do.								
THEME: Delivering good quality care									
Adhering to activity	The extent to which an activity is completed according to a pre-set criteria of duration, frequency and intensity.	x	x	x	x	x	x	x	x
Improving care delivery	For families, good care meant care that offers opportunities for activities that reflect the needs of the person with dementia. For organisations, offering activity for people with dementia meant staff retention and staff satisfaction. An association is made between providing activity and offering good quality of care, that is cost effective in relation to	x	x	x	x	x	x	x	x

Outcome domain identified:	Scope	Stakeholder		Stage			Setting		
		Prof. and researchers	**Patients and carers**	**Mild to moderate**	**Moderate to severe**	**Severe**	**Community**	**Institution**	**Hospital**
	health and social care for patients and carers. It includes defining the longevity of any positive effects of activity.								

Appendix C Supplementary material Paper IV (Delphi survey)

A Core Outcome Set to evaluate physical activity interventions for people living with dementia

"This is a pre-copyedited, author-produced version of the supplementary material of an article accepted for publication in The Gerontologist, following peer review. The version of record Gonçalves, A.C..; Samuel, D.; Ramsay, M.; Demain, S.; and Marques, A. 2019. The Gerontologist, 100, is available online at: [*https://doi.org/10.1093/geront/gnz100*](https://doi.org/10.1093/geront/gnz100)

Supplementary material:

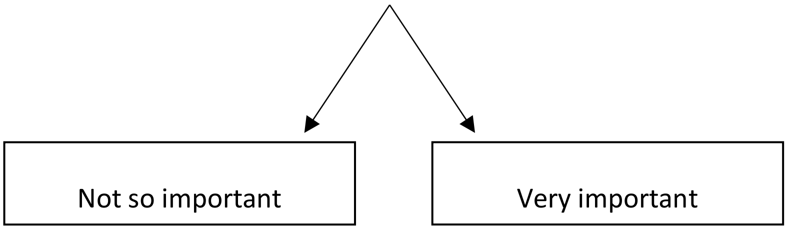
I. A step by step approach to the card sorted strategy, used by participants living with dementia in Delphi round two.
II. List of outcomes and levels of agreement per Delphi survey round.
III. Sample characterization of people living with dementia
IV. Demographic characteristics of participants who dropped out after round one.

I. Card sorting strategy, step by step.

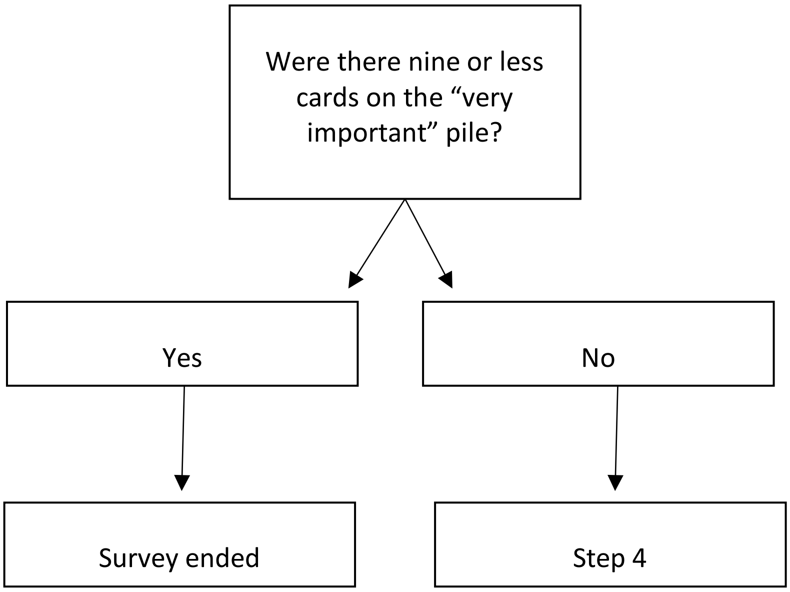
Step 1: Participants living with dementia were shown the pile of cards and the procedure was explained.

Figure A1: Pile of 36 cards representing the outcomes included in the Delphi round two.

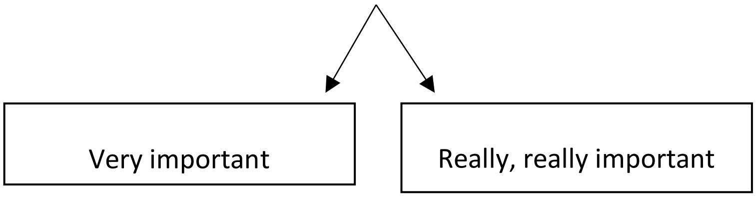
Step 2: The participant was shown one card at the time and asked to put it on one of the following piles:



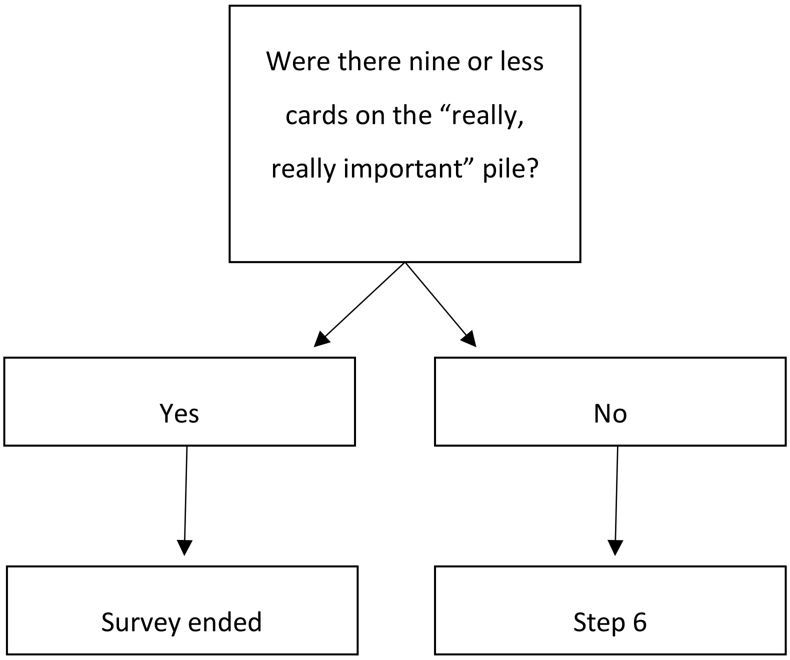
Step 3: The cards on the "not so important" pile were excluded. The cards on the "Very important" pile were counted.



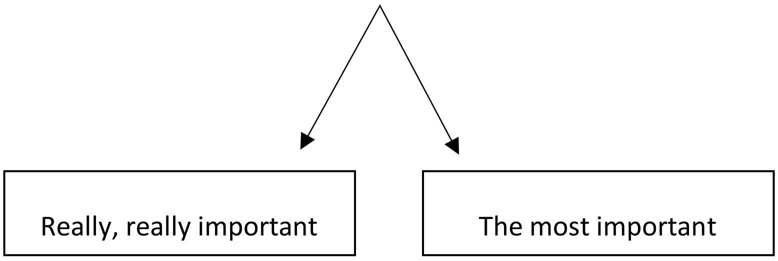
Step 4: From the "very important" pile, participant was shown one card at the time and asked to put it one of the following piles:



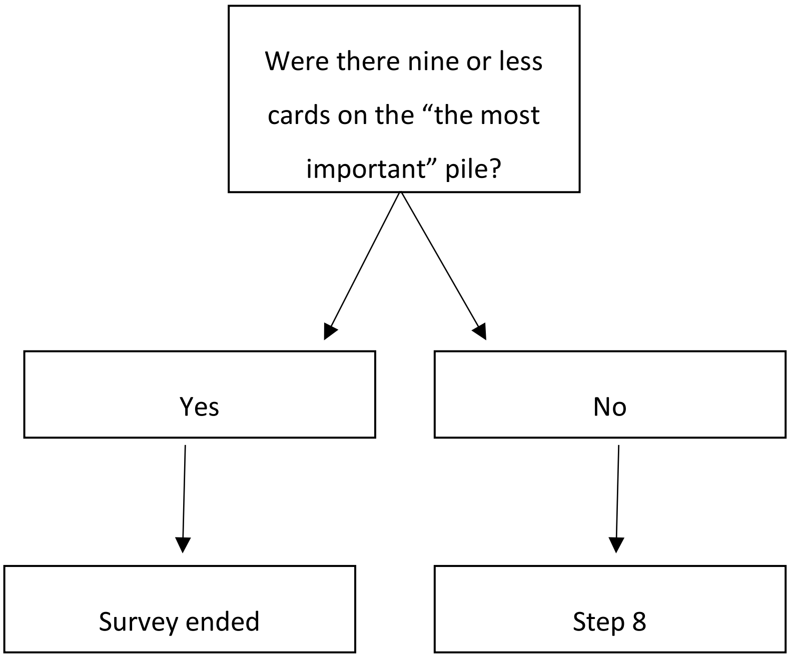
Step 5: The cards on the "very important" pile were excluded. The cards on the "Really, really important" pile were counted.



Step 6: From the "really, really important" pile, the participant was shown one card at the time and asked to put it one of the following piles:



Step 7: The cards on the "really, really important" pile were exclude. The cards on "the most important" were counted.



Step 8: The participant was shown a white board with the words "Top 9" on it. The board could only fit 9 cards. The participant was asked to select, from "the most important" pile only the top nine outcomes.

Figure A2: Pile of top nine cards as an example of use of the white board on step 8. The pictures on the following cards are free to use but require attributions:

Walking better being able to stand up and climb stairs: Designed by Freepik

Doing what you can do: Created by Asierromero - Freepik.com

Staying healthy and fit: Created by Pressfoto - Freepik.com

Feeling brighter: Created by Asier_relampagoestudio - Freepik.com

Enjoying the moment: Designed by Bearfotos / Freepik

Feeling useful and having a purpose: Designed by Pressfoto / Freepik

Reducing anxiety: Created by Asierromero - Freepik.com

Seeing the person not the dementia: Designed by Asier Relampagoestudio / Freepik

Further details on the cards and the process of development of this card sorting strategy are available on request via the corresponding author.

II. List of outcomes and levels of agreement per Delphi survey round.

Legend: * indicates outcomes that had been selected by less than 15% of the participants in the round one Delphi, but were kept on round two, as they had been mentioned by at least one person living with dementia in the interview study that preceded this Delphi. This was to safeguard the views of people living with dementia, as they were not included in the first Delphi round. # indicates an outcome that was excluded at the consensus meeting, but the panel of experts included its concept in the definition of a wider outcome that was included (e.g. "improving balance" was included in the definition of "preventing falls"). ‡ indicates an outcome that was not included in the Core Outcome Set, but a recommendation was made about activity delivery with regards to this outcome.

Note: the breakdown of percentages of agreement per stakeholder group is available on request.

Outcome domain in lay terminology	**Percentage of agreement round one, all participants (n=91)**	**Excluded after Delphi round two**	**Percentage of agreement round two, all participants (n=95)**	**Excluded after Delphi round two**	**Excluded at the consensus meeting (percentage of agreement to include)**
Preventing falls	40.7 %	No	67.4 %	No	No (100%) – Included in the Core Outcome Set

Outcome domain in lay terminology	Percentage of agreement round one, all participants (n=91)	Excluded after Delphi round two	Percentage of agreement round two, all participants (n=95)	Excluded after Delphi round two	Excluded at the consensus meeting (percentage of agreement to include)
Walking better, being able to stand up and climb stairs	38.5 %	No	56.8 %	No	No (100%) – Included in the Core Outcome Set
Seeing the person, not the dementia ‡	37.4 %	No	61.1%	No	Yes (0%)
Feeling useful and having a purpose	34.1 %	No	51.6 %	No	No (100%) – Included in the Core Outcome Set
Enjoying activity and feeling good about it	33.0 %	No	56.8%	No	Yes (0%)
Enjoying the moment	33.0 %	No	38.9%	No	No (100%) – included in the Core Outcome Set

Outcome domain in lay terminology	Percentage of agreement round one, all participants (n=91)	Excluded after Delphi round two	Percentage of agreement round two, all participants (n=95)	Excluded after Delphi round two	Excluded at the consensus meeting (percentage of agreement to include)
Having better wellbeing	33.0 %	No	46.3 %	No	Yes (0%)
Taking part and being included #	31.9 %	No	50.5%	No	Yes (0%)
Being more active	27.5 %	No	31.6 %	No	Yes (0%)
Feeling less depressed and avoiding depression	25.3 %	No	34.7 %	No	Yes (0%)
Having better self-esteem	25.3%	No	22.1 %	No	Yes (0%)
Reducing anxiety ‡	25.3 %	No	25.3 %	No	Yes (0%)
Improving balance #	24.2 %	No	28.4 %	No	Yes (0%)

Outcome domain in lay terminology	Percentage of agreement round one, all participants (n=91)	Excluded after Delphi round two	Percentage of agreement round two, all participants (n=95)	Excluded after Delphi round two	Excluded at the consensus meeting (percentage of agreement to include)
Keeping movement in the joints #	24.2 %	No	22.1 %	No	Yes (0%)
Being more sociable and chatty	23.1 %	No	18.9 %	No	Yes (64%)
Slowing down the dementia	23.1 %	No	22.1 %	No	Yes (73%)
Feeling brighter	22.0 %	No	18.9%	No	No (100%) – Included in the Core Outcome Set
Living life to the full	22.0 %	No	14.7 %	Yes	Not applicable
Feeling less bored	19.8 %	No	11.6 %	Yes	Not applicable

Outcome domain in lay terminology	Percentage of agreement round one, all participants (n=91)	Excluded after Delphi round two	Percentage of agreement round two, all participants (n=95)	Excluded after Delphi round two	Excluded at the consensus meeting (percentage of agreement to include)
Doing what you can do	18.7 %	No	17.9 %	No	No (100%) – Included in the Core Outcome Set
Having a healthy and balanced routine	18.7 %	No	10.5%	Yes	Not applicable
Improving brain health	18.7 %	No	13.7 %	Yes	Not applicable
Eating and drinking better	17.6 %	No	8.4 %	Yes	Not applicable
Moving more easily	17.6 %	No	14.7 %	Yes	Not applicable
Being able to do every-day tasks without help	16.5 %	No	12.6 %	Yes	Not applicable

Outcome domain in lay terminology	Percentage of agreement round one, all participants (n=91)	Excluded after Delphi round two	Percentage of agreement round two, all participants (n=95)	Excluded after Delphi round two	Excluded at the consensus meeting (percentage of agreement to include)
Catching moments of the “old” person	16.5 %	No	4.2%	Yes	Not applicable
Keeping going as normal	16.5 %	No	8.4 %	Yes	Not applicable
Making friends and building relationships	16.5 %	No	12.6 %	Yes	Not applicable
Meeting individual needs of patient and family	16.5 %	No	7.4 %	Yes	Not applicable
Easing pain	15.4 %	No	8.4%	Yes	Not applicable
Staying healthy and fit	15.4 %	No	25.3 %	No	No (100%) – Included in the Core Outcome Set

Outcome domain in lay terminology	Percentage of agreement round one, all participants (n=91)	Excluded after Delphi round two	Percentage of agreement round two, all participants (n=95)	Excluded after Delphi round two	Excluded at the consensus meeting (percentage of agreement to include)
Avoiding going to hospital or being discharged quicker	14.3%	Yes	Not applicable	Not applicable	Not applicable
Communicating through movement	14.3 %	Yes	Not applicable	Not applicable	Not applicable
Managing behaviours that challenge	13.2 %	Yes	Not applicable	Not applicable	Not applicable
Remembering happy times *	13.2 %	No	10.5 %	Yes	Not applicable
Feeling free and in control *	12.1 %	No	4.2 %	Yes	Not applicable

Outcome domain in lay terminology	Percentage of agreement round one, all participants (n=91)	Excluded after Delphi round two	Percentage of agreement round two, all participants (n=95)	Excluded after Delphi round two	Excluded at the consensus meeting (percentage of agreement to include)
Feeling more relaxed *	9.9 %	No	2.1%	Yes	Not applicable
Improving posture	9.9 %	Yes	Not applicable	Not applicable	Not applicable
Taking less medication	8.8 %	Yes	Not applicable	Not applicable	Not applicable
Achieving activity goals	7.7 %	Yes	Not applicable	Not applicable	Not applicable
Having more opportunities *	6.6 %	No	2.1 %	Yes	Not applicable
Keeping a healthy weight and a healthy heart *	6.6 %	No	11.6 %	Yes	Not applicable
Continuing with the same care	4.4 %	Yes	Not applicable	Not applicable	Not applicable

Outcome domain in lay terminology	Percentage of agreement round one, all participants (n=91)	Excluded after Delphi round two	Percentage of agreement round two, all participants (n=95)	Excluded after Delphi round two	Excluded at the consensus meeting (percentage of agreement to include)
Delivering better care	4.4 %	Yes	Not applicable	Not applicable	Not applicable
Managing pressure ulcers (bed sores)	4.4 %	Yes	Not applicable	Not applicable	Not applicable
Opening bowels	4.4 %	Yes	Not applicable	Not applicable	Not applicable
Preventing incontinence	4.4 %	Yes	Not applicable	Not applicable	Not applicable
Living longer	2.2 %	Yes	Not applicable	Not applicable	Not applicable
Becoming interested in a cause and getting others interested in what you can still do	1.1 %	Yes	Not applicable	Not applicable	Not applicable
Reducing swollen legs	1.1 %	Yes	Not applicable	Not applicable	Not applicable

III. Sample characterisation of people living with dementia

Participants living with dementia (n=20)	**Mean (SD) or n (%)**
Age (Years)	81.3 (±8.4)
Gender (Female)	10 (50%)
Years post diagnosis	3.8 (±2.8)
Physical Activity Scale for the elderly	52.0 (±45.1)
Living with the following dementia types (self-reported):	
Alzheimer’s Disease	5 (25%)
Vascular Dementia	5 (25%)
Mixed Dementia	4 (20%)
Dementia with Lewy Bodies	1 (5%)
Frontotemporal Dementia	1 (5%)
Not known	4 (20%)
Mini-Mental stage examination score	21.1 (±5.3)
Setting:	
Home or other community settings	11 (55%)
Sheltered accommodation	3 (15%)
Care or nursing home or assisted living	6 (30%)
Hospital	0 (0%)

IV. Demographic characteristics of participants who dropped out after round one. Please note that one participant may have supported people living with dementia in multiple settings and stages of disease progression.

Group 1: Family carers (n=8)	Mean (SD) or n (%)	Group 2: professionals (n=8)	Mean (SD) or n (%)
Age (Years):		*Age (Years):*	
18-29	0 (0%)	18-29	1 (12.5%)
30-39	2 (25%)	30-39	3 (37.5%)
40-49	0 (0%)	40-49	2 (25%)
50-59	3 (37.5%)	50-59	2 (25%)
60-69	1 (12.5%)	60-69	0
70-79	1 (12.5%)	70-79	0
80-89	1 (12.5%)	80-89	0
Gender (Female)	6 (75%)	*Gender (Female)*	8 (100%)
Years post diagnosis of the person with dementia he/she cares for	5.7 (±3.9)	*Years of experience in dementia care*	18.2 (±12.3)
Experience of d supporting physical activity per stage of disease progression (self-reported):		*Experience of d supporting physical activity per stage of disease progression (self-reported):*	
Mild to Moderate	5 (62.5%)	Mild to Moderate	5 (62.5%)
Moderate to Severe	4 (50%)	Moderate to Severe	1 (12.5%)
Severe	1 (12.5%)	Severe	0 (0%)
All stages	0 (0%)	All stages	3 (37.5%)

Group 1: Family carers (n=8)	**Mean (SD) or n (%)**	**Group 2: professionals (n=8)**	**Mean (SD) or n (%)**
Not known	1 (12.5%)	Not known	0 (0%)
Experience of supporting physical activity per setting:		*Experience in supporting physical activity per setting:*	
Home or other community settings	6 (75%)	Home or other community settings	7 (87.5%)
Sheltered accommodation	2 (25%)	Sheltered accommodation	1 (12.5%)
Care or nursing home or assisted living	3 (37.5%)	Care or nursing home or assisted living	4 (50%)
Hospital	1 (12.5%)	Hospital	3 (37.5%)

Appendix D Supplementary material Paper V (Prioritisation of negative side effects and carer outcomes)

Physical activity for people living with dementia: a prioritisation of carer outcomes and side effects from the perspectives of professionals and family carers

Gonçalves, A.C..; Demain, S.; Samuel, D.; and Marques, A. 2019. Submission being currently planned.

List of all carer outcomes considered during the prioritisation exercise round two, with ranking positions per stakeholder group.

All carer outcomes in lay terminology	Final overall ranking position (n=75)	Ranking position considering the carer stakeholder group only (n=36)	Ranking position considering the professionals stakeholder group only (n=39)	Definition, as in the glossary made available to participants
Carer feeling positive and satisfied	1	1	2	Carers feeling positive about the person living with dementia being active, improving and having a fulfilling time. Carers feeling proud of the person living with dementia and seeing them doing activities they used to do in the past. Carer having better self-esteem. In the literature this was

All carer outcomes in lay terminology	Final overall ranking position (n=75)	Ranking position considering the carer stakeholder group only (n=36)	Ranking position considering the professionals stakeholder group only (n=39)	Definition, as in the glossary made available to participants
				linked to confidence in their care abilities and carers' satisfaction with the intervention.
Carer improving wellbeing	2	2	1	Carer wellbeing and quality of life. Carer having fun.
Making the lives of carers easier	3	3	3	Physical activity may reduce the burden of care in the long-term by: maintaining functional independence of the person with dementia and finding the person living with dementia more agreeable to tasks, lightening the workload that need to be done by the carer; carer accessing support from professionals; and carer experiencing less challenging behaviour, including less sun downing from the person living with dementia. In the short-term, carers' lives can be made easier by: giving the carer a break; time and space to themselves or some respite,

All carer outcomes in lay terminology	Final overall ranking position (n=75)	Ranking position considering the carer stakeholder group only (n=36)	Ranking position considering the professionals stakeholder group only (n=39)	Definition, as in the glossary made available to participants
				while the person with dementia is involved in activity and needing less input from the carer.
Carer making friends and getting support	4	5	4	Family carers meeting other relatives of people with dementia and developing a network of friendship and peer support. Carers meeting other carers and having their carer role recognised.
Carer feeling less depressed	5	6	5	Lack of activity and engagement for the person living with dementia was linked to carer depression.
Carer improving mood	6	4	7	Carer feeling happy or being in better mood.

All carer outcomes in lay terminology	Final overall ranking position (n=75)	Ranking position considering the carer stakeholder group only (n=36)	Ranking position considering the professionals stakeholder group only (n=39)	Definition, as in the glossary made available to participants
Carer feeling less worried	7	9	6	Carer feeling less worried because the person living with dementia has taken part in activity.
Carer having better health	8	8	8	Includes weight management and overall wellbeing and health. In the literature this was measured as the carer's use of health and social care services and therefore linked to costs.
Carer getting better sleep	9	7	9	Improved carer sleep quality (specific sleep parameters not specified).
Carer being more active	10	10	10	Joining in with the person with dementia, adding to their habitual levels of activity.
Carer walking better	11	11	11	Carer mobility and balance, ability to walk with less joint pain.

All carer outcomes in lay terminology	Final overall ranking position (n=75)	Ranking position considering the carer stakeholder group only (n=36)	Ranking position considering the professionals stakeholder group only (n=39)	Definition, as in the glossary made available to participants
Carer living longer	12	12	12	Carer living longer as a result of the person living with dementia taking part in activity.

List of all side effects of physical activity, considered during the prioritisation exercise round two, with ranking positions per stakeholder group.

All side effects of activity for patients and carers in lay terminology	Final overall ranking position (n=75)	Ranking position considering the carer stakeholder group only (n=36)	Ranking position considering the professionals stakeholder group only (n=39)	Definition, as in the glossary made available to participants
Becoming agitated and confused	1	2	1	Becoming challenging, frustrated, rude or overstimulated during the physical activity. Refusing to go back into a care setting after a physical activity in a different environment. In some cases, physical activities with these effects were considered not appropriate for the person living with dementia and are often interrupted.
Falling over	2	1	4	Sustaining falls or increasing falls risk by being active. Sustaining injuries after a fall (e.g. fractures) and having to attend emergency care because of falls. Being about to fall, but being able to save oneself. Increasing fear of falling and reduced confidence in walking due to fear of a fall.

All side effects of activity for patients and carers in lay terminology	Final overall ranking position (n=75)	Ranking position considering the carer stakeholder group only (n=36)	Ranking position considering the professionals stakeholder group only (n=39)	Definition, as in the glossary made available to participants
Feeling discomfort and pain	3	3	2	Includes joint pain, muscle soreness or stiffness after exercising. Complaining of pain or experiencing physical discomfort during activity. Not being able to be as active as usual in the day(s) after the physical activity.
Having a bad experience	4	4	3	Triggering negative emotions or feelings (e.g. embarrassment or unhappiness). Having an unsatisfying experience. Physical activity reinforcing illness and loss of roles (e.g. letting the team down) and therefore having a negative impact on perceptions of wellbeing.
Feeling tired or exhausted	5	5	6	Feeling tired, drained, exhausted. Described as something that would stop the physical activity and that should be used to monitor the intensity of the physical activity.

All side effects of activity for patients and carers in lay terminology	Final overall ranking position (n=75)	Ranking position considering the carer stakeholder group only (n=36)	Ranking position considering the professionals stakeholder group only (n=39)	Definition, as in the glossary made available to participants
Getting hurt or accidently hurting others	6	6	5	Includes injuries to muscles and joints. May result from doing the wrong exercise or overdoing it; or from “bumping into” objects or fellow participants.
Making lives of carers harder	7	8	8	Carers may face an increase in workload: being/feeling responsible to offer physical activity and sometimes persuade them to join in activities. For carers, physical activity can imply effort, fatigue and negative emotions of guilt, frustration, embarrassment and worry about patient safety. It may increase risks of negative health outcomes for the carer.
Becoming unwell or having to go to hospital	8	10	9	Being hospitalised or admitted to an emergency department. Experiencing a deterioration of overall health or becoming too unwell to continue activity.

All side effects of activity for patients and carers in lay terminology	Final overall ranking position (n=75)	Ranking position considering the carer stakeholder group only (n=36)	Ranking position considering the professionals stakeholder group only (n=39)	Definition, as in the glossary made available to participants
Feeling dizzy, sick or fainting	9	9	10	Feeling dizzy, nauseous or light-headed. Having a syncopal episode (losing conscientiousness for a moment).
Becoming uncomfortable because of shortness of breath	10	7	11	Shortness of breath that is uncomfortable or more exacerbated than in normal exercise. Needing to “catch one’s breath” and interrupting the physical activity because of it.
Creating a conflict between the carer and the person living with dementia	11	13	7	Triggering an argument during a physical activity. Sometimes the conflict can start from the carer trying to guide the person living with dementia to do a physical activity that he/she refuses to do. Impacting on the relationship between the carer and the person living with dementia,

All side effects of activity for patients and carers in lay terminology	Final overall ranking position (n=75)	Ranking position considering the carer stakeholder group only (n=36)	Ranking position considering the professionals stakeholder group only (n=39)	Definition, as in the glossary made available to participants
				where the person living with dementia is “being told” what to do and has less choice.
Getting lost	12	12	12	Not being able to find the way back after a physical activity. Having to use technology to find the person with dementia.
Having a heart problem while exercising	13	11	13	Suffering a Transient Ischemic Attack or developing cardiac pathology. This is also a reason for caution when involving a person with dementia in physical activity.
Not being able to sleep after an activity	14	14	15	This was linked to possible changes of environment or routine caused by the physical activity (e.g. coming back to care after having been on holiday).

All side effects of activity for patients and carers in lay terminology	Final overall ranking position (n=75)	Ranking position considering the carer stakeholder group only (n=36)	Ranking position considering the professionals stakeholder group only (n=39)	Definition, as in the glossary made available to participants
Becoming more disabled	15	17	16	Becoming or feeling more disabled while doing physical activity after the diagnosis of dementia.
Increasing risk of death	16	16	17	Mortality. Number of deaths during physical activity or within the time period while the person with dementia was a participant in a physical activity intervention.
Going to a care home or nursing home	17	15	19	Being placed into an institution for permanent full-time care as a consequence of deteriorating health during the physical activity, or resulting in an interruption of participation in physical activity.
Carer feeling "heartbroken"	18	20	14	Carer having a sense of loss while seeing the person living with dementia being physically active.

All side effects of activity for patients and carers in lay terminology	Final overall ranking position (n=75)	Ranking position considering the carer stakeholder group only (n=36)	Ranking position considering the professionals stakeholder group only (n=39)	Definition, as in the glossary made available to participants
Forgetting the activity	19	18	18	Offering physical activity that the person with dementia is then not able to recall.
Accidently eating or drinking something harmful	20	19	20	Eating or drinking something that should not be ingested, by confusing it with real food or drink while being physically active (e.g. mistaking a cleaning product with an energy drink while in the gym; eating something from the garden while outdoors).
Finding cysts	21	21	21	Right ganglion cyst discovered in research participant of a physical activity intervention. The link between this and the activity was not made clear but the discovery of the cyst was considered a non-serious adverse event possibly related to study intervention.

Appendix E COS-STAD statement completed checklist

The following checklist is based on the COS-STAD publication by Kirkham et al. (2017).

SECTION/ TOPIC	ITEM No.	CHECKLIST ITEM	Location in the thesis document
Scope specification	1	The research or practice setting(s) in which the COS is to be applied COS developers should consider the details of the setting (e.g., for application in research studies or for use in routine care) that will be covered by the COS.	Page 27 under “scope”
	2	The health condition(s) covered by the COS COS developers should consider the details of the health conditions (e.g., treatment of rheumatoid arthritis or screening for cancer) that will be covered by the COS.	Page 27 under “scope”
	3	The population(s) covered by the COS COS developers should consider the details of the population (e.g., patients with advanced disease or children) that will be covered by the COS.	Page 27 under “scope”
	4	The intervention(s) covered by the COS COS developers should consider the details of the interventions (e.g., all interventions, drug therapy, or surgical interventions) that will be covered by the COS.	Page 27 under “scope”
Stakeholders	5	Those who will use the COS in research	Page 27-28 under “stakeholders”.

involved		COS developers should involve those who will do the research that will use the COS (e.g., clinical trialists or industry).	Final characteristics of the included stakeholders in pages 70, 95-96 and page 114
	6	Healthcare professionals with experience of patients with the condition COS developers should involve those healthcare professionals who would be able to suggest important outcomes (e.g., clinical experts, practitioners, and investigators with particular experience in the condition).	Page 27-28 under “stakeholders”. Final characteristics of the included stakeholders in pages 70, 95-96 and page 114
	7	Patients with the condition or their Representatives COS developers should involve those who have experienced or who are affected by the condition (e.g., patients, family members, and carers).	Page 27-28 under “stakeholders”. Final characteristics of the included stakeholders in pages 70, 95-96 and page 114
	8	The initial list of outcomes considered both healthcare professionals' and patients' views COS developers should consider the views of healthcare professionals and patients (most likely identified from literature reviews or interviews) when generating an initial list of outcomes for inclusion in the consensus process.	Paper III shows how the initial list of outcomes was generated – see particularly flowchart on page 71
	9	A scoring process and consensus definition were described a priori.	Consensus defined in the protocol – page 33

	Although different consensus methods may be employed in different studies, to avoid any potential biases, COS developers should describe their consensus method a priori.	
10	Criteria for including/dropping/adding outcomes were described a priori.	Consensus defined in the protocol – page 33
	COS developers should also prespecify criteria for including, dropping, or adding new outcomes to avoid potential biases.	
11	Care was taken to avoid ambiguity of language used in the list of outcomes.	All outcomes were described in lay terms – a process completed through PPI – see page 94 “use of lay terminology during the Delphi”.
	COS developers should consider the language used when describing outcomes in front of different stakeholder groups. An example of 1 approach taken is to include both lay and medical terms, with these previously piloted with the stakeholders.	

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