Alzheimer Europe

Overcoming ethical challenges affecting the involvement of people with dementia in research: recognising diversity and promoting inclusive research

The report entitled “Overcoming ethical challenges affecting the involvement of people with dementia in research: recognising diversity and promoting inclusive research” received funding under an operating grant from the European Union’s Health Programme (2014–2020)

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Preface

I believe my involvement in research saved my life following my diagnosis of dementia and I am passionate about public involvement.

Research ethics is a positive challenge; it forces us all, clinicians, academics and people affected by dementia, to think about dementia research in a critical and holistic manner. This paper by Alzheimer Europe is a timely response to that challenge.

I am immensely proud that the European Working Group of People with Dementia (EWGPWD) were key contributors to this work – in particular that our input allowed an improved understanding of a broader view of the benefit of research participation.

Research is about more than the topic being researched; it is about placing a value on our voice, realising that people with dementia have insights and worthwhile contributions. Research ethics are important but we must not get so concerned about ethics that we take an overly paternalistic view and exclude people living with dementia.

Researchers must understand that people living with dementia don't simply want to answer questions, we would like to set the research question and to do that we must have robust and ethical public involvement.

I am particularly delighted to see the issues of diversity and inclusion being addressed in this paper. There are millions of people living with dementia throughout Europe; people living in rural areas with no transport, people living alone with no support to prepare for and travel to meetings; people who do not use email, or whose sight is poor and need support with documents; people who live in a country where they do not speak the language. And they have lost their voice. I hope that this paper will support researchers to help them find their voice once again.

Alzheimer Europe is a leader in promoting a diverse dementia voice and this paper further supports that. Thank you to members of the ethics working group, chaired by Dianne Gove, for their work on this project and for drafting this paper, and to the members of the EWGPWD and the external advisors for their valuable input.

Helen Rochford-Brennan,
Chair of the European Working Group of People with Dementia

On behalf of the European Working Group of People with Dementia

European Working Group of People with Dementia
ALZHEIMER EUROPE

Introduction

What is research and why is dementia research important?

Research consists of a systematic, organised inquiry to find answers to worthwhile questions, using predefined methods or procedures which are clearly documented. The answers to such questions should contribute towards a body of knowledge or theory and it should be possible for other people to understand exactly what researchers did to arrive at their conclusions and any limitations there may have been to the study.

Research into the care, treatment and support of people with dementia is essential for the lives and wellbeing of millions of people who have or at some point will develop dementia. Such research often provides a means to develop new drugs and treatments, promote good health, find out what kinds of services and care people want, explore the needs and experience of professional and informal carers, and better understand what it means to live with dementia. Very often, research provides a basis for government policies, the development of guidelines and the creation or improvement of products, services, support, medication and procedures.

This discussion paper is about all kinds of research involving people with dementia. Involvement in research should be understood as including 'Public Involvement' and involvement as a research participant (formerly known as a research subject). Both forms of involvement are equally important and raise ethical issues, which are the focus of this discussion paper.

The relevance of different research paradigms for inclusive research

For quite some time, there were two main research paradigms[1], namely the positivist/post positivist paradigm (linked to quantitative research) and the constructivist (or interpretivist) paradigm (linked to qualitative research). A few decades ago, there were huge methodological debates as to which of the two paradigms was 'right'. These debates were based on disagreements about ontology (the nature of knowledge and what counts as knowledge or fact), epistemology (the source of knowledge and how knowledge is determined) and axiology (the aims of research and how researchers' own values are understood in relation to the research process). Some researchers argued that only the positivist/post positivist paradigm was 'real science'.

Others argued that it was not suited to the study of complex human and social phenomena. This debate came to be known as the paradigm wars[2] and some posited that the two approaches were irreconcilable due to their very different underlying philosophies.

Researchers have largely moved beyond this dispute, adopting a more pragmatic approach. This recognises the strengths and limitations of each and accepts that it is the type of question that determines/justifies which method is appropriate and valid. Pragmatists emphasise that the focus should be on the research problem and that pluralistic approaches should be adopted to derive the necessary knowledge about it (Creswell 2009). Nevertheless, perspectives and concerns reminiscent of these early debates are still sometimes evident amongst researchers, funding bodies and research ethics committees and may sometimes hamper efforts to render research more inclusive with regard to people with dementia. The reason for this is that the definition of science and hence of scientific research was, historically, developed within a positivist context and even now, some definitions still reflect positivist assumptions. Methods used to make research more inclusive, on the other hand, tend to draw on qualitative research methods (i.e. reflecting assumptions from the constructivist paradigm), which are still considered by some as not being 'scientific'. Hence, attempts to be more inclusive may be met by resistance from researchers, funders and research ethics committees who have a narrow perspective of research.

Another important issue of relevance to the promotion of inclusive research is that some researchers feel that the two key paradigms mentioned above do not adequately or appropriately address social injustice or do not go far enough in advocating for marginalised groups. The two key paradigms have been criticised for not actively pursuing the interests of minority groups with the result that members of such groups all too easily fall through the net and consequently are underrepresented in research. There was also a criticism that lay people were involved in research solely as 'subjects' or 'participants' and that they could be involved throughout the research process so as to ensure that their needs and preferences were taken into consideration.

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[1] "constellation of beliefs, values, techniques and so on shared by members of a given community" (Kuhn 1970, p. 175)

[2] See Baumgarten M (2012). *Paradigm Wars – Validity and Reliability in Qualitative Research. Grin Verlag and Tashakkori and Teddlie (1998). Mixed methodology. Combining qualitative and quantitative approaches.* London: Sage Publications

This has led to the development of the advocacy and participatory research paradigm in which researchers are often guided by a theoretical perspective (e.g. racialised or disability-based, feminist, critical or queer theory) and seek to engage people from marginalised groups actively in the whole research process as collaborators in research as well as research participants. The key issue is not whether a particular method is used but whether there is a shared commitment to bringing about social change and an emphasis on participants (usually from marginalised communities) benefiting in some way from having participated, particularly if this is not the norm, and from accessible findings (Pyett 2002). Whilst the participatory aspect of this paradigm shares certain goals with Public Involvement (see Part 1 of this paper), the advocacy and participatory paradigm is based on the premise that "research inquiry needs to be intertwined with politics and a political agenda", should address issues such as inequality, oppression, domination, suppression, and alienation, create political debate and bring about social changes (Creswell 2009, p.9). These are not necessarily aims shared by all researchers who nevertheless strive for a greater involvement of people with dementia in research.

Different research paradigms, types of research and research methodologies represent opportunities to develop an understanding of issues of relevance to the lives and wellbeing of people with dementia. They also come with certain challenges, an important one being how to ensure the ethical involvement of people with dementia in research. Such challenges may be greater with regard to certain issues or to the involvement of minority or marginalised groups. One paradigm is not better or worse than another. The key challenge is to ensure that the research is carried out ethically and in accordance with established procedures, standards and principles, whilst challenging and adapting these if and when necessary in order to involve a diverse set of people with dementia in research.

- **What springs to mind when you think of the term 'scientific'?**
- **What is your world view about research and about what counts as 'knowledge'?**
- **Do you think this has any impact on your approach to inclusive research?**

What is meant by the ethical involvement of people with dementia in research?

The involvement of people with dementia in research is part of a more global aim to ensure that research *per se* is ethically sound. All research must be worthwhile, effective and conducted in an ethical manner. Throughout history, there have been numerous examples of this not happening and this has led to the publication of various guidelines as well as the obligation for researchers to obtain ethical approval from research ethics committees in order to carry out their research. The Nuremberg Code (1948) was the first internationally agreed set of guidelines on the good conduct of research. It was published shortly after the end of the Second World War, mainly in response to public outcry over medical research carried out under the Nazi regime on people, by force and/or involving a lack of due concern for their wellbeing. However, unethical studies were not limited to the war period and did not stop with the Nuremberg Code. Other cases eventually came to light such as the Tuskegee syphilis study (1932 – 72), the human radiation experiments (1945–72), the Jewish Chronic Disease Hospital New York Study (1963), the Willowbrook study (1956) and several hundred others which were identified in reports by Beecher and Pappworth published in the 1960s (Savelescu and Hope 2010). The protective measures for research participants, laid out in the Nuremberg Code, were later incorporated by the World Medical Association into the Declaration of Helsinki (1964). These protective measures were also incorporated by the Council of Europe into the European Convention for the Protection of Human Rights and Dignity of the Human Being with Regard to the Application of Biology and Medicine (the Oviedo Convention) and supplemented in its Additional Protocol on the Convention of Human Rights and Biomedicine concerning Biomedical Research (2005).

Whilst much of the early emphasis was on protection from harm and respect for autonomy, behaving in an ethical manner towards people with dementia engaged in research is also about empowerment, rights, respect, equity and wellbeing. The biomedical ethical principles described by Beauchamp and Childress (2012) are now widely used by researchers from many disciplines and contexts. These principles represent an important means to help ensure the ethical involvement of people with dementia in research, emphasising not only respect for persons (their autonomy and dignity), beneficence (which could also cover the promotion of wellbeing and empowerment) and non-maleficence (i.e. protection from harm), but also the need for justice/equity, which is particularly important with regard to the issue of equal inclusion. A possible drawback to this 'principlist' approach is that more than one principle may be relevant and that the four principles do not necessarily capture all moral concerns. However, as suggested by Hunter (2010), such moral decision making on the basis of principles can also be supported by people with different ethical perspectives (e.g. derived from consequentialism or duty-based ethics). Abstract principles cannot be considered in strict isolation of the context and the people involved but may form a useful starting point for a broad ethical consideration incorporating, for example, elements of:

- virtue ethics (which focuses on the character of the person carrying out an action),
- ethics of care (which focuses on people within a complex net of relationships and the importance of empathy and solidarity),
- communitarism (which focuses on the maintenance of the community based on recognition that people are embedded within culture and society) and
- discourse ethics (which emphasises the need to ensure that moral norms are established through a fair dialogue in which everyone's perspectives and viewpoints are heard and taken seriously).[3]

The ethical criteria for clinical research described by Emanuel et al. (2000) are also widely cited and provide guidance to help ensure that clinical research is robust, has value and is meaningful, providing society with accurate answers to questions that were worth asking. They cover: 1. value (linked to the enhancement of health or knowledge), 2. scientific validity (methodologically rigorous), 3. fair subject selection (guided by scientific objectives not vulnerability or privilege, with a fair distribution of risks and benefits), 4. a favourable risk-benefit ratio, 5. independent review (with the possibility to approve, amend or terminate studies), 6. informed consent and 7. respect for enrolled research participants (protection of privacy, possibility to withdraw and monitoring of participants' wellbeing). Emanuel et al. (2000) claim that the requirements are universal but that they must be adapted to the health, economic, cultural and technical conditions in which research is conducted.

Alzheimer Europe promotes a rights-based approach to the involvement of people with dementia in its work and subsequently in all research in which it involves people with dementia. This falls within the scope of a deontological approach based on the notion that there is something morally important about being human, that this gives rise to certain rights shared by all humans and that other people have a duty to respect those rights (Hunter 2010). Article 31 of the United Nations Convention on the Rights of Persons with Disabilities (CRPD)[4], states that appropriate information, including statistical and research data, must be collected to formulate and implement policies. In keeping with the guiding principle of the Convention, which is that of inclusion, a broad range of people with dementia should be involved. Moreover, reference to the right of people with disabilities[5] to reasonable accommodation (Article 2) means that appropriate adaptations must be made to ensure that people with dementia, for example, have the same opportunities to take part in research as other people. Under the CRPD, people with disabilities include "those who have long-term physical, mental, intellectual or sensory impairments which in interaction with various barriers may hinder their full and effective participation in society on an equal basis with others" (Article 1). Ethical involvement of people with dementia in research can also be linked to the PANEL principles of Participation, Accountability, Non-Discrimination and Equality, Empowerment and Legality, to the International Ethical Guidelines for Biomedical Research Involving Human Subjects[6] and to the EU Clinical Trials Directive[7].

The ethically sound conduct of research is further regulated and monitored by national, local and university ethics committees throughout the world in relation to medical and non-medical research involving human participants. Members of research ethics committees are likely to have different approaches and this is important in ensuring that some ethical issues are not overlooked as a result of an over-emphasis on a single approach to ethical decision making (Hunter 2007). Guidelines at national level may also apply in specific or multiple research domains, such as the Code of Human Research Ethics of the British Psychological Society (2014)[8] or the Code of Ethics for Scientific Research in Belgium (2009)[9].

Ethically sound involvement in research is also about who sets the research agenda, who is involved, at what stage and in what capacity. These issues have been influenced by broad historical developments such as the feminist, disability, intellectual disability and black civil rights movements, and consumerist debates (Gradinger et al. 2013). According to Beresford (2019), feminists and disabled people in particular challenged some of the deeply engrained foundations of positivist research such as distance, neutrality and objectivity, and questioned the independence of mainstream research with regard to funding often based on commercial priorities and reflecting the values of healthcare systems. Awareness of the need to involve people affected by various medical conditions in decisions related to the research agenda has led to some initiatives, such as the James Lind Alliance (funded by the National Institute for Health Research in the UK), which bring together people directly

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[3] A clear overview of different ethical perspectives of relevance to the ethics of research can be found in Chapter 1 of the European Textbook on Ethics in Research (European Commission 2010).

[4] Please see: https://www.un.org/development/desa/disabilities/convention-on-the-rights-of-persons-with-disabilities/convention-on-the-rights-of-persons-with-disabilities-2.html

[5] For a discussion about dementia as a disability, please see Alzheimer Europe's 2017 report on this topic: https://www.alzheimer-europe.org/Publications/Alzheimer-Europe-Reports

[6] Council for International Organizations of Medical Sciences (2002)

[7] This will be replaced by the Clinical Trials Directive which will come into application in 2020. See: https://ec.europa.eu/health/human-use/clinical-trials_en

[8] https://www.bps.org.uk/sites/bps.org.uk/files/Policy/Policy%20-%20Files/BPS%20Code%20of%20Human%20Research%20Ethics.pdf

[9] https://www.law.kuleuven.be/canon_law/education/scientific_integrity/ethischecodeen.pdf

affected by the condition, carers and clinicians to influence the choice of issues to be addressed and where possible to influence the funding of such research[10]. The aim of the James Lind Alliance is to create Priority Setting Partnerships (PSPs) to identify and prioritise key unanswered questions in a wide range of medical conditions, thereby ensuring that health research funders are aware of the issues that matter most to the people likely to be affected by research in their everyday lives. This corresponds to the ethical goal of ensuring that research addresses worthwhile questions (including questions that are worthwhile to people with dementia).

Any research involving human participants should have anticipated social value for society. In relation to clinical research, this is defined as being "the nature and magnitude of the improvement [*that*] the intervention is expected to have on the wellbeing of patients, individuals in society or society" (Habets, van Delden and Bredenoord 2014, p.3). However, the necessity to involve people with dementia in decisions related to priority setting for research should not deny or undermine the importance of researchers' right to scientific curiosity. As pointed out by Flinterman, Broerse and Bunders (2007), basic research initiated on this basis with no direct reflection about its relevance to the current interests of patients has sometimes led to important breakthroughs in knowledge development and innovations. Important knowledge gained from research does not have to have immediate practical ramifications for it to be deemed to have value (Emanuel et al. 2000). In a series of articles on increasing value and reducing waste in biomedical research published in the Lancet[11], Chalmers et al. (2014) suggest that it is in keeping with the nature of science to study things that are uncertain and which do not always result in worthwhile achievements for patients. They add, however, that funders should be more transparent about how they prioritise important uncertainties and how they take into account the needs of people for whose lives the findings of research may be relevant.

The ethically sound involvement of people with dementia in research needs to be considered in relation to Public Involvement (see Part 1 of this paper) and in relation to people who are research participants, stretching from the very beginning of the research process (e.g. during the conceptualisation and design phase) up to and even beyond the end of the research project when the findings are disseminated (see Part 4 of this paper). The people involved should be respected and treated as equals. This means that their input should be taken into consideration and valued, and measures should be taken to empower, where possible, and promote the wellbeing and safety of all involved (i.e. people with dementia and researchers alike). The ethically sound involvement of people with dementia in research is not limited to the protection of vulnerable research participants but also to the empowerment of people with dementia based on a reciprocal relationship of respect. In this discussion paper, we will be looking at such involvement both in the context of Public Involvement (PI) and as research participants.

Why is further reflection on the ethical involvement of people with dementia in research needed?

Much of the widely used ethical guidance on research was developed in the context of biomedical and clinical research. The various documents have influenced the actions and thoughts of researchers, research ethics committees and funding bodies for several decades and have been meaningfully applied in numerous other research contexts and domains. However, research is constantly evolving, along with knowledge, understanding of dementia (as a medical condition and a personal experience) and attitudes and practices linked to the involvement of people with cognitive difficulties, such as people with dementia, in research.

Research ethics committees also have a key role to play in promoting the ethically sound involvement of people with dementia in research but concerns about promoting wellbeing and avoiding harm are not always conducive to the effective and meaningful participation of people with dementia in research. Many people with dementia continue to be excluded on the basis of measures actually designed to protect them from harm, on the basis of structural discrimination or inadvertently through lack of awareness of their needs, interests, circumstances or difficulties. This is unfortunate as they have important knowledge to share.

Consequently, it is important to be open to different ways of interpreting and applying ethical guidance to new situations, challenges and goals. It is necessary to look critically at existing guidelines and for research ethics committees, researchers and funders to consider novel ways to involve people with dementia in research, balancing their needs and interests with requirements for good quality research. This should at best promote and at least not hinder the ethically sound involvement of people with dementia in a broad range of research settings and domains.

In accordance with the principle of justice, the benefits of research should be equally distributed amongst all members of society (Smith 2008). Research findings

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[10] In America, the Patient-Centred Outcomes Research Insititute (PCORI) promotes 'PCOR' research. This identifies evidence gaps with patients, researches issues which matter to patients and uses the findings to improve healthcare in a constant cycle in which research and practice constantly inform each other. See https://ahrq.gov/pcor/dissemination-of-pcor/index.html

[11] These documents can be found at: https://www.thelancet.com/journals/lancet/article/PIIS0140-6736(13)62329-6/fulltext

must correspond to the needs and interests of everyone with dementia in order to ensure an equal chance of good health, well-being and quality of life for people with dementia from minority groups in Europe. This should also help ensure that the strengths and valuable contribution of people from minority groups to societies are visible and acknowledged. Research findings are used either directly (e.g. in healthcare professionals' daily practice) or indirectly (e.g. in policy or education). However, people from minority ethnic groups, as well as other people in socially vulnerable situations, are underrepresented in research, both in the context of shaping and conducting research, and as research participants. Their lack of involvement in research cannot be dismissed on the grounds that they are not interested in research, would not have the capacity to take part or are 'hard to reach'.

There are numerous factors which hinder inclusive research such as invitations which are full of complex sentences and difficult words, proposing meetings in unfamiliar places, certain inclusion criteria and complicated consent procedures. Even when people are included, research materials, data collection tools and procedures may be culturally inappropriate and not validated, thus reducing the potential value of the involvement of people from some groups. Many of these factors can be addressed directly by researchers, research ethics committees and funders. Some may require a more substantial effort over time (e.g. being inherent in existing structures and procedures of organisations and deeply engrained in attitudes and perceptions).

People with dementia do not form a homogeneous group. They come from all walks of life and may have different needs and experiences based on the underlying conditions and causes of their dementia (e.g. the underlying disease or condition, whether they have early onset, familial or late onset), how far advanced their condition is and different symptoms related to their condition. There are also numerous sub-groups of people with dementia whose experiences and needs should be viewed as the result of dynamic intersections between different shared characteristics and collective experiences (Jubany, Güuell and Davis 2011). Vertovec (2007) highlights the "super diversity" of minority ethnic populations, drawing attention to the rising diversity within groups in relation to country of origin, socio-economic status and religious and cultural traditions. Such differences can, according to Uppal, Bonas and Philpott (2014), impact on defining individual norms, values and experiences. The diversity of the experience of dementia needs to be reflected in research.

About this discussion paper

Purpose, authors, contributors and target group

The purpose of this discussion paper is to promote reflection about the ethical involvement of people with dementia in research. The paper was drafted in 2019 by a group of researchers with expertise in inclusive dementia research and ethics (hereafter referred to as 'the ethics working group'), some of whom also have experience as members of research ethics committees. This working group was set up and chaired by Alzheimer Europe. Please see Appendix 1 for a brief bio of each of the co-authors of this paper and a list of the names of external experts who commented on the paper.

The European Working Group of People with Dementia (hereafter referred to as the EWGPWD) supported the ethics working group by providing input on various sections of the discussion paper and reviewing the recommendations. There were group discussions before and during the drafting of the text which influenced the issues addressed and the recommendations made. This method of involvement was chosen by the members of the EWGPWD. It consisted of a bottom-up approach whereby people with dementia were not provided with a summary of the issues considered as important to the ethics working group but rather were asked about the issues that they considered important. Their views were then fed back to the ethics working group who added their own contribution, debated these and additional issues and reviewed the relevant scientific and grey literature. In view of the length and complexity of the resulting paper, different members of the EWGPWD were invited to contribute further to different parts of the text.

The two groups agreed that in order to promote inclusive research, it was important to reach those who plan, conduct, evaluate and fund research. This discussion paper is therefore targeted at researchers, research ethics committees and funders.

The members of the ethics working group and the EWGPWD recognise that we, like many researchers, people with dementia, funders and members of research ethics committees, are looking at the issue of the ethical involvement of people with dementia in research through a particular research paradigm or lens. This reflects to some extent dominant Western-orientated values, assumptions and priorities and risks blinding us to the experience of many people with dementia from minority groups. For this reason, we have tried to be objective, to put ourselves in the shoes of other people and to be reflexive about our own influences, assumptions and attitudes. Part 3 of this paper addresses the issue of reflexivity and positionality and questions for reflection have been included at various points throughout the text. We have also included testimonials from members of the EWGPWD who provide a personal perspective on some of the key issues addressed, which they highlighted in discussions prior to and during the drafting of this paper.

Scope

In order to avoid duplication of effort and for the ethics working group to be able to address the many possible issues related to the ethical involvement in research within the timeframe of the project, the scope of this discussion paper has been deliberately limited to people who have dementia. We will therefore not be addressing issues linked to the involvement of people with normal cognition, pre-clinical Alzheimer's disease or mild cognitive impairment in dementia research. This should not in any way be interpreted as overlooking the important contribution that people from these groups who do not have dementia make to dementia research. However, a considerable body of reflection has and is still being carried out into ethical issues linked to the involvement of people from these three groups in dementia research by ethics working groups in the context of recent and current European projects (e.g. EPAD, AMYPAD and ROADMAP, to name but a few[12]). Similarly, the issue of data protection (covering privacy, confidentiality and other issues relevant to the ethical management of personal data) will not be addressed in this discussion paper as considerable reflection on these topics is currently underway in the context of a 3-year, IMI-funded project called 'Neuronet'.[13]

The paper is about the involvement of people with any kind of dementia in research, not just Alzheimer's dementia. It is relevant to researchers, funders and research ethics committees involved in any kind or branch of research and in any discipline (e.g. quantitative, qualitative, mixed methods, clinical etc.). Whilst much of the focus will be on dementia research, it should not be assumed that the involvement of people with dementia should be limited solely to dementia research.

Frequent reference will be made to minority groups. This should be interpreted as referring to groups of people having one or more characteristics in common which puts them at risk of exclusion in the context of research. This might, for example, be based on age, ethnicity, health status, gender identity, complex support needs, level of education, socio-economic status, disability, intellectual disability, difficulties with language, learning or literacy (including computer literacy and health literacy), place of residence (including care homes, prisons, being homeless and being a member of traveller communities) and communication issues such as having combinations and degrees of deafness and blindness. The list is not exhaustive. Other terms are occasionally used such as marginalised and disadvantaged (in this paper mainly in relation to discussions about intersectionality), both of which have slightly different connotations. Marginalised, for example, suggests that people are living on the boundaries of or outside society, and this may sometimes be through choice. As Pyett (2002) points out, they are still members of society and may actually consider mainstream society as marginal to their values and lives and there are some powerful minority groups who are not disadvantaged[14].

Finally, we recognise that most people live within a social web of interdependent relationships with others (e.g. relatives, friends and wider communities). They are not isolated entities. For many people, the views and experience of others are important to them. Moreover, their participation in research may have a psychological, emotional, financial or other impact on close friends and family. Relatives of people with dementia often play an important role in research as participants in their own right, in the context of PI and through the support they provide, which enables people with dementia to participate in research. It is important that they are also engaged in research in an ethical manner. The emphasis of this paper is on the involvement of people with dementia in research, but respect for individuals and families from different ethnic groups also entails recognition of the importance for many people of the family (e.g. in decision making and supporting involvement).

Structure of the discussion paper and how to use it

This discussion paper is divided into four sections:

1. ethical challenges linked to Public Involvement (PI)[15]
2. ethical challenges linked to recruitment and to informed consent
3. ethical challenges during participation in research
4. ethical challenges linked to involvement after the end of research

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[12] See http://www.alzheimer-europe.org for information about these projects and links to their respective websites.

[13] The Neuronet project will run until 2022, will consolidate the ethics work from 15 IMI-funded neurodegeneration projects, specifically relating to patient privacy, data protection and confidentiality.

[14] Our focus is not on powerful minority groups as they are unlikely to be excluded from research.

[15] Please see Part 1 for a discussion about this term.

The paper addresses a large number of issues and refers to the situation of a broad range of people with dementia. Examples are provided about the experience of people with various characteristics or from specific minority groups. This should not be interpreted as implying that only these groups are concerned. It was not possible to mention every minority group or person affected by non-inclusive research practices. It is nevertheless possible that we (i.e. the authors) have overlooked a particular issue that is important to the topic of this paper. We would welcome feedback from readers about specific issues of relevance to the promotion of inclusive research with people with dementia, which have not been addressed.

Readers may not read each part of the discussion paper or all the sections and sub-sections with the same level of interest and attention. Some may prefer to dip in and out of the different parts of the discussion paper. With this in mind, we have included several summaries as well as cross references in the text, partly to avoid repetition and partly as readers may have missed the initial discussion of an issue which is revisited later in the text. On the other hand, and in keeping with the concepts of reflexivity and positionality (see Part 3), the lack of interest in particular issues is perhaps revealing in itself and may have some relevance to the issue of inclusive research.

Throughout the text, readers will find testimonials from people with dementia and a few questions to encourage reflection. A glossary can be found in Appendix 2 containing some of the key concepts covered in the paper and there are recommendations interspersed throughout. These are, where appropriate, separated into recommendations for researchers, recommendations for research ethics committees and recommendations for research funders.

Part 1: Ethical Challenges Linked to Public Involvement

What is meant by Public Involvement?

People with dementia can contribute towards research in different ways. The organisation 'INVOLVE'[16] makes a useful distinction between three possible ways.

Involvement	This consists of the active involvement of people in research projects and in research organisations other than as research participants.
Participation	This consists of people taking part in a research study as a research participant (formerly often referred to as a research subject).
Engagement	This consists of people receiving information and being informed about a research study.

The term 'public' is understood as including patients and potential patients, informal (unpaid) carers, parents and guardians, people who use, or have used, health and social care services, people with disabilities, but not people who are employed as health or social care professionals, or academics (INVOLVE 2017).

In Part 1 of this discussion paper, we focus on the contribution of people with dementia to research in the context of Public Involvement and in Part 2 on their contribution as research participants (i.e. providing researchers with data for their studies).

The concept of Public Involvement

The term 'Public Involvement' (PI) is usually understood as meaning carrying out research and developing policies with or by members of the public and patients rather than on or for them. This is how the term is used in much of the literature on this topic. Although the term covers a broad range of people, the scope of this discussion paper is limited to the involvement of people with dementia in PI. Consequently, discussions surrounding PI in this paper relate to the involvement of people with dementia. Please see the sub-section 'a word about terminology' for more details.

PI is not about merely raising awareness or providing information about ongoing or completed research (sometimes referred to as patient engagement) or about being a research participant. Rather, it is about creating a partnership between researchers and the public/patients, whereby all contribute collaboratively in varying degrees towards the research process or the research output. It is not a specific method but an approach to involving people in research other than as research participants. PI could be considered as an overarching term which groups together a wide range of approaches and methods (see Figure 1).

> ▶ ***"I share this disease with millions of other people – so why should I be ashamed of it? On the contrary, I want to speak out about how it feels to live with dementia. You might all have a long history of research in the field of dementia but we, who live with dementia, can tell you much better about our demands, needs and wishes. We are the real experts of our condition! Don't use me but involve me!"***
> **(Angela, person with dementia, Austria).**

PI is not rigidly defined according to the extent of the contribution to a particular research project or to the nature of the task. The nature and extent of the involvement may differ from one research project to the next. It can occur along a continuum from involvement in an isolated task, through involvement at several or all stages of the research process up to full involvement as a core member or leader of the research team. Furthermore, different people may be involved at different stages of the research or in different tasks.

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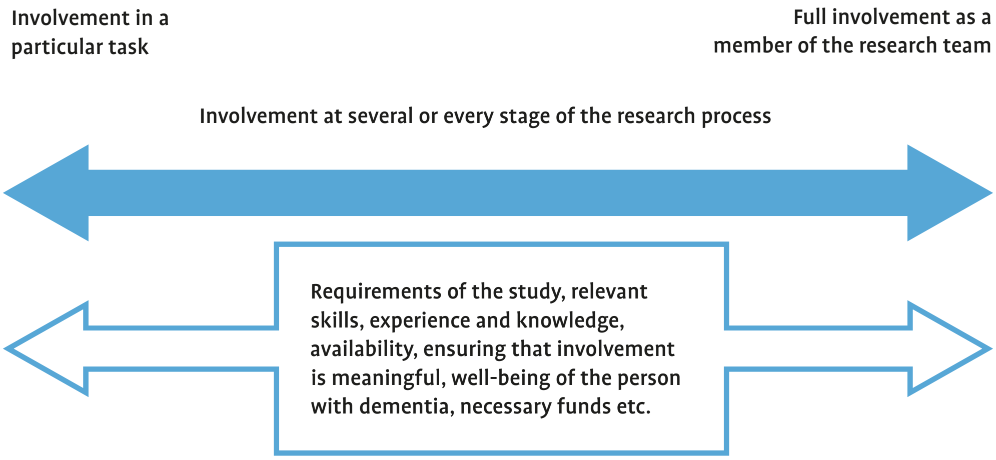
[16] Please see the INVOLVE website which contains a wealth of information on this topic: https://www.invo.org.uk/

Figure 1: Extent of involvement in PI and issues affecting involvement



Methods used in PI may sometimes resemble those used in qualitative research such as interviews and focus group discussions. The use of specific methods does not determine whether something is PI or research. Rather, it is the reason for using a particular method and how the information is used that are the determining factors. If the information is used as data to answer a research question, then it is research; if the information is used to develop or improve the research process, or if involvement promotes democracy, the right to voice, justice and fairness, then it is PI. Hoddinott et al. (2018, p. 3) summarise this as follows:

> *"Discussion with patients at a workshop can seem similar to collecting data in a focus group, because both involve listening to patients' perspectives, but the context and outcomes from listening differ. PPI means that researchers are in a continuing and reciprocal relationship with patients and make decisions with them about the research. In qualitative research, researchers listen to patients in order to improve their understanding of a topic".*

The following provides a brief overview of some of the key areas in which people with dementia might typically be involved in PI. These activities are ideally planned and organised in such a way that they are accessible to a broad range of people and do not require prior knowledge of research. This is not an exhaustive list.

- Providing insight into the experience of living with dementia (in the context of a consultancy role, not as research data),
- Monitoring plans and the conduct of research in order to help prevent the exploitation and maltreatment of people with dementia,
- Influencing decisions about which studies to fund,
- Helping to shape and define research questions so that they reflect the needs, priorities and interests of people with dementia (e.g. with individual researchers or through priority setting initiatives such as the James Lind Alliance in the UK mentioned in the introduction),[17]
- Sitting on ethics panels and project steering/advisory committees,
- Improving recruitment (especially of people from groups which appear difficult to reach),
- Ensuring transparency and accountability of the research process,
- Offering the perspective of a person with dementia about the topic, research question or methods,
- Encouraging reflexivity amongst researchers (see Part 3 of this discussion paper),
- Challenging set views and restrictive paradigms (either explicitly or simply by their presence),
- Contributing towards the design of methods and tools which are more appropriate and accessible to people with dementia,
- Contributing towards the collection of data and the interpretation of findings,
- Helping ensure that outcomes will be relevant to people with dementia (e.g. that services will be appropriate and hence used),
- Contributing towards accessible findings,
- Helping disseminate accessible information about the study.

How can researchers best describe what PI is about and attract people with no prior experience or knowledge about research to it?

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[17] More more information and a guidebook, please see: http://www.jla.nihr.ac.uk/jla-guidebook/

The rationale for Public Involvement

The above list of areas in which people with dementia contribute towards research through PI provide some indication as to why PI is important for research and researchers. The rationale for PI is broadly based on two key arguments (Ives, Damery and Redwood 2013, Gradinger et al. 2015) which also highlight its importance for people with dementia, members of the public and society in general.

- The first, based on normative arguments, is sometimes described as ideological or process orientated. It emphasises ethical, social and political concerns. PI is portrayed as 'an end in itself', linking involvement to democracy (e.g. democratic decision making, public accountability, legitimisation and transparency), people having rights (e.g. a right to voice, a right to be involved in research relevant to one's own condition) and to ethical principles of justice and fairness. It has also been argued that research 'done to' rather than 'with' people could be considered a breach of autonomy even though research participants provide informed consent to participate in research and those involved in PI are seldom also participants in the same study (Ives et al. 2013).
- The second, based on substantive arguments, is sometimes described as pragmatic, portraying PI as a 'means to an end', linked to attempts to improve the quality, validity, relevance and/or utility of research (both from a research and user perspective). It can also be considered as instrumental in providing knowledge that might otherwise be missing. This includes, for example, highlighting issues and asking questions about things that researchers have perhaps not considered. Tritter and McCallum (2006) suggest that key contributions often arise from personal experience and a non-medical or technical frame of reference.

☞ **Do values such as 'democracy' and 'a right to voice' have different meanings for different people?**

Some researchers argue that the rationale for involving 'patients' is different to that for involving the public (McCoy et al. 2018). They suggest that people who have experience of a particular disease or condition are well suited to being involved in health research or policy related to their own treatment or care. The public, or so they argue, should be involved in decisions related to research or policy with wider implications (e.g. the allocation of scarce resources or for matters which require the ability to make trade-offs between competing values and interests) for which a degree of neutrality, with no established perspective and a willingness to consider competing perspectives. They associate public involvement with the goals of democratic representation, accountability and transparency (similar to some of the goals described in the first argument above) and patient involvement with some of the arguments described in the second argument above (e.g. sharing their unique experience of a condition) (McCoy et al. 2018).

However, the two arguments/rationales described above, in the context of involving people with dementia in PI, are equally valid. It may be important to involve a broader set of people (i.e. not restricted to those with a certain condition) in research for some issues (which are of relevance to a broader range of people) but this does not detract from the goal of involving people with dementia in all kinds of research as a means to ensure transparency, legitimacy and accountability. Similarly, it would be wrong to assume that people with dementia (or any other condition) are incapable of considering competing perspectives fairly just because they have a condition which is particularly relevant to the debate.

In the past, it was largely assumed that people with dementia were unable to express their opinions or share their experience. This often resulted in them being silenced and in some cases represented by informal or professional carers who do not always have the same perspectives or fully understand their views and experience. The modern disability movement, which started in the 1960s in America, brought people with disabilities together to fight for a common cause. The people with disabilities, mainly physical disabilities and sensory impairments at the start, argued that they were best placed to determine their needs and voice their interests. They coined the phrase 'nothing about us without us'. People with dementia are now part of this movement and it is important that they have a seat at the table and are listened to.

PI represents a step towards recognition of the importance of hearing the voices of people with dementia and addressing what Flicker describes as 'epistemic injustice'. This consists of 'testimonial injustice' whereby less credibility is granted to people on the basis of social prejudices (e.g. consisting of unjustified beliefs that they are incompetent) and 'hermeneutic injustice' whereby marginalised groups are deprived of the opportunity to contribute their social experiences to a comprehensive understanding of a particular event or condition and are harmed as a consequence (Jongsma, Spaeth and Schicktanz 2017). The harm arising from epistemic injustice in the context of research is that when research is not inclusive of people with dementia (particularly from minority and less heard groups), those people are silenced and thus discriminated against. It is now increasingly accepted that people with dementia have an important contribution to make to research and PI is increasingly required for research funding or ethics approval in some countries. However, whilst the concept is gradually becoming more common in the field of dementia research, the practice is not yet widespread across the whole of Europe.

How Public Involvement differs to other approaches to involvement in the research process

We mentioned earlier possible confusion about the difference between PI and research in relation to the various methods used. There is also sometimes confusion about how PI differs from other related concepts such as co-production, co-design and co-creation which are specific approaches to PI.

To complicate matters, the term ‘engagement’ is used by some organisations and individuals to describe what others would call ‘involvement’ and vice versa. In the United States of America and Canada, and in Europe, for example in research linked to the Innovative Medicines Initiative (IMI), the term ‘engagement’ is typically used to denote what we describe in this paper as ‘involvement’.

There are some similarities between participatory research and PI, such as seeking to include and give a voice to people who are typically marginalised. A major difference is that whereas participatory approaches typically seek to involve *all* relevant stakeholders from a particular community, both as advisors and as participants with the aim of bringing about social change (Knowles, Voorhees *and* Planner 2015), PI focuses on people with a direct experience of a particular condition as distinct from people being research participants. Knowles et al (2015) nevertheless suggest that researchers might benefit from looking at areas where one approach could learn from the other (e.g. PI could perhaps learn more about going out into relevant communities and meeting people in their own spaces, doing things their way and on their terms and conditions).

Sustainability

Researchers often experience difficulty recruiting a sufficient number of participants for their studies and PI means seeking to involve an even greater number of people in research. It could be argued that there is a need to consider the sustainability of PI and the possible synergetic burden. In other words, does the goal of involving people in research through PI and as research participants risk placing too many demands on people with dementia and would people with dementia who might otherwise have become research participants opt for PI instead? Whilst this issue is worthy of attention, it should not be assumed that the same people are attracted to either role in research or that involvement in the context of PI would rule out being a research participant at another point in time (or vice versa).

On the other hand, PI is a relatively new concept, not only for people with dementia but also for many researchers. Researchers therefore need to be clear about their aims and needs in relation to their research and to make it clear to people with dementia what these are and what it might bring to those people to contribute to research either through PI or as a research participant. Some people with dementia will be more drawn towards participation in research involving little responsibility, interaction or reflection beyond what is asked of them as research participants, and would not be interested in getting involved in discussions with researchers, for example, about research design and procedures. Some might simply be interested in the topic or the goal of a particular study (e.g. to improve care or test a new drug) or agree to be involved in either PI or as participants based on having been asked by a respected person or gatekeeper. Others might find PI activities more rewarding or motivating but may assume that they need to have experience in or knowledge of research (which should not be necessary). It is therefore important for sustainability to ensure that people with dementia fully understand that there are different ways of contributing towards research, that they have options. The co-existence of both approaches should not be seen as competition for a limited set of people but rather as an opportunity to enable more people to contribute towards research in a way that they consider meaningful and personally rewarding.

▶ ***“Six months ago, I was diagnosed with dementia and really wanted to do something constructive. I wanted to play a role in finding a solution to this terrible disease. I volunteered for a clinical trial but unfortunately had a bad reaction against the drug and eventually withdrew from the study. Then I heard they were looking for people with dementia to do ‘PI’ for another study. This was completely new to me but I decided to give it a try. Now, I really feel I’m doing something useful and am pleased to be contributing towards research in this way”*** **(Juan, person with dementia, Spain).**

A word about terminology

The term ‘involvement’ is sometimes preceded by the term ‘public’, ‘public and patient’ or just ‘patient’. The reference to patients is increasingly considered as problematic in that a lot of people with dementia do not see themselves as patients and are voicing their concerns about being ‘positioned’ in this way. Everyone who consults a medical doctor is at that moment a patient but this is not the sum total of a person’s identity. A ‘person first’ approach to the concept would suggest emphasising the person rather than the fact that they have a medical condition. This, however, would include the entire population, whereas Public Involvement, and variations of this term, is about involving people either because they have lived experience of a particular condition and/or because they are members of the public (i.e. not health or social care professionals or academics).

In 2017, when Alzheimer Europe published its position paper on this topic, co-authored by INTERDEM[18] and the EWGPWD, the term 'public and patient involvement' was preferred. The topic of terminology was raised in a parallel session at Alzheimer Europe's annual conference in The Hague in 2019, which was chaired by the Vice Chair of the EWGPWD and included presentations from people with dementia about working groups of people with dementia involved in PI. Presenters and members of the audience, including representatives of some Alzheimer Associations, emphasised the desire to move away from 'patient-focused' language. We are therefore using the term Public Involvement in this discussion paper. The fact that public involvement is a term which covers quite a broad range of people should not be interpreted as implying that the involvement of one set of people is interchangeable with that of another. Our focus is on the involvement of people with dementia.

Recommendations for researchers

- Be clear with people with dementia what PI is about, why it is important and how it differs from being a research participant.
- Bear in mind that different levels of involvement are possible and that the level and type of involvement should be dependent on a range of requirements linked to the research itself and to the needs, wishes and interests of the people with dementia involved. See section on 'Levels of involvement and the issue of power'.
- Be clear about which groups of people are involved in PI (e.g. just people with dementia or also other people) and about the contribution of each (which should be specified in any subsequent reporting).

Recommendations for research ethics committees

- Be clear about the role, purpose and value of PI in research and how this differs from research participation.
- Seek clarification if necessary about how PI is embedded in the study and how those involved will be supported.
- Suggest the implementation of PI activities at different stages of research, including the design of the study.
- Include a person with dementia as an external expert if not already an ordinary member of a research ethics committee.
- Consult a person with dementia when reviewing a protocol and the informational materials.

Recommendations for funders

- Invite people with dementia to have a say in prioritising research proposals.
- Integrate patient relevant research questions or outcomes in open calls.
- Assign a score to PI activities when selecting projects for funding.
- Provide a dedicated budget for PI activities.
- Recognise in funding applications the range of roles that PI may play.
- Ensure that PI is explained in enough detail to determine meaningful involvement.

Summary

Public Involvement (PI) is about involving people with dementia in the research process, but not as participants. It means doing research with rather than on or to people with dementia. This is based on their right to voice their needs and perspectives and to democratic processes such as legitimisation and transparency. It is also a means to an end in that it enables researchers to benefit from the lived experience and perspectives of people with dementia, thereby fulfilling the criteria for good and hence ethical research. There is not one 'right' way to do PI. Indeed, it must be a flexible process which responds to the needs, possibilities and interests of a diverse set of people with dementia. In this discussion paper, we are using the term 'Public Involvement' (albeit with a unique focus on the involvement of people with dementia) as a move away from 'patient-focused' language.

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[18] http://interdem.org/

Promoting and valuing diversity

Capturing diversity versus seeking representativeness

Through PI, people with dementia can contribute towards research by sharing their unique insight into living with dementia and how this relates to the research topic and to the methodology with researchers. However, concerns about representativeness may block attempts to begin to involve them (Tritter and McCallum 2006) and call into question the value of their contribution. In a synthesis consisting of a mapping of values found in diverse literature on PI in health and social care research, frequent concerns were detected about whether people engaged in PI were representative of the community being studied because of selective recruitment or difficulty reaching certain groups of people (Gradinger et al. 2015).

A systematic review of PI in research[19] revealed that most of the 142 studies included had used convenience sampling[20] and the authors suggested that this was acceptable, adding that random sampling is the least biased approach but can fail for very small numbers (Domecq et al. 2014). The authors commented that despite the risk of convenience sampling and volunteering resulting in “a sample of patients that are not truly representative of the target population”, researchers should use the approach best suited to the availability of ‘subjects’ and based on the research topic. The use of terminology related to quantitative research when referring to people engaged in PI may contribute towards confusion and lack of clarity about their role.

People engaged in PI should not be described in terms of their ‘representativeness’ in the statistical sense (i.e. with associated assumptions that their views and experience can be generalised to the wider population of people with dementia). People with dementia have knowledge and experience of their condition and should not be considered as representing everyone with dementia. Any information they share about their experience is valuable and it is important to involve a sufficiently broad range of people in order to obtain a sufficiently comprehensive and nuanced understanding of the experience of dementia. However, for some issues just a few people might be sufficient. PI should not be confused with qualitative research for which it is often important to reach a point of saturation (see section on sampling in Part 2 of this paper). In PI, each person is expected to share his/her expertise and unique perspective with the research team (albeit it with appropriate encouragement and support). The perspectives, thoughts, feelings and experience of people living with dementia are valuable. These may be quite different from one person to the next and this does not make them any less valuable. Rather it enables research to benefit from a greater understanding of the diversity of people’s experience of living with dementia and of the complex nature of the condition.

On the other hand, in keeping with the principle of equity/justice, everyone should have an equal opportunity to be involved in PI. Consequently, if people with very similar characteristics are involved to a much greater extent than others, this suggests that a large proportion of society is being excluded and that PI is not capturing the diversity of experience of people with dementia across Europe. Researchers are sometimes criticised for involving people who are the easiest to find. Indeed, the term ‘hard to reach’ is increasingly being recognised as serving to blame people from minority groups for low levels of involvement (e.g. reflecting assumptions that they have made themselves inaccessible or are unwilling to get involved) (INVOLVE 2012). Clearly, some forms of PI are more intensive and demanding than others, which requires a trade-off between inclusiveness and 'demandingness' (Jongsma and Friesen 2019). There is a risk that some people with dementia, especially those who are often marginalised, may find the kind of participation proposed too challenging and hence not get involved. Some symptoms of dementia may also result in some people needing extra support or not being able to take part in certain activities (e.g. because of restlessness). Attention must be paid to ensure that different methods and opportunities for inclusion are appropriate and adapted to the needs of a broad range of people with dementia.

Some people have more opportunities to engage in PI than others because they are asked more. Possible reasons for being more frequently solicited include, for example, having connections with researchers or organisations, having certain skills and moving in circles that are familiar to many researchers. The contributions of people who are frequently involved are no less valuable for that reason. Nevertheless, people who share certain characteristics (e.g. linked to level of education, ethnicity and socio-economic status) and are frequently involved in PI may sometimes find themselves referred to in a disparaging manner (INVOLVE 2012) (e.g. as ‘the usual suspects’ or as ‘super patients’) (Black et al. 2018, Beresford 2019). Such people are often more experienced, confident and assertive and more likely to make points which the researchers might not want to hear (Beresford 2019). Criticisms have also come from other people engaged in PI that researchers are perhaps too dependent on ‘super patients’, take their perspectives into account more often and miss the perceptions of a more diverse set of people, including people from marginalised groups (Black et al. 2018). This may also lead to concerns from other people with

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[19] The review was of Patient Engagement but the authors’ definition corresponded to that of PI adopted in this paper.

[20] The issue of sampling for participation in research is addressed in Part 2 of this report.

dementia. Jongsma, Spaeth and Schicktanz (2017) report criticism from some people with dementia of a lady who was very vocal in PI, but whose experience, whilst acknowledged as being valuable, was not considered by other people with dementia as reflecting their experience. Whilst the views and experience of people engaged in PI are not expected to represent those of other people, if one or a few people dominate discussions, their views may be the only ones heard.

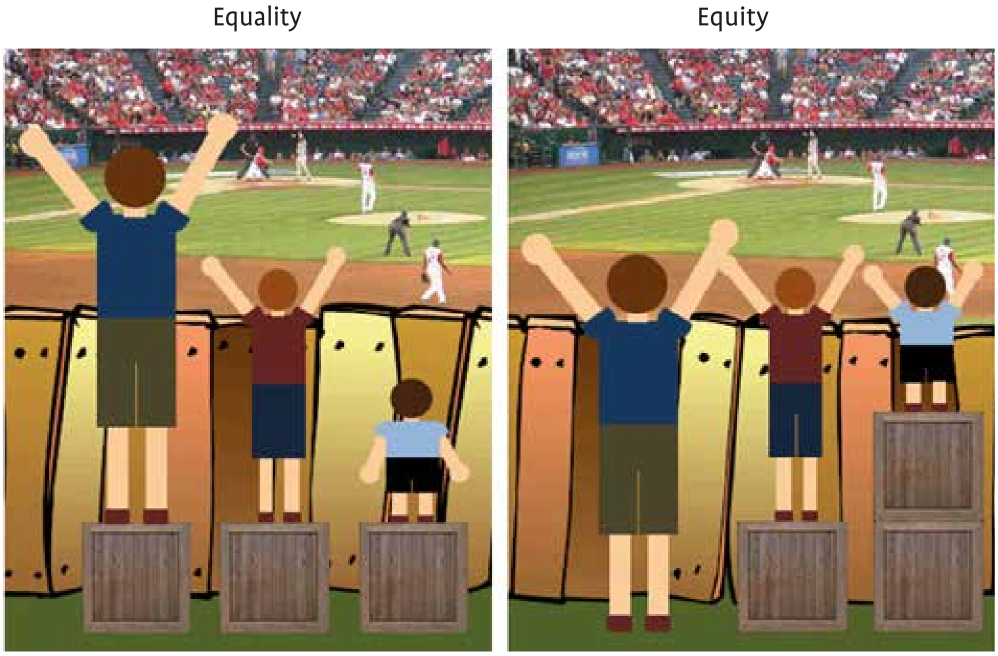
The problem, however, lies not in the people with dementia who are currently involved in PI but in the fact that researchers are often only drawing on people from a narrow section of the population with a valuable but limited range of experience and perspectives, and failing to be inclusive (e.g. overlooking the insights and experience of other people who are also very much affected by the topic of the research and may have a lot to offer) (INVOLVE 2012). This is an issue of relevance to the principle of fairness and to the ethical requirement to conduct good quality research. It should, however, be noted that diversity is not always visible. Assumptions are sometimes made based on ignorance and stereotypical beliefs about what people from minority groups 'look like' without really knowing about those involved (e.g. their ethnicity, life experience, religious beliefs, level of education and gender identities). Moreover, it should not be assumed that people with the same condition or from a particular sub-group have by definition/default the same opinions and interests.

Equality and equity

There are also several factors which may make it more difficult, and hence less likely, for some people to contribute to PI, some of which are addressed in the next section on 'Understanding multiple and intersecting identities'. An understanding of the difference and relationship between equality and equity is important in this context. In 2012, Craig Froehle produced the following graphic (Figure 2) to illustrate the point that "equal opportunity alone wasn't a satisfactory goal and that we should somehow take into consideration equality of outcomes (i.e. fairness or equity)" (Froehle 2016).

Gillon (1994) emphasises that justice is more than mere equality in that people are sometimes treated unjustly even if they are treated equally. He argues that it is important to treat equals equally and unequals unequally in proportion to morally relevant inequalities. An approach based on equality involves treating everybody in the same way without taking into account differences between people, which may be inherent, linked to circumstances or structurally determined. In the context of PI, this might involve organising PI sessions on a holy day when people from a particular faith would be unlikely to participate, discussing highly technical documents which require a level of literacy that some people do not have or having meetings on the 5th floor of a building with no lift, which would exclude people with mobility problems. These are all examples of equality, which when coupled with lack of sensitivity for

Figure 2: The relationship between equality and equity



Source: https://medium.com/@CRA1G/the-evolution-of-an-accidental-meme-ddc4e139e0e4 individual needs and preferences, are basically unfair and not conducive to attracting and involving a diverse set of people with dementia in PI. Equity is therefore not about simply providing everyone with same opportunities but about fairness and equality in outcomes. Issues related to inequity therefore need to be addressed when striving for equal opportunities and outcomes.

Perceptions of the value of Public Involvement

Ensuring meaningful and valuable PI touches on respect for the individual in many ways and if carried out properly contributes towards good quality research of relevance to people with dementia. If approached in a tokenistic way, it involves deception, contempt and exploitation, perhaps providing a seat at the table, but failing to take into consideration people's contribution. Many researchers carry out PI on the basis of ethical, pragmatic and methodological motives (Ball et al. 2019). However, as recognition of the importance of involving people with dementia in research increases, so too does the risk of tokenism (Brett et al. 2014, Hardavella et al. 2015). In the context of mental health research, Patterson, Trite and Weaver (2014) point out that incorporating lived experience into the research process may improve the quality, relevance, acceptability and ethical status of research but is by no means universally accepted. Critics have claimed that conducting PI is time-consuming, challenging and often tokenistic. Researchers sometimes plan for PI simply because it is a necessary requirement for funding or in some cases to obtain ethical approval. It is not uncommon for organisations working with people with dementia to be contacted, often at the last minute shortly before submission of a proposal, by researchers wishing to involve people with dementia in their research without any clear plan or goal, and without appropriate funding. This would most likely result in a half-hearted, tokenistic involvement of people with dementia in PI as these researchers may not be intrinsically motivated to conduct PI or may underestimate the need for the planning of such activities.

☞ **How could funders encourage researchers to conduct meaningful PI?**

People with dementia, particularly people from various minority groups, may sometimes feel that their involvement is simply 'to tick the box' or tokenistic in the sense that they are, for example, the 'representative' Black person or Sikh person or person with an intellectual disability in the PI team. In other words, they feel that they are only there so that researchers can say that someone from their group was involved in the research and that what they have to say is perhaps of little or no importance to the researchers. Researchers should try to ensure that people are not made to feel this way as a result of their treatment or attitude because it is a form of harm and injustice, and may also affect people's willingness to share valuable information about their experience of dementia. This may sometimes be because researchers do not yet have sufficient cultural awareness or skills to reach out to a more diverse set of people in an appropriate manner so as to make each person feel valued as an individual. As more people with a broader range of characteristics start to take part in PI, their complex identities will hopefully be recognised and their contributions no longer reduced to a narrow, stereotypical understanding of a particular category into which they might otherwise have been placed.

Some researchers may have practical concerns about the time or skills needed to conduct PI properly or about specific challenges linked to the involvement of people with dementia. Some may be unclear about the concept of PI and what it entails. These are valid concerns which need to be addressed for PI to be effectively and meaningfully included in a research project. Lack of enthusiasm or resistance to the concept of PI by some members of the research team, however, not only risks a constant struggle to legitimise and promote the importance of PI in research but also jeopardises its effectiveness and risks tokenism.

Negative attitudes about PI and lack of responsiveness by researchers may also be detrimental to the wellbeing of the people with dementia involved and a major source of frustration. In some cases, people have experienced stigma, prejudice and discrimination. Examples from people involved in PI for mental health research include excessive scrutiny or, conversely, over-valuation of their contribution, undermining of their validity and status as an expert by experience if a research activity is performed too well, being considered as one of 'the usual suspects' (as mentioned in the previous section) or as 'non-representative' and experiencing resistance from researchers wishing to retain traditional research hierarchies (Patterson et al. 2014). There are also reports of positive experiences of PI and of mixed feelings within the same project. Some people with dementia contributing to PI in brain donation projects, for example, have reported feeling valued and involved whereas others, at different sites, described their involvement as tokenistic and frustrating (Hayes et al. 2018).

Tacit assumptions and expectations of people with dementia, carers, researchers, funders and members of research ethics committees about PI need to be addressed and a way forward negotiated (Poland et al. 2019). More research into the impact of PI would also be helpful in convincing some researchers about its value. In addition, it is important to consider what makes people feel valued and respected in some studies or at some research sites and not in others. It may be linked not only to attitudes but also to practices and procedures which do or do not contribute towards a respectful, equal partnership and opportunities to contribute meaningfully to research through PI.

Recommendations for researchers

- When involved in specific studies, discuss how you and your co-researchers feel about PI, the concept of lived experience and the goal of creating equal partnerships. Acknowledge and discuss different perspectives to agree on how to present PI to the people with dementia who may eventually contribute to your research in this way.
- Communicate your perception of the role and value of PI in research in the most appropriate way to the people with dementia involved and listen to their perspectives.
- Consider how invitations to people with dementia from marginalised or minority groups are interpreted by those concerned and try to ensure that they feel valued as individuals (and not simply 'to tick the box' or as a token gesture).
- Try to ensure that activities are not overly demanding and are inclusive of a variety of perspectives of people with dementia.
- Ask people with dementia directly (e.g. in invitation letters or pictorial adverts for PI) to let you know if there is anything that would make it easier for them to be involved.
- Include the concept of PI in training on research methods.

Recommendations for research ethics committees

- Assess whether researchers have a specific training/the required expertise to carry out PI activities.
- Require a detailed description (and not just general statements) of PI activities in research protocols.
- Assess whether methods, times and opportunities for PI are appropriate to promote equity in PI.

Recommendations for funders

- Encourage researchers to allocate part of their budget and adequate time to attempts to support ongoing involvement of a more diverse set of people with dementia.
- Take into consideration that more inclusive PI activities may require a higher budget.
- Take into consideration that more inclusive PI activities will increase the social value of research.

Summary

Promoting and valuing diversity is about seeking a diverse group of people with dementia for Public Involvement (PI) activities. It is not about representativeness, at least not in the statistical sense (e.g. people with dementia think X, Y or Z) but about listening to the perspectives and learning from the lived experience of very different people. It is too easy to categorise people as being 'hard to reach'. We need to be looking at why people from some groups within society or with certain characteristics are being excluded and at how practices, procedures and structures within society contribute to such exclusion. If we want to promote inclusive research, it is important first to address the issue of inequity.

Hierarchies and power

Levels of involvement and the issue of power

Research is about producing worthwhile knowledge, yet knowledge is often associated with power[21]. This perhaps partly explains the popularity of Arnstein's (1971) ladder of citizen participation which is frequently evoked as a means to evaluate and in many cases to judge different levels of PI in research, with some forms of PI being considered as better than others. The typology of the ladder was initially proposed by Arnstein in 1971 to be deliberately provocative with each rung of the ladder corresponding to the alleged extent of citizens' power in determining three US federal social programmes, namely urban renewal, antipoverty and Model Cities. Nevertheless, it is frequently reproduced, slightly adapted or used uncritically (in terms of its original purpose, focus and historical context). As a result, consultation (labelled in Arnstein's article as tokenistic) has become almost a dirty word. However, Arnstein only specified that consultation should be considered tokenistic if proffered by power holders as the total extent of participation whereby citizens hear and are heard but lack the power to ensure that their views are heeded "by the powerful". Moreover, as suggested by Maier, "the process of increasing participation should not be simplified to a one-dimensional parallel of climbing a ladder" (2001, p.716) and it should not be assumed that people with dementia engaged in PI are all preoccupied with power.

People with dementia can engage in PI in different ways and to different degrees such as by being involved in consultations about various issues, sharing perspectives and providing advice, collaborating with researchers more directly on specific tasks or by carrying out research. Co-production is one approach to PI which particularly emphasises the sharing of power and responsibility from the start to the end of the project, including the generation of knowledge (INVOLVE 2018). This tends to be perceived in terms of joint ownership of projects and working together in the pursuit of a joint understanding. The term 'empowerment', which is also often associated with co-production, reportedly challenges embedded knowledge hierarchies 'of the expert versus the lay subject', and recognises that communication is not 'a one way transfer from a knowing subject to a supposedly ignorant one' (Porter, 2010) and that there are 'experts by training' and 'experts by experience' (Cheffey, Hill, McCullough and McCullough 2017). Tritter and McCallum describe this as being,

> *"not a hierarchy of knowledge – relevant professional versus irrelevant lay – but rather a complementarity between forms of knowing, set within a willingness to acknowledge differences" (2006, p.164).*

However, whilst certain aspects of involvement are perhaps more central or emphasised in co-production, they should also be present in any form of PI. Key principles such as sharing power, embracing diversity, striving for inclusivity, respecting and valuing the knowledge of all those involved, reciprocity, and building and maintaining relationships (INVOLVE 2018) should be reflected in all PI, without power necessarily being the focal point, the sole measure of participation or the criterion for evaluation of the value of a particular contribution.

As Tritter and McCallum (2006, p.164) suggest in relation to Arnstein's ladder, involvement should not be perceived as a "contest between two parties wrestling for control of a finite amount of power", thereby excluding opportunities for collaboration and shared decision making. People with dementia can contribute towards research in different ways according to their interests, experience and abilities. Not everyone is interested in the same level of involvement and capable of the same level or kind of participation in decision making. One person may wish to contribute to a small part or particular aspect of the research, whereas another might prefer to play a much more active role.

At the level of society, it has been suggested that the very goal of working together in a positive and collaborative manner with some marginalised groups is idealistic and naïve. This is because of assumptions and philosophies underlying much research, particularly in the field of medicine and psychiatry, and because people with mental disorders can still be deprived of their freedom, lose certain rights, be subjected to coercive measures and not be taken seriously. Rose and Kalathil (2019)[22] state that most current research in the fields of medicine and psychiatry still has remnants of Enlightenment thought and Eurocentricity, with a focus on rationality, the thinking, reasoning subject and the alleged 'racial' and cultural superiority of White Europeans and that this undergirds much of health research. For Rose and Kalathil, such a situation is not conducive to the goals of co-production. They suggest that a change in thinking is needed before this can be achieved.

Equality and clarity of roles

People engaged in PI and researchers tend to be perceived and defined in dichotomous terms. This is reflected in the social meanings attributed to members of each group, in expectations about who will conduct various tasks, in legal and financial responsibilities (irrespective of the extent to which people with dementia contribute towards key decisions) and in the use of language and titles (Morrow et al. 2010). The constant positioning of the various people

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[21] The phrase "knowledge is power" can be traced as far back as 599–661 CE to Imam Ali

[22] The context of Rose and Kalathil's argument is co-production involving researchers, policy makers and non-White service users with mental disorders, the latter described as the "mad" and the "racialized mad".

contributing to research through language and other means serves as a constant reminder of differences and may raise questions about equality and the role of each.

People with dementia are usually invited by researchers to engage in PI. In many cases, the researchers determine the timing, duration, frequency and nature of involvement of the latter. Flexibility and sensitivity to the needs and interests of people with dementia (e.g. in terms of the timing and preferred mode of involvement) are important but it is often inevitable that PI follows the pace of the development/progress made in a particular research project. For this reason, unless people with dementia have an extremely high level of involvement in the research (e.g. a co-researchers), the researchers will largely determine the timing of PI and the kind of involvement required. Researchers also usually manage the budgets necessary to pay for meeting rooms, travel, accommodation, materials and any out-of-pocket expenses. Academic discussions often revolve around whether PI is valued by the researchers and in many cases, researchers take the lead and have final responsibility for certain tasks (e.g. linked to accounting, safety issues, applications for ethical approval, reporting obligations and statistical analysis), however much shared decision-making there may be. It is therefore important to be aware that terms such as PI, co-researcher and researcher (to name but a few) reflect acquired roles which are actively negotiated and sustained within social contexts and that various constraints may affect how people do and do not act (Morrow et al. 2010).

Respect for equality should not be interpreted as the need to deny differences in people's roles and responsibilities. At the same time, attention should be paid to how people's roles and responsibilities are portrayed and communicated so as to avoid social positioning and the creation of unnecessary friction between people who are all investing their time, skills, energy and experience in the research. Terms such as 'expert by experience' and 'expert by training' may reflect different types of expertise but if each type of expertise is truly valued on an equal basis, this has to be demonstrated through actions not just words. Moreover, the emphasis of these two terms is on expertise, albeit of different kinds. This confirms the belief that different kinds of knowledge can exist side by side but in making this distinction there is also a risk that meanings come to be associated with each and that these do not necessarily reflect an equal value.

Tailoring Public Involvement to the interests and abilities of all people with dementia

Ethical involvement of people with dementia in PI calls for mutual respect, different opportunities for involvement and possibilities for shared decision making, which correspond to individual interests and abilities, and clarity about the roles and responsibilities of everyone involved. Every effort should be made to ensure that all people with dementia with an interest and ability to contribute to PI have an equal opportunity to do so. With regard to ability, efforts should be made to address inequity and to promote inclusion. The concept of 'reasonable accommodation', mentioned earlier, is relevant in this respect. This means that reasonable adjustments/adaptations should be made to enable people with dementia to contribute to research.

Measures to promote inclusiveness need to be balanced against the need for PI to be effective (in terms of contributing towards improving research and giving a broad range of people a voice), the need to promote the wellbeing of those involved and to work within the context of certain constraints (e.g. linked to available funds, group dynamics, progress with a specific study and academic, ethical and administrative formalities). This requires a certain degree of flexibility (in terms of what is considered possible), acceptance of different needs and differences, a readiness to consider various forms of adaptation/adjustment as 'reasonable', and above all, a desire to promote diversity within research[23].

Researchers should have a clear idea about why and how they wish to involve people with dementia in their research in the context of PI, but a discussion between researchers and the people providing PI about the goals and expectations of each at the start of the project would be helpful (Black et al. 2018). It may also be helpful to consider different types and levels of involvement of people with dementia in PI as occurring along a continuum (see Figure 1 on page 12), perhaps with different people intervening at different points and on different tasks. This would ensure the involvement of people with dementia, in accordance with each person's interests and what they are able to contribute, at every stage of the research. It is not helpful to denigrate anybody's contribution on the grounds that it does not represent a certain type or level of involvement. Furthermore, if a person with dementia wishes to contribute to a certain part of a study and has valuable experience to share, he or she should be encouraged to do so. For some people, participation is the goal of their involvement and that is what they find rewarding and empowering. Others may have a keen desire to take on responsibility and to play a role in decision making.

It should not be assumed that everyone is capable of or interested in the same kind or level of involvement. Parveen et al. (2018) propose a person-centred and culturally sensitive approach to working with minority ethnic communities in PI (which could be equally valuable to the involvement of people with dementia in general). This approach involves providing people who are potentially interested in engaging

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[23] Also with regard to the trade-off between inclusiveness and demandingness mentioned earlier (Jongsma and Friesen 2019).

in PI with information about the study and opportunities for involvement and encouraging them to reflect on their possible level of involvement and the aspects of the study they would like to be involved in. This helps avoid judgemental and value-laden reflections about different levels of involvement and implicit power relations. This more tailored, person-centred and cultural approach would help ensure that people with the appropriate skills and interest (not necessarily the same ones) are involved throughout the whole research process in a continuous manner. This also creates more of a balance in that the researchers seek the support of people with dementia and people with dementia decide in what way and to what extent they would be willing and able to provide that support.

This was exemplified in a recent IMI-funded research project for which Alzheimer Europe asked members of the European Working Group of People with Dementia (EWGPWD) if they would like to be part of a Patient Advisory Board (PAB). Some members of the group expressed an interest in contributing towards certain tasks periodically and others were keen on a greater involvement (more regularly, on a wider range of tasks). The latter had a particular interest in the topic of the study. The EWGPWD proposed having a general PAB and a Core PAB. The members of the Core PAB agreed to be contacted at relatively short notice and to provide feedback rapidly by various means (e.g. email, teleconference or through additional meetings). This has enabled a more interactive approach between people with dementia and the researchers within tight timeframes reflecting the demands of the study. This approach has not undermined the continued work of the general PAB on issues which can be planned more in advance and for which more support can be provided to the people with dementia who might need it.

Recommendations for researchers

- Formulate a tailored, person-centred and cultural approach to PI.
- Discuss the goals and expectations of PI with people with dementia at the beginning of the process of involvement and revisit as needed.
- Be clear about roles and responsibilities of people engaged in PI and researchers, while avoiding devaluing the contribution of people with dementia.
- Ensure the involvement of people with dementia in accordance with each person’s interests and what they are able to contribute.
- Have clarity from the outset about how PI will be included in the study and build this into a project timeline.
- Regularly update people involved in PI on progress with the research and plans for their ongoing involvement.

Recommendations for research ethics committees

- Assess whether research protocols clearly describe roles and responsibilities of people engaged in PI and researchers.
- Assess whether research protocols envisage different possibilities of involvement at the different stages of the research process to ensure that the persons’ interests and possibilities of contributing are respected.

Summary

Public Involvement (PI) needs to embrace diversity, strive for inclusivity and be based on respect for everyone’s involvement (irrespective of the type and level of involvement of each person). Flexibility and a readiness to make reasonable adjustments/adaptations are essential to enable a broad range of people with dementia to engage in PI activities. It is important to build and maintain mutually respectful relationships between people with dementia and researchers. An over-emphasis on power and the portrayal of involvement in the form of a hierarchy devalue the meaningful and valuable involvement of many people with dementia. Decision making should be shared wherever possible but the roles and responsibilities of researchers and people with dementia may differ depending on the type and level of involvement of each person in a particular study. It is important to ensure that this does not impact on the perceived value of the contribution of each.

Understanding multiple and intersecting characteristics/identities

There are numerous factors which interfere with the willingness and ability of people with dementia to contribute towards PI. In this section of the report, we look at some of these factors. Most challenges are experienced to a greater or lesser extent by many people but some people face considerable challenges and often more than one. Different people, depending on their history, personal situation and the level of support available to them, are better or worse placed to address various challenges they may face. For example, the challenges and experience of taking part in PI in a foreign language of a white, 55-year old, highly-educated, man with dementia are likely to be different to those encountered by an Asian woman of 85 who is illiterate in her own language. Similarly, people who are living in care homes, who have an intellectual disability or who have problems with substance abuse, to name just a few examples, may be affected differently by various challenges which need to be addressed in order to include them in PI activities. Sometimes, symptoms of dementia, inappropriate support and unmet needs may result in people with dementia acting in ways that other people find challenging and this may also represent a barrier to their possible involvement. In this section of the paper, we first examine the concept of intersectionality and then look at some concrete examples of related issues.

About intersectionality

People with dementia form a diverse, heterogeneous group made up of people with different characteristics such as different ethnicities, ages, genders, disabilities, levels of education and socio-economic backgrounds etc. There is not 'one dementia' and not just one experience of it. If researchers want to involve a more diverse group in PI (and as participants in research), it is important to consider these differences and how they impact on involvement in research or, more specifically, on exclusion which is a form of discrimination[24]. However, focusing on differences often involves categorising people into what come to be perceived as neatly defined, separate groups. This process of categorisation, when combined with drawing a boundary between 'we' (usually members of what has come to be considered as the majority group, often consciously or unconsciously seen as the norm) and 'them' (usually members of what has come to be considered as the minority group), may serve as a mechanism for constructing 'otherness' (Torres 2015). Through dialogue and social interaction, meanings are constructed around the different categories which reinforce and perpetuate perceptions of normality and deviance, and contribute towards power imbalances and discrimination[25].

Whilst it is important to identify characteristics which are commonly associated with discrimination, it is also important to look at ways in which socially constructed categories intertwine and may create new forms of discrimination which are not immediately obvious (Angelucci 2017). This is often described as intersectionality, a term coined by Kimberlé Crenshaw in 1989 to describe the different lines of oppression and marginalisation in society that can affect a person, based on their ethnicity, age, gender, ability, sexual orientation, class or the intersection of two of more of those aspects. It is a lens to explain the underlying mechanisms of the multidimensionality of marginalised subjects' lived experiences (Nash 2008). Intersectionality therefore provides a lens to address complexities that different identities entail and the position of these identities, as an advantage or disadvantage, on a structural level.

Intersectionality is about exploring the relationships between socio-cultural categories and identities. Knudsen (2006) describes it as something that can be used to analyse the production of power and of social and cultural hierarchies within different discourses and institutions, whereby some people come to be positioned not only as different or 'other' but as troublesome. These categories can be considered as socially constructed because, as Pickering (2001, p.72) points out, "conceptions of the Other and the structures of differences and similarity which they mobilize do not exist in any natural form at all… the location of the Other is primarily in language". In other words, minority groups 'exist' because societies have chosen to focus on specific attributes (or features or experiences), name them and label people as belonging to groups reflecting those attributes. The task then becomes one of seeking to understand their difference (the assumption being that these differences reside in 'them') and "the logic behind their peculiarities" (Torres 2015, p.941). Locating 'the problem' in the individual detracts attention from the way that structures, organisations, procedures and systems create problems and blames individuals and groups for not being able to adapt (e.g. to Western culture, language and traditions). There is a risk of people considered as 'other' being blamed, for example, for not being able to adapt to Western culture or to 'age successfully', and of serious forms of discrimination (e.g. racism and ageism) being attributed to bias and prejudice alone, and overlooking the role of institutions and systems in creating and promoting oppression, discrimination and marginalisation (Torres 2015).

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[24] e.g in the form of epistemic injustice (see p.18).

[25] This has some similarities to Link and Phelan's (2001 and 2006) conceptualisation of stigma involving the labelling of socially salient attributes shared by a group and a process of cognitive separation (i.e. making a distinction between 'us' and 'them'), whereby those with the attribute are considered as in some way deviant.

It has been argued that failing to consider intersecting identities may even render certain characteristics or vulnerabilities invisible. Writing about the intersection of 'race' and 'sex', Crenshaw (1989) warned against thinking about discrimination as occurring along a single categorical axis. She argued, taking the example of sex discrimination in the work place, that discrimination against a white woman and discrimination against a black woman is not comparable and that failing to recognise the impact of the combined discrimination against black women by reducing the issue to the category of sex and taking the situation of white women as the norm renders the impact of 'race' invisible. This contrasts with identity politics, which encourages mobilisation around a single axis of human traits (such as the illness as identity, or gender as identity). Similarly, Torres (2015) points out that fairness and non-discrimination does not necessarily result from treating men and women exactly the same because it amounts to treating a woman as if she were a man (and vice versa), which is not the case. The risk in putting pressure on people to classify a particular axis as their single defining feature is that they may see themselves as complex individuals who cannot be represented so selectively or reductively. People may, however, identify more strongly with some personal characteristics than others at certain times or in certain situations. The important point is to recognise the possible impact of how different identities intersect.

Unquestioning acceptance of certain 'norms' can also contribute towards intersectional invisibility. Purdie-Vaughns and Eibach (2008) have described the impact of androcentrism (the tendency to consider male experience as the norm), ethnocentrism (the tendency for members of the socially dominant ethnic group as the norm) and heterocentrism (the tendency to consider heterosexuality as natural/the norm and other forms as deviant or lifestyle choices) on people with intersecting identities. This contributes towards people being defined as non-prototypical members of marginalised groups. The impact of where or how a person is positioned is also affected by the context:

> *"Capitalist women would be identified as oppressed when situated in patriarchy and as oppressors when situated in capitalism. Working class men would be identified as oppressed when situated in capitalism and as oppressors when situated in patriarchy. The situation would obviously get more complicated once we added race to the schema of oppression" (Smiley 2008, p.99).*

What does this mean in relation to the involvement of people with dementia in PI? Firstly, it suggests the need to consider social and cultural categories as dynamic and fluid, in a permanent process of construction, deconstruction and renegotiation of meanings affecting people's identities, social roles and relationships, and with implications for marginalisation and potential discrimination. This does not mean that categories *per se* should be denied or rejected but rather that broad and sweeping categorisations (bordering on stereotypes) should be avoided (Knudsen 2006). Secondly, it necessitates a focus on "the complexity of relationships among multiple social groups and across analytical categories and not on complexities within single social groups, single categories, or both" (McCall 2005, p. 1786). Marginalised intersectional identities amongst people with dementia can be taken as the starting point to explore the complexity of the lived experience of people with dementia (through participation in research) and for that diverse experience to influence the research process in the context of PI. Finally, it highlights the importance of reflexivity on the part of researchers as a means to increase awareness of their own assumptions, possible restricted or biased world views and potential intersectional blindness at the individual and structural level (please see the section on reflexivity and positionality in Part 3 for more details).

The categorisation of people into sub-groups on the basis of perceived difference along a unique axis is one way of making sense of the world and of taking short cuts so as to make the vast array of information available more manageable but it has social consequences. Torres (2015) encourages researchers to reflect on the implications of using broad social categories such as class, gender, sexuality, ethnicity and age which typically result in assumptions. She asks whether and if so why a particular category matters, what assumptions are attached to it, whether these are helpful and what this says about individual responsibility versus structural factors.

- **Which social categories do you notice most?**
- **Do you make assumptions about people based on these (e.g. this is a woman or this is a person from a traveller community, so this probably means X, Y or Z)?**
- **What do you think the advantages and disadvantages are (for the people who are so categorised) of doing this?**
- **What is your intersectional societal position? How does it impact on your research (research question, methodology, and writing)? How does it impact on your collaboration with people with dementia with their intersectional societal position?**

Language and communication difficulties

Difficulties with language and communication are frequently experienced by people with dementia. This may, for example, involve having difficulty finding words, pulling ideas together, keeping track of conversations, remembering what has already been said and formulating sentences. Documents may become hard to read and understand. Such difficulties often result in people feeling uneasy, embarrassed or awkward when interacting with other people, and having problems at work. This may lead to loss of confidence and self-esteem, to confusion and misunderstandings and even to withdrawal from situations which require literacy and communication skills. These difficulties with language and communication tend to increase as dementia becomes more advanced. Consequently, people with more advanced dementia may find tasks which rely heavily on language and communication more challenging. This may result in them being excluded from PI, either by researchers or as a result of self-exclusion.

> ▶ *"Two years ago, I was diagnosed with dementia and have been participating in many surveys and studies. I think that patient involvement is essential for a good study and I am happy to answer. But it is getting difficult for me now to read and write, although I was an educated engineer and communicated in both English and Flemish without any problems"* **(Geert, person with dementia, Belgium).**

Lack of fluency in the main language used in a particular country can also be a major barrier to involvement in PI. Many people from minority ethnic groups have difficulties understanding and communicating in the national language of the country in which they live. Impairments affecting communication (e.g. linked to hearing, vision and speaking) may also result in the exclusion of people with dementia.

Most PI work is conducted in the national language of a particular country and involves reading, writing and speaking at some point. The consequence of conducting PI in this way is that large numbers of people are prevented from contributing to research. It is estimated, for example, that 60% of first-generation immigrants in the Netherlands lack proficiency in the Dutch language (Uysal-Bozkir, Parlevliet and de Rooij 2013). In the South Asian community in the United Kingdom, only 35% of older people over 65 years of age can speak English and only 21% can read and write English, often communicating in their first language, which for many is Urdu (Blakemore et al. 2018).

In some minority ethnic groups, there is also a greater likelihood of older people being illiterate in their primary language (i.e. their mother tongue), especially older women (Bhattacharyya and Benbow 2013, Nielsen and Jørgensen 2013) and women in some communities have fewer opportunities than men to learn the language of the country (Beresford 2019). To complicate matters, in some languages, there is no word for dementia. This is the case in many Asian languages including, for example, the Punjabi language (Lawrence et al. 2011, Uppal and Bonas 2013, Mohammed 2017, Sagbakken, Spilker and Ingebretsen 2018). Similarly, it cannot be assumed that there is a clear understanding about dementia within each country's Deaf community because of the lack of access to information in their own language. Some countries have a sign for dementia in their own signed language, but this is dependent on their understanding about dementia (Young, Ferguson-Coleman and Keady 2018). It has also been suggested that being bilingual or multilingual may delay the onset of dementia by a few years. However, as dementia progresses many people revert to their primary language or experience 'intrusion' which consists of mixing the two languages (Khan 2011).

Some people with dementia have additional difficulties because they do not have adequate reading and writing skills in the national language of the country in which they live. These difficulties are not limited solely to people from minority ethnic groups. According to UNESCO, "A person is literate who can, with understanding, both read and write a short statement on his or her everyday life," whereas UNICEF emphasises the ability "to use reading, writing and numeracy skills for effective functioning and development of the individual and the community" (UNESCO 2006).[26] However, a person may lack literacy skills but be able to communicate fluently or be literate but communicate non-verbally.

Digital literacy

People who are not 'digitally literate' (able to use computers, tablets and smart phones well) face similar difficulties to people who have difficulties reading and writing (Beresford 2019). This may include using email, Word, Excel or social media and searching for information on the Internet. Figure 3 below, from Eurostat (2018), shows the Internet use of adults (aged 16–74) in the European Union in 2016 in percentages. This clearly demonstrates the digital divide by age and educational attainment. Moreover, some groups of people are likely to have less access to Internet than others such as care home residents and some people know how to use it but are wary of it (Beresford 2019). In the UK, the Office for National Statistics reported that in 2014 only 37% of adults aged 75 years and over had ever used the Internet (compared to almost 99% of 16 to 24-year-olds). There were only small differences in Internet use between ethnic groups. The use of computers and the Internet for webinars or teleconferences may be a way to involve people who are not able to attend meetings (e.g. who have limited mobility or live in rural areas) but can also exclude people who are not able to use them or do not have access to them. It should also be borne in mind that people with dementia may develop difficulties writing and using computers.

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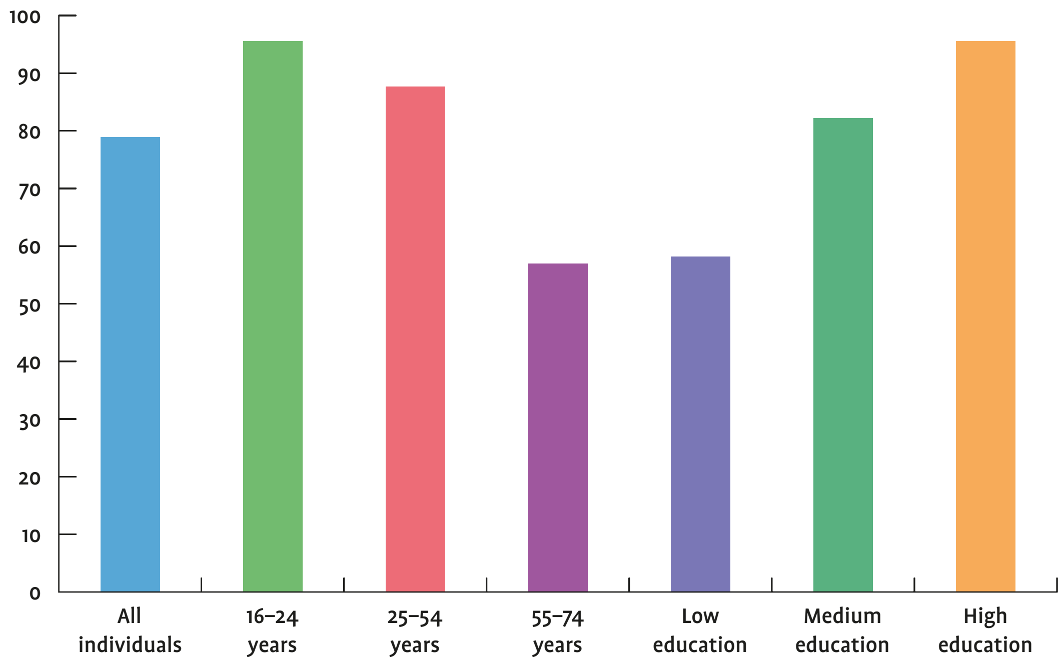
[26] For further information about literacy and illiteracy, including definitions from several countries, please see Chapter 6 of the 2006 report commissioned by UNESCO: http://www.unesco.org/education/GMR2006/full/chapt6_eng.pdf

Figure 3: Percentages of Individuals who used the Internet on average at least once a week, by age group and level of formal education (Source Eurostat 2016)



In many cases, people who have problems with language and literacy are also living in socially vulnerable situations (e.g. in poverty and in poor housing conditions etc.). They have different needs and are at greater risk of health disparities. It is therefore important that their views are taken into consideration by researchers when designing and conducting research.

Years of education

Socio-economic status, education, language ability, health literacy, geographical location and being a member of the main ethnic group are all factors which may affect whether a person with dementia knows about research and how to get involved in a particular study in the context of PI or as a participant. According to Wilson (2000), people with higher levels of education sometimes have a greater awareness of societal problems and may be more likely to be asked to volunteer. This could be because they are often already involved in organisations, have high literacy levels and may have developed certain useful skills such as running meetings. They may therefore also feel more confident about their ability to contribute.

Older people on average tend to have had fewer years of formal, compulsory education compared to their younger counterparts. In the past, children could leave school at a relatively earlier age than the children of today and were sometimes allowed to leave school to take up employment, either permanently or periodically. Some went into apprenticeships and some had their schooling interrupted by world wars. Nowadays, in member states of the European Union (EU-28), the leaving age is between 15 and 18[27], with this age sometimes being dependent on certain conditions being fulfilled (European Commission 2016). People from some minority ethnic groups have had little or no formal education (Berdai Chaouni and De Donder 2018). Historically, and even today in some parts of the world, children with intellectual disabilities are excluded from the general education system, whether or not through choice, and placed in 'special schools' where the quality of educational attainment is often less than that provided in mainstream education (Right to Education 2018). People from traveller communities also tend to have low literacy and educational levels (Liégeois 2007, Condon et al. 2019). A 2011 census of England and Wales further revealed that only 40% of people from traveller communities had any formal qualifications.

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[27] In 5 of the German 'Länder', the leaving age is 19. For a comparative table, including additional countries in Europe, please see: https://www.anefore.lu/wp-content/uploads/2016/10/EURYDICE-COMPULSORY-EDUCATION.pdf

The issues of self-confidence and how PI is portrayed and conducted therefore need to be addressed. People with lower levels of education may be reluctant to take part in activities which are perceived as highly intellectual or academic (e.g. working alongside researchers, helping develop research materials and being advisors in the research process). Approaches are needed which do not focus on written texts and preparatory reading, which take into account different abilities and ways of communicating and which are culturally sensitive and appropriate for the people involved.

English as the working language for Public Involvement

At the European or international level, researchers often have a single working language and this is often English. In some studies, PI involving people with dementia is carried out at different levels. For example, there may be local/national groups in various countries and a broader group which brings representatives of these smaller groups together. There may, in addition, be one or two people who sit on a project steering committee. The people who contribute to PI in such projects other than at local/national level, and in many cases have more direct contact with the leaders of various aspects of the study, tend to be either native English speakers or people who have acquired a high level of competency in English as a second language. Some researchers and organisations go so far as to set a minimum level of English (e.g. B2) needed to join a patient PI pool or Patient Advisory Group, even stating a preference for people with a scientific or research background. B2 is the ‘vantage’ or ‘upper intermediate level’ of language competency described in The Common European Framework of Reference for Languages (CEF or CEFR). It applies to a person who is able to:

- “understand the main ideas of complex text on both concrete and abstract topics, including technical discussions in their field of specialization.
- interact with a degree of fluency and spontaneity that makes regular interaction with native speakers quite possible without strain for either party.
- produce clear, detailed text on a wide range of subjects and explain a viewpoint on a topical issue giving the advantages and disadvantages of various options”.

Some researchers, in addition, have a preference for people who already have knowledge or experience of research, or experience interacting with different relevant stakeholders (e.g. in the domain of drug development, health technology assessment (HTA) bodies, regulators or payers).

As mentioned earlier, many older people did not have as many years of formal education as their younger counterparts and in some countries, English is not the foreign language prioritised in schools. Moreover, a survey commissioned by the European Commission found that 62% of people in Europe over the age of 55 do not speak any language other than their mother tongue well enough to have a conversation (Eurobarometer 2012). In Hungary, Italy, the United Kingdom and Portugal, over 60% do not speak any foreign language and in Spain, Romania, Bulgaria, the Czech Republic and Poland at least 50% do not. This way of organising PI results in large numbers of people being prevented from taking part in research.

Recommendations for researchers

- Try to create an atmosphere of trust and acceptance (favour approaches which encourage continuing involvement so that this atmosphere can be built up over time but do not discourage people who prefer more limited involvement).
- Consider going out into the community in order to build rapport and trust with people who might feel intimidated about meeting researchers and attending meetings at a university.
- Ask people in advance what they would like to know or feel they might need in order to make a decision about getting involved.
- Be ready to propose possible support and information if people are not sufficiently aware of PI to know what might be helpful.
- Recognise the value of augmented and alternative communication to supplement or replace speech or writing in order to facilitate PI.
- Be proactive in trying to find people from different backgrounds rather than immediately approaching whoever is available. This may require extra time and funds (e.g. for translated materials or support with communication), which should therefore be planned for.
- Plan for resources and time for finding people to involve in PI.
- Avoid, where possible, the unnecessary use of academic titles (e.g. in German, Frau Professor Doktor Schmidt) in joint meetings between people engaged in PI and researchers (whilst respecting cultural differences in approaches to the use of titles and customary levels of formality).

- Strive for flexibility in the promotion and organisation of PI so as to accommodate as far as possible the interests, needs and priorities of a diverse group of people with dementia. This may involve some of the following:
    - Ensure that a high level of education, language or literacy is not needed to engage in PI by avoiding overly focusing on texts (e.g. develop interactive activities and small group discussions), bearing in mind that some people may find it easier to express themselves in writing.
    - Encourage people with dementia who are not highly educated and who may have problems with language and literacy to engage in PI (e.g. emphasising that high levels of education are not necessary and the importance to the researchers of their contribution, stating the wish not to exclude them on the basis of difficulties with language and literacy and asking what kind of support they might find helpful).
    - Aim for accessible documentation and materials (e.g. use plain language, encourage people to ask if anything is unclear, and avoid jargon, abbreviations and unnecessarily complicated language).
    - Provide interpretation and encourage people to bring along a supporter. Bear in mind that some people may need more than one supporter (and budget for this).
    - Develop together with one or more people with dementia accessible materials to be used in PI. Discuss also planned activities and clarity of instructions or questions.
    - Test/pilot materials on a few people to ensure that they are in fact accessible, regardless of people's literacy, before using them with a PI group.
    - Adapt existing documents to make them more accessible such as providing clear explanations about what certain terms mean, about the topic of the research or about research goals and methods and using a relatively low level of language competence (e.g. B1 level).
    - Send relevant documentation well in advance and be available for questions or to meet in advance to discuss agenda items.
    - Plan for sufficient time during meetings to present the key issues in the documentation and for any questions.
    - Try to ensure that methods of communication are suited to the people involved. Where appropriate, for example, use verbal methods of communication rather than written or computerised techniques (e.g. in small, informal group discussions or one-to-one exchanges rather than written feedback of computerised documents by means of track changes).
    - Do not assume that everyone has access to Internet or computers and knows how to use them.
    - Recognise the added value of visual or pictorial communication where appropriate in the agenda, action points or minutes (e.g. through use of photosymbols for people with intellectual disability), bearing in mind that some people with dementia have difficulties understanding abstract images.
    - For large-scale European research projects, for which the working language is often English, take measures to increase possibilities for people who are not fluent in English to contribute to PI.
    - Challenge the reliance on English. Seek alternative approaches which do not rely on people engaging in PI having high levels of fluency in English.
    - Bear in mind the possible impact of involving people who are unable to contribute to discussions directly (in terms of group dynamics, maximising the contribution of everyone involved and the wellbeing of the individuals concerned and the other people involved). Look for approaches and methods of support which are beneficial to the group, the individuals concerned and the PI process.
- Seek feedback about people's experience of PI and how it could be improved.
- Consider the value of seeking feedback via pictorial or non-verbal communication methods in addition to verbal or written ones.

Recommendations for research ethics committees

- Assess whether developed methods and materials have the potential to include people with different characteristics such as different ethnicities, ages, genders, disabilities, levels of education and socio-economic backgrounds.
- Ensure that PI methods are appropriate to the study and will facilitate appropriate involvement of people with dementia.

Recommendations for funders

- Ensure that PI methods are appropriate to the study and will facilitate the involvement of people with dementia and promote diversity.

Summary

People with dementia are often considered as being members of a homogeneous group but people experience dementia differently. Recognising such difference is important but categorising people with dementia into different sub-groups risks over-emphasising difference, drawing a boundary between ‘us’ and ‘them’ and thereby constructing ‘otherness’, which may be disempowering and lead to discrimination. In keeping with the concept of intersectionality, it is important to recognise the complexity of people’s identities and the dynamic and fluid nature of social and cultural categories so as to avoid make broad and sweeping categorisations.

Such categorisations may reflect certain personal characteristics but they also result from the way society is organised, often unfairly. Careful attention, consideration and appropriate methods are needed to prevent inequity in the context of PI. This also calls for researchers to reflect on their assumptions and possible restricted world view in order to gain insight into interpersonal and structural factors which might, in this case, hinder the promotion of diversity and inclusive research.

Ageism (public and internalised)

Older people are sometimes actively sought as participants for a particular study because of their age which actually puts them in the target population, but in many cases older people (especially the oldest old – see below) are excluded from research, both as participants and in the context of PI. Whereas there may be some challenges to involving older people in PI, a systematic review of older people’s patient and public involvement in health and social care research found that the benefits to research and for the older adults themselves outweighed the challenges (Baldwin et al. 2018). The prevalence of dementia increases with age so it is particularly important to involve a broad range of older people with dementia in PI. However, there is a lack of consensus or awareness about who counts as ‘older’ (i.e. older than what?).

In many countries, people aged 65 and over are defined as older people (Ouchi et al. 2017[28]), whereas the United Nations considers people over the age of 60 or 65 as older (or elderly persons) and those over the age of 80 as the ‘oldest old’[29]. AGE Platform Europe defines the 50+ age group as older[30]. Targeted measures to encourage greater involvement of older people with dementia in PI need to be clear about the target group. 50+ is perhaps too broad a target category to be effective in attracting a balanced range of people from 50 to over a 100. It would cover over 200 million people in the European Union extended over at least five decades[31].

The exclusion of older people from research, including PI, may be partly a reflection of ageism (i.e. defined by the World Health Organisation as stereotyping, prejudice and discrimination against people on the basis of their age[32]). Ageism, like racism and sexism, reinforces social inequalities by legitimising and sustaining inequalities between groups, and is more pronounced towards women, people with a low socio-economic status and people with dementia (Ayalon and Tesch-Römer 2017, WHO 2019).

The internalisation of ageist attitudes, whereby older people themselves see age in a negative light and adhere to negative stereotypes of older people, may affect their self-confidence and willingness to take part in PI activities. Beresford (2019) highlighted the influence of confidence and self-esteem for people of all ages involved in PI and this can be further hampered by negative stereotyping. The internalisation of various forms of negative stereotyping has been shown to affect people’s confidence in and actual performance on certain tasks. Barber (2017), points out, for example, that when older people are placed in

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[28] In Japan, proposals have been made to redefine old age, suggesting the term ‘pre old age’ for people aged 65 to 74 and old age for people aged 75+

[29] Please see http://www.searo.who.int/entity/health_situation_trends/data/chi/elderly-population/en/

[30] Please see https://www.age-platform.eu/about-age

[31] According to EUROSTAT figures from 2018 (see Age Platform Europe above)

[32] For more information, see WHO section on website about ageism in healthcare https://www.who.int/ageing/ageism/en/

situations which they fear might confirm the stereotype that older people are not cognitively capable, their level of performance often falls below their own potential. It is not known to what extent people with dementia are affected by possible internalised stereotypes such as the stereotype that they are not capable of expressing their views (i.e. simply because they have dementia).

Training and support

In some cases, people with dementia engaging in PI have prior or even current experience of conducting research. When this is not the case, some researchers feel that people engaged in PI should be offered training so that they can better understand the topic, the issues and implications of adopting various methods and what different tests and procedures involve, and thereby contribute more meaningfully to discussions. Brett et al. (2014) argue that training in research methodology would empower people to contribute to discussions surrounding the research design and to ask questions about the study rather than limiting their contribution to accounts of their lived experience.

By way of example, a study which set out to evaluate the impact of involvement of older people with dementia and age-related hearing and/or vision impairment in a multi-site European research study provided six hours of training (reduced from a six-day programme) broken up into manageable chunks of one hour “to support their input to the research” (Miah et al. 2018). The training covered research awareness, understanding the research process (including making sense of published papers), qualitative and quantitative methods, developing and evaluating interventions, health economics, ethics and governance.

Some researchers have found the Participatory Learning and Action research approach helpful in situations where they have no familiarity with the languages or culture of the target population, where the targeted minority groups lack trust in the researchers who are perceived as representing ‘the establishment’ or dominant cultural group, and where some of the potential participants have low levels of language and literacy (O’Reilly-de-Brún et al. 2016). However, this may necessitate fairly extensive capacity building skills and training, and a high level of involvement which would not correspond to the wishes and abilities of every person interested in PI.

There may be considerable differences regarding the content, duration and type of training offered to people with dementia engaged in PI. Such training may be helpful and beneficial if it is the right level for the people involved but if perceived in terms of training people with dementia ‘to bring them up to the level of researchers’, it could be construed as failing to value the knowledge that they already bring to the research process.

One of the key aims of PI is for researchers to benefit from the experience and insight that people with dementia have in relation to the research topic and the conduct of their study. Training is sometimes perceived as a threat to the input from people with dementia. Hayes et al. (2018), for example, highlight the threat of lay representatives over time becoming so highly informed that they identify with researchers to the extent that they become distanced from what would normally have been their own viewpoint. Goffman (1963) mentioned a similar situation in his seminal work on stigma in which he described situations in which people with a particular attribute (albeit a discrediting one), have a new career thrust on them, that of representing their ‘category’ (i.e. people who share the same attribute). In so doing, he suggests they break out of a relatively closed circle and start to have dealings with people from other categories, gradually ceasing to be representative of the people they represent (1963, p.39). Caron-Flinterman et al. (2007) use the term proto-professionalism to refer to the internalisation by patients of biomedical scientific language and principles. They point out that in the context of biomedical research, these people come to be regarded and treated as ‘real partners’ but are considered by some as having lost sight of the patient perspective.

As mentioned earlier, people with dementia are not expected to be representative (in the statistical sense) when contributing to PI. One of the aims in involving them in research in this way is to hear their views and learn about their experiences and perspectives as a person with dementia (in relation to all areas and stages of a particular study), not as a professional researcher. Ives, Damery and Redwood (2013) call this the ‘professionalisation paradox’ which they claim represents a fundamental tension between the moral and pragmatic motivations of PI in health research. They emphasise the importance of PI providing researchers with access to the lay perspective and suggest that even if researchers also had that perspective personally (i.e. themselves having the condition which was the topic of research), they would lack ‘outsider’ status and the ability to be a critical observer, free from the concerns and pressures of publication, generating income and building up an impressive CV.

Ives and colleagues also ask with regard to PI, “Why would we want to put unskilled lay people in a position where they can direct a process about which they know and understand little?” (2013, p.183). They go on to reflect on the benefits of training in empowering people engaged in PI and ensuring that they can legitimately enjoy an equal position of power alongside professional researchers but conclude that this jeopardises their ability to act as lay representatives. They do not claim that training causes people engaged in PI to lose their experience of illness or disability but that they become less able to authentically reflect the lay perspective and that their contributions become tamed and more in line with those of the researchers.

As a counter argument, Staley (2013) points out that PI is a complex activity involving many different kinds of involvement, not all of which require specific training. For some forms of involvement, such as advising on how to reach or better motivate people to take part in research or how to make documents more accessible, a person who is 'closer to the ground' may be highly effective and would not necessarily benefit from formal training in research (Staley 2013). She further remarks that even when training is provided, in most cases, the aim is not to equip people to do research but rather to provide them with sufficient knowledge to understand the basics of research and to be confident that they can contribute to the study effectively.

A possible exception is where people engaged in PI actually 'do' research (e.g. help recruit participants, gain informed consent and collect and analyse data). This is in keeping with the concept of PI occurring along a continuum and with the possibility of people with dementia being co-researchers but they must have the necessary scientific rigour to conduct high quality research. In addition, researchers may need to obtain informed consent from research participants for their anonymised data to be shared with people engaged in PI and supervision would be needed (Hoddinott et al. 2018). It may also be necessary to address certain governance issues (e.g. in relation to terms of employment, legal issues and the right to payment (Hoddinott et al. 2018). Such involvement would need to be supervised because a full grasp of all the relevant issues and assimilation of knowledge required to conduct high quality research would normally take several months or years to acquire, not just several hours.

> ▶ ***"Training in photovoice and time to get to know others in the research team made all the difference. I liked that it was at a slower pace to start with. There was lots of time to understand it before we started using it"* (Co-researcher with intellectual disability in Scotland).**

It could be argued that training should be mainly about enabling people to contribute more to the research process than solely accounts of their personal experience (Brett et al. 2014) and about helping them to communicate their opinions and perspectives more effectively to researchers (Morrow et al. 2010). In many cases, it would be helpful to reframe training so as to focus on the issue of providing support, which could include providing accessible information and helping people to develop skills that they might find helpful for PI. If researchers present PI as something for which training is necessary, this may give the wrong message to people who might otherwise be interested in it. It may be perceived as indicating that PI is only for highly educated people or for people who are good at and willing to study. People who do not fall into that category may feel discouraged or disempowered. The term 'co-learning' suggests a more constructive approach. This term is sometimes used to describe collaborative learning for which the goal is not to turn people into researchers but to help them to understand the research process and at the same time enable researchers to learn about a person-centred approach and PI (Hoddinott et al. 2018).

- ☞ **Could it be that the need for training to do PI indicates the lack of ability of researchers to make PI accessible?**
- ☞ **Should the concept of training for PI be replaced by that of appropriate support and the opportunity to acquire useful skills?**

Recommendations for researchers

- Make it clear when looking for people to engage in PI activities that there is no age limit on involvement and that everyone's perspectives and experiences are important (e.g. those of younger and older people, those of people with mild, moderate and advanced dementia etc.).
- Where training is a requirement for PI (for example in a co-researcher role), ensure this is done appropriately over an extended timeframe if required and with ongoing support and reinforcement of learning.
- Avoid making training a requirement for others forms of PI.
- Ensure that any training offered, is in keeping with the nature of the proposed activities, the requirements of the study and last but not least, the abilities and desire of the people involved to receive it, taking into consideration also the challenges that many people with dementia face.
- Recognise that the suggested need for training may be off-putting to some people, especially those with lower levels of education or who lack confidence in their ability to learn, who have intellectual disabilities or who have difficulties with language and literacy.

- Create the conditions in which people who wish to share their experience of dementia with researchers can do so without having to do any training.
- Clarify the roles and responsibilities of the key researchers involved in the study as well as those of people engaged in PI (and how this corresponds to the possible need for support).
- Learn how to make PI accessible to people who do not have prior experience or knowledge about research or who are new to PI (e.g. offer support to develop certain skills or confidence to express views to researchers or ask questions).
- Bear in mind what different people who are interested in contributing to PI can realistically be expected to learn in the time frame available and in keeping with their individual capacities and interests.
- Consider the possibility of offering, and seeking support for, appropriate and accessible support (including explanatory information) in another language.

Recommendations for research ethics committees

- Assess how relevant support is organised in the context of PI and whether it is suited to the needs and abilities of people with dementia.
- Consider the role of people engaged with PI and ensure they are fully and appropriately supported, including for any essential training required.

Recommendations for funders

- Offer the possibility to apply for costs to cover the promotion and provision of appropriate skills and information materials for people engaged in PI activities where appropriate.
- Do not make training compulsory for most PI activities.

Summary

The prevalence of dementia increases with age so it is particularly important to involve older people with dementia in Public Involvement (PI) as well as younger people. The continued exclusion of older people from research, including PI, may reflect ageism which, like racism and sexism, reinforces social inequalities by legitimising and sustaining inequalities between groups. The internalisation of ageist attitudes, based on negative stereotyping and devaluing older people, may have a negative impact on confidence and self-esteem and interfere with readiness to take part in PI.

PI enables researchers to benefit from the experience and insight that people with dementia have in relation to the research topic and the conduct of their study. It is sometimes suggested that people with dementia should be provided with training so as to have a sufficient understanding of research to be able to contribute meaningfully to discussions. A counter argument is that training may result in an over-identification with the position of researchers, make people with dementia less able to authentically reflect the lay perspective and jeopardise their unique outsider status and position as critical observers. We are not aware of evidence that this occurs. We suggest that the key issue is that PI work should be organised and presented in such a way that training is not necessary because it may be off-putting to some people and make them feel inadequate. Opportunities to develop relevant skills and to obtain clarification about relevant issues should be provided if and when requested.

Acting on input from PI activities

Managing different perspectives

A key principle of ethical research is that it needs to be carried out in accordance with rigorous scientific standards, but not everyone engaged in PI will necessarily be familiar with or fully understand those standards. This could be considered a limitation to the involvement of people with dementia in certain PI activities. It could also be a strength in that they can look at various aspects of the research with a critical eye, without subjecting themselves to the kind of self-censorship that might come with a more in-depth knowledge of research methods and methodologies. This brings us back to the issue of whether training is appropriate.

In their systematic review of the impact of PI on health and social care research, Brett et al. (2014) report two examples of researchers adapting their research design (i.e. not having a placebo arm) in response to PI feedback about what constitutes a more ethical study design from the lay/user perspective. They state,

> *"While this may have impacted on the robustness of the study design for researchers, it provided a more ethical study design for users. (…/…) This can provide an important challenge for researchers and highlight potential tensions between academic criteria of good quality research compared with the user perspective of what constitutes appropriate research. It is the researchers' role to ensure any study is of high quality scientifically and to explain and justify the research design and negotiate changes that make the study more acceptable to service users without compromising its robustness or validity" (2012, p.641).*

Drawing on the CIOMS guidelines for biomedical research involving human subjects[33], Emanuel et al. (2000) remind researchers that unsound research (i.e. not conducted in a methodologically rigorous manner) is unethical as it exposes people "to risks or inconvenience to no purpose". Researchers therefore need to be able to balance the desire to respect and include input from the public and patients in their research against ethical concerns about the scientific validity of the research. Procedures are needed to manage such situations in a collaborative way that values the perspectives of all involved and, where possible, leads to a decision that is acceptable to all, but someone has to have ultimate responsibility. In most cases, that someone is the Principal Investigator although he or she may seek advice from an internal steering committee and people with dementia may be sitting on that committee.

> *"Lay people need to know enough about the research process to appreciate which aspects cannot be altered without harming scientific quality. But ultimately it is always the researchers' responsibility to draw on their technical expertise to ensure the robustness of the research" (Staley 2013, p.1).*

Ensuring involvement at a sufficiently early stage

Commissioning structures can be very complex and bureaucratic with technically defined subject areas and methods which may preclude meaningful involvement, and often a lack of awareness and resistance from professionals (Tembo, Morrow, Worswick and Lennard et al. 2019). Not surprisingly, people with dementia are rarely involved in research commissioning (e.g. priority setting and developing calls for funding). According to Tembo et al. 2019), significant changes are needed regarding communication, practices, systems, structures and cultures which all hinder meaningful contribution of patients and the public to research commissioning. They point out that concerns have been reported, for example, about the lack of impartiality of members of the public, suggesting that the public lack objectivity, may be biased and may be influenced by personal interests (Tembo et al. 2019), thus calling into question the 'rationality' of involving the public in decisions about the allocation of funds for research. While Tembo and colleagues raise an interesting point, the accusation seems to be misplaced and overstretched. Surely other stakeholders and the researchers themselves are not impartial; this is not only a concern about the public. It thereby fails to balance these concerns against others linked to the potential bias and self-interest of researchers (e.g. linked to scientific curiosity, rivalry for scarce funds, the desire for peer recognition and the need to publish for job security), which might contribute towards tokenistic involvement of people with dementia.

People with dementia are also often absent at the stage of development of research proposals. In many cases, their involvement starts when the project starts because funding is not yet available for PI until funding has been awarded. Consequently, it may be difficult for them to shape the nature of their involvement and certain aspects of the research design if these have already been described in detail in the proposal and the accompanying description of activities, and for which specific funds have been allocated in the budget. If and when it becomes apparent that certain decisions have already been made, people engaged in PI may be reluctant to raise questions or suggest changes, anticipating that their input will be opposed, ignored or not considered a realistic option (Morrow et al. 2010). Some

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[33] CIOMS stands for the Council for International Organisations of Medical Sciences (1993)

people with dementia who contribute in the very early stages of research may find that they are unable to continue once the research officially starts because their health or circumstances may have changed in the time between the application for funding and the start of project (which can be quite lengthy e.g. up to a year).

Reflecting on the issue of payment

Volunteering

In the vast majority of cases, people contribute to PI (and/or become participants in research) on a volunteer basis. According to Wilson (2000, p. 215- 216):

> *"Volunteering is any activity in which time is given freely to benefit another person, group or cause. Volunteering is part of a cluster of helping behaviors, entailing more commitment than spontaneous assistance but narrower in scope than the care provided to family and friends. (…/…) Some think the desire to help others is constitutive of volunteering. Others subscribe to the view that volunteering means acting to produce a "public" good: no reference to motive is necessary."*

Volunteering is common and somewhat of a tradition in some countries. A longitudinal survey of volunteering amongst some 30,000 people over the age of 50 in 11 Continental European countries found that it was most common in Sweden, Denmark, Germany, the Netherlands, Belgium, France and Switzerland (ranging from 14.4% to 21%) and less so in Spain, Greece, Italy and Austria (2.9% to 8.3%) (Hank and Erlinghagen 2010). People who volunteer to be involved in research do so for a variety of reasons. Some of these reasons may result in personal benefit such as increased self-esteem, social contact or learning new skills, without this necessarily being the driving force for such activities.

There is a complex relationship between volunteering and a range of factors such as the number of hours that people spend in paid employment, overall income, employment status and other responsibilities, such as caring (Wilson 2000). One European study found that people with higher levels of education and those not in paid employment were more likely to take up volunteering (Hank and Erlinghagen 2010) but again this is complex, not always linear and may be linked to certain types of volunteering. Data is lacking on how people with dementia hear about and come to be involved in PI and how they can be facilitated (i.e. what do they need to be on board, which methods of involvement are appropriate?). As the concept and recognised value of PI in research becomes more common, it will be important to reflect on how to attract people with dementia from diverse backgrounds and from countries where volunteering is not common, and to explore the challenges and barriers which hinder the participation of some groups of people.

Payment for Public Involvement

In the last few years, there has been a movement towards paying 'patient representatives' for their expertise, time and effort based on concerns about fairness and equality. An argument in favour of payment is that the expertise provided by people with dementia ('experts by experience') is of equal importance to that provided by experts in other domains ('experts by training'). In this way, payment for involvement (as opposed to merely covering out-of-pocket expenses) is seen as formal acknowledgement of the equal value of the expertise provided by people with dementia and people with expertise in other domains.

Payment for PI is gaining ground particularly in the area of research into the development of new drugs as witnessed by policies and guidelines being developed by the EFPIA[34] (European Federation of Pharmaceutical Industries and Associations), as well as in the 'Patient Pool' currently being set up by the Innovative Medicines Initiative (IMI) which funds a lot of dementia research in Europe[35]. Aside from the issue of equally valuing the expertise provided by people with dementia, it is perhaps particularly important for pharmaceutical companies, who invest millions in dementia research and if successful make considerable financial profits, to demonstrate fairness in properly compensating everyone who contributes towards making the clinical trial a success.

An accessible report explaining to the general public and patients what they might want to know about payment for PI National Health Service-related, public health or social care research was produced in the United Kingdom in 2016 by INVOLVE[36]. The authors emphasise the importance of payment for PI in terms of recognition of time, skills and expertise, equality and helping ensure that a wide range of people have the opportunity to influence the research process.

> *"It is good practice for organisations to pay you for your involvement in research. This is one way in which you can be rewarded for the time, skills and expertise that you contribute to the research process. Paying people for their involvement in research helps to support more equal partnerships between researchers and members of the public. It helps to support the*

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[34] For more information about EFPIA, see: https://www.efpia.eu/

[35] For more information about the IMI, see: https://www.imi.europa.eu/

[36] INVOLVE is a national advisory group funded by the National Institute for Health Research (NIHR) to support public involvement in NHS, public health and social care research.

> *inclusion of people who might not otherwise be able to get involved, whether for financial or other reasons relating to access. Consequently, it widens the potential pool of people who might influence the course of research" (INVOLVE 2016, p.8).*

Payment of out-of-pocket expenses (e.g. for travel, accommodation and meals), including swift and easy reimbursement of costs, may make it possible for people on a low income to engage in PI and without this affecting their wellbeing. Many researchers/funders also cover the costs of supporters to enable people with dementia who need assistance for travel or during meetings to engage in PI.

Challenges linked to payment

There are mixed views in the literature and amongst those directly involved about payment for PI (Black et al. 2018). Many people with dementia have limited opportunities to earn money and people with limited financial resources might appreciate the opportunity to be paid for their contribution to research. Payment might also encourage people from more diverse backgrounds to engage in PI (i.e. people for whom payment would be an incentive, people who are perhaps less motivated by an awareness of social and health related issues and/or people would not normally do volunteer work).

There is also the issue of payment being interpreted as a sign of respect, quality and value. This has also been reported in relation to participation in qualitative research (Head 2009). However, offering payment for PI can also be perceived as devaluing volunteering. In an article on incentives for research (including financial payments), Grant and Sugarman state,

> *"Incentives may induce people to do the right thing, but for the wrong reason, and thus undermine responsibility, altruism and other important values" (2004, p. 722).*

Moreover, payment does not automatically convey or guarantee respect for the person receiving it and their contribution. Likewise, voluntary involvement does not preclude respect for a person's contribution or value.

It is important to consider whether payment represents a financial incentive to motivate people to contribute towards research, thereby making the invitation more attractive, or amounts to coercion (an exercise of power resulting in people doing things against their principles or better judgement) (Grant and Sugarman 2004). In the context of research, payment is sometimes considered as a potential means of coercion. Payment may be considered as involving 'undue influence' and thereby undermining consent, but this would not apply to PI which does not normally require informed consent as the people involved are not research participants. Moreover, as PI does not tend to involve the same risks as participation in research, especially in clinical trials, Millum and Garnett (2019) argue that payment for research participation is not a form of coercion that undermines consent, which necessarily involves threat, but that payment constitutes coercion as subjection[37]. As the issue of informed consent is not relevant to PI, it should not be necessary to be as strict about payment for PI as for participation in research.

In PI, payment tends to be understood within an economic paradigm, almost as a form of trade or payment in exchange for a service. Were it to become an obligation to involve people with dementia in PI, financial incentives might start to be used primarily to attract them to PI rather than in recognition of a fair exchange. Some similar issues might then need to be considered as for participation in research. However, the issue of risk and burden is not comparable. Payment might nevertheless also run counter to the principle of justice in that more people with dementia who would welcome payment might eventually be involved in PI, their willingness to be involved exploited and a further imbalance/lack of diversity created. Nevertheless, it is probable that financial incentives to engage in PI would not consist of inducing people to do something to which they are adverse but more to do something that they might otherwise have had no particular interest in.

Moreover, not everyone who contributes to PI wishes to be paid. Payment suggests a commercial exchange of goods or services, accompanied by obligations to provide what the other party wants in return for the payment. People with low self-esteem may be wary of getting involved if payment is offered as they may worry about whether they will be able to provide what is required (Beresford 2019).

With regard to people who are on a low income or receiving benefits from the State and who might benefit from payment for their contribution, such payment may be problematic. It may, for example, jeopardise their rights to benefits, result in them having to make complicated or costly tax declarations or involve a risk of being accused of fraud (e.g. based on travelling to and contributing towards meetings and discussions despite having been declared unfit to work on the grounds of disability).

In smaller projects such as those conducted by PhD students or researchers in institutions with limited funds, the obligation to pay people for PI activities may be problematic

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[37] the state of being dominated, controlled or influenced

and hinder potentially valuable research. Payment may result in competition, with people interested in engaging in PI being attracted to projects offering a better rate of payment. Competition can be positive but may not necessarily attract people with dementia interested in PI to the most worthwhile projects. Researchers involved in small-scale research projects who can perhaps only offer EUR 20 to EUR 50 per person per meeting may find it difficult to compete with offers of EUR 200–500 for involvement in a certain task. If PI is conducted, as generally recommended, throughout the whole research process, even small payments can add up and necessitate sizeable funds.

The significance of the amount paid also needs to be considered. Small payments may be considered insulting, especially when people are working alongside professionals who are perceived as being highly paid. The amount paid may also depend on who is conducting the research, the amount of funds available and the scale of the project. In its position paper on PI, drafted in collaboration with INTERDEM and the EWGPWD, Alzheimer Europe argued that if funds are available for the payment of external experts (e.g. fees to attend a meeting or daily allowances), the same money should be offered to people with dementia engaged in PI on an equal basis (Gove et al. 2017).

Input from people with dementia would be helpful on this issue to determine, for example, whether and if so on what basis people with dementia should be paid for PI, what amount they would find acceptable and reasonable, and whether they would prefer a gift voucher or a donation to be made to a good cause in their name. It is unlikely that a decision could be made for every group of people with dementia involved in PI or for every research project but people with dementia need to have their say in the matter. When trying to decide what would be fair, this is sometimes phrased in terms of compensation according to 'fair market value'. Some organisations have clear guidelines. The American Patient-Centred Outcomes Research Institute (PCORI) (2015, p.1), for example, recommends that payments for people engaged in PI[38] should reflect "the level of expertise, commitment, responsibility, the type of work involved, and the degree of participation contemplated" and should also take into account a range of associated factors such as the effort involved, a person's experience, skill level, time for preparation, comparable levels of pay and responsibility of participating professionals, local wages and national restrictions and that this should be informed by principles of equity.

Recommendations for researchers

- Involve people with dementia in PI as early in the process as possible (e.g. during the development of the research proposal and protocol).
- Give people with dementia involved in PI prior to the official start of a research project an estimate of how long it may take before the research starts. Also, inform them of the risk that the project may not receive funding and hence eventually not go ahead.
- Explain at the start of their involvement how input from people with dementia will contribute towards the research and about any restrictions which might already exist (e.g. linked to funding, ethics approval, available resources etc.).
- Explain that all ideas are welcome, that the researchers welcome criticism and suggestions, that not every suggestion will necessarily be acted on but that there will be transparency about how decisions are made and that people with dementia engaged in PI are key players in the decision making process.
- Avoid making global assumptions about whether or not people want to receive payment.
- Clearly state whether PI for a particular project is on a volunteer basis or in return for payment (and in the case of the latter, if it is possible to decline payment).
- Irrespective of whether participation in PI is on a voluntary or paid basis, provide a clear and understandable explanation of what costs can be reimbursed and/or what payment people are entitled to receive, how to go about obtaining such reimbursements or payments, and ensure that reimbursement or payments are made promptly.

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[38] Described as engaged research partners in their document. See also "Working together with patients: principles for remunerating patients, patient organisation representatives & carers for work undertaken with the pharmaceutical industry" produced by EFPIA (June 2019).

[39] For an example of a document providing advice on payment to people in receipt of welfare benefits (INVOLVE 2018), please see: https://www.invo.org.uk/wp-content/uploads/2018/11/INVPayInfSheet-Nov2018.pdf

- Develop a clear and transparent payment policy (if payment is envisaged) in collaboration with people with dementia.
- If payments are made to experts by training for the same kinds of tasks as those being carried out by experts by experience, offer the same payment to each.
- Consult relevant guidance on fair remuneration.
- Ask people involved in PI, who were offered payment, whether they considered the amount fair and take this into consideration when discussing any future possible payments.
- If payment is offered for PI, also provide information about the possible impact this may have with regard to taxes, benefits and certain rights or entitlements that a person might have (e.g. linked to disability, mobility or tax exemptions).[39]
- Try to ensure that people engaged in PI can have available payments transferred directly from the research funds to a charity or worthwhile cause of their choice, without this necessitating any money passing through their own accounts.

Recommendations for research ethics committees

- Ask researchers to provide details of pre-study involvement of people with dementia (e.g. in the topic selection, plans for PI and possible suitability of various methods for involving people with dementia).
- Assess whether the dedicated budget and criteria for the reimbursement of people involved in PI are fair.

Recommendations for funders

- Involve people with dementia in discussions about funding and commissioning.
- Assign a score to the involvement of people with dementia in the writing of the research protocols when selecting projects for funding.
- Provide a dedicated budget for the reimbursement of people involved in PI.

Summary

PI is carried out within the context of research which must fulfil certain criteria (e.g. for social value, safety, rigour and scientific validity). However, all aspects of research, including the PI component, must be ethically sound and this requires some degree of flexibility from all involved. Researchers nevertheless stand accountable for their research and need to be able to justify their decisions. PI needs to start from the very beginning, before key decisions have been taken about the study. People get involved in PI for different reasons. Some people with dementia are interested in doing PI work on a voluntary basis and others would appreciate being paid. In some countries, volunteer work is more common than in others but volunteering is an individual choice. Arguments in favour of paying for PI are often focused on concerns about fairness, equality and respect but this should not be taken to imply that people who do PI on a voluntary basis are being used or disrespected or that payment automatically translates into respect. Ideally, people should have a choice and any policy for payment (or reimbursement of costs) should be clear, understandable and transparent.

Monitoring and reporting

Planning PI right at the start of a research project is important but does not in itself guarantee success. As PI is still in its early days, more work is needed to evaluate its effectiveness, justification, the appropriateness of methods used and its conceptual aspects, including ethical implications. With regard to the issue of involvement being meaningful, some form of monitoring and evaluation is needed (e.g. to check whether people with dementia feel that their voices are being heard, to check whether researchers are acting on the input they receive and to evaluate in what way and to what extent PI is having a positive impact on the research process).

Researchers are increasingly taking measures to report on PI and to evaluate it. However, whilst there may be some pressure on researchers to engage in PI, there are often little or no requirements for detailed reporting beyond the initial plans. In 2011, Staniszewska and colleagues developed the GRIPP (Guidance for Reporting Involvement of Patients and the Public) checklist, which represented the first international attempt to strengthen the quality of PI in research. In 2017, this was revisited and resulted in GRIPP2, a short form and long version of a reporting checklist for reporting of the context, process, and impact of PI in health and social care research (Staniszewska et al. 2017). They highlight the ethical imperatives of reporting research in such a way that it is understandable and possible for other researchers to learn from and build on the experience acquired by their peers.

Staniszewska et al. (2017) suggest that failure to report PI in study reports may be seen as a form of misreporting and misrepresentation. With regard to the ethical involvement of people with dementia in research in the context of PI, they also emphasise the need to develop a patient or service user version of GRIPP2. This would not only enable people with dementia engaged in PI to play a more active role in committees responsible for reviewing research reports but also to develop assessment criteria which reflect their priorities and concerns.

Ethics approval and legal agreements

The issue of ethical approval for PI is often unclear. Hardavella et al. (2015) raise the issue as to whether PI raises any ethical concerns for those involved. They state:

> *"They are actually acting as specialist advisers, providing valuable knowledge based on their experience of a health condition or public health concern; therefore ethical approval is not needed for the active involvement in research, e.g. helping to develop a protocol, questionnaire or information sheet, or for being a member of an advisory group or co-application" (2015, p.224).*

Many researchers have reported being obliged to obtain ethical approval for PI activities. Some have voiced the opinion that it would be necessary to seek ethical approval if discussions were audio-recorded or if they would like to publish an article about the PI activity in a peer-reviewed journal. There is even anecdotal evidence of ethics approval being requested for a person with dementia to co-author an article. Fortunately, there are also good examples of research ethics committees having reviewed outlines for planned PI work and confirmed that this did not constitute research and did not require ethical approval (Poland et al. 2014).

In an article by Di Lorito et al. (2017), a small number of studies were reviewed on the topic of 'peer research' and the European Working Group of People with Dementia (EWGPWD) contributed towards the interpretation of the findings. The members of the EWGPWD had no objection to meetings and discussions being audio-recorded and felt that it was important to contribute towards articles in peer-reviewed journals if and when appropriate. Their focus was more orientated towards empowerment than protection from harm or methodological issues linked to actual participation in research (as opposed to PI). They emphasised the importance of:

> *"human rights (e.g. autonomy and respect), justice and equity (e.g. opportunities for people with dementia to be invited to participate in research) and tokenism (e.g. relevance of involving people with dementia from the beginning of the research and not just in the process but also in the decision making e.g. around which areas of research should be prioritised)" (Di Lorito et al. 2017, p. 66).*

Researchers who submit applications for ethical approval usually have to demonstrate that they have reflected on any issues which pose a potential threat to their own wellbeing (e.g. psychological, physical or other) as well as that of research participants. The involvement of people with dementia in research in the context of PI should in principle also not result in harm to themselves. Consequently, Principal Investigators responsible for seeking ethical approval should also reflect on the wellbeing and protection from harm of people engaged in PI in the project. This should not be taken to imply that ethics approval should be obtained for their involvement but that their wellbeing and safety should be equally considered.

As mentioned earlier in the sub-section on training, people with dementia can contribute to PI in different ways. In situations where people with dementia are directly involved in research tasks (e.g. conducting interviews, moderating focus groups or accessing data), they should have the necessary and appropriate skills and should be able to conform to the same ethical requirements as other researchers, namely with regard to the conduct of good science and the promotion/protection of the wellbeing of research participants.

Some researchers and organisations ask people who take part in PI activities to sign contracts covering issues such as confidentiality, intellectual property, data protection, financial compensation and travel expenses. This is particularly common in the field of biomedical research in which pharmaceutical companies are involved. These documents may be difficult to understand and quite daunting in general, but even more so for people who have cognitive and other impairments or with low levels of language and literacy. In addition, some contain clauses about potential conflicts of interest and propose exclusivity clauses. Exclusivity clauses, especially when based on a narrow definition of conflict of interests, may seriously limit opportunities for PI and the independence of people with dementia involved. Guidelines on this issue were developed in 2018 by Myeloma Patients Europe, WECAN, PFMD and experts of pharmaceutical companies.[40] Some of these have been incorporated into the recommendations below.

Recommendations for researchers

- Include details in the research protocol about how PI will be conducted (e.g. when, in relation to which aspects of the research, methods for involvement, funding required for relevant activities etc.).
- When reporting on studies and in peer-reviewed articles, include details of how PI was conducted, with whom and what the impact of it was.
- Be specific, when reporting, about the origin of various input from mixed groups (e.g. was a particular point which led to an important change or decision made by a person with dementia, a carer or a member of the general public? Did different stakeholders disagree on particular issues or have different perspectives?).
- Mark documents as ‘confidential’ if they are and remind people with dementia at the beginning and end of discussions if these were confidential (in addition to the usual requirement not to repeat issues raised in the context of PI outside of that context).
- Offer or at least be prepared to talk through any documentation that people with dementia may be asked to sign and to address any concerns they may have.
- Summarise key requirements (e.g. related to codes of conduct or practice) in any contracts that people with dementia are asked to sign, and refer the signatory to a readily available full document so as to keep the contract as concise as possible.
- Plan and budget for pre-meetings if required. For example, PI involving people with an intellectual disability can include a meeting in advance of steering/advisory group meetings to talk through the agenda and plan involvement, resulting in increased confidence and active participation.

Recommendations for research ethics committees

- Make it clear to researchers that although ethical research should include PI and that PI should be performed in an ethical manner, PI does not require ethical approval from a research ethics committee *per se* (and explain any exceptions to this rule).
- Ask in feedback how researchers will involve people with dementia in PI prior to the start of studies if this is not stated in applications
- Ask researchers to include in applications details of PI which was conducted prior to review by the research ethics committee.

Recommendations for funders

- Make detailed reporting of PI a requirement for funding (e.g. link it to a deliverable).
- Do not request or expect exclusivity from people with dementia for PI activities.

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[40] The guidelines are available at: https://www.mpeurope.org/legal_agreements/wp-content/uploads/2019/03/Legal_Agreements_A5_3mm-bleed_PRINT_v2.pdf

Summary

It is important that Public Involvement (PI) is rigorously monitored and evaluated. Often, there are no formal requirements for this. People with dementia need to be involved in this under-developed aspect of PI work (i.e. in the development of monitoring, evaluation and reporting tools). However, the concept of PI is not yet widely understood across Europe. Ethical approval is not required for PI because people engaged in PI activities are not research participants and are not providing data. People with dementia engaged in PI should nevertheless not come to any harm as a result of their involvement. Whilst ethics approval should not be required, their wellbeing and safety, as well as that of the researchers, should be given due consideration.

Contracts covering issues such as confidentiality, intellectual property, data protection, financial compensation and travel expenses are often long, complicated and difficult to understand in general and especially for people with cognitive difficulties and/or difficulties with language, literacy and education. Some contracts contain exclusivity clauses which limit opportunities for PI. Short accessible summaries and necessary support should be provided, and exclusivity clauses avoided for PI activities.

Part 2: Ethical Challenges Linked to Recruitment and to Informed Consent

The importance of involving a diverse set of participants in research

In Part 1, we looked at issues related to the contribution of people with dementia to research through Public Involvement. This involves them sharing their perspectives, opinions and insight with researchers, thereby having a voice and helping improve the research process. We now turn to issues related to the contribution of people with dementia to research through participation in research studies (i.e. as a research participant) which also raises several ethical challenges in relation to the participants, the researchers and the researcher process.

In accordance with the requirements for good science, researchers must recruit participants who are best suited to providing a response to their research question or hypothesis. Depending on the methodology adopted, this means involving people who are representative of the group targeted by the study and/or who can together produce a broad range of perspectives and experience on a particular issue. Populations are made up of very different people and there is a risk of people with certain characteristics being excluded. All too often, the very people who are most in need of research to improve their condition or situation are the ones who are excluded (Sin 2005). In addition, and in keeping with the principle of justice, measures must be taken to ensure 'fair subject selection' whereby the most stigmatised and vulnerable individuals in society are not selected for participation in risky research and the rich and socially powerful for research more likely to entail some personal benefit (Emanuel, Wendler and Grady 2000).

Historically, a first obstacle was to ensure that people with dementia were involved as participants in dementia research. It was often assumed that they were incapable of communicating their thoughts and feelings and that data obtained from them would be unreliable (Hubbard, Downs and Tester 2003). This was combined with an emphasis on biomedical research (Lepore et al. 2017). Regarding clinical trials, people with dementia are the ones who will eventually be using dementia drugs and therefore they need to be involved in their development. However, some groups of people with dementia (e.g. those who are no longer living at home, those without a partner and those with comorbidities) are often excluded from clinical research. This means that the study populations are often not representative of the broader population of people with dementia (Jongsma, van Bruchem-Visser, van de Vathorst and Mattace-Raso 2016). The informed consent process and the inclusion and exclusion criteria for studies are often stumbling blocks in terms of involving a diverse set of people with dementia in research. Behavioural and psychological symptoms of dementia (BPSD) may also sometimes result in exclusion from various studies (e.g. if researchers do not feel able to cope or have concerns that a potential participant might not comply with research requirements).

People with dementia are best placed to provide information about their experience of living with dementia and their reactions and preferences on a range of issues (with all necessary care, attention and support having been provided to make this possible and to obtain informed consent, sometimes using non-written, non-verbal options). Unfortunately, the perspectives and experiences of people with dementia are still sometimes unnecessarily expressed via the intermediary of informal carers or health and social care professionals. At the same time, it is important to bear in mind that for some ethnic minority groups, the consent of the family is also very important. Similarly, country-specific guidance and legislation should be followed when including people with dementia who lack capacity and where opinions are sought on their behalf from those legally permitted to do so. Restricting the informed consent process solely to the person with dementia may be too individualistic an approach within some communities (Berdai-Chaouni, Claeys and De Donder 2018).

Finally, many people with dementia find themselves excluded from research into issues that are not specifically related to dementia or into medical conditions other than dementia. This seems to reflect an assumption that having dementia is the sum total of a person's identity, overshadowing everything else, including experiences, hopes and perspectives linked to other aspects of people's lives, including other diseases.

Dementia is experienced in different ways by different people. People with dementia have different levels of support, different social relationships and roles, and different capacities. They also have a range of different cultural backgrounds with many people identifying with a number of sub-groups including ‘communities within communities’. The diversity of people with dementia should be reflected in research samples but often isn’t.

As mentioned earlier, people who have lower levels of education, intellectual disabilities, lower health literacy, more advanced dementia or are from a minority ethnic group are frequently excluded, as are many older people living with dementia who may also have other health conditions (e.g. depression, cancer or diabetes). People with dementia should not be excluded or miss out on opportunities to take part in research solely on the grounds of characteristics they may or may not have. Ignoring or being ignorant of the needs and interests of large proportions of people with dementia, resulting in them not being involved in research, runs counter to the principle of equity and involves epistemic injustice. It means that some people with dementia are not being granted the same opportunities as other people to contribute towards science (with the potential burdens, risks and benefits that this entails) and thus to society. With regard to epistemic injustice, they are not being recognised or considered as knowers, which is harmful for them in that it leads to discrimination and in researchers missing the opportunity to hear their voice, resulting in limited and biased knowledge about the topic.

Whilst the aim of research is not to benefit individual participants, many participants do feel that they gain something personally out of participation such as a feeling of personal satisfaction from pursuing altruistic motives, increased contact, a feeling of hope, of having a purpose and of empowerment, and access to ‘state-of-the-art’ consultation and disease monitoring, information and management. Potential benefit must of course be balanced against potential harm. However, an overemphasis on harm may lead to the possible benefits of participation being overlooked. Hellström et al. (2007) conclude that there are serious consequences to not including people with dementia in research and suggest that the question should not so much be about whether people with dementia should be included in research, but rather how we can best achieve this and how we can afford not to. Excluding some groups of people from the opportunity to experience such perceived benefits could be considered as discriminatory and institutional racism (Hussain-Gambles 2003).

From a scientific perspective, failing to involve a diverse set of people with dementia in research means that the responses to research questions and hypotheses are incomplete, misleading and at worst wrong. Konkel suggests that not only does the failure to involve a more ethnically diverse set of people in research exacerbate health disparities but it also represents “a missed scientific opportunity to fully understand the factors that lead to poor health and disease” (2015, p. 298). Bhopal and Sheikh further suggest that the exclusion of people from minority ethnic groups “promotes inequality, maintains inequity and is unethical and institutionally racist, if not illegal” (2009, p. 2). From a disability and human rights perspective, appropriate measures should be taken to ensure that people with dementia are enabled and empowered to take part in research on an equal basis as people without dementia. The following general principles from Article 3 of the CRPD, which were developed in the context of attempts to ensure the equal involvement of people with disabilities in society, could be equally helpful when considering the involvement of a diverse set of people with dementia in research:

- Respect for inherent dignity, individual autonomy including the freedom to make one’s own choices, and independence of persons,
- Non-discrimination,
- Full and effective participation and inclusion in society,
- Respect for difference and acceptance of persons with disabilities as part of human diversity and humanity,
- Equality of opportunity,
- Accessibility,
- Equality between men and women.

Recommendations for researchers, research ethics committees and funders

Strive to address inequity so as to ensure that a broad range of people with dementia are involved in research as participants for the following reasons:

- People with dementia should all have an equal opportunity to be involved in research (in keeping with the principles of justice, solidarity and respect for autonomy).
- Excluding certain groups of people or people who possess or lack certain characteristics would be discriminatory.
- Having advanced dementia, or lacking capacity to consent, does not necessarily mean that a person may not participate in research.
- Findings cannot be generalised (in the context of quantitative research) to people with dementia if participants were from a small sub-section of people with dementia.
- Findings cannot be considered to represent the diversity of people with dementia if participants were from a small sub-section of people with dementia.
- Failure to do so may result in treatment, services, care and support which only corresponds to the needs of a sub-section of society with the needs of other members of society going unmet.
- Failure to do so results in societies failing to benefit from the wealth of experience and of potentially different ideas, perspectives and solutions.
- Failure to do so prevents the creation and maintenance of an inclusive society.

Additional recommendation for research ethics committees

- Recognise that much research is still required into supporting people with advanced dementia and this population should not be excluded from participating in research provided that appropriate processes have been followed.

Summary

Researchers must recruit participants who are best suited to providing a response to their research question or hypothesis. The principle of justice should be respected, which means that everyone should have the same opportunity to take risks, enjoy the benefits and generally contribute towards society through participation in research. Some groups of people with dementia continue to be excluded with the result that the diversity of people with dementia, which should be reflected in research samples, often isn’t. The people who are excluded often have lower levels of education and health literacy, intellectual disabilities, more advanced dementia, co-morbidities or are older or from minority ethnic groups. This exclusion is unethical and unacceptable, leading to some groups of people with dementia being disempowered and their needs and wishes ignored, and representing missed scientific opportunities and incomplete or misleading results.

Attracting interest and addressing barriers to research participation

A key determining factor for the involvement of any person in research is that he or she knows about the study and realises that it would, subject to the fulfilment of certain conditions, be possible to be involved. Even though members of the general public are frequently informed about the latest research findings in newspapers or on the Internet, or hear about breakthroughs on the television, they do not necessarily make or understand the connection between research and those breakthroughs, hear about studies which are recruiting participants or know how to find out about them. Awareness of research and willingness to participate does not, however, necessarily translate into people becoming research participants. Other factors may affect participation such as, for example, a person's current medical condition, cultural and linguistic factors, financial and time constraints, employment constraints, geographical proximity to research sites and mobility/travel issues (Wendler et al. 2005, Konkel 2015). The timing and effects of various medication may also affect the desire to participate (e.g. people feeling groggy in the morning, needing support to take medication). In this section, we look at some of the key issues linked to the initial task of informing and arousing the interest of a broad range of people with dementia in research, and how this in turn may affect the participation of a diverse set of research participants with dementia.

Language and literacy

In Part 1 of this report, the issue of literacy and language was raised as a barrier to the involvement of some people with dementia in PI. Difficulties with language, literacy, as well as the ability to use computers, may also result in some groups of people not being aware of research for which they may be suitable participants. Increasingly, Internet platforms are being set up to inform the general public about research opportunities and to encourage people to participate in research[41]. However, this caters for people who are literate, able to use computers and who have Internet access, which is not always readily available or affordable. As these approaches become increasingly popular, there is a risk of more traditional methods of contact becoming less common and of knowledge about (and hence access to) research being ever more restricted to people with a similar set of characteristics.

Differences in levels of health literacy may also affect people's interest in participating in research. Health literacy is commonly defined as "the capacity to obtain, process, and understand basic health information and services needed to make appropriate health decisions" (Brach et al. 2012, p.1). In this sense, it is associated with healthcare decision making but failure to understand information about health may also affect motivation to contribute towards health-related research (e.g. to improve diagnosis and quality of life, develop treatments, better understand how people are affected by certain conditions and develop non-pharmaceutical therapies and approaches to disease management). In other words, if people do not understand the topic of research and why it is important to address it, it is unlikely that they would be motivated to participate.

Linguistic, educational and cultural issues may also be significant barriers to the participation of people with dementia in research because many of the tools used to diagnose dementia or measure disease progression, as well as instruments to measure research outcomes (all of which are essential components of many studies), are not suitable for people with dementia from linguistically and culturally diverse groups. Some have been translated but many have only been validated in populations from the Western culture. Moreover, language is not the only obstacle as the tools and instruments may in addition be culturally biased[42]. The fact that these have not been developed is an example of inequity and a structural barrier to inclusive research which needs urgent attention. Moreover, the tendency to focus predominantly on written informed consent is also a barrier (e.g. for some older migrants and people with language difficulties) and recorded verbal consent should be considered as an alternative (Berdai-Chaouni, Claeys and De Donder 2018).

Trust and legitimacy

One possible approach to raising awareness about opportunities to participate in research is through the intermediary of health and social care professionals who have access to a broad range of people. However, distrust of such professionals and the fear of racism are common amongst some minority ethnic communities (Shah 2007, APPGD 2013, Kenning et al. 2017, Condon et al. 2019). The fact that scientists have not always been trustworthy in the past and their general lack of public accountability may hamper the willingness of people to participate in current scientific research. Where there have been healthcare research scandals involving scientific misconduct (such as the Tuskegee study in the United States as described in the introduction), trust in researchers may be particularly low, in general and in particular amongst some minority ethnic groups (Daigle 2019). However, it should not be assumed that all members of a particular minority

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[41] Examples include ResearchMatch, the Alzheimer's Prevention Registry, and the Alzheimer's Association's TrialMatch (Source: Lepore et al. 2017)

[42] For more information on this topic, please see the section on "Challenges surrounding the development of culturally sensitive assessment and diagnostic tools section" in Alzheimer Europe's 2018 report on Intercultural care and support: https://www.alzheimer-europe.org/Ethics/Ethical-issues-in-practice/2018-Intercultural-care-and-support

ethnic group share these concerns or that awareness of such historical abuse is the sole reason for distrust in healthcare professionals and researchers (Wendler 2005).

With specific reference to people from gypsy[43], Roma and traveller communities, Condon et al. (2019) point out that frequent mistrust of authorities occurs within the context of a long history of genocide, banishment, discrimination and rejection by mainstream society. There has also been a long history of abuse, discrimination and persecution against people from the LGBTI[44] community, with homosexuality until quite recently having been illegal and considered in the medical profession as a form of sexual deviation (Peisah, Burns, Edmonds and Brodaty 2018). Many older people will have hidden their sexual orientation, gender identity or intersex status for many years and will be hesitant to engage in research which might result in it becoming known.

People with intellectual disabilities have also throughout history been marginalised. According to Rosner (2015), this may have a negative impact on their willingness to participate in research in that they may have low levels of trust in their own capabilities. Such self-selection/restriction may not always be evident. For some people, it may be linked to language use (the way things are communicated). It may be possible for researchers to build up trust by using the same language. It should not be assumed that everyone who seems to be able and free to participate in research shares that opinion. Low levels of education, lack of confidence in one's own abilities, difficulties communicating, internalised negative stereotypes and assumptions that participants would not be allowed or welcome (e.g. for people in group living environments or in situations of dependency) may all contribute to some people with intellectual disabilities not following up on opportunities to participate in research.

The issue of distrust may also be related to the perceived legitimacy of research projects in the sense that a lack of trust in the medical profession and researchers may raise questions about the value and purpose of the study. People from groups which are typically excluded from research may be suspicious about why they are being invited to participate or, conversely, may not believe that the goals of the study are in the interests of people from their communities. Indeed, much of the research which they have the opportunity to participate in does not necessarily reflect the interests or priorities of people from their communities (although there is no reason why such research should be their sole interest).

> *"Many researchers have been using our community people, our culture, and our knowledge to do business. For example, lots of student researchers get their academic degree through using our knowledge……. I see this research as a business making use of our community's people, culture and practice" (comment from a person from an indigenous community, cited in Datta 2017, p. 10–11).*

Key persons or figures from various communities may therefore be crucial in providing access to people from various communities. It is important to develop a trusted and respectful relationship with these key people and not to 'use' them. Sometimes, potential participants from such communities do not even want to have full details about the research as they trust the judgement of the key person. If that person says that the research is good and that the researchers are trustworthy, then people will follow.

In contrast, it is important to be aware of some pitfalls related to trust. Research into the motivations of people to participate in research shows that trust is an important factor influencing decision making (Tromp, Zwaan and van de Vathorst 2016). This trust is very diverse, including, for example, personal trust in researchers or care professionals, institutional trust, trust in research in general and trust in the overarching system (Tromp and van de Vathorst 2018). It is crucial not to take advantage of these types of trust when approaching people for research participation.

Racial/ethnic bias

In most cases, the exclusion of certain groups of people in research is unintentional but unseen racial biases[45] may sometimes affect who gets recruited into studies (Konkel 2015). Researchers may, for example, assume that a person from a particular ethnic group would not want to participate or unconsciously overemphasise negative aspects of the study which may deter the person from deciding to participate. Bias against studies which would have involved more people from ethnic and other minority groups (e.g. on issues of particular relevance to people from those groups) may also occur at the funding stage. According to Konkel (2015), studies in the United States which focus on health disparities and minority populations are less likely to receive federal funding. This may be partly explained by the fact that only approximately 10% of those responsible for reviewing funding applications are from minority groups.

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[43] The term 'gypsy' is commonly used in the United Kingdom (officially and by members of the gypsy community) but not in continental Europe where it is generally considered offensive.

[44] LGBTI stands for lesbian, gay bisexual, transgender and intersex

[45] Whilst Alzheimer Europe challenges the concept of race and promotes the concept of ethnicity, the term race is sometimes used in this discussion paper to refer to bias and discrimination resulting from persisting beliefs within society about 'racial' differences.

As with the involvement of people from minority ethnic groups in PI, it is often assumed that people from those groups do not participate in research as participants because they are not interested. There is nevertheless evidence to suggest that people from minority ethnic groups are just as willing to participate as their counterparts from majority ethnic groups when given the opportunity and provided that the research objectives are presented in a way which is culturally meaningful (Wendler 2005). This means that it is clear what the benefits of the research could be for them or their community and that this is presented in a way that is familiar to them. Wendler concludes that measures need to be taken to facilitate equal access to health research for all minority groups, rather than focusing on trying to change attitudes of people from minority ethnic groups.

The perception and portrayal of dementia and of people from various minority groups

Dementia has, over the centuries, and still is perceived in numerous different ways (e.g. as a divine punishment or test for wrongdoings, an organic brain disorder, a mental disorder, a natural consequence of ageing, something that is contagious and a sign of neglect and poor care of the person concerned) (Alzheimer Europe 2018). The different ways that dementia is perceived and portrayed has been long associated with barriers to help seeking, diagnosis and the uptake of services, and may also affect readiness to contribute towards research. People with dementia are not always treated as equal, respected and valued members of society. Society in general still has a long way to go in overcoming stigma and prejudice associated with dementia. Many people report feeling marginalised as a result of having dementia (Alzheimer's Disease International 2012). In some minority ethnic groups, an additional factor to consider is the 'newness' of dementia as a condition. Members of those groups are less familiar with dementia. This may, for example, be due to recent migration, the population being younger or lower levels of education.

Diagnostic labels can be useful in the clinical context and help people understand difficulties they may have been experiencing for some time. However, Garand, Lingler, Conner and Dew (2009) suggest that some people with dementia may be reluctant to take part in research because they (or in some cases their families) wish to avoid the stigma frequently associated with the diagnostic label, which could take the form of public stigma or self-stigma[46]. Public stigma involves the identification and labelling of people with a socially salient attribute (such as having dementia), attaching negative stereotypes, considering people with that attribute as being 'other' (i.e. not like 'us'), devaluing them and discriminating against them (including denial of equal opportunities as well as social distancing) (Link and Phelan 2001). The attribute is not inherently stigmatising but comes to be considered as such because of the meanings attached to it which are socially salient and generally involve a perception of some kind of threat (e.g. to individuals, society or morality) (Stangor and Crandall 2000). Self-stigma involves the internalisation of negative stereotypes, resulting in low self-esteem and the anticipation of discrimination. Having a lower level of understanding of a particular condition has been associated with greater stigma (Corrigan and Watson 2007). Lower levels of education and difficulties with language and literacy are common amongst (but not restricted to) people from some minority ethnic groups. This may also partly explain higher levels of stigma within certain communities.

Close relatives and friends of people with dementia sometimes experience courtesy stigma (or 'stigma by association') (Blum 1991, MacRae 1999, Werner and Heinik 2008). Courtesy stigma involves perceived loss of social status and devaluation of a person because of his or her association with someone who is stigmatised (Goffman 1963). This experience is not limited to specific ethnic groups but may be particularly relevant for groups which are less individualistic. In some communities, the way that dementia is perceived or understood may have implications for the family as well as for the individual, and this in turn may have a negative or positive effect on participation in research. For example, the belief that dementia is a mental disorder that runs in the family may have implications for the status and honour of the family and hence be relevant to marriage negotiations. Consequently, there may be a certain reluctance to take part in dementia research. However, in some minority ethnic communities and in certain religions, people with dementia maintain their status of 'wise old person' even in an advanced stage of dementia which engenders more openness about dementia. This differs from the dominant Western discourse which tends to idealise youth and results in older people losing their respected status because of ageing. Islam, for example, emphasises the need to respect all older people (Mohammed 2017).

It is important to bear in mind that many people with dementia already experience stigma in relation to other attributes. Some of these people may be reluctant to participate in research which focuses on that particular attribute (e.g. a comorbidity or personal characteristic) or in which they will be categorised on that basis. The means by which potential participants are identified and categorised, even if used with good intentions, can sometimes be misguided and stigmatising. On the other hand, it should not be assumed that reluctance to take part in PI or research is because of stigma or that people, even in communities where dementia is commonly perceived as a stigma, all consider it as such or would avoid participation for that reason.

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[46] Also in relation to research related to mild cognitive impairment.

Another issue related to diagnostic labels is that if people don't identify with a particular label or relate to it, they might not feel that they are being targeted to participate in PI or research. Terminology is constantly evolving and there are cultural variations in the use of various terms. Some people are confused about the meanings of the terms 'Alzheimer's disease' and 'dementia' as they are used inconsistently by some researchers and healthcare professionals who mix old and new conceptualisations (for more on this issue, see also page 60–61). In some counties, the term 'dementia' (as directly translated from English) is considered inappropriate if not offensive and therefore avoided (e.g. in Finland where the term 'memory disorders' is mainly used). Consequently, people with dementia who might otherwise be interested in contributing to research may not recognise themselves as being eligible or not wish to participate in a study where they are referred to in such a way. Moreover, terminology which reflects a strictly biomedical perspective of dementia does not correspond to some people's own perspective and experience (Jongsma, Spaeth and Schicktanz 2017). This may have a negative impact on their participation in research.

Some terms used to describe other conditions or people from minority groups are also considered derogatory or demeaning. A term which was acceptable and the norm at one point in time (e.g. mental retardation) is sometimes replaced or largely falls into disuse but is still heard from time to time in some geographical areas and can be found in various publications (Nash, Hawkins, Kawchuk and Shea 2012). Lack of familiarity with a particular group of people may result in overlooking distinctions made by people in those groups that are important to their self-identity and how they wish to be considered. In the literature, this is also framed as hermeneutic injustice (Jongsma, Spaeth and Schicktanz 2017). For example, Young, Ferguson-Coleman and Keady (2018, p.1002) point out that Deaf people (Deaf with a capital D) "constitute a cultural–linguistic minority for whom to be Deaf is not a marker of deficit but rather a feature of individual and collective identity". This is different from people who lose their hearing later in life. Uncertainty or ignorance about the correct terminology to use in order to be respectful towards potential participants may sometimes result in people feeling devalued and not wishing to contribute towards research. This is therefore one area in which the involvement of people from the targeted groups, perhaps with the involvement of gatekeepers, could be particularly helpful.

Recommendations for researchers

- Be sensitive to the different possible ways that dementia is perceived when trying to recruit people with dementia for research.
- Be ready to question your own assumptions about dementia and people with dementia.
- Help people to avoid the stigma they may feel as a result of being seen in a memory disorders clinic by making it possible for data collection to occur in their own home (may also help with compliance and retention).
- Pay attention to language and respect the linguistic preferences of potential participants.
- Check how people would prefer to receive information (e.g. not in headed envelopes of an Alzheimer's disease centre or on documents with letterheads which refer to mental health, dementia or memory clinics).
- Check when it would be convenient for potential participants to be contacted or visited (e.g. at a particular time of the day or when a certain relative is not likely to be home).
- Build up trust with marginalised communities by speaking or involving a co-researcher who speaks their language or shares other characteristics of the group.

Recommendations for research ethics committees

- Try to ensure diversity on research ethics committee panels.
- Assess whether methods and materials used to advertise research projects are such as to be accessible and respectful towards people from different socio-cultural backgrounds.
- Ensure country-specific guidance and legislation has been followed to ensure capacity, but also recognise the valuable contribution to research by participants with advanced dementia.
- Assess whether the spaces where the study is carried out and the research procedures are such as to avoid any stigmatisation of participants.
- Seek clarification of additional support that may be required for data collection (e.g. the key worker of a person with intellectual disability).

Recommendations for funders

- Try to ensure diversity on funding committees.
- Be sensitive to how symptoms or conditions are framed in calls for proposals.

Summary

For some people with dementia, a range of factors stand in the way of them hearing about research, weighing up the pros and cons of participating and potentially volunteering to participate. Examples include difficulties with language and literacy, lack of trust of the people proposing the research or of researchers, not feeling that the research is relevant to people from their communities, not wanting to be associated with studies about dementia because of the way that it is perceived and portrayed and the negative labelling of sub-groups of the population of people with dementia. These barriers are often a result of the way that researchers raise awareness about their studies, the means that they use to attract potential participants, personal racial/ethnic assumptions or bias and lack of sensitivity to perceptions of dementia as a stigma. As was mentioned earlier in relation to PI, informational materials may sometimes be culturally inappropriate and disseminated in places that do not correspond to the cultural diversity of people with dementia. This calls for greater sensitivity and awareness from researchers and emphasises the importance of ensuring diversity on research ethics committees and funding committees.

The financial costs of involvement

Costs linked to being a research participant may put some people off participating, especially those from a lower socio-economic background. Such costs could, for example, include public transport, taxis or petrol and parking fees for trips to study centres, food, accommodation, minding fees for dependent relatives, loss of earnings, loss of holidays, and refreshments if the study visit takes place in the person's home. Researchers seeking to involve people with difficulties with language, literacy and learning would need to budget for extra costs and allow for extra time for outreach efforts, interpretation, translation, bilingual experts, researchers with expertise in intellectual disability, adapted information materials, transport to people's homes for home visits, additional transcription and translation costs for data analysis and costs linked to the development and validation of culturally sensitive data collection tools.

Some of these costs would be incurred at the recruitment stage and others would arise later but all such costs and the process for reimbursement should be made clear from the outset in order to avoid the risk of influencing decisions about the recruitment of a more diverse population in research. Researchers and funders need to take such financial costs into consideration when drawing up and approving budgets for research that is truly inclusive. Cost should not be used as an excuse for failure to involve a diverse population or exclude certain groups of people. Writing in the context of the involvement of people from minority ethnic groups in clinical trials, Hussain-Gambles concludes,

> *"Economic considerations should not, however, be used as an excuse for the exclusion of minority people from clinical trials. Good science is expensive, and since the distribution of resources depends on priorities, it is a question of how high up the priority list is the problem of unequal representation in clinical trials" (Hussain-Gambles 2003, p.141).*

Gatekeeping

Gatekeepers are people who formally or informally control researchers' access to certain groups of participants. They may, for example, be respected members of minority ethnic groups, managers of care homes, legal guardians or informal carers. Gatekeeping can be empowering as well as restrictive with regard to the opportunities that people from various sub-groups of society have to participate in research and the possibilities that researchers have to reach them. It is therefore an important phenomenon to consider in relation to the recruitment of a diverse group of people with dementia in research. Taking the example of research involving the widely neglected group consisting of older homeless people with dementia, Manthorpe et al. (2019) found that staff in hostels for older homeless people were quite good at identifying hostel residents with memory difficulties, some of which may be linked to dementia. They were frequently in contact with them and were able to recognise some of the difficulties they were experiencing in carrying out everyday activities.

Gatekeeping may serve the following purposes, which could be considered in a positive or negative light, depending on one's perspective:

- To enable people from different sub-groups to hear about research opportunities, and understand the importance of a particular study for society and for themselves,
- To enable researchers to access groups of people whom they would otherwise have difficulty reaching (e.g. because of language difficulties, trust issues, cultural and religious traditions governing interaction etc.),
- To protect people perceived as vulnerable from exploitation or from research activities which may be detrimental to their wellbeing or that of their entourage,
- To facilitate or promote research that is likely to be beneficial to people from the communities or sub-groups of society to which the gatekeepers have access,
- To restrict access to potential participants in order to avoid extra workload or responsibilities (e.g. for health and social care professionals).

Gatekeepers have some degree of power to benefit people from the communities they represent by promoting particular research and facilitating the participating of people from those communities in such research. There are, nevertheless, a few challenges that need to be considered when seeking the support of gatekeepers.

When resulting in the restriction of researchers' access to broader groups of people with dementia who might otherwise have been interested in participating in research, gatekeeping could sometimes be considered as paternalistic and even an abuse of power. It may, for example, deny people with dementia the right to decide for themselves whether or not to take part in research (Lepore et al. 2017), thus failing to respect their autonomy. Nordentoft and Kappel (2011), for example, describe organisational restrictions which make it difficult to carry out research involving vulnerable groups in and outside of hospitals in Denmark. For gatekeepers who do not necessarily know the potential participants personally, there is a risk of them making assumptions based on stereotypes and inadequate knowledge (e.g. about the needs, interests or perceived vulnerability of individuals from various sub-groups such as people in residential care, with advanced dementia, in the oldest old group or with intellectual disabilities). In some contexts (e.g. in many German nursing homes), it is mainly legal guardians who decide whether or not residents will take part in research and they may be hesitant to allow this because of concerns about the research being burdensome to residents (Palm et al. 2016). It may also be the case that they do not know the resident sufficiently well to make the decision about participation.

Doody (2018) describes how gatekeepers may, when trying to promote the inclusion of people with intellectual disabilities in research, actually block their inclusion with their 'protective power', which can be positive and nurturing when used appropriately (Witham, Beddow and Hagh 2015), but may also serve to silence and oppress potential participants. This represents an ethical dilemma in which gatekeepers may be caught between the desire to protect people from harm and the desire to respect and promote their autonomy. Doody (2018) suggests that it may be helpful for gatekeepers to have access to feedback given to researchers by the research ethics committees who review study proposals.

It should also be borne in mind that there are different levels of gatekeeping. Researchers may sometimes need to get past several layers of gatekeepers to recruit older people with dementia for research (Hellström et al. 2007). There may, for example, be directors of care homes who act as initial gatekeepers and different members of staff who must respect the directors' decisions but may have the power to block or hinder access to potential participants. There may also be legal guardians and informal carers. In the residential care setting, care staff are likely to be most affected by the involvement of people with dementia in research (Doody 2018) and may sometimes have personal reasons for refusing access.

Witham, Beddow and Haigh (2015) describe the need to understand attitudes and perceptions surrounding dementia and ageing, which are common in some settings in which researchers seek to access various marginal groups to participate in research. This includes social constructions of ageing as dependency and vulnerability, the 'positioning' (Harré and van Langenhove 1999) of people with dementia as incompetent and examples of what Kitwood described as 'malignant social psychology', including infantilisation and objectification of people with dementia (Kitwood 1990, 1997) and of a general hesitancy and uncertainty in dealing with people seen as 'other' or having 'special needs'. The infantilisation of people with dementia is a cultural cliché that is persistant and problematic. It silences people with dementia, thereby diminishing their autonomy and can be harmful in terms of not providing them with appropriate care and opportunities (e.g. to participate in research). It should therefore be avoided/challenged (Jongsma & Schweda 2018). Witham et al. (2015, p. 35) ask how some groups of people come to be positioned in such a way that they are not even asked in the first place and thus denied choice and autonomy. They suggest the "need to remain reflexive in situating the patient as 'vulnerable' and positioning them in such a way that choice and decision making become compromised".

Researchers who are not members of the communities or groups from which they are seeking to recruit participants may not always understand the extent to which a particular key person represents the interests of the whole community or just a part of that community. Fisher warns against "the unwarranted assumption that opinions of minority scholars and community leaders reflect or override those of the less educated and more vulnerable community members who may be the target of investigation" (2009).

Some communities are very heterogeneous, hierarchical and/or with significant cultural, social or religious sub-divisions. According to Bhattacharyya and Benbow (2013), there are hierarchies and caste systems in some minority ethnic groups. The South Asian community in the Bradford area of the UK, for example, has been described as culturally and socially fragmented with divisions based on caste, status, gender and generational hierarchies. People with intellectual disabilities also form a very heterogeneous group made up of several sub-groups (Doody 2018). People may sometimes object to being placed in a broader or seemingly inappropriate group by researchers. Condon et al. (2019) report a study (not related to dementia) in which Show People[47] were offended at the suggestion that their views would be similar to those of Gypsies, Roma and Travellers and not to those of the general population. This suggests the need for sensitivity to inter group differences that are prominent in many groups, including that of people with dementia.

Once identified and involved in a particular study, gatekeepers may find that researchers are keen to approach them about other research. This could be interpreted positively as evidence of researchers' interest in involving a more diverse set of people in research. Some gatekeepers may, however, find themselves inundated with requests, have difficulty managing the increased workload and have to reject some of them (Doody 2018). Having successfully negotiated access to a minority group, there may be a tendency for researchers to stick to that group because it has become 'easy to reach'. This does not represent fairness or progress in reaching out to a wider group of people as other perspectives may still be excluded.

Finally, gatekeepers often play a vital role in recruiting people with dementia from minority groups and invest a lot of time and effort in this task. Researchers may wish to offer them a token of their gratitude. Alternatively, based on the concept of reciprocity, some gatekeepers may expect or request something from researchers (e.g. payment, a service or a contribution to a programme that would be beneficial to people from that group). There are differences of opinion amongst researchers as to whether recognition of the reciprocal relationship with gatekeepers should be based on a personal gesture or token of gratitude or on a more formal arrangement. Irrespective of the approach adopted, it is important to ensure that anything offered in return for the support of gatekeepers does not have an adverse effect on the choice of research projects to which they lend their support.

In addition to external gatekeepers, researchers need to bear in mind that they themselves might also be gatekeeping (Sharkey, Savulescu, Aranda and Schofield 2010). They themselves might have implicit inclusion and exclusion criteria that lead to them not approaching all eligible research participants (Tromp and van de Vathorst (2015). For example, they might have a prejudiced belief that someone would not want to participate, think that participation would be too burdensome or think that someone would be non-compliant in the research. Although this practice of individual gatekeeping by researchers is understandable, it is not desirable (Tromp and van de Vathorst 2015), because:

- It denies personal choice,
- It may deny a person a possible benefit resulting from participation in the research,
- It may reduce inclusion rates,
- It can cause selective inclusion (i.e. bias) and therefore endangers generalisability (in the case of quantitative research),
- It can result in an unfair distribution of burden, risk and benefit among people with dementia.

Different agendas/priorities

Research into issues relating to various minority groups often reflects topics chosen by or identified as being problematic by people who are not members of those groups and often for economic or political reasons (Tyack 1995). The actual topics selected may also be influenced by researchers' lack of awareness of the priorities of minority groups and partly due to hidden agendas of researchers and research institutes (e.g. linked to career advancement, the desire for recognition from peers and society, competition for funds and scientific curiosity). Increasing the involvement of a diverse set of people in research not only requires equal opportunities to participate in research but also equal opportunities to influence the research agenda. This point was made earlier with the regard to the importance of PI at the earliest possible stage, namely when exploring different possible research topics, prior to seeking specific funding. Involving people from minority groups in research should also extend to the choice of research topic.

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[47] Show People are a cultural minority that have owned and operated family-run funfairs and circuses for generations. They often work on rides and attractions at fun fairs in the summer months and settle in the winter to repair the machinery (Horsfield 2017).

There are different forms of dementia, the most common one being Alzheimer's dementia, followed by vascular dementia and then dementia with Lewy bodies, and mixed dementia is also common (Livingston et al. 2017). However, research into dementia is not closely aligned with the prevalence of these different forms. This means that some forms of dementia, such as Lewy body dementia, familial dementia and vascular dementia, are under-researched (Jongsma et al. 2016). Consequently, people with dementia do not all have the opportunity to participate in research related to their specific condition.

> ▶ *"As I have frontotemporal dementia I am not eligible to participate in much of the research that is going on. I realise there is not as much being done on FTD but I am often even excluded from studies on the topic of living with dementia. I have dementia so why is a lot of this research just about people with Alzheimer's?"* **(Petri, person with dementia, Finland).**

Relevant questions and hypotheses must be addressed by researchers. It is therefore important to consult members of different minority groups, such as minority ethnic communities, people with intellectual disabilities or members of the LGBTI communities, so as to identify and respond to their interests and concerns (Konkel 2015). Promoting and respecting the values and interests of different communities and protecting communities from harm is what Weijer and Emanuel (2000) described as a matter of 'respect for communities'.

Recommendations for researchers

- Create a realistic study population with adequate and appropriate inclusion and exclusion criteria.
- Budget for the cost of involving a diverse set of people with dementia as participants in research when applying for research funds.
- Ensure that the time required for informed consent is reflected in project timelines.
- Discuss with people from the relevant communities about possible participation, the kind of support they might need and the financial cost of this (e.g. travel, interpretation, dietary requirements, additional cost for one or more support people etc.).
- Invest in a diverse research team. The diversity within a research team enhances sensitivity to possible invisible issues related to the gatekeeper/research team relationship and can help avoid possible conflicts and mis-use of this relationship either by the gate keeper or by the team, and thus enhance access to communities.

Recommendations for research ethics committees

- Be aware of too restrictive inclusion and exclusion criteria, which cause an irrelevant and impossible research population and diminished generalisability.
- Ask researchers to describe the measures that they envisage to involve a diverse set of people with dementia in research, including ways to approach possible gatekeepers.
- Be willing to consider necessary adaptations to existing procedures and open to novel approaches which may be necessary to achieve inclusive research.
- Assess (in relation to particular payment for particular research) whether reimbursement for participation in research would encourage the participation of people with dementia with limited financial resources and how ethically justifiable this would be.
- Ensure that anything offered to gatekeepers in return for their support is not such as to influence their choice to support research projects.

Recommendations for funders

- Involve relevant individuals and communities in decisions related to funding.

Summary

It costs money to attract and involve a diverse set of people with dementia in research. Research funders need to bear this in mind and researchers need to ensure that they consider all the potential costs involved and draw up a comprehensive budget. People with dementia may be less enthusiastic about participating in research if they don't feel that it is relevant to them (e.g. to people with their type of dementia, to people from their communities or to people who have things in common with them). Gatekeepers often play an important role in enabling researchers to communicate the importance of their study to specific groups of people with dementia which the researchers might otherwise find it difficult to reach or engage with. They may also provide valuable insight into what is important to certain groups and even negotiate something for members of a group in return for the personal investment of some of its members. However, gatekeepers may at times hinder access to certain groups of people as a result of being over-protective and may not always reflect the interests of everyone in what is believed to be the group they represent. Researchers themselves may also gate keep in a way by ruling some people out of participation on the basis of assumptions about what they would or would not want or be likely to do.

Sampling issues

In the previous section, we explored some of the key issues related to the involvement of a wide range of participants in research, with a focus on practical issues, cultural and societal challenges and ethical considerations. In this section, we reflect on issues which are closely related to the methodological requirements of research sampling, although these also have practical, cultural and ethical implications.

Sampling bias and failure to capture diverse perspectives and experience

As it is rarely possible, in terms of practicalities, cost and time, to include all possible cases in a study, researchers typically select a sample of people to participate in their research. There are several different approaches to sampling in research, both for quantitative and qualitative approaches. Both approaches, including the many variations of each, emphasise the need to involve a broad cross-section of people. This is essential for truly inclusive research but despite concerted efforts to involve people with dementia from particular sub-groups of the population, for example, it often does not happen.

One of the difficulties when seeking to identify people from specific minority groups within an overall population for research (e.g. to try to ensure that the sample is more inclusive) is that details of the characteristics needed are not always readily available. Indeed, in many cases, it would be considered unethical or even illegal to record various details about people. For some characteristics, such as ethnicity or having impairments, it might be possible to ask if participants would be willing to provide such information (i.e. on a voluntary basis). This then poses the problem of providing pre-determined options, including 'other' or 'non-stated' or classifying open responses afterwards. Either way, this imposes a classification by researchers which does not necessarily reflect real-world characteristics as experienced by or meaningful to the people concerned. For example, people who appear (to others) to belong to the same ethnic group may describe their ethnicity in a number of different ways. In the United Kingdom, for example, some people might describe themselves as BAME or BME or alternatively as Black, Afro-Caribbean, South Asian, Pakistani or Sikh, with many adding 'British' (e.g. Black British).

In quantitative research, good quality research (which is a pre-requisite for ethical research) relies on appropriate sampling strategies to avoid selection bias. According to LaMorfe (2016),

> *"Selection bias can result when the selection of subjects into a study or their likelihood of being retained in the study leads to a result that is different from what you would have gotten if you had enrolled the entire target population."*

Selection bias would result in findings obtained from an over-represented selection of people from a particular group being generalised not only to other similar people from that group but also to the underrepresented group of people (Simundic 2013). A classic example of this would be the generalisation of the findings of a study only involving White ethnic groups to people from Asian or Black ethnic groups, possibly leading to the development of culturally inappropriate services. Biased sampling results in inaccurate, misleading findings, which are not truly generalisable and the abuse of participants' time and effort, as well as a waste of funding.

Many quantitative researchers would argue that samples should ideally be random to help ensure that they are statistically representative, thus permitting generalisation from the sample to the larger population. Contrary to popular usage of the term, 'random' does not imply that the selection of participants was haphazard but that strict measures were applied to try to ensure that the sample was not systematically biased (see below). Non-probability (i.e. non-randomised) sampling techniques are often considered inferior to probability (i.e. randomised) sampling techniques even though random sampling cannot guarantee the representativeness of a particular sample. This issue is discussed further in the sub-section on sampling hierarchies.

Qualitative research typically involves smaller numbers of participants, often purposefully selected to obtain rich, in-depth and meaningful information which illuminates the research question whilst capturing the diversity of experience. The terminology may differ in that the findings of qualitative research are not derived from statistical analysis and not intended to be generalisable but the goal of ensuring that the sample is sufficient and suited to answer the hypothesis or research question is similar.

Self-selection may occur in some quantitative and qualitative studies depending on the approaches adopted. For example, an Internet-based survey to measure or explore mobility issues of people with dementia would be likely to attract people who are able to use computers, perhaps with support, and have access to one, and exclude those who aren't and have not. Self-selection may also be influenced by the topic and by cultural differences with regard to readiness to discuss the issue with researchers or people who are not from the community. The opposite may also be the case in that people may sometimes be reluctant to discuss or provide information about a particular issue to someone from the same community (e.g. for fear of gossip or rumours, or to protect the honour and dignity of the family or individual). For quantitative studies, this would risk participants who choose to participate in a study not being equivalent (in terms of the research criteria) to the people who opted out. In qualitative studies, it would risk the people participating having very similar perspectives or experiences and not capturing the full diversity of perspectives and experience.

Sample size

Obtaining a sufficient number of people with dementia to participate in research can often be difficult. For research involving quantitative analysis, research ethics committees and funders may be justified in asking for clarification of the number involved (e.g. based on the population size, margin of error, confidence level and standard of deviation) in order to check that this would offer sufficient statistical power, and avoid wrongly failing to reject the null hypothesis. In quantitative research, if a sample size were too small, it could be assumed that the efforts of the research participants, and in some cases their discomfort or burden, were in vain because the goals of the study, namely to obtain a valid response to the research question which can be generalised to the broader population, were not achieved. It could also be argued that financial and time resources invested in the study

> *"were squandered since ultimately it will contribute absolutely nothing to improve clinical practice or quality of life. The situation becomes even worse if the research involves public funding: A total waste of taxpayer money" (Faber and Fonseca 2014, p.28).*

Faber and Fonseca (2014) point out that sample sizes which are larger than necessary are also unethical because this would indicate that some people were needlessly exposed to risk or burden, or that their time, efforts and public funds were wasted. Larger-than needed sample sizes are rarely the problem though. When striving to include people from minority groups in research, attracting a sufficient number of participants can be a real problem and in quantitative research, simple random sampling is often not the best approach. Moreover, actively seeking to include a sufficient number of people from various sub-groups in representational proportions necessitates the involvement of a much larger number of people, with the financial and practical implications that this may incur (Hussain-Gambles 2003). The need to be able to identify people from a more diverse background can also be problematic for qualitative research.

In qualitative research, relatively small numbers of participants are quite common. The principle of qualitative research is that researchers seek to obtain data, often regarding quite complex social phenomena, with a focus on meaningfulness, the diversity of experience and rich descriptions. Researchers are nevertheless often expected by research ethics committees and funders to give some indication of the possible number of participants to be involved in a study even though in many cases the number is determined by the saturation of the data obtained. Once the point of saturation has been reached, further collection of data would be unethical. As with excessive numbers involved in quantitative research, continuing to recruit participants and to collect data beyond the point of saturation would amount to a misuse of human and financial resources, with people sometimes undergoing burdensome tests or questioning for no benefit at all. Asking researchers to justify the number of participants in advance may be based on an inadequate understanding of certain qualitative research approaches, may put pressure on researchers to involve more people than necessary and may be interpreted as questioning the validity of qualitative research by trying to apply standards and criteria for quantitative research.

As a means to involve people with more advanced dementia in qualitative research, it has been suggested that larger samples than usual may be required in order to obtain sufficiently rich data. Reporting on the experience of a few studies, Beuscher and Grando (2009) state that it may also be helpful to conduct multiple interviews, to have multiple readings of the transcripts and to triangulate the findings of the interviews with observational and field notes.

Recommendations for researchers

- Describe in the research protocol the rationale for the sampling strategy and how this is conducive to the promotion of inclusive research involving people with dementia.

Recommendations for research ethics committees

- Consider the need for inclusive research when evaluating the appropriateness of the sampling strategy in the context of judging the scientific validity of research projects.
- Ensure that the assessment of sampling strategies is appropriate to the research design and proposed methodology.

Recommendations for funders

- Recognise that small sample sizes do not necessarily mean a less effective study provided that there is methodological rigour and reliability in approach.

Summary

Because of the time, effort and money it would cost to include all possible cases in a study, researchers typically select a sample of people to participate in their research. The risk of selection bias is particularly important in quantitative research and may result in the results of an over-represented selection of people wrongly being generalised. Random sampling is probably not the best approach to gain a representative sample of people with dementia living in multicultural and diverse societies. In qualitative research, smaller numbers of participants are often purposefully selected to obtain rich, in-depth and meaningful information which captures the diversity of experience. Self-selection may occur in some quantitative and qualitative studies and may contribute towards an unrepresentative and characteristically restricted sample. Obtaining the right number of participants can often be difficult. Too small a sample in quantitative research may result in inconclusive results that lack sufficient power to be generalisable, and too many people in qualitative research (sometimes based on a lack of understanding of the principles of qualitative sampling) may lead to an abuse of people's time and effort. In both cases, inappropriate sampling leads to a waste of financial and human resources which could have been put to better use.

Inclusion and exclusion criteria

Many of the factors which represent challenges for inclusive research with people with dementia are quite subtle and not intended to prevent people from participating in research. Inclusion and exclusion criteria for research are part of standard research practice for the conduct of good quality scientific research. They are also not designed to be discriminatory. The aim of inclusion criteria is to determine the key features of the target population that will enable researchers to answer their research question or hypothesis. The aim of exclusion criteria is to define features of the target population (who meet the inclusion criteria) which might interfere with the success of the study (e.g. increase the likelihood of them being lost in follow up, missing study visits, providing inaccurate data or resulting in an unfavourable outcome) or put the participants at risk (Patino and Ferreira 2018). A key question for researchers and research ethics committees to answer is whether the rationale for inclusion and exclusion for particular studies is valid (i.e. is there a valid justification for them?).

Care nevertheless needs to be taken when developing inclusion and exclusion criteria to ensure that people are not unnecessarily deprived of the opportunity to take part and that they do not jeopardise the external validity of the study. For example, classing every possible characteristic which might lead to an unfavourable outcome (e.g. in a clinical trial) as an exclusion criterion would mean excluding a huge number of people who might like to be involved and the findings having little relevance or generalisability to the real-world population. Inclusion and exclusion criteria therefore have a key role to play in the ethical conduct of research but must be carefully scrutinised to ensure that they are applied in an ethical manner. In this section, we look at several characteristics commonly used as inclusion or exclusion criteria.

Age

Age is usually classed as an inclusion criterion in that it describes the age range of the people the researchers would like to include in their study. There are some good examples of research involving older people in research such as the Alzheimer's Disease Neuroimaging Initiative (ADNI) (Weiner 2010) and the "nun study" into healthy ageing and dementia involving 678 Catholic sisters aged between 75 and 107 (Snowdon 2003). However, upper age limits exclude many people with dementia from research who could equally contribute towards answering a particular research question or hypothesis. Younger people with dementia may also find themselves wrongly excluded from various types of dementia research, including clinical trials (e.g. because they do not correspond to stereotypes of people with dementia). If the reasons for their exclusion cannot be justified, it should be considered as discriminatory.

> ▶ *"There is usually a minimum and maximum age limit for participating in dementia research. I don't really understand why this is necessary. I have dementia, the study is about people with dementia and yet I am excluded. I find this frustrating and unfair."*
> **(Miha, person with dementia, Czech Republic)**

It is a fundamental principle and a legal and ethical requirement of drug development and treatment in Europe that drugs be tested on people with the same characteristics as those who will eventually use them (if proven effective and safe). In 1994, the European Medicines Agency (1994) stated that patients entering clinical trials should be reasonably representative of the population that will be later treated by the drug. Yet earlier, in 1989, the American Food and Drugs Administration commented that there is no good basis for the exclusion of patients on the basis of advanced age alone.

According to Beswick et al. (2008), the literature on the age of older people in clinical trials for Alzheimer's disease is limited but for clinical trials to be truly representative, they should include a large percentage of people between the ages of 75 and 90. In a review of 109 clinical trials of therapeutic interventions in adults assessing morbidity or mortality, it was revealed that 20% of studies automatically excluded patients above a certain age (Zulman et al. 2011). Clinical trials therefore often involve participants who are not representative of those for whom the medication will most likely be used if the trial is successful (Heait et al. 2002, Jongsma, van Bruchem-Visser, van de Vathorst and Mattace-Raso 2016). Many clinical drug trials still have inclusion criteria which prevent older people with dementia from participating even though the largest proportion of people with dementia are aged 80 and over (Brayne and Davis 2012). This means that many older people with dementia are taking medication, which has not been proven safe or effective for them because similar-aged people were excluded from the trials, and that their views and experience are frequently not sufficiently taken into consideration in dementia research. There may be differences between younger and older people with regard to:

- adverse reactions,
- pharmacokinetics and pharmacodynamics (biological effects in this case linked to age e.g. affecting metabolism and sensitivity to medication),
- comorbidities (having additional acute illnesses and chronic conditions – such as dementia),
- polypharmacy (increased use of medication/older people taking several different drugs each day) (Orwig et al. 2011).

These age-related differences, sometimes expressed as exclusion criteria, may contribute towards the exclusion of older people from clinical trials. Another possible reason for excluding older people from clinical trials is that their inclusion might dilute the active treatment effect, thereby resulting in results which are not statistically significant (Cherubini et al. 2010). This has obvious implications for the development of marketable drugs but also with regard to fairness to the hundreds or thousands of participants who dedicate their time and energy to a study which may be inconclusive. On the other hand, the characteristics of the majority of older people who will eventually take the drugs are different to those of the people who took part in the trial.

Sox and Greenfield (2009) criticise randomised clinical efficacy trials for attempting to create near ideal circumstances in order to determine whether the intervention could possibly work which do not reflect the conditions in real clinical practice. The EFGCP GMWP[48] (Diener et al. 2013) advised in their guidance synthesis on medical research for and with older people in Europe that participants in clinical trials, should be reasonably representative of the population that will be later treated by the drug (and hence to include more participants in the older age range) and to avoid the direct extrapolation to older patients of efficacy/safety data and of the drug benefit/risk profile observed in younger adults, as this may lead to safety issues and iatrogenic disorders.

Possible barriers to the involvement of older people with dementia in research in general include, for example, physical and cognitive impairments, difficulties with mobility, lack of transport, a lower threshold for burden, distrust, higher costs linked to the recruitment and retention of participants, polypharmacy and various co-morbidities. However, such factors do not apply to all older people and are not limited to older people. In addition, age is often associated with a more advanced stage of dementia. Hubbard et al. (2003) caution against categorising people into different stages, suggesting rather the need to focus on specific impairments and to use methods suited to the individual, irrespective of the duration or severity of dementia.

It is important to be aware of possible factors which may affect the ability or willingness of older people to participate in research and of various challenges that researchers face when seeking to involve older people in research. However, these factors and challenges should not be unquestioningly seen as legitimate grounds for exclusion. With the necessary time, effort and financial investment, many of the challenges to including older people with dementia in research can often be overcome by proportionate measures. Adherence to recruitment methods and study protocols, which do not take into account the specific needs of older people, with or without dementia, as well as failure to build in additional costs into the design of studies for the recruitment of older people, could be considered as examples of ageism and structural discrimination. This is also not in keeping with the principles of dignity, respect and social inclusion. With regard to the issue of age discrimination in clinical trials, Cherubini et al. (2010) concludes:

> *“Older people who are more characteristic of those seen in clinical practice must be included in clinical trials to achieve the goal of safe and effective drug therapy for this growing patient population. (…/…) The design of clinical trials that include older adults more typical of those seen in clinical practice should be reconsidered for inclusion of complex older adults with multiple comorbidities and polypharmacy, addressing concerns about the feasibility of multiple measurements; the risk:benefit ratio of the proposed interventions; the determination of adequate sample sizes, taking into account higher attrition and heterogeneity; the complexity of multicenter trials; and finally, the higher costs of such clinical trials.”*

Place of residence

Trial delivery sites for clinical trials tend to be located in and around large towns and cities. This is practical when trying to attract a sufficient number of participants but results in the exclusion of many people who live in rural areas or small islands, especially people with dementia who may have difficulty planning trips, finding their way around unfamiliar places and who need to be accompanied by another person (who in turn may have other obligations and commitments and therefore not have enough free time to assist).

> ▶ ***“I am Idalina and have dementia. I live in Madeira and although this is Portuguese, there is not much research being done here. Living on an island means that people are excluded from a lot of research because studies are all on the mainland. I would be more than willing to travel to mainland Portugal to participate in a study. But I don’t really get to hear about possible research opportunities over there and I’m not sure I would be accepted anyway, and that is unethical in the sense of it being discriminatory.”***
> **(Idalina, person with dementia, Madeira)**

People with dementia living in nursing homes are sometimes explicitly excluded from studies. In many studies, recruitment simply targets people living independently or no mention is made about living situation as an eligibility criterion (Jongsma et al. 2016). In such cases, people in nursing homes may be excluded because of structural discrimination resulting in unequal access to dementia research and inadequate or unproven care for those living in nursing homes.

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[48] European Forum for Good Clinical Practice Geriatric Medicine Working Pary

People serving prison sentences and homeless people are typically excluded from research (unless a study is specifically about people in those groups). Manthorpe et al. (2019) suggest that older homeless people with memory problems, including those associated with dementia, are often reluctant to engage with services and are difficult to assess and help because of persistent heavy drinking, behaviour that other people find challenging (perhaps in some cases linked to substance abuse), self-neglect and physical health problems. Little is known about the problems and needs of older homeless people with memory problems and it is likely that some of the challenges affecting their help seeking would also be challenging for researchers considering their involvement in studies. There may be issues which many researchers find too challenging and it may be that these are often simply forgotten groups.

In some group living situations, dementia is quite common but seldom mentioned (e.g. in some assisted living or nursing homes for older people). Dementia is harder to diagnose in people with an intellectual disability due to lack of appropriate diagnostic tools. Staff and families may therefore be reluctant for residents to take part in dementia research for fear of raising concerns about dementia amongst other residents and act as gatekeepers, thus denying them opportunities that other people have. People living in various types of residential homes may also be excluded from research because they don't get to hear about studies, they may have mobility or transport problems or participation might be considered by others as interfering too much with the routines of the home. If formal diagnosis, including imaging, is required for recruitment, older people living in nursing homes may sometimes be excluded because they are considered too frail to transport or because nursing homes are not affiliated with a clinic (Palm et al. 2016).

Having a study partner

In most clinical trials and sometimes also in non-pharmaceutical research involving people with dementia, it is a necessary requirement to have a study partner[49]. Study partners are typically chosen by research participants. Usually, they attend study visits with the person with dementia and may be asked to complete questionnaires and provide information. They may also be asked to give independent reports about the participant's mental status. They are often a spouse/partner, friend or sibling as they need to know the person quite well and be aware of his or her behaviour or condition. Study partners are generally considered as research participants and thus subject to research ethics provisions. This differs from people who simply have a supportive role (e.g. a key worker for a person with an intellectual disability or a spouse who may remind the participant about meetings or accompany him/her travelling) who are not participants in the study themselves. Study partners should also not be considered as proxy-consenters for people who are unable to consent to participation. These two roles are sometimes combined in one person but are fundamentally different. A study partner is a research participant who acts as an informant, whereas a proxy-consenter has a formal role in which he or she consents to research participation on behalf of a person who is unable to consent.

The reasons for having study partners include concerns about the condition of the person with dementia deteriorating over the course of the study, about the person with dementia needing assistance to attend study visits and comply with the research requirements and, as stated above, to provide additional information (an informed observer's perspective) about his or her behaviour, progression or mental status during the course of the research.

Requiring a study partner, irrespective of a person's actual needs, involves making assumptions about people with dementia (e.g. that all participants need assistance, that their perspective or the information they provide is not reliable and that their condition will deteriorate in the course of the study) and could therefore serve to reinforce negative stereotypes about people with dementia and contribute towards stigma. On the other hand, some studies even require a study partner for healthy controls but this is less common.

Some people with dementia may have difficulty finding a trusted person to act as their study partner, particularly for research stretching over several years (e.g. they may live alone[50], have no relatives who live close by or do not know someone with the necessary skills, time or interest).

> ▸ ***"I would normally agree to take part in lots of different studies but some state you have to have a study partner and even if they don't, I know I would need support. My wife would gladly help me but she has other commitments. It's a shame but that's the way it is."*** **(Geert, person with dementia, Belgium)**

People who live alone and people whose potential study partners are either opposed to their participation in research or have commitments which make it difficult for them to take on this role may, for example, be excluded.

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[49] This is also the case in some clinical trials involving people with preclinical or prodromal Alzheimer's disease but this is beyond the scope of this report.

[50] The number of older people, including people with dementia, who live alone, is constantly rising (Eurostat 2017)

> *"Being a study partner necessitates the commitment of time, effort, and insight into the research participant's cognition and function. In short, being a study partner is work. If an older adult doesn't have someone who's able or willing to do this work with her, then the study partner requirement is a barrier to her research participation" (Largent, Karlawish and Grill 2018, p-1–2)*

Largent et al. (2018) suggest the need to look for ways, within the limits of methodological and scientific rigour, to make the participation of research partners more palatable (e.g. having study visits outside office hours, offering home visits or transport to the study centre, covering parking costs and allowing remote participation by Skype to complete some study instruments).

There may also be reasons not to involve a partner for fear of drawing attention to a relationship which either the person with dementia or the potential study partner would prefer to remain private. Many people with dementia are to some extent dependent on their relatives and close friends for support or care. According to Davis (2017), the relationship between research participants with dementia and their study partners may sometimes be harmed as a result of their joint participation in research. For example, participants may feel dependent on their study partners for their continued participation in the study, have the unpleasant feeling of being observed or reported on or start to question whether certain interactions are authentic or scrutiny prior to reporting to researchers.

The requirement to have a study partner may reduce the number of eligible research participants and skew the research population. It therefore needs to be considered whether a study partner is really necessary, whether the function they serve could be managed in another way and if not, to look for ways to remove obstacles to their participation.

Literacy levels and language difficulties

Difficulties with language and literacy, which have already been discussed with regard to PI, may also prevent some people (e.g. from ethnic minority groups or with intellectual disabilities) from contributing towards research as research participants. Participant information sheets, informed consent forms and various research tools such as questionnaires, rating scales and computerised texts, are all key documents which must be accessible and suitable for all research participants. Some documents, such as informed consent forms, are extremely long and complicated. If the level of language is too high or complex and if various documents and research instruments are not culturally appropriate (i.e. have perhaps been translated but not validated on people with similar characteristics as the intended research participants), this will exclude some groups of people, provide ambiguous data (because responses might not correspond to the intended question without the researcher necessarily realising this) and/or involve people as research participants without them having given fully informed consent. Some people (with and without dementia) may even sign without having read the document at all (e.g. due to its complexity and length). Plous and Holm (2015) also highlight the problem of 'routinisation' (albeit in relation to online consent) whereby few people read the information provided and often give consent as an act of routine (a kind of formality). They suggest that 'routinisation' threatens the protection of personal autonomy as it involves people not taking the opportunity to reflect on the implications of providing or refusing consent.

Necessary measures are likely to incur extra time and costs and it would be unacceptable, as mentioned earlier, to use this as grounds to exclude some people with dementia from participating in research unless the measures needed would be clearly disproportionate. In multicultural societies, minority ethnic groups in a particular geographical area may speak over 100 different languages and dialects, and some people may not be literate in their own mother tongue. Including a diverse group of people with dementia in research necessitates the availability of appropriate research instruments to use once recruited. It would be impossible to translate research materials into every possible language. Moreover, materials would ideally have to be translated, back-translated and properly validated. This would sometimes be a study in itself. Certain well-known tests may already exist in other languages and already be validated but may be difficult or costly to obtain. Sometimes, certain minority ethnic groups have a common language, which can be used to address these groups and involve them in research.

Level of education

In some studies, a certain level of education is required (e.g. expressed as a specified number of years' schooling or as having completed high school or college). In cases where this requirement is not directly linked to the research question or hypothesis, this could be construed as discrimination as it serves to exclude people who do not have that level of education. Whereas some people from majority ethnic groups, especially of the older generations, may have had little or no formal education, this is more common amongst some minority ethnic groups. Many people with intellectual disabilities would also find themselves excluded from research on the grounds of lower levels of educational attainment.

It might be argued that a specific level of education is needed to be able to comply with the requirements of the research, such as understanding materials and completing tests. However, a lower number of years' schooling or the absence of educational diplomas does not necessarily mean that a person lacks the ability to participate. Moreover, if research is to be rendered more inclusive, the onus should be on researchers to make their materials and instruments suitable/appropriate for the population they aim to study (bearing in mind that a target population of 'people with dementia' should ideally include a wide range of people, some of whom will have low levels of education, literacy and language abilities etc.).

Mobility

Provisions can often be made to ensure that people with dementia who have difficulties with mobility or transport can attend study visits. This could, for example, involve having some or all of the study visits in the person's home or organising and paying for a taxi. Possible solutions to promote the inclusion of people with mobility problems would be likely to involve additional costs and time. These should be budgeted into the study. In addition, as people may be inclined to exclude themselves based on the assumption that they would be unable to get to study sites, measures to promote their inclusion should be stated upfront in any information about the study.

Stage/severity of dementia

Unless there are strict scientific reasons, participation in research should not be limited to people with very mild dementia, but this is often the case. Sometimes, there are clear eligibility criteria which rule out the participation of people with advanced dementia but sometimes exclusion criteria are indirect (based on conditions such as living independently or having a certain level of capacity) or ambiguous (based on "investigator's uncertainty about willingness, ability, or medical status of the patient to comply with the protocol requirements" – Jongsma et al. 2016, p. 5).

The experiences and contributions of people with more advanced dementia to research are important. Their unjustifiable exclusion from research (i.e. based on a label and associated assumptions rather than on whether they comply with justifiable inclusion and exclusion criteria) contributes towards the lack of an evidence base for the treatment of people with more advanced dementia. Exclusion also results in a gap in understanding of the aetiology of the underlying illnesses. Furthermore, it deprives a sub-section of people with dementia of the right to contribute towards research and to reap any possible benefits linked to participation. There may be methodological and ethical challenges linked to involving people with more advanced dementia in research, especially if people also have some behavioural and psychological symptoms of dementia. However, such challenges can often be addressed successfully, thereby promoting more inclusive research. They should not simply be considered as a justification for exclusion.

Co-morbidities

As pointed out by Tinetti and Studenski (2011), there are an infinite number of combinations of diseases and treatments which makes the identification of a truly representative population very difficult. Comparative effectiveness research involves the comparison of different available treatments or examines their impact on particular sub-populations. This can be meaningful to doctors when trying to make a decision about or with a specific patient, particularly when treatments which are effective for one condition might exacerbate another condition in the same patient (Institute of Medicine of the National Academies 2009, Tinetti and Studenski 2011).

In a review of 103 dementia research protocols in the Netherlands covering drug trials, intervention studies and observational studies, 54% were found to exclude people with a psychiatric disorder and 56% people with a neurological disorder. 22% mentioned visual or hearing impairments, 38% medication use and 54% somatic comorbidities as exclusion criteria (Jongsma, van Bruchem-Visser, van de Vathorst and Raso 2016). In 9% of the studies, all five exclusion criteria were mentioned. Jongsma et al. (2016) point out that the average person with dementia has several comorbidities and that excluding people with comorbidities limits the external validity of studies and does not represent the full spectrum of people with dementia typically seen in clinical practice. They argue in favour of conducting pragmatic (i.e. real life) intervention studies involving a large number of people and a limited number of eligibility criteria, with the aim of obtaining more applicable and generalisable findings.

It is important not only to involve people with dementia in research but to involve them in research which results in knowledge that is meaningful to them. People with dementia who have comorbidities may have different priorities than researchers. In a review of over 100 studies into conditions which are common in older people, only 27% mentioned outcomes which were of relevance to the older people such as quality of life or functional status (Covinsky 2011). Whereas researchers may place their emphasis on disease specific outcomes, older people with multiple conditions may prioritise outcomes of a more individual or personal nature. Jongsma et al. (2016) also mention the importance of including patient-centred outcomes in pragmatic intervention studies.

Specific sub-groups of the population

Sampling strategies must ensure that the people who take part in research are the ones whose data will enable researchers to answer their hypotheses/research questions. Difficulties arise when definitions and conceptualisations of some groups of people are inconsistent, ambiguous or too broad. This may also result in some people being excluded from research, and conclusions being drawn or assumptions made about people from certain minority groups that are incorrect (i.e. over generalising and overlooking important contributions from excluded members of those groups).

Different definitions of the term 'migrant', for example, may lead to some people having a reduced chance of being involved in a study. It is generally agreed that a migrant is a person who has moved away from his/her country to take up residence in another country, temporarily or permanently, for one of more of a range of reasons. However, there is no universally accepted definition of 'migrant' (International Organization for Migration/IOM 2019, United Nations 2019). Precise definitions may vary considerably with some requiring a person to have crossed an international border, others not; some requiring a minimum or maximum period of time in the host country, others not. One could also ask at what point does a person cease to be classified as a migrant (after 10 or 20 years, never?). Sometimes a distinction is made between migrants and expatriates (expats). For many people, the latter[51] are not considered as migrants. This distinction most probably reflects negative stereotypes and value judgements about different groups of people and may also affect who volunteers to participate in studies.

Some terms which have different meanings are used interchangeably. This is the case for the terms 'migrant' and 'person from a minority ethnic group'. Many people from minority ethnic groups are not migrants. Some were born and grew up in the same country where they currently live and have no history of migration. Depending on the research question or hypothesis, if the two terms are used interchangeably, some people would be wrongly included or excluded and this would have an impact on the findings.

Terms used to define various groups of people who may at some point have dementia (i.e. people with intellectual disabilities, from BAME communities, with sensory impairments or from LGBTI communities) may change over time and differ with regard to whether and if so, how they are used across Europe. The term 'homosexual' for example, has largely fallen out of use since the 1970s, having been replaced by the term 'gay'. It is increasingly considered to be an outdated medicalised term with negative connotations (Shaw et al. 2019). However, it is still used in some countries and in some circles and can be found in books, magazine articles, sermons and everyday conversations. The use of the term would nevertheless be off-putting to many gay people who might find it disempowering. The terms used may also result in some people being eligible for recruitment, others being excluded and the results of different studies being difficult to compare. The following extract describes issues related to the use of terms for Gypsies, Roma and Travellers:

> *"Amongst the range of people referred to as 'Gypsy/Travellers' are English, Welsh and Scottish Gypsies, Irish Travellers, New Age Travellers, Boat People and Show People, as well as Roma from a variety of central and eastern European countries. (…./…). In continental Europe, Roma people are no longer officially described as Gypsies as the word has come to be associated with racial abuse. Many different groups are described as Roma (e.g. Manouches, Ashkali, Sinti and Boyash people)" (Condon et al. 2019, p.2).*

Condon et al. (2019) point out that membership of this broad category is fluid with people marrying into it, leaving it and not wishing to be associated with it if asked for official purposes. Moreover, the official UK census of 2011 only included Gypsies and Irish Travellers (combined as a single ethnic group).

For some researchers, there may also be uncertainty and a blurring of the distinction between terminology linked to intellectual disability (the most common form being Down's Syndrome), developmental disorders (such as autism which may or may not include an intellectual disability) and terms such as learning difficulty and 'mental retardation'. There are some cultural variations in the use of some of the terminology surrounding intellectual and developmental disabilities and differences in use between healthcare professionals and lay people (Nash, Hawkins, Kawchuk and Shea 2012) and this could lead to confusion and ambiguity in relation to recruitment.

Some studies recruit participants from a fairly broad population, such as people with 'cognitive impairment', 'memory problems' or 'cognitive difficulties'. This typically occurs in settings in which the specific characteristics of the population are not known (e.g. in residential care homes where many people with dementia do not have a formal diagnosis) or where a clear distinction is not necessary as it is not relevant to the research question or hypothesis (Palm et al. 2016). Such broad categories, when appropriate, facilitate recruitment, avoid researchers embarking on diagnoses which would otherwise not have been made and avoid people being informed that they have dementia solely for the purpose of the study (see next sub-section). However, terms should not be used as if they were interchangeable if this is not the case.

The inconsistent or incorrect use of terminology surrounding Alzheimer's disease may result in some people who have been recruited to studies wrongly assuming that they have dementia. Originally, Alzheimer's disease was more or less synonymous with Alzheimer's dementia. Recent conceptualisations of Alzheimer's disease, developed by

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[51] Examples of expats include people of Scandinavian, German or British origin who have moved to Spain on retirement.

the IWG and Dubois et al., propose a definition based on Alzheimer's disease pathology, extending along a continuum from a pre-clinical state, through a prodromal state and on to a state of advanced dementia (Alzheimer Europe 2016). The issue regarding the new conceptualisation of Alzheimer's disease is not that some people will be excluded from research but rather that some of those who are included may experience harm as a result of a lack of clarity surrounding terminology.

People may interpret information about the new definitions of Alzheimer's disease within the framework of their current knowledge about Alzheimer's disease and dementia. Consequently, there is a risk, in the context of research, that they might interpret categories such as preclinical Alzheimer's disease, prodromal Alzheimer's disease (IWG) or MCI due to Alzheimer's disease (NIA-AA) as forms of dementia (Alzheimer Europe 2016). This may have ethical implications linked to informed consent (i.e. it could not be considered as informed if based on a misunderstanding). This is an issue linked to the involvement of people who do not actually have dementia in research which is beyond the scope of this paper but it is nevertheless closely related. In a recent paper on this topic, Frisoni et al. (2019) concluded that the disease narratives of researchers and society need to be better aligned to ensure that the new conceptual framework is in tune with the social representation of Alzheimer's disease. Even if there is no mention of dementia, current lay understandings of Alzheimer's disease, which are still used by many healthcare professionals, combined with insufficient explanation by researchers, may increase the risk of such assumptions being made. An apparent discrepancy between information provided by different healthcare professionals and researchers may also result in a loss of trust in the healthcare and research professions.

Diagnosis

Progress in diagnosing dementia is being made but this is not uniform across Europe. In a European five-country study into carers' perceptions of the timeliness of dementia diagnoses, 30.7% to 45.5% of carers estimated that diagnosis had occurred at the middle to late stages of dementia (Woods et al. 2018). People in the middle to late stages of dementia have limited opportunities to be involved in research and late diagnosis deprives them of opportunities to be involved earlier.

Not being diagnosed at all (i.e. ever) is even more problematic with regard to the pursuit of inclusive research. Some studies suggest that more than half of community-dwelling people with dementia have not been diagnosed and this corresponds with the findings of a recent systematic literature review and meta-analysis of the prevalence and determinants of undetected dementia in the community (Lang et al. 2017). In this study, the rate of undetected dementia in Europe was 53.7%. A recent study of residents with dementia in German nursing homes found that 30% to 40% had not been accurately diagnosed and that inaccurate diagnoses had also been reported in studies in Norway, Israel and Ireland (Palm et al. 2016). Findings from the European systematic literature review were mixed regarding the number of cases of undetected dementia in relation to place of residence (community or residential setting) or ethnicity (Lang et al. 2017). However, the authors highlighted certain factors which were associated with a higher risk of not being diagnosed such as low socio-economic status, not speaking the local language, living alone, gender (male) and younger age.

Other factors which have been identified as rendering timely and accurate diagnosis of some groups of people more problematic include difficulties with language and communication, cultural perceptions of dementia and health, stigma and taboo, prejudice, structural discrimination and lack of referral by GPs (Beattie et al. 2005, Nielsen at al. 2011a and 2011b). These factors are common amongst people from minority ethnic groups but also apply to many other people with dementia. Nielsen et al. (2015) point out that in Denmark, only 11% of the expected number of older people from minority ethnic groups with dementia receive a formal diagnosis of dementia. It has been suggested that many GPs lack the cultural and linguistic skills to diagnose people from minority ethnic groups as well as the appropriate diagnostic instruments. Also, many people with dementia do not receive a formal diagnosis, either from their GP or from a specialist (Beattie et al. 2005, Diaz, Kumar and Engedal 2015). In some studies, it may be possible to include people who, for a range of reasons, do not have a clear diagnosis of dementia on the basis of reported symptoms. This would enable people who have dementia but do not have a clear clinical diagnosis to be involved in research. However, in such cases, it would be necessary to be consistent (e.g. to describe the study using the same terms and on the basis of the same inclusion criteria. Otherwise, the findings could be misleading and people would not be motivated to participate (i.e. not seeing the link to their own condition or lived experience).

Relying on recorded medical diagnoses of dementia can be problematic if they have not all been made on the basis of the same criteria. Diagnoses made in some settings and using some criteria may be more thorough than others (e.g. including imaging and complex cognitive testing). There is evidence in some countries that the use of imaging techniques and neuropsychological testing is lower in the primary care sector (Palm et al. 2016). Researchers who need to determine sub-types of dementia on the basis of existing diagnostic findings would therefore be unable to include some people with dementia in their studies. Moreover, Palm et al. (2016) point out that in the late stage of dementia, a differentiation between dementia subtypes is difficult. This means that people in the later stage of dementia who did not initially receive a differential diagnosis would not be able to participate in some studies. On the other hand, whereas the vast majority of research into dementia is about Alzheimer's dementia, non-drug interventional studies often do not specify the type/cause of dementia (Jongsma et al. 2016).

Despite a well-established and documented relationship between Down's Syndrome and Alzheimer's disease, with the first clinical signs of dementia usually occurring around the age of 50, the diagnosis of dementia in people with Down's Syndrome remains highly problematic (Cipriani et al. 2018). This is partly because of pre-existing levels of intellectual disability, lack of a consistently used diagnostic tool and a huge variation in levels of performance on cognitive tasks, combined with communication difficulties. It may also be because early signs of dementia differ from those experienced by people who do not have Down's Syndrome (Cipriani et al. 2018). Such difficulties at times result in 'probable' rather than confirmed diagnosis. A probable diagnosis of dementia, in people who do, and do not, have an intellectual disability should not be viewed as a reason to exclude them from research participation but should be noted.

Recruiting people with dementia using proxy measures of diagnosis (i.e. carers' or healthcare professionals' opinions rather than a formal diagnosis) is one way of including people who might otherwise be excluded but raises both scientific and ethical challenges. First, it touches on the validity and generalisability of the findings in that they cannot be generalised to people with dementia as it is not known whether all the participants actually had dementia. Second, it raises questions about trustworthiness and respect in that the researchers may strongly suspect that the participants have dementia (based on the information they have been given), but have not informed the people concerned. Indeed, doing so could be considered unethical due to the risk of causing harm but it also could also be considered a form of deception and an invasion of people's privacy. Finally, with regard to respect for autonomy, people should be informed amongst other things about the purpose of the study and why they have been invited to participate. If this does not occur, the volitional component of consent (i.e. the exercise of free will) would not have been fulfilled (Faden and Beauchamp 1986).

Recommendations for researchers

- Reflect on the justification and possible discriminatory assumptions behind the selection of inclusion and exclusion criteria.
- Take measures to ensure that very old and frail people with multi-morbidities (which includes many people with dementia) are not unfairly excluded from research.
- Only require a study partner if essential for the research or the participant's well-being.
- Consider research approaches for which a study partner would not be required.
- Explain to research ethics committees the rationale for including population groups that may at times have 'suspected' rather than confirmed diagnoses (e.g. people with Down's syndrome).
- Explain to research ethics committees the rationale for exclusion criteria which may exclude whole sub-groups of people with dementia from participating in research.
- Consider whether certain exclusion criteria could not be avoided if appropriate support was provided (e.g. ensuring that materials are accessible rather than excluding people with less than a certain number of years of education).
- Avoid inconsistent, ambiguous or too broad definitions and conceptualisations of groups of people with dementia.

Recommendations for research ethics committees

- Challenge ambiguous inclusion and exclusion criteria in research protocols, which limit the internal and external validity of research.

Recommendations for funders

- Cover expenses such as PI, logistics support and recruitment of diverse groups to allow for more inclusive research.
- Foster studies that have relevant outcome measures for people with dementia.
- Foster studies that contribute towards quality of care, including care for those living alone or in nursing homes and non-pharmacological studies whilst recognising that these studies may have small sample sizes.

Summary

Having inclusion and exclusion criteria enables researchers to involve people who will, collectively, be able to provide an answer to their research question or hypothesis and not jeopardise the success of their study or result in harm. A key question is, nevertheless, whether the rationale for inclusion and exclusion is scientifically justifiable and fair. Criteria typically include age, place of residence, literacy and language ability, level of education, mobility, co-morbidities and having a specific diagnosis. Sometimes they are unjustifiable (e.g. based on common practice, assumptions, reluctance to challenge research ethics committees and fear of anything that might risk a particular result not being found). Care needs to be taken when developing inclusion and exclusion criteria to ensure that people are not unnecessarily deprived of the opportunity to take part and that the criteria do not jeopardise the external validity of the study (i.e. resulting in findings from quantitative research studies being generalised to people who have little in common with those who participated in the study.

Issues related to the informed consent procedure

The principle of informed consent is one of the most fundamental foundations for the conduct of ethical research and, in addition, a legal prerequisite for the conduct of biomedical and some forms of clinical research. The aim of informed consent is to protect participants from harm (in keeping with the principle of non-maleficence) whilst respecting their autonomy and avoiding deception or coercion (Smith 2008, Beauchamp and Childress 2012). It is also linked to the principle of justice (or rather to potential injustice) in that people who seem unable to consent, but were not provided with accessible information and appropriate support (e.g. suited to impairments they may have), may be wrongly deprived of the same opportunities to participate in research that other people have. People with dementia may also be able to rely on proxy decision makers and/or advance directives as a means to express their right to self-determination at a time when this would otherwise be difficult or impossible. The use of such options may be considered empowering or restrictive and their ethical, practical and legal implications need to be considered. In this section, we examine possible challenges surrounding informed consent from people with dementia (including those for whom informed consent may be particularly difficult), the role of proxy decision makers and the use of advance directives for research involving people with dementia. But first, we will briefly examine the concepts of capacity and competence.

Capacity and competence

Informed consent offers participants the opportunity to weigh the possible benefits against the burden and risk of participating in research. It has a volitional component. This means that research participants must be sufficiently informed and that consent must be given voluntarily and without any coercive measures being used. There is also a cognitive component which is that research participants must have the capacity to make the decision at hand and must be competent (Faden and Beauchamp 1986, Beauchamp and Childress 2012). Decisional capacity and competence are sometimes used interchangeably, but their meaning differs slightly. Decisional capacities are the capacities needed to make a decision. Competence is the dichotomous legal judgement whether the capacities are enough to make the decision at hand. This must be in relation to a particular task, at a particular moment in time and under specified conditions (Buchanan and Brock 1990). Competent persons have sufficient decision-making capacities, while incompetent persons have insufficient decision-making capacities.

The area of competence of relevance to the involvement of people with dementia in research is that of decision-making capacity. People may have varying degrees of decision-making capacity. In the case of dementia, it may fluctuate not only because of dementia but also due to a range of psychosocial, situational, medical, psychiatric and neurological factors (Holzer et al. 1997). However, whereas capacity is not an all or nothing matter in that people may possess varying degrees of capacities (with regard to a particular task), competence (i.e. the legal concept) is. A person either reaches the established threshold for competence with regard to a particular task or does not (Buchanan and Brock 1990). So a person is judged either competent or incompetent to make a decision about participating in a particular study. The importance of making such a distinction is to determine whether a potential participant should consent to participation in a particular study or whether somebody else should make that decision on his/her behalf. Lack or loss of the capacity to consent to research should not systematically result in a person's exclusion from research (Hérault, Bravo and Trottier 2018).

Part of the process of obtaining informed consent involves determining whether a person has the required capacities for the decision at hand. Competence does not equate with perfect rationality in that people have the right to make decisions which others may consider unwise or irrational provided that they fulfil the criteria governing valid informed consent (DuBois 2008). This is particularly important in relation to people with dementia from various sub-groups in society who may have different values, expectations and beliefs to those of the researchers. Other factors such as level of education, socio-economic status, language and communication difficulties and distrust may also affect assessments of capacity and ultimately influence whether a person with dementia is deemed competent to participate in a particular study.

Assessment of the capacity to consent

Informed consent should not be understood as a signed piece of paper or a one-off requirement at the beginning of a study. Rather, it is a process based on verbal, non-verbal and behavioural cues, which needs to be revisited regularly throughout the research (Dewing 2007, Hellström, Nolan, Nordenfelt and Lundh 2007) and accompanied by ongoing reflection about the ethical involvement of participants.

> *"Consent-seeking should not be thought of merely as an event. The securing of approval from research ethics committees or the signing of a consent form by respondents do not, in themselves, absolve researchers of further need to engage in ethical concerns and reflexive practice. Researchers should refrain from treating such ritualistic enactment of consent-seeking as the be-all and end-all of ethical considerations but should instead engage in a more reflexive approach to the types and levels of consent required before, during and after the act of research" (Sin 2005, p. 290).*

This is applicable to all research designs but a degree of flexibility and the possibility for adaptation, which may result in new ethical issues arising which were not previously anticipated, is particularly relevant for longitudinal studies and some qualitative research designs (Hellström et al. 2007). A range of approaches can be used to obtain informed consent for participation in research such as clinical interviews, discussions and measures of capacity, including neurological test batteries (Beattie 2009).

The MacArthur Competency Assessment Tool for Clinical Research (MacCAT-CR) (Appelbaum 2007, Appelbaum and Grisso 2001) is sometimes described as the 'gold standard' for the assessment of capacity to consent to clinical research, although not necessarily on its own (Howe 2012). It covers understanding, appreciation of the nature of the situation, reasoning and expressing a choice. Although the tool provides a score, the end decision about capacity to consent is made by the physician administering the tool and this reflects a judgement about the appropriateness of the decision rather than the person's capacity and hence is a normative judgement (Jongsma 2016). This means that people who make decisions about whether to participate in research which reflect different values, atypical reasoning, eccentric views or perspectives which in some way are interpreted as falling outside the norm, may be deemed as lacking the capacity to consent to research. As mentioned earlier, this would be unethical but may well be the case for many people from sub-groups in society with which researchers are not familiar. On the other hand, it could also be argued that by not providing a cut-off score, the MacCAT-CR provides greater flexibility and, if used sensitively, could be a more inclusive tool for use with people whose scores do not fully reflect their actual capacity to consent.

Examples of other instruments include the University of California San Diego Brief Assessment of Capacity to Consent (UBACC) and the Evaluation to Sign Consent (ESC) (Beattie 2009). The Mini Mental Status Exam (MMSE) is not generally considered as a suitable or reliable measure of the capacity to consent to research (Howe 2012). Moreover, as with diagnosis, care should be taken when drawing conclusions about capacity to consent using instruments which have not been validated for people from various minority groups.

The ability of researchers from different domains (i.e. including non-medical research) to assess capacity to consent to research varies dramatically and some researchers may need to seek assistance from their peers, or at least to reflect on the possibility of having an external assessor of the capacity to consent. Non-medical research does not necessarily entail the same risks to participants as clinical drugs trials and the level of understanding required by participants may sometimes be lower. Nevertheless, it may result in some form of harm and still touches on a range of ethical issues such as respect for autonomy, voluntariness, justice, confidentiality and dignity.

Some organisations and institutions have developed checklists for researchers of things to consider when assessing the capacity to consent to participation in research. The British Psychological Society (Dobson 2008)[52] in the United Kingdom, for example, has a form to guide researchers when participants do not have capacity to consent. This covers whether efforts have been made to maximise the capacity to consent, whether impairments exist which might affect decision-making capacity, whether the person understands the voluntary nature of the research, what it is about and is able to understand and weighed up the benefits and risks, and finally whether he or she has communicated his/her decision regarding participation. Depending on the responses given,

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[52] Developed in collaboration with the Royal College of Speech and Language Therapists and the Royal College of Psychiatrists.

researchers are advised to inform themselves about how to enable decision making, discuss the issue with the Principal Researcher or conclude that the person does not have the capacity to grant or withhold consent to take part in the research project.

The ongoing assessment of consent can be challenging in relation to people who already have dementia and whose capacity to consent may deteriorate during a particular study. Hellström et al. (2007) argue that most approaches to consent rely too heavily on cognitive ability, including the recollection and manipulation of facts, do not take feelings and experiences sufficiently into account and therefore place people with dementia in a threatening and unpleasant situation. The performance of people with dementia on tests of cognitive performance may be influenced by a range of factors such as the time of the day, level of concentration and motivation. Cognitive processing may also be affected by a person's state of general well-being (Damasio 2000). Consequently, tests which are carried out with minimal regard for the individual may affect the results obtained and lead to the unnecessary exclusion of some people with dementia from research.

Flexible and adapted informed consent procedures need to be developed or elaborated in collaboration with members of the groups concerned. This might, for example, include ensuring that information for informed consent is presented in a way that makes it easier for people from specific groups to understand and retain it sufficiently long to make a decision. Dewing (2008) points out that traditional competency-based approaches to informed consent fail to take adequate account of the situational aspect of capacity and the importance of interdependence and relationships. Some researchers have proposed alternative methods such as verbal and behavioural consent (Bamford and Bruce 2000, Bartlett and Martin 2002, Cowdell 2006). Possibilities for alternative forms of consent must also be considered in the case of people with dementia with additional impairments (e.g. visual impairments and intellectual disabilities).

Many people with dementia are able to give informed consent if appropriate adjustments are made and necessary support provided. Process consent is an approach and method to obtain ongoing consent from people "who have an extremely limited capacity, who would generally be thought to be incapable of legally informed consent by others, but on observation can communicate and express their wishes in other ways" (Dewing 2008, p.63). It is a person-centred approach which is also influenced by the concept of social justice and the importance of relationships. Process consent has been implemented, following ethics approval, in the United Kingdom, Ireland and Australia for several qualitative research projects involving people with dementia in the field of gerontology. Dewing (2008) describes the approach as a way to work towards inclusion rather than exclusion of people with dementia in research but recognises that there will still be a cut-off point at which some people will not be able to make or communicate even small choices.

Process consent offers an alternative to the current over-emphasis on cognitive capacity, is flexible and focuses on individuals and their residual capacities and other abilities. It is not yet known whether this approach could be further expanded beyond the specific area of gerontological research and there are a few challenges surrounding the observation and interpretation of states of wellbeing (see Part 3 for more on this topic). However, Dewing (2008) draws attention to the risk of researchers too readily accepting the gold standard of informed consent and emphasises the need to seek alternative methodologies for consent.

Being informed vs understanding

Information provided to potential research participants with dementia should cover various issues related to a particular study (e.g. what the study is about, why the person is being asked to participate, what participation would involve, potential risks and possible benefits if any, who is responsible for the study, the timeframe of the study and whom to contact for more information, further explanation or in case of complaints) and general issues related to participation in research (e.g. the voluntary nature of participation, the right to withdraw at any time, confidentiality and privacy issues). Participants must be able to comprehend the information provided, demonstrate that they have understood, assess the relevance of information to them and make a reasoned decision.

Some research can be particularly complex, requiring researchers to provide additional information to help people understand various concepts and the purpose of the study. In order to promote inclusive research, it is important to ensure that every effort is made, even for relatively straightforward studies about everyday issues, to ensure that participant information sheets and informed consent forms are accessible to people with varying degrees of cognitive impairment, intellectual disabilities and language and communication difficulties, as well as being appropriate for a culturally diverse population. It may be necessary to develop more than one version of relevant documentation, to provide alternative materials and tools, and to provide support. Watchman (2014) also emphasises the importance of adopting a flexible and individualistic approach to facilitate the active engagement and participation of people with intellectual disabilities and dementia in research.

Assessment of burden, risk and potential benefit

In the context of research, risk is often discussed alongside burden and balanced against possible benefit. Ethical research aims to minimise risk and enhance potential benefit so that the risks to the participant are proportionate to the potential benefits to that person and society (Emanuel, Wendler and Grady 2000). However, the definition of benefit is often too narrow to take fully into account the kinds of benefits that are possible and important to people with dementia participating in research, such as spiritual, emotional or psychological benefit. Moreover, there are ethical and legal limits to the degree of risk and burden that is considered acceptable for research involving people who are unable to consent. For example the Council of Europe's Additional Protocol on Human Rights and Biomedicine (2005) only allows research without a direct benefit with people who are unable to consent if the research entails only minimal risk and minimal burden.

Additionally, article 15 of this document contains a series of conditions in the case of people without the capacity to consent to research. These include conditions such as that the results have the potential to produce real and direct benefit to their health, that the research could not be carried out on people capable of giving consent, that the people have been properly informed of their rights and of safeguards, that authorisations from legal representatives have been obtained, that previously expressed wishes taken into account and that they have not objected to participation.

The issue of direct benefit may be side-stepped subject to fulfilment of the above-mentioned conditions provided that, amongst other conditions:

- "the research has the aim of contributing, through significant improvement in the scientific understanding of the individual's condition, disease or disorder, to the ultimate attainment of results capable of conferring benefit to the person concerned or to other persons in the same age category or afflicted with the same disease or disorder or having the same condition;
- the research entails only minimal risk and minimal burden for the individual concerned; and any consideration of additional potential benefits of the research shall not be used to justify an increased level of risk or burden."

Assessments of risk, benefit and burden cannot easily be made for whole groups of people and there is a risk of paternalism, leading to a lot of people being overprotected and excluded from research without having any say in the matter. Drawing on a relational ethics perspective, Fisher (2009) suggests that failure to include the perspectives of those directly concerned in such discussions may lead to the rejection of scientific research procedures which potential participants and their relatives might perceive as benign and/or worthwhile considering. She states:

> *"Formulating regulations and ethical judgments solely on the bases of opinions expressed by experts in the scholarly community and IRB members risks treating subjects as 'research material' rather than as moral agents with the right to judge the ethicality of investigative procedures in which they participate" (2009, p.5).*

As unique individuals, people with dementia have their own perceptions of danger and of the level of risk that they are willing to take. Having dementia may even be an additional motivation to take part in research (e.g. to benefit relatives and other people who may develop dementia in the future or as a means to contribute to society in some way). There is therefore a risk of overprotecting people with dementia, infantilising them and depriving them of their autonomy, which is problematic for the reasons discussed earlier in the sub-section on gatekeeping (Jongsma and Schweda 2018).

With regard to benefits which are not directly linked to health, Casaret, Jason and Karlawish (2000) point out that when people are approaching death, for example, the things that they find important may change. They may, for example, place a greater value on dignity, meaning, control, strengthening relationships and addressing unfinished business of a personal nature. In the case of people with advanced dementia, the expression of such concerns or priorities may be difficult but this does not mean that they do not exist. Participating in research may, for some people with dementia, correspond to these new priorities (e.g. by providing meaning or enabling them to feel part of something). For others, participating in research might interfere with current goals and priorities (e.g. by taking up valuable time they might prefer to spend with relatives or friends). Similarly, a person who is very close to death might not assess the possibility of serious risk in the same way as a person who has not yet reached that stage. Some risks may be considered as being more significant and others less so (Casarett, Jason and Karlawish, 2000). This would depend greatly on the individual as well as on his/her awareness of his/her prognosis.

People with dementia, and those whose opinions they consider valuable, should be involved, to the greatest extent possible, in assessing the level of risk they are willing to take and the level of burden they are willing to accept. Details of the responses from participants (or in some cases their proxies) about the perceived risks, burdens and benefits of participation should be taken into consideration by researchers when designing subsequent studies. It is important to realise that in practice, researchers focus a lot on lowering risk when designing their studies and often forget about the burden that participation may entail. While burden is also a very important factor in potential participants' decision making, burden for them does not only entail specific procedures. It also includes logistical burden (e.g. travelling to hospitals, time spent participating) and burden for the family and carers. These types of burden are very important in relation to decision making and should already be dealt with by researchers in the research protocol.

Voluntariness and the therapeutic misconception

Participation in research of any kind must be voluntary. There are different degrees of lack of voluntariness ranging from being subjected to research without having had any choice in the matter, being forced to participate on the basis of some kind of threat or deciding to participate on the basis of undue influence (DuBois 2008). Pressure to participate can be quite subtle and may be unintentional. An example would be a person being asked by a healthcare professional or staff in a residential care setting and fearing upsetting that person due to a feeling of dependency on him or her, respect for his/her position or not wanting to jeopardise an existing positive relationship with that person.

People's voluntariness could also be influenced by their interpretation of what they are being asked to do and why. The decision about whether or not to participate in research may depend on how research is understood and most importantly, whether a person understands the difference between research and treatment or care. This might not always be as obvious as it first seems, especially in situations where a person is informed about a study by a healthcare professional in a healthcare setting (Lewens 2006). There is some evidence to suggest that people with dementia considering participation in research frequently mistake proposed research for treatment options (Dresser 2001). The risk of this happening may be even greater in the case of people with mild dementia who write combined advance directives for treatment and research (Pierce 2010). Medical researchers and healthcare professionals involved in recruitment for research should not take it for granted that people with dementia understand this difference and also bear in mind other factors which may make it more difficult for some people to understand (e.g. people who are not familiar with the way the healthcare system works and the connection to medical research, who have difficulties understanding the language used, who are stressed about their condition or who feel intimidated by the perceived power status of the person providing them with the explanation).

The provision of treatment and care is generally consented to on the basis of an assumption that it is necessary and will hopefully in some way be personally beneficial. If this distinction is not clear, it cannot be said that a person has given informed consent to research. Medical researchers should try to ensure that potential participants realise that the overall aim of research, including biomedical research, is not to improve their health or contribute to their personal wellbeing.

It is also important to ensure that healthcare professionals responsible for recruiting research participants, who are sometimes extremely enthusiastic about the study, do not 'oversell' the virtues of the study or communicate their assumption that it will be beneficial in some way to potential participants. Research participants will not necessarily benefit from better monitoring, for example, and even if this were the case, this should not be a selling point but rather an indication of the need to improve standard diagnosis and care.

Financial incentives to contribute to research in the context of PI were discussed in Part 1 of this discussion paper and linked to arguments surrounding recognition of the value of a person's contribution to research. With regard to potential research participants being offered financial incentives, the principle of justice needs to be considered because payment could contribute towards the exploitation of some groups of people and skew samples in much the same way as their systematic exclusion would. In addition, for some people sufficient payment might result in them accepting risks or burdens that they would otherwise not have been willing to take.

Vulnerability and disability

Researchers may be faced with difficult decisions about how to comply with rigidly defined informed consent procedures, including additional measures to protect vulnerable participants, whilst not excluding certain groups of people (such as some people with intellectual disabilities, with more advanced dementia or from various minority groups) or including them without their consent (Doody 2018). Raudonis (1992) describes a fine line between "protecting vulnerable research participants and making paternalistic decisions supposedly in their best interests" (Nordentoft and Kappel 2011, p. 368). Alongside the clear-cut perception of competence as something a person has or does not have, Sin (2005) emphasises the fluidity of consent and the dynamic nature of research, which calls for a more reflexive approach and acknowledgement of the socially constructed nature of assumptions surrounding the concepts of 'normal', 'competent' and 'informed consent'.

A lot of the protective measures governing the participation of people with dementia are linked to perceptions that they form a vulnerable group of people. Such vulnerability is an acquired status compared to other groups in society who may have been considered as vulnerable all their lives, which is the case for many people with physical or intellectual disabilities. The perceived vulnerability and in many cases over-estimation of vulnerability may partly have its origins in the long history of institutional care of some groups of people and the focus on looking after or caring for them

(Doody 2018). The normalisation movement which began in the 1970s, based on the work of Wolfensberger, Nirje and Bank-Mikkelson amongst others, was linked to intellectual disability and promoted 'the dignity of risk' rather than protection. Its focus on integration in community life is perhaps also relevant to the goal of enabling people with dementia to participate in research and for researchers, funders and research ethics committees alike to avoid overly focusing on protection and denying freedom of choice to people with severe, profound or significant disabilities (NHS Health Scotland 2016).

The first three paragraphs of article 12 of the United Nations Convention on the Rights of Persons with Disabilities (CRPD 2006) may also be helpful when considering the issue of consent to research by people with dementia, namely,

5. States Parties reaffirm that persons with disabilities have the right to recognition everywhere as persons before the law,
6. States Parties shall recognize that persons with disabilities enjoy legal capacity on an equal basis with others in all aspects of life,
7. States Parties shall take appropriate measures to provide access by persons with disabilities to the support they may require in exercising their legal capacity.

Consequently, it should not be concluded that a person lacks competence (i.e. legal capacity) to consent to research unless necessary adaptations and efforts have been made to minimise the possible impact of various impairments (e.g. linked to language, communication, memory and abstract reasoning) on decision-making capacity. Similar adaptations should also be considered in relation to other factors, not necessarily physical, mental, intellectual or sensory impairments, which may impact on the capacity of a person with dementia to give informed consent.

Assent and dissent

A key aspect of the informed consent procedure, also linked to the requirement of voluntariness, is that people understand that they have the right not only to refuse to participate in a study but also to withdraw at any time without having to provide any justification for their decision. When a person with dementia continues to participate though unable to confirm his/her informed consent (e.g. based on a proxy decision maker having taken over this responsibility), special attention should be paid to signs of possible assent and dissent. These should equally be respected in the case of people who still have the capacity to consent (Jongsma, Bos and van de Vathorst 2015).

Black, Rabins, Sugarman and Karlawish (2010) suggest that both assent and dissent can be expressed or indicated verbally (e.g. saying 'no'), behaviourally (e.g. acting agreeably or trying to move away) or emotionally (e.g. through facial expression or posture) and advise researchers to consult with informal carers to help them understand individual participants' typical means of expression in relation to assent and dissent. This means that even if unable to understand the ins and outs of the study or give reasons for wanting to take part or withdraw, a person with fairly advanced dementia may be able to communicate his/her wish to withdraw or refuse a particular research activity and this may contribute towards decisions to involve more people with advanced dementia in research. However, verbal and non-verbal signs of dissent may be subject to individual and cultural variations. Moreover, Young, Ferguson-Coleman and Keady (2014) point out with reference to Deaf people, much of the focus on non-verbal communication of people with dementia equates non-verbal with non-linguistic, whereas with Deaf people the non-verbal is actually language. Facial expression, raising or lowering the eyebrows, puffing out the cheeks and movement of the eyes all contribute towards the meaning of an utterance. Similarly, averting one's gaze is not simply a non-verbal marker of indifference but a means of disengagement as no further communication can take place (Young et al. 2014).

Black et al. (2010) describe the concept of assent as a means to respect a person's remaining autonomy and to enable cognitively impaired people to be involved, to the extent that this is possible, in the decision making process. Careful attention to signs of assent or dissent throughout the research process may also help keep the person with dementia and his/her needs and wishes at the centre of research and avoid him/her being treated as a mere means to an end.

The term 'dissent' is used somewhat ambiguously in the literature. One definition refers to objection during a trial ('the wish to discontinue participation'), also sometimes referred to as objection, distress or resistance. This notion of dissent draws on respect for autonomy whereby dissent should be closely monitored, given that some people who lack decisional capacity are not able (cannot formulate the wish) to withdraw during the trial (Jongsma, Bos and van de Vathorst 2015). Another definition describes dissent as the opposite of consent or assent, consisting of a measure of protection from risks and burden that a person might not want to accept, a means to respect his/her dignity and a shield from distress or from unwanted research activities. Black et al. suggest that the need to respect dissent is, ethically-speaking, hierarchically more important than obtaining assent due to its protective function but nevertheless argue in favour of including people with dementia who are unable to consent and also unable to dissent. They recommend that research ethics committees determine whether any protection beyond proxy consent (e.g. such as an advance directive for research and independent monitor) is needed for people who lack the ability to assent and/or the ability to dissent.

Recommendations for researchers

- Ensure that informed consent does not consist of a one-off event if the research is ongoing over a period of time.
- Clarify the process for determining capacity on an ongoing basis.
- Bear in mind that although a diagnosis of dementia may justify an evaluation of a person's capacity to consent to research, it does not automatically mean that a person lacks such capacity.
- Reflect on appropriate processes and legislation where judgement of capacity is difficult.
- Develop and propose to research ethics committees alternative methods of obtaining informed consent which are neither exclusionary nor paternalistic.
- If people are unable to write, check whether your research ethics committee would accept oral consent (e.g. in the presence of at least one witness and repeated at regular intervals).
- Provide details about how and by whom capacity will be assessed.
- Where possible, arrange for ongoing assessments of capacity to be carried out by those with the necessary expertise who are independent from the research so as to avoid bias.
- Be transparent about the role of medical researchers so as to prevent therapeutic misconception (e.g. do not sign letters about possible research trials as 'your treating doctor').
- Where possible, try to ensure that healthcare professionals (where relevant) proposing research participation are not also responsible for a person's treatment or care, so as to avoid putting the person with dementia under pressure to consent and also to avoid the therapeutic misconception (i.e. mistaking a proposal to participate in research for an offer of treatment).
- Take measures to include people with dementia in the informed consent process, including when they are deemed to lack capacity (e.g. provide information and ask for assent).
- Closely monitor participants during research and check whether they are still willing to participate and understand what participation involves.
- Use validated tools, wherever possible and when available, to obtain consent from people from minority groups or with specific characteristics which necessitate special measures.
- Consider developing or using different methods and tools, where required, to provide people with dementia with information about the proposed research.

Recommendations for research ethics committees

- Require researchers to describe in research protocols the method used to identify a person who could support the patient in the informed consent process, if needed.
- Recommend that the healthcare professional proposing participation in research is not also responsible for a potential participant's treatment or care.
- Require researchers to provide details of their plans to seek ongoing informed consent and to provide justification if this is not planned.

Recommendations for funders

- Ensure that any reimbursement or payment proposed to research participants is in line with available national or local guidance on rates, or in keeping with the principle of fair market value.
- Encourage researchers to incorporate costs related to obtaining ongoing consent in their requests for funding.
- Ensure clarity about the process to monitor ongoing capacity if research continues over an extended period of time.

Summary

The informed consent procedure is fundamental to the conduct of ethically sound research and closely linked to the principles of respect for autonomy, non-maleficence and justice. Approaches are needed which provide appropriate protection from harm whilst not overriding the right to self-determination (e.g. through blanket judgement of vulnerability). Informed consent should be seen as an ongoing process, continuing throughout the whole research process. It is important to be attentive to signs of assent and dissent which may be expressed differently by people with dementia from different sub-groups of the population. The capacity to provide ongoing consent and the way that assent and dissent is communicated may vary for each person as dementia progresses.

The issues of burden, risk and benefit, and the importance that each person attributes to them, need to be considered, as well as the potential confusion between research and treatment, especially with regard to clinical research. Creative and innovative methods are needed to make it possible for a diverse population of people with dementia to be involved in research (e.g. people with dementia from minority ethnic groups, with lower levels of education, language or literacy difficulties, with intellectual disability or with more advanced dementia). Procedures, attitudes and the environment should promote rather than hinder the abilities of people with dementia to provide consent.

Consent with and on behalf of people with dementia: shared, supported and proxy decision making

Researchers often suggest to potential participants that they discuss their possible participation in a study with their close family and friends, and state that they are more than welcome to contact them for further clarification or with any questions. However, discussions about the involvement of other people in the decision-making process related to informed consent to research are often linked to concerns about a person lacking the capacity to provide informed consent. There has also been a move away from the concept of proxy (i.e. substituted) decision making towards shared decision making and supported decision making. This is sometimes seen as a way to promote autonomy and enable people with dementia who have limited capacities to continue to play a role in decision making. The concepts of shared decision making and supported decision are similar in some respects and may be complementary to some extent. They developed separately but in parallel and the origin of each leads to a slight different in how they are perceived.

Shared decision making, for example, focuses on a range of skills and practices that can be used to promote a collaborative decision making process. This has been typically used in the context of healthcare decision making (Elwyn et al. 2012). The emphasis is on the decision-making process with the actual decision, especially the issue of who made it, being considered as far less important. The process, which typically involves healthcare professionals as well as patients, involves the use of decision aids, the provision of information and ongoing discussions, all of which are aimed at increasing knowledge, helping people to feel less confused about a range of options and resulting in more informed and values-based choices, better communication and people being more involved in making decisions (Simmons and Gooding 2017). Shared decision making has, however, been criticised for failing to take sufficiently into account the power imbalance which often exists between patients and healthcare providers or researchers and for decision aids often failing to accommodate for the needs of people with low health literacy (Simmons and Gooding 2017).

Shared decision making can also be understood as an individual or cultural choice or philosophy irrespective of cognitive ability. For many people with dementia, participating or sharing in the decision-making process is as important as, if not more important than, making the actual decision (Daly, Bunn and Goodman 2018). In some cultures, there is a deeply rooted set of ethical and cultural traditions which reflect familism rather than the individualist approach common to informed consent procedures in Western culture. Writing in the context of recent attempts to promote a more individualistic approach to informed consent in China, Bian (2015) states:

> *"Familism is the Confucian family-based and family-oriented way of life embodied in the Chinese mode of decision-making in which all close family members play an important role. Close family members in contemporary Chinese society typically include the patient's spouse and parents (especially when one's children are not yet adults), as well as adult children. When facing major decisions for any family member, all close family members come together to make a collective decision" (Bian 2015, p.375).*

According to this philosophy, the family is prioritised over the individual, which runs counter to the taken-for-granted practices of individualism, which are dominant in many modern Western societies (Bian 2015). Familism and the practice of shared decision making is common amongst several minority ethnic communities in Europe and is not related to lack of decision-making capacity. Enabling a less individualistic approach to decision making in the context of research may be an important means to promote the ethical involvement of a more diverse set of people with dementia in research, irrespective of the reason for the choice of this approach.

Supported decision making is about providing the necessary support to enable a person to make a decision, including one that is legally recognised. Whilst the term does not appear in the UN Convention on the Rights of Persons with Disabilities (2006), supported decision making very much reflects the ethos of that convention and is often associated with it. Simmons and Gooding (2017, p.276) describe the rationale for supported decision making as follows:

> *"In crude terms, supported decision-making promotes the idea that just as people who use wheelchairs are entitled to ramps in order to access buildings, so too people with mental health-related disability – 'psychosocial disability' – are entitled to support to exercise choices and their lives. (The same idea holds for people with intellectual disability, or any other disability that may affect decision-making, including the way other people perceive and/or denigrate a person's decision-making ability)."*

In their recommendations on research (and data sharing) involving people with dementia, Thorogood et al. (2018) point out that research ethics guidelines generally promote supported or shared decision-making. They suggest that this might include "simplifying consent forms, providing visual or memory aids, taking interactive or educational approaches (where persons with dementia are asked to explain their understanding of consent elements), re-explaining misunderstood information, or involving familiar carers to facilitate explanation and communication of a decision" (Thorogood et al. 2018, p.1338). Such measures could perhaps be further extended to address any barrier experienced by people with dementia, regardless of whether it is linked to a health condition (e.g. such as having a lower level of literacy or being a member of a group that is stigmatised etc.).

In situations where a person has been appointed who is authorised to decide on behalf of a person with dementia about participation in research (i.e. a 'proxy' or 'substitute' decision maker), it is increasingly excepted that that person should base his/her decisions on the known will and preferences of the person with dementia and not on the basis of 'best interests' or 'welfare' (Thorogood et al. 2018). Where possible, people with dementia should, as far as possible, be included in the decision-making process. In addition, even people evaluated as unable to give consent for research may preserve the capacity to appoint a research proxy (Kim et al. 2011).

It is increasingly recommended, if not stipulated (European Directive 2001), that proxy consent should represent the person's presumed will. In reality, this is often not the case. Proxies often do not know a person's will and preference and make decisions that the person appointing them would not have made (Kim et al. 2013, Thorogood et al. 2018). Proxies may sometimes be too restrictive, thus depriving people with dementia of the opportunity to be involved in research or, on the contrary, involve people with dementia in research when this does not correspond to their wishes, values and interests (Jongsma 2016).

An important area to consider is that of risk because decisions about participation in research involve considering potential risks, burdens and benefits. According to Landau et al. (2010), a person's position regarding the relative importance of safety and autonomy may depend on whether he or she is personally responsible for the safety and wellbeing of the person with dementia. People may be more protective of others, than they would be of themselves (Nuffield Council on Bioethics 2009), particularly in cases where the person being 'protected' is considered in some way vulnerable. Proxy consent is often described as paternalistic but as Hellström et al. (2007) point out, it may also serve as a means to enable people with dementia, who would otherwise be excluded from research, to engage in an activity which can, in many cases, boost self-esteem, be enjoyable and enriching, and provide a means to validate feelings and experiences.

The recommendation or obligation for proxies to take into account current or previously expressed wishes of a person with dementia who lacks the capacity to consent to research could be considered as a factor contributing towards respect for autonomy, beneficence and non-maleficence, but the phrase 'taken into account' does not guarantee that the proxy will respect the wishes of the latter. If the known or supposed wishes of the person with dementia are not acted on (unless there is a morally justifiable reason for not doing so), it cannot be said that his or her autonomy has been respected. This suggestion should not be interpreted as undermining the goodwill or underestimating the difficulties that many proxies experience when making decisions on behalf of people with dementia. Proxies can be very beneficial in combination with advance directives because they may be well-placed to help interpret/identify the kinds of studies a person might have wanted to participate in, drawing on the information provided in an advance directive, their knowledge of the person and possible information about his/her wishes and interests. Some people with dementia grant their proxies the power to override their previously expressed wishes (e.g. in the light of scientific advances which result in unanticipated consequences for participation or on significant changes in the person's situation). Others may want their advance directives to be followed, without leaving leeway for their proxy to decide.

Some people may be hesitant about taking on the role of proxy. Such hesitancy may be linked to a range of factors such as not understanding the concept of proxy decision making, not understanding the possible legal implications of being a proxy, worrying about the level of commitment needed, being unsure about what knowledge and skills they may need and whether they have that, having language difficulties, finding explanations and documentation difficult to understand, having concerns about having to discuss personal or complex issues with researchers or lacking trust in researchers. In some communities, there may be issues of respect and clearly defined responsibilities which influence who should take on that role. That person might not be the one who understands best the needs and wishes of the person with dementia. In some communities, it may be common to involve several people in such important decisions and not to appoint one person as proxy.

Recommendations for researchers

- Show willingness and be proactive in discussing the issue of shared, supported and substitute decision making with potential participants with dementia and other relevant people. Some people will not be familiar with the different options.
- Ensure that people with dementia are encouraged, where possible, to make their own decisions relating to participation in research, bearing in mind that some may prefer to appoint a proxy.
- Do not seek the opinion of a proxy decision maker if the person with dementia has the capacity to decide for him/herself.

Recommendations for research ethics committees

- Ask researchers to describe how they intend to involve people with dementia who lack the capacity to provide informed consent to research without assistance or support.
- Be open to potential participants' possible wishes to involve significant others in the informed consent process.

Summary

When promoting inclusive research, it is important to respect priorities and traditions related to decision making which are more family or group orientated (as opposed to the more individualistic approach common in much of Europe). As dementia progresses, people may in any case need more support in order to make and express decisions related to their potential participation in research. Shared decision making helps ensure that when people with dementia lack the capacity to make decisions on their own, every attempt is made to enable them to participate in the decision-making process. In some cases, where they are not able to make a decision themselves, such participation may be just as important as the decision that is eventually made. Supported decision making is about providing sufficient support to enable the person with dementia to make a decision, including one that is legally recognised. Proxy decision making does not simply involve making a decision on someone else's behalf because proxies are expected to make decisions which represent a person's will and preferences (and their presumed will if they are currently unable to express preferences). Researchers need to play an active role in whatever form of decision making is legally appropriate and adopted so as to ensure that people with dementia are meaningfully involved to the greatest extent possible and according to their preferences.

Advance directives for research

In the context of inclusive research, it is important to consider the involvement of people with more advanced dementia in research, including those who do not have the necessary capacity to consent and who do not have a representative with the legal right to make such decisions on their behalf. It may be possible to determine what a person's wishes would have been by consulting close family and friends or an advance directive for research. Advance directives were developed in the 1960s in the United States of America and were originally written to express in writing wishes regarding medical treatment in the event of an accident or illness which made it impossible to exercise self-determination (Vollmann 2001). The moral authority of advance directives resides in the principle of respect for a person's precedent autonomy (Berghmans 2000, Vollmann 2001), by permitting such autonomy to be extended into the future, well beyond the point that he or she would otherwise have been able to exercise it. The concept of the advance directive has been further extended to the research situation in some countries. Advance directives may be legally binding or simply advisory depending on their legal status in each country and sometimes on the nature of the decisions to be made (Andorno, Gennet, Jongsma and Elger 2016).

Several ethical concerns have been raised about the use of advance directives in the context of dementia (in general, not necessarily for research). These are mainly linked to arguments about personal identity and changing interests. There is a broad and complex philosophical debate surrounding personhood and dementia. An issue of particular concern and of relevance to the debate about advance directives for research is whether the person who wrote an advance directive is the *same* person as the one for whom it may later be applied, and if not, why the advance directive should be respected.

Parfit (1984), for example, takes the view that personal identity is constituted over time by varying degrees of continuity between former and later selves in terms of a wide range of psychological and physical features. The psychological aspect of personal identity is constituted by the degrees of similarity between two temporally separate selves with regard to a person's personality, belief structure and desires, which may in certain cases, depending on the degree of similarity and continuity, move from being intra-personal to being inter-personal. In line with this argument, if psychological continuity were to become so deeply disturbed that someone became 'another person' (e.g. based on perceived changes in personality due to advanced dementia), would this mean that the advance directive should have no more moral force in relation to that person than it would have, had it been written by a stranger, friend or relative? According to Dresser (1986), a person with dementia may lose the 'properties' necessary for retaining personal identity over time, thereby becoming a different moral person. This claim relies on the assumption that people with dementia could lose so much mental capacity that their past preferences and interests no longer seem attributable to them (Dresser 1995, Buchanan and Brock 1989). This claim, that dementia causes a disruption in identity, draws on Parfit's theory of identity. Based on the assumption that the writer of the advance directive and the person with dementia are different people, it is argued that there is no reason to assume that the directive has any moral authority over the person with dementia.

Dworkin (1994), on the other hand, argues in favour of respecting advance directives. He views autonomy as a reflection of a person's integrity, whereby people express their character through the lives they lead based on values, commitments, convictions and critical as well as experiential interests. Critical interests include the kinds of things that give meaning to people's lives and make a person think, had it not been so, their life would have been worse or wasted. Experiential interests cover things that people do simply because they enjoy doing them. Dworkin's view would seem to imply that having dementia at a particular stage in a person's life, would just be one stage in a complete life which has already involved different stages. The stage someone is currently in, is affected by interests and concerns which transcend that stage and are important for their life as a whole. The competent and incompetent selves are thus one and the same person. It could therefore be concluded that advance directives should be respected because they are expressions of the critical interests a person has, and that they should therefore take precedence over current experiential interests. However, Jongsma (2016), whilst not rejecting the idea that advance directives should be respected, argues that research participation is not a critical interest for most people and that advance directives for research would be better understood quite simply as 'declarations of willingness'. Moreover, in keeping with ethically sound research practices, any clear objection to the initiation or continuation of a research procedure should result in the withdrawal of a research participant (Jongsma and van de Vathorst 2015). Consequently, aside from precedent autonomy, researchers also have an obligation to protect people with diminished capacities from undue harm and burden.

Porteri (2018) interprets claims about the disruption of psychological continuity and personal identity as implying that when people with dementia become incompetent, they may become a different and a new person, and that the person they used to be ceases to exist. She points out that potential changes in personality, in beliefs and interests, as a consequence of dementia, are feared precisely because people feel that these changes will affect themselves and not that they will give rise to a different and new person. People with dementia, at whatever stage of the disease, are still recognised as mothers, fathers, partners, sisters, brothers and friends. Relatives and friends have a personal commitment to them and feel that they have responsibilities and duties towards them not just because they are part of the larger human family, but because they are the very same person they used to be (Porteri 2018). Moreover, taking the identity argument to the extreme, if the person is no longer the same person, then the relatives of that person should theoretically have no say in matters relating to him or her. Porteri concludes that body is an important concept in that as long as the body is there, so is the person, adding that bodily continuity as a criterion for considering the person with dementia as the same person better guarantees respect for that person's rights and wellbeing prior to and after loss of capacity (in this context in relation to participation in research).

The legal status of advance directives for research in Europe (at least for those covering biomedical research) remains unclear. In 2001, the Clinical Trials Directive, to be replaced eventually by the new Regulation on Clinical Trials when it comes into force, makes reference to "incapacitated adults who have not given or not refused informed consent before the onset of their incapacity". This reference is of relevance to whether additional measures of protection are considered necessary, without there being any further elaboration of the concept of advance directives (Jongsma 2016). The current European legal framework regarding the use of advance directives for research in the context of biomedical research neither explicitly mentions the possibility of their use not forbids it (Andorno et al. 2016). This ambiguity/lack of clarity may fuel unease and uncertainty surrounding the use of advance directives as the sole instrument by which researchers might determine whether or not people with dementia who lack the capacity to consent and have no proxy can participate in research. Finally, the desire to participate in most clinical trials is dependent on participants having a study partner (usually a partner, adult child or close friend). Consequently, without the agreement of a close relative or friend, a person with dementia having expressed the wish to participate in research in an advance directive for research may be prevented from doing so.

Consenting to future research in an advance directive is also problematic because, as pointed out by Berghmans (1998), it is difficult to give consent for a future experiment which has not yet been devised and which, by the very nature of research, is likely to be innovative. Researchers may find it easier to accept a negative advance directive for research as this does not raise issues about the ambiguity surrounding the risks and burdens people might be exposing themselves to. The risks of research as well as the potential burdens and benefits can only ever be estimated but in the case of decisions made for research that might occur several years later, procedures and methods may have advanced beyond what was initially imaginable. The 'informed' aspect of consent is therefore missing in the case of advance directives for research. However, people can be informed about and helped to understand the implications of this lack of prior knowledge and then make as informed a decision as possible. Those who have a trusted person could be encouraged to appoint and allow the involvement of a proxy (see next sub-section) in future decisions about participation in research.

Recommendations for researchers

- Involve proxy decision makers, if they have been granted that power, in determining whether the research that is eventually proposed is in line with the wishes expressed in the advance directive.
- Do not resort to the use of an advance directive for research if the person with dementia currently has the capacity to consent or refuse to participate in research.
- Consider the current wishes of a person with dementia with regard to continuing with or withdrawing from a study (in terms of indications of assent or dissent). Clear signs of the wish to withdraw from a study should be respected even if the person's current participation is based on a valid advance directive.
- Reflect on personal beliefs about personhood and how this relates to advance decision making.

Recommendations for research ethics committees

- Recommend that researchers consider previously expressed wishes and preferences when proposing participation in research.
- Accept preferences expressed in an advance directive as a valid expression of interest in participating in the research.

Summary

Advance directives for research provide another way for people to express their wishes about future participation in research, in most cases drawn up before they develop dementia. Ethical concerns around advance directives for research tend to focus on issues related to personal identity, changing interests and that fact that the requirement for people to make 'informed' decisions is problematic because relevant and necessary information about particular studies is not available when the directive is drawn up. The notions of a loss of self or personhood and of a complete change of identity, as obstacles to the use of advance directives for research, are rejected by the ethics working group. They emphasise the need to respect any current signs of objection to the initiation or continuation of a research procedure and the importance of involving people who are authorised to do so in determining whether proposed research is in line with the wishes expressed in the advance directive.

Part 3: Ethical Challenges during Participation in Research: promoting wellbeing and avoiding harm

Much of the discussion paper so far has focused on how to attract the interest of a diverse set of people with dementia to research and how to go about ensuring a fair and just selection of those people in the context of research. The primary goal of research is not to benefit participants but researchers must consider participants' wellbeing and not focus on acquiring valuable data at all costs. Also, once people with dementia have agreed to participate in research, it is important that researchers ensure that they are treated with respect and due consideration for their wellbeing. Furthermore, researchers can be considered as having a duty of care towards their participants, which includes taking necessary measures to avoid them coming to any harm.

▶ ***"When I participate in a research project, please explain what the research is about in clear words and give me a printed version in simple language. Make it easy for me to understand the purpose and aims of this study, and in what way I can contribute. On the one hand, I want to be treated like any other person but on the other hand, I need assistance to participate. I want to be seen holistically as a person with special needs but also as a person who is capable, creative and responsible. To feel well despite my disease, I need empathy, respect, kindness and the opportunity to be heard and understood"*** **(Angela, person with dementia, Austria).**

The fact that research participants provided informed consent does not release researchers from the moral obligation to take measures to try to ensure that they have as positive and safe an experience as possible. Researchers' responsibilities can be perceived not merely as negative obligations (i.e. to refrain from causing them harm) but also in terms of positive obligations towards research participants (Richardson 2008). In some situations, it may be best to take concrete action to promote participants' wellbeing and protect them from harm, whereas in others it may be best to refrain from doing something[53].

Our interest in Part 3 of this discussion paper is on the wellbeing and safety of people with dementia whilst participating in research, and on any lasting impact of research on wellbeing. People who participate in research are voluntarily contributing to society and irrespective of the importance that they assign to their own wellbeing and safety, researchers have a duty and responsibility to promote their wellbeing and protect them from harm.

Promoting wellbeing and minimising the risk of harm

A positive and safe environment for participants

Perceptions of wellbeing are socially constructed and there may differences from one person and cultural group to the next with regard to how wellbeing is interpreted and the importance attached to it. Wellbeing is a fairly broad concept as the following quote from Naci and Ioannidis (2015) suggests.

> *"Wellness refers to diverse and interconnected dimensions of physical, mental, and social well-being that extend beyond the traditional definition of health. It includes choices and activities aimed at achieving physical vitality, mental alacrity, social satisfaction, a sense of accomplishment, and personal fulfilment."*

When people with dementia volunteer to participate in research, they often do not know exactly what to expect. Many will be stepping outside their usual comfort zones, going to unfamiliar places and interacting with people they don't know.

Researchers should treat participants with dementia with the same level of respect as that afforded to other people (e.g. being polite, showing acceptance and behaving in an appropriate and courteous manner), take necessary measures to

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[53] Some of the issues, especially in relation to wellbeing, will also be relevant to people with dementia contributing to Public Involvement activities.

ensure continuing respect for participants' right to self-determination (see sub-section on ongoing consent and the right to withdraw) but in addition, provide all necessary support linked to the specific needs that participants may have as a result of having dementia. Some challenges and needs, for which adaptations and flexibility may be required, may be directly linked to cognitive impairments associated with dementia (e.g. related to memory, attention span, handling information, logical thinking, orientation in time and space, calculating, writing, reading and communicating). However, drawing on the concept of disability, the extent to which such impairments become barriers to participation in research also depends on procedures, attitudes and practices within society and to lack of reasonable accommodation. Careful consideration of how, when and where study visits are planned, the adaptation of tools, instruments and activities, the provision of additional time, the use of aids and props to facilitate communication and promote understanding, and the development of creative methodologies are all important issues to consider in order to promote the wellbeing of people with dementia involved in research.

It is important that researchers take into account physical, emotional, relational and interpersonal factors in the context of multiple and complex identities and challenges encountered by a diverse set of participants with dementia. Researchers may need to develop their cultural awareness and sensitivity (knowledge and acceptance of people from different cultures and of cultural differences and similarities between people) and cultural competence (knowledge combined with appropriate attitudes and skills (e.g. openness, respect and acceptance). The acquisition of cultural competence requires some degree of self-reflection (e.g. awareness of one's own background and a readiness to question one's own assumptions) and is therefore an important aspect of reflexivity and positioning (see sub-section on this topic). Many researchers would benefit from involving a diverse set of people with dementia in PI and from contacting representatives from different communities so as to ensure that the positive environment they seek to create for research participants is inclusive and responds to the needs, wishes and challenges of a broad group of people with dementia.

The research environment is made up of researchers with different beliefs and attitudes, possessing different virtues which may also impact on participants' wellbeing. Velasquez, Andre, Shanks and Meyer (1988), like other theorists of the ethics of virtues, argue that the fundamental question should not always be "what should I do?" but "what kind of person should I be?" They suggest that virtues such as honesty, compassion, generosity, fidelity, integrity and fairness, to name but a few (which are also attitudes, dispositions and character traits) can be learned, practiced and developed. This is something that happens through interaction with others within a community and may eventually result in a predisposition to act in ways which reflect those virtues. Certain virtues are likely to be considered more important than others in different cultures and in different contexts, hence the importance of cultural awareness and sensitivity.

- ☞ **Do you consider acceptance of diversity and a desire to promote inclusion as virtues?**
- ☞ **Are there other characteristics you would consider as virtues which would be important for researchers to have or cultivate?**

The wellbeing of informal carers and supporters

The focus of this discussion paper is on people with dementia but for many people with dementia involved in research, their participation can have an impact on relatives and close friends who support them. Informal carers and supporters may sometimes find themselves with responsibilities they did not necessarily choose (e.g. ensuring that the person with dementia attends study visits, has read and understood relevant documentation and complies with research procedures). Some of them may have specific needs (e.g. linked to language, literacy or mobility etc.), they may have competing obligations (e.g. professional, family and personal responsibilities) and some may lack trust in researchers (especially if from a marginalised group) or simply prefer to spend their time on other things. In cases where study partners are required for research, this not only excludes people with dementia who don't have one but also risks putting pressure on informal carers and supporters who are not able or do not wish to get involved. Such pressure and the impact that this can have on people's lives, wellbeing and relationships should be considered and alternatives found. Informal carers and supporters should not be taken for granted as without them, much research involving people with dementia would not happen. Neither should they be ignored or denied the possibility to support the person with dementia. Terminology such as relatives, families and friends may sometimes exclude people from the LGBTI community, who may not relate to such terms which are not consistent with that of the 'family of choice'. Similarly, for some people with intellectual disability long-term social care support staff may be more of a constant in their lives than family.

An area of particular concern is that of interpretation. This issue has been discussed in relation to the diagnosis of dementia and assessment for services (Alzheimer Europe 2018) but it is also relevant to participation in research (e.g. in relation to informed consent, interviews and focus group discussions and completing outcomes measures). There are pros and cons to the use of relatives as interpreters. On the positive side, relatives are often familiar with the person's situation and better able to understand what the person is trying to communicate, they are in many cases trusted (i.e. they will not reveal what the person says outside of the family) and in some cases may be familiar with a particular language or dialect that is not spoken by many people. Their involvement may be essential to the wellbeing of a person with dementia and to their participation in a study. There are, however, some issues to consider in relation to the wellbeing of all concerned. The involvement of relatives may mean overriding the general principles of respect for privacy and confidentiality. Younger people (e.g. second or third generation migrants who sometimes have better language skills than their older relatives) may be uncomfortable with certain issues (e.g. sensitive topics addressed in qualitative research or medical details about the participant). Some people with dementia might also be uneasy talking about issues which could call into question their standing within the family. It could be argued that the use of relatives and friends for interpretation in research increases the risk of obtaining unreliable data.[54]

The prevention of harm

A safe environment also means a place or situation in which people with dementia will not be harmed. Some tests that research participants undergo can be experienced as burdensome, stressful, uncomfortable and even painful. Research participants must be informed of these risks as part of the informed consent process. However, in cases where proxy consent has been provided or consent has been provided in an advance directive, the participant might not currently understand what is being asked of them and why. In addition, pain and discomfort are to some extent subjective. What bothers or is painful to one person, may be experienced quite differently by someone else. The following table provides examples of a few issues linked to various research activities which may result in some people with dementia having a less than positive experience of participating in research.

Action	Potentially negative consequences/experience
Doing tests of performance (e.g. doing a paper and pencil test or a computer test)	• Performance anxiety (more so in the case of people with low levels of education or difficulties with language and literacy). • Loss of self-esteem or concerns about deterioration of condition because of known or imagined score. • Culturally biased questions. Feel discriminated against.
Being involved in a focus group discussion	• Feeling of not having sufficiently contributed. • Feeling of not having been equally valued by the researchers. • Feeling of not being sufficiently knowledgeable or eloquent. • Concerns about having disappointed the researchers.
Having a scan	• Claustrophobic feeling. • Being disturbed by the noise. • Having a complication or lasting side effects.
Having a spinal tap	• Painful or unpleasant.
Giving a blood sample	• Painful or unpleasant.
Being interviewed	• Feeling intimidated by highly educated researchers. • Concerns about performance in relation to other people who were interviewed. • Concerns that other people will find out what was said (e.g. maybe was too critical about someone). • Revival of memories of unpleasant things from the past. • Unease linked to the discussion of sensitive topics.
Survey	• Difficulty understanding some of the questions. • Fear of making mistakes, of letting the researchers down or of not responding in a way that the participant thinks the researcher is looking for. • Arousal of unpleasant emotions and feelings such as anger, frustration and inadequacy.
Observation	• Concerns about who has access to the information. • Concerns about doing or saying something embarrassing. • Feeling exhausted. No break from observation, no privacy.

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[54] For a discussion about issues related to the use of relatives and close friends for interpretation, please see Alzheimer Europe's report on intercultural care and support (2018).

Information provided about what is involved (in terms of the procedure and known risks) is not necessarily sufficient preparation or protection from harm. People may, for example, evaluate whether or not a risk is acceptable on the basis of information provided but burden, has an experiential component (Jongsma and de Vathorst 2015). People do not necessarily know how they will feel about a particular intervention or procedure until they have experienced it, particularly in the case of repeated invasive procedures like blood tests but also for non-medical procedures which some people might find disturbing (e.g. in-depth interviews on sensitive issues). Having dementia may also alter someone's experience of certain research procedures because of increased difficulties with language, communication and comprehension. These may be further compounded for people with low levels of education, additional difficulties with language and literacy or from communities which are typically marginalised. A person might, for example, not fully understand or remember what the procedure involves, be unable to express his/her concerns or ask questions, or feel confused, disorientated or overwhelmed. For these reasons, it is vitally important that researchers take an interest in how participants experience what they are asked to do, that researchers provide appropriate support and consideration and that they are attentive to whether participants are happy to continue or wish to end their participation in their research.

With regard to the duty of care mentioned earlier, it is generally accepted that researchers should take precautions to ensure that their participants do not come to any harm during their study, but also to take any necessary measures if they discover that participants in their study are, for example, currently being abused, exploited or suffering from neglect. In some qualitative studies which address sensitive topics and where a relationship of trust is built up over time, interactions may sometimes be mistaken by participants as having a therapeutic quality (i.e. researchers being committed to improving their personal health or situation rather than seeking data to answer a particular research question). Some researchers, especially in the context of advocacy research, argue that researchers have a personal responsibility to resolve such issues. Others argue that it is not the role of researchers to intervene directly in matters for which they lack the necessary expertise and could do more harm than good, and that their role should be limited to alerting relevant professionals. Responses to unethical practice should also be in line with regulatory bodies where appropriate (e.g. in the United Kingdom, this would be the Nursing and Midwifery Council when conducting research with National Health Service patients).

Recommendations for researchers

- Be sensitive to and respect different interests and needs of participants.
- Recognise the equal value of different types and levels of contribution towards research.
- Build on advice obtained from PI and from relevant gatekeepers about how to create a supportive, respectful and culturally appropriate environment and manner of interacting with people with dementia from diverse backgrounds. Be aware of expectations surrounding participation in research (including the management of hopes, fears and possible disappointment in relation to people with dementia, carers and the wider network).
- Be attentive to possible perceptions of a power differential and of actual differences in power (interpersonal and structural).
- Ensure that you have sufficient experience conducting research with people with dementia and if not, seek the support of researchers who have.
- If people with dementia are unable to communicate their wishes, consult informal carers/supporters in order to gain a better understanding of what would promote the wellbeing and respect the known wishes of the former with regard to their participation in research.
- Ensure that procedures and interventions are not unnecessarily or over burdensome and ask for feedback about levels of burden during and after a study.
- Ensure that sensitive topics are approached with sensitivity and care.
- Ensure that participants with dementia have someone they can contact after the study should they need support or have any questions.
- If unsure about where the boundaries lie with regard to intervention in issues related to the wellbeing of research participants but not directly linked to the research, check with your ethics committee.

Recommendations for research ethics committees

- Ensure that researchers provide details of provisions to promote wellbeing and protect participants from harm.

Summary

Measures to promote wellbeing, respect autonomy, treat people with dementia with respect, and value cultural differences and diversity must continue throughout the whole research process. The aim should be to ensure that participants have a positive experience in return for their personal investment in research and that they are not harmed. This may also contribute towards the retention of participants which is beneficial to the research process. Researchers need to bear in mind that the same research procedure may be experienced differently by different participants and that pain and discomfort are to some extent subjective. The wellbeing of informal carers and supporters should also be considered because not only are they important in their own right but the continued participation of people with dementia in research is often dependent on their continued support.

The ongoing right to self-determination

Regardless of how informed consent was initially provided (e.g. by the participant, by a proxy or in an advance directive), it is important, in terms of respect for autonomy, beneficence and non-maleficence, that informed consent is perceived as a continuous process. The concept of ongoing consent, whereby consent is sought at regular intervals throughout the study, is becoming increasingly common.

Confirmation of the wish to participate in research

The concept of ongoing consent (including indications of assent) when conducting research with people with dementia is particularly important because:

- participants with dementia who initially provided informed consent themselves may at some point forget that they did so,
- participants with dementia may not have provided consent themselves prior to the actual study (as consent was either provided by a proxy or by means of an advanced directive some time ago), and
- participants with dementia might be confused and not realise that they are currently involved in research and/or that they are entitled to withdraw at any time.

The process of ongoing consent provides an opportunity for researchers to verify that participants with dementia still have the capacity to consent to continued participation. Where this is not the case, it provides an opportunity to reflect on whether and if so how, this affects their continued participation and the possible need for additional protective measures. It also provides a regular opportunity to discuss with participants how they feel about various procedures and interventions that they have undergone or that are planned.

The concept of 'process consent', which was mentioned earlier, involves re-visiting and re-establishing consent regularly throughout the study with a particular focus on residual capacity and on adapting methods to correspond to participants' individual abilities (not necessarily cognitive) and their preferred ways of receiving information (Dewing 2008). This might, for example, involve the use of modified, simplified forms, key words with or without pictures, web-based information, images, photosymbols, 'talking mats', videos to contextualise the information, extending the evaluation of consent over an extended period and judging consent on the basis of how a person responds and the feelings they express.

Dewing (2008) emphasises that process consent is meant for use with persons with extremely limited capacity who would generally be thought to be lacking competence (i.e. the legal capacity to consent) but that it can also be used in conjunction with other forms of consent, including proxy consent, as a means to include people with dementia in research. However, greater attention needs to be paid to how to maximise the potential of a broad range of people to consent to participate in research. The level of capacity is not the only factor interfering with the ability of people with dementia to consent. Various supports and approaches, including the establishment of a trusted relationship and environment, also need to be culturally appropriate and to correspond to the needs and abilities of people with dementia from various marginalised groups. Dewing (2008) describes the first step of process consent as consisting of establishing basic biographical knowledge of the person and having some 'clues' about how each person presents when in a state of relative wellbeing, so as to be able to approach that person for ongoing consent in a favourable context/environment and to build up a relationship of trust.

It is unclear to what extent such an approach would be feasible or accepted by research ethics committee for some types of research (e.g. for randomised controlled trials or in studies involving large numbers of participants). Nevertheless, the general concept of ongoing consent can and is applied in a wide range of research projects in the form of re-assessing informed consent or verifying assent at various stages of the research process, thereby ensuring that the principle of voluntariness is still applicable and that people with dementia can express their self-determination.

The continued right to withdraw

The continued right to withdraw from research is an integral part of the ongoing consent process. Whereas participants need capacity to consent, they do not need capacity to withdraw from research. This can be done at any point and participants should not be asked to justify their reasons for withdrawal. This does not prevent researchers from discussing withdrawal with participants (mainly in case of research that has a high potential to benefit participants) so as to ensure that withdrawal from the study is not based on misunderstandings or on contingent reasons that could be overcome. People with dementia may, however, at some point lack the ability to communicate their desire to withdraw, as mentioned above not understand that they have the choice, and even not be aware that participation is causing them distress. For this reason, it is important that researchers are attentive to signs of distress, discomfort or harm throughout the whole research process, during which participants' ability to understand and communicate may deteriorate.

Apart from obvious signs of resistance such as turning away, verbal and emotional expressions and non-verbal indications of the desire to withdraw, there may also be very subtle signs which are difficult to interpret, especially in the light of the multiple intersecting identities of both participants and researchers. For this reason, informal carers and supporters may have an important role to play in helping researchers to identify relevant signs of distress, harm or the desire to withdraw.

Recommendations for researchers

- Consider creative, person-centred approaches to ongoing consent which recognise and understand the needs of a diverse group of people with dementia.
- Be attentive to possible resistance from participants with dementia (gestures, body language, facial expression, emotional expression and verbal indication).
- Try to gain cultural awareness so as to recognise such signs from people from different cultures and with diverse characteristics.
- Consult informal carers about possible signs of dissent of people with dementia who are unable to consent. Do not proceed with an intervention in case of doubt.
- Ensure that approaches and attitudes to ongoing consent and withdrawal are culturally appropriate.
- Do not make withdrawal dependent on proof of capacity (as in consent); it should be possible at any time and with no justification necessary.
- Be aware that people with dementia may forget that they are involved in research and not understand that they have a choice to refuse a particular intervention or act.

Recommendations for research ethics committees

- Ask researchers to describe the measures that they will take to verify whether participants wish to continue or withdraw from the study after initial informed consent has been given.

Recommendations for funders

- Recognise the value of small-scale studies where priority is given to seldom heard voices and where capacity may fluctuate.

Reflexivity and positionality

The importance of self-reflection

When striving for more inclusive research, the concepts of reflexivity and positionality are particularly relevant. Reflexivity consists of a continuing process of self-scrutiny on the part of researchers through which they attempt to acquire a greater awareness of the relationship between themselves and research participants. Positionality is about where researchers stand in relation to the people with and on whom they are conducting research.

Through reflexivity and positionality, researchers reflect on the personal factors which may in some way affect their choice of research topic and appropriate methodology, how they analyse their findings, which findings they emphasise and how they frame and communicate their conclusions (Malterud 2001). Reflexivity at a more structural level is also important. This involves reflecting, for example, on the social position of the researcher or on what the researcher represents (e.g. structures like universities or hospitals). Researchers need to be aware of their own degree of power and privilege compared to that of people with dementia involved in PI or as research participants. They need to reflect on how these possible different positions influence their collaboration and the information or data, and on how they can reduce the power gap in this collaboration. In other words, is this research or collaboration a reproduction of structural lines of oppression or have steps been taken to move towards inclusive research?

This involves researchers paying attention to how their perceptions as well as cognitive, theoretical, linguistic, political and cultural characteristics or circumstances, as well as structural factors, might affect their interpretations (Alvesson and Sköldberg 2009). There is no single accepted procedure for reflexivity but it typically consists of reflecting on one's own background, history and other relevant factors, as well as writing memos of one's thoughts and observations about the data collection and analysis.

Reflexivity is usually associated with qualitative research methods in that it is grounded in the belief that researchers can never be totally separate/detached from their research participants. Some quantitative researchers, influenced by the positivist paradigm, would be more inclined to consider self-reflection within the research process as too subjective, and as a problem which with the right research design could hopefully be eradicated (Newton 2009). However, according to Lakew (2016), attention to subjectivity may actually increase the likelihood of achieving objectivity by obtaining a more complete and inclusive account of 'reality'.

> *"...models do not build themselves any more than they interpret themselves [...] choices are still to be made, and these are frequently based on intuitions, hunches and ideas of what is needed that have not yet been fully rationalized" (Greiffenhagen, Mair and Sharrock 2011, p.103).*

Positionality is closely linked to reflexivity in that it rests on the assumption that people have multiple overlapping identities and that a person's position in society, including their cultural background as well as a range of factors and characteristics which shape and reflect their identities, affects the way they make sense of the world. In the past, it was often assumed that a researcher was either an insider or an outsider (with regard to different groups of potential participants) and that there were certain advantages and disadvantages to being either. Drawing on critical and feminist theory, postmodernism, multiculturalism and participatory and action research, insider/outsider issues are increasingly being reframed in terms of one's positionality with regard to ethnicity, class, gender, culture and other factors. It is claimed that this is a better approach to understanding the dynamics of research within and across different cultures and sub-groups (Merriam, Johnson-Bailey, Lee, Kee, Ntseane and Muhamad 2001). Aguilar (1981, p.25) emphasises the internal variation and multiplicity of social and cultural characteristics in all cultures and sub-cultures and asks, "What is it that an insider is insider of?" This suggests that there are different levels of insider and outsider status which may be different at different times (Merriam et al. 2001).

Lack of awareness of one's positionality may affect how certain things are understood, research priorities, sampling strategies and the analysis of results, not only conclusions that are drawn but which specific issues are analysed in the first place. Researchers who fail to address the concepts of reflexivity and positionality may end up with a selective perception, miss intricacies (which fall outside the realm of their experience) and fail to understand the meanings that research participants and people contributing to PI give to their lives.

Knowledge claims and objectivity

The issue of what counts as knowledge represents a fundamental question underlying various research paradigms and is also relevant to the concept of reflexivity. Corbin and Strauss (2008) suggest that knowledge arises through the action and interaction of self-reflective beings, with past memories and recollections entering directly or indirectly into actions. Drawing on a pragmatic approach to the acquisition of knowledge, they suggest that

> *"....acts of knowing embody perspectives. Thus, what is discovered about 'reality' cannot be divorced from the operative perspective of the knower, which enters silently into his or her search for, and ultimate conclusions about, some event" (Corbin and Strauss 2008, p.4).*

There may also be differences within a similar methodological approach. Kvale and Brinkmann (2009) describe a key difference of relevance to the issue of reflexivity, in the context of different philosophical approaches to qualitative interviewing, through the metaphors of the miner and the traveller. The miner metaphor describes knowledge as being like a buried valuable metal which the miner unearths without contaminating it in any way. It is there, intact, waiting to be discovered. The traveller metaphor describes knowledge as something that is constructed as a result of the traveller walking along with people, listening to their experience of the world, trying to make sense of it and retelling it on his or her return.

The focus of the positivist paradigm was initially on the discovery of an 'absolute truth' which could be objectively observed and measured, and on the need to avoid 'contamination' of the data by the researcher (i.e. resulting from attitudes, beliefs or behaviour). It is now widely accepted that researcher reactivity (i.e. the possibility that the researcher might influence the research situation or the participants) and total neutrality or objectivity (in the sense of freedom from bias) cannot be completely ruled out. Researchers may strive for objectivity but people can never be truly divorced from their own subjectivity.

> *"We can strive to remain objective, but must be ever mindful of our subjectivities. Such is positionality. We have to acknowledge who we are as individuals, and as members of groups, and as resting in and moving within social positions" (Bourke 2014, p.3).*

Ritchie and Lewis (2009) suggest that reflexivity is actually important when striving for objectivity and neutrality. It provides a way to reflect on different ways in which bias might occur and can also be useful when seeking to understand and portray the perspectives of research participants.

Power relations and matching

In some feminist approaches to interviewing, reflexivity and positionality are incorporated into the data collection process, which is perceived as a reflective and interactive activity that is non-hierarchical and avoids objectifying the participant. Such approaches reflect a more collaborative approach to research whereby researchers are encouraged to step outside the traditional formal and neutral role, and to share information about themselves. This can also be perceived as a form of reciprocity (Ritchie and Lewis 2009).

It has been suggested that gender matching or matching based on social class, ethnic group or other key socio-demographic characteristics may help provide greater insight into participants' experience, help address perceived power imbalances and encourage people from such groups to contribute to research (Ritchie and Lewis 2009). Self-reflection on this issue may lead to decisions about whether or not to interview people with different key characteristics to those of the researchers (e.g. whether female researchers should interview women in a particular study or whether researchers should be matched with participants from the same minority group, who speak the same language or with similar key socio-demographic criteria). Sometimes, having certain characteristics in common (e.g. based on ethnicity, gender, disability or age) may mean that researchers have experienced similar forms of oppression and unequal power relations. The perception of an unequal relationship of power in the context of a study would probably not be conducive to open discussion, particularly if issues of oppression or discrimination were highly relevant to the research questions (Ritchie and Lewis 2009). However, as pointed out by Berdai-Chaouni, Claeys and De Donder (2018), presumed proximity, based for example on having a shared ethnicity, may result in overlooking other factors such as social class, which create a distance from the research participants and in failing to seek other ways of challenging barriers.

Reflection and subsequent decisions about possible matching need to be made very carefully to avoid making assumptions and researchers limiting themselves to only conducting research with certain participants. It is important to bear in mind here that there may be power imbalances within certain groups (e.g. based on class, gender, religion, language spoken and ancestry) and that consequently, it will not always be considered as a disadvantage for a researcher to conduct research with people from a group with which he/she does not personally identify.

> *"Can Whites study Blacks? Straights study gays? The colonized study the colonizer? (…/…) More recent discussions of insider/outsider status have unveiled the complexity inherent in either status and have acknowledged that the boundaries between the two positions are not all that clearly delineated. In the real world of data collection, there is a good bit of slippage and fluidity between these two states" (Merriam et al. 2001, p.405).*

Similarly, generalisations about researchers with certain characteristics being either suited or not suited to conduct research with certain groups of participants would amount to negative stereotyping, prejudice and discrimination. Participants may also be inclined to assume that a matched researcher understands certain issues or customs (i.e. wanting to avoid stating the obvious, based on an assumed understanding), and therefore not describe their experience in detail. For some participants, the matched criteria may not be central to their self-identity and some participants may feel more comfortable discussing certain issues with an 'outsider'.

Self-reflection, with regard to key socio-demographic characteristics, should therefore aim for an increased awareness of one's own characteristics and how these might be perceived by and have an impact on participants, but without this leading to actions based on generalisations. Ritchie and Lewis (2009) conclude that researchers need to assess the potential risks of cultural collusion versus unhelpful power dynamics. This might result in greater sensitivity to the needs and wellbeing of participants, measures being taken to promote trust and perhaps seeking support from fellow researchers, members of the communities or groups in question or gatekeepers. However, being a member of a particular community does not automatically make someone an expert on the lives of everyone from that group or grant them immediate access to the thoughts and feelings of people from that group (Shaw et al. 2019).

Assumptions, beliefs and attitudes

Researchers, like everyone else in society, have a wide range of beliefs and assumptions about various issues which may impact on their attitudes and behaviour towards people with dementia. This could in turn have an impact on how they conduct research and on the experience that people with dementia have of PI and of participating in research. Areas which would be particularly useful to reflect on include perceptions of dementia, personhood and vulnerability.

Dementia

Reflexivity and positioning enables researchers to become aware of their beliefs and assumptions about dementia and people with dementia, of how these may differ to those held by the people who may contribute to their research and to be attentive and respectful of other ways of making sense of dementia.

There may be considerable variation amongst researchers, research ethics committees and funders with regard to the extent to which dementia is perceived in a holistic[55] as opposed to a strictly biomedical manner. Literature on lay people's perceptions of dementia reveal, in addition, beliefs about the origin of dementia, such as it being a natural part of ageing, a divine test or punishment, something that is contagious, a mental disorder (that runs in families) or a result of lack of activity or of neglect by families (Mukadam, Cooper and Livingston 2011, Alzheimer Europe 2013, Parveen et al. 2017). In the context of research, perceptions and beliefs about dementia may affect the opportunities that are offered to and taken up by people with dementia, the choice of research questions which are deemed worthwhile, the methodologies considered possible and appropriate, and the way that people with dementia are treated, which may in turn have an impact on their wellbeing and involvement in research.

Perceptions are often expressed through words and may be associated with images, including metaphors. Words and images can be powerful and affect the way that people think about the concepts and people to which they apply. George (2010) describes this in terms of a moral challenge linked to semantic choice, whereby subtle alterations in the way that people talk about certain conditions may contribute to more humane approaches to them. The term 'demented', for example, is still used by some researchers (who often have a more medical background) despite frequent objections from people with dementia. Such objections are linked to the term being considered insulting due to its association in Latin with being 'out of one's mind' and in some countries, the term being used in a pejorative manner in much the same way as terms such as 'crazy' or 'mad'. Similarly, some French and Dutch terms (e.g. dément, Alzheimérien and dementerende) are nouns which denote what a person "is". This is similar to the practice of calling people with schizophrenia "schizophrenics" in that it reduces a person to a disease state. Referring to people with dementia as patients may make sense in clinical research settings but outside of that context may sometimes be perceived as a focus on the disease and not the person.

Personhood

Perceptions of personhood were mentioned earlier in relation to advance directives with regard to the moral justification to respect them. They are also important with regard to respect, dignity, wellbeing and equity.

For some people, personhood is dependent on the possession of certain capacities. Others would argue that personhood more or less equates with simply being a human being, whereas the role of interaction has also been highlighted, as has the importance of the physical body. Kitwood (1997, p.8), for example, described personhood as "a standing or status that is bestowed upon one human being, by others, in the context of relationship and social being". With regard to perceptions of personhood which are linked to the possession of certain cognitive capacities, including the capacity for logical reasoning and memory, Post (2006) criticises what he calls a hyper-cognitive approach which risks denying some people 'person status'. He also describes the belief that some people have of a kind of "non-material soul that still exists intact underneath all the neurological losses of dementia" (Post 2006, p.231). However, some people believe that this unique inner essence continues beyond human life (e.g. through reincarnation or an afterlife), whereas for others, it is something that can be lost.

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[55] For example, reflecting a more bio-psycho-social model proposed by Kitwood, including the perception of dementia as a disability.

> *"But you have to remember, that body is not who the person is. The person is gone."*
>
> *"There's no sense is asking her.... She's not there..." (Dunham and Cannon 2008, p. 49).*

Perceptions of the loss of a human essence are often associated with terms and images such as "off/away with the fairies", "lights on, no-one home", an "empty shell", "not being there", "sitting there like vegetables", the person just being "a body", "vegetables in a parallel world", a "speechless shadow" (Devlin et al. 2007, Dunham and Cannon 2008, Piehl 2009, Swane 1996) and with metaphors evoking monster-like characteristics, a ghostly disappearing self, a living death, natural floods and disaster, fights and crusades, and epidemics. These metaphors, along with others such as a return to childhood, challenge the notion of individuality, human dignity and personhood and disregard people's individual life course and their values, thereby promoting inadequate understandings of dementia and hence also influencing attitudes and behaviour towards them in ways that are problematic (Jongsma & Schweda 2018).

The changed behaviour and appearance of the person are emphasised at the expense of their personality, character, individuality and identity. Some people may find such metaphors helpful as a means to avoid blaming the person with dementia. Perceiving them as something different may serve to lessen their perceived responsibility for behaving in a certain way and enable carers to retain a positive image of them. However, it is unlikely that this would be helpful to people with dementia in terms of their being treated with consideration and respect. The use of metaphors may affect the way that people relate to others who have dementia and may result in negative stereotyping. Through metaphors, the disease can be given an identity (e.g. of a flood, a monster, an attacker etc.) which can be visualised and emotions projected onto it. It can be blamed, hated and avoided. Stereotypes and images based on metaphors can be fairly powerful and prevent people from recognising the individuality of each person with dementia and their personhood. Beliefs about personhood may have a significant impact on how researchers treat people with dementia. They may affect how researchers relate to people with dementia participating in their research, for example, whether they treat them as individuals with feelings, needs and a unique history or as objects to scrutinise and study. In the case of the latter, there is a risk of people with dementia being used as a means to an end, their dignity not being respected and their needs and physical and emotional well-being overlooked.

Not all researchers will have reflected on the issue of personhood but may nevertheless have beliefs about it which are based on taken-for-granted assumptions. In such cases, it may be difficult to know where to start and some background reading about various philosophical approaches may be useful in gaining self-awareness as well as a broader perspective on this issue.

Vulnerability

The issue of vulnerability was discussed earlier in relation to the informed consent procedure. However, it also has implications, more generally, for attitudes and behaviour towards people with dementia involved in research. Some definitions of vulnerability focus on people lacking characteristics or attributes (e.g. insufficient power, education, resources or strength) that are considered necessary to protect their interests (CIOMS 2002) or in a more political sense on people lacking "basic rights and liberties that make them particularly open to exploitation" (Zion 2002).

In the case of dementia, the main issues which might result in people with dementia being considered as vulnerable are age, cognitive impairment, being dependent on others for care, being a resident in a long-term care institution or having end-stage dementia. Additional factors may co-exist such as being a member of an ethnic minority group, gender, physical or intellectual disability, sexual orientation and living situation (e.g. being homeless or nomadic) (Alzheimer Europe 2011). Categories have also been proposed which focus on the characteristics or situations believed to render people vulnerable, such as cognitive, communicative, legal, deferential, institutional, linked to infrastructure, medical, economic and social (Kipnis 2001, National Bioethics Advisory Commission 2001).

However, as Levine et al. (2004) point out, not everyone who fits into any of these categories is vulnerable, and some people who do not fit into any of these categories could nevertheless be vulnerable in some way. Group-based approaches to vulnerability detract from the perception of all human beings as potentially vulnerable (as being capable of suffering), as well as of the vulnerability of people for reasons which are not immediately evident. Focusing on vulnerable groups may also contribute towards seeing certain groups of people as 'the problem' and overlooking similarities and differences between groups and individuals. For example, research participants all require some degree of protection, some people belong to more than one group and some members of a group may need additional protection with regard to certain issues but not others (DuBois 2008). There is a risk of stereotyping vulnerable research participants, devaluing them and this leading to unfair treatment or discrimination rather than appropriate protection of their rights.

An alternative non-group based approach to vulnerability involves assessing people for specific vulnerabilities and heightened risk. A specific condition or diagnosis (e.g. dementia) would alert researchers and justify them in assessing people with that condition for different types of vulnerability insofar as they relate to their study (DuBois 2008). The process of reflexivity and positioning could help researchers to recognise aspects of their own identity and situation for which they might be considered vulnerable, either currently or at some point in the future, how their position might affect someone with dementia who is considered as being vulnerable and the potential impact of their understanding of vulnerability on the research process and on the people with dementia contributing to their research.

Recommendations for researchers

- Take time to reflect on your own perceptions and beliefs with regard to dementia, people with dementia and the involvement of people with dementia as research participants (also in PI).
- Reflect on your positionality in relation to potential participants with dementia from diverse backgrounds and marginalised groups.
- Take necessary measures to act on the results of such reflection to improve the research process and/or the experience of participants.

Summary

The conduct of inclusive research calls for researchers to examine their own beliefs, perceptions and assumptions about people who may have very different characteristics, lives, histories and cultures to their own. Although researchers may realise that they have certain things in common with participants and may be respectful of possible differences, reflection is often needed to gain insight into how their own social position and personal characteristics might reflect or communicate differences of power and in some way impact on participants' wellbeing and on the research itself. The solution is not necessarily to match participants with researchers who have similar characteristics. There are advantages and disadvantages to this practice. Similarly, beliefs about the origin, cause and lived experience of dementia, as well as taken-for-granted assumptions about related concepts such as vulnerability and personhood, may affect how researchers behave towards people with dementia and decisions they make about research design and methodology. The concepts and practice of reflexivity and positionality are often associated with qualitative research but we encourage quantitative researchers to also consider their potential value (e.g. as a means to foster respectful relationships with research participants, avoid the potentially harmful impact of perceived power imbalances and reduce bias).

Part 4: Ethical Challenges Linked to Involvement after the end of research

Introduction

The involvement of people with dementia in research is not only important during research but also after it ends. People with dementia who contribute towards research through PI or as research participants should not be simply dropped or disregarded even if researchers need to move on to other studies. People have often invested a great deal of time and effort in research projects and may be interested in knowing what changed as a result of their investment. Regardless of the nature or level of their involvement, out of a sense of respect, gratitude and reciprocity, and in keeping with the relationship of trust hopefully already established, researchers should carefully plan how the involvement of people with dementia in a particular study ends. The aim should be to ensure that people with dementia feel appreciated, valued and hopefully motivated to repeat the experience.

Feeding back findings to relevant individuals and communities

In keeping with the principle of reciprocity, researchers have a moral duty to provide feedback to those who contributed to the research and to relevant lay communities (e.g. to the general public, minority communities for whom the topic was particularly relevant). As stated by Taylor (2019, p. 1),

> *"Research subjects contribute to the greater good and expose themselves to risk of harm. They have a right to know the outcome of the research. This is especially important for participants with a direct interest in the findings, for whom knowledge can be power."*

The ethical requirement to provide feedback is stated in the Declaration of Helsinki (World Medical Association 2013) but only in relation to medical research. In addition to reciprocity, which involves an element of gratitude and respect, involving people with dementia in the period following the end of the study also provides an opportunity to ensure that the findings are communicated in an accessible way to the people and communities for whom they are particularly relevant and who could benefit from them. Some findings could have a direct impact on people's lives and some findings could be used to influence policy and service development.

> *"…. social scientists do not have a mandate to guarantee justice, but we do have a responsibility to our research participants who have consented to let us into their worlds, to treat them with respect and understanding and to ensure that the results of our research are appropriately disseminated" (Shaw, Howe, Beazer and Carr 2019, p.14).*

Through effective and meaningful feedback, this may also raise awareness about the importance of research and encourage people to get involved. Measures to inform people with dementia about the results and significance of the findings can be planned and costed in advance and form part of dissemination activities. The role of PI continues here for example in supporting the development of easy-read or plain language findings that enable the sharing of findings and recommendations in an accessible manner. In large-scale research projects (e.g. European, cross-country studies), some members of the consortium may be allocated responsibility for this and have funds to accomplish this work. Such work may also be part of a broader initiative aimed at creating and maintaining interest in the research topic and work amongst the general public. In smaller scale research projects, principal investigators may need to do this themselves. Social media may support this process by increasing the impact and reach of work and positively influencing relevant communities. The obligation to provide feedback to participants should not be considered as being dependent on having dedicated or remaining funds.

▶ ***"We want researchers to come back and tell us the outcome of research that we have been involved in. Please be honest and don't bury 'bad' findings"*** **(Scottish Dementia Working Group 2014).**

Structural barriers

There may often be challenges to fulfilling the duty to provide feedback which appear to be structural. For example, researchers may have to account for the hours they spend on particular projects. If after-study communication/feedback was not accounted for in the initial budget (which they might not necessarily have influenced), they may have difficulty dedicating the necessary time to this important task. In such cases, it would be necessary to use their own time to fulfil this duty, to challenge funding practices and to ensure that future funding covers lay communication and feedback. There may be personal reasons and situations which make some researchers more successful than others in achieving this. However, the publication of the findings of research in scientific, peer-reviewed journals is not always covered by budgets (i.e. in terms of time and fees for open access journals) and yet researchers typically invest their time and energy in submitting articles for publication because it is in their interest to do so (e.g. for their academic careers).

Some researchers make a generalised and patronising assumption that participants would not be interested in the results, would not understand them or would misinterpret them (perhaps raising unrealistic hopes, or on the contrary shattering hope), whilst others raise concerns about the logistics of tracking people down and about data protection issues (Taylor 2019). For some, it may be a challenge to present the results in a clear and understandable way. However, even legitimate concerns about providing the right kind of feedback in the right way do not justify failure to respect this moral obligation. Structural barriers are created, maintained and perpetuated by individuals and therefore need to be challenged. Efforts to ensure that lay people also benefit from knowledge resulting from research have led to a few important changes. Some funders of research, for example, now require researchers to publish their findings in open access journals. This is a step in the right direction but as pointed out by Taylor (2019, p.1), "Results might be in the public domain but it doesn't follow that people know about them, can find them, have access to them, or make sense of them".

In the context of clinical trials, lay summaries of research findings have become obligatory under the Clinical Trials Directive, moving from a 'nice to have' to a 'need to have' status. Similarly, the British Medical Journal has recently introduced a requirement for researchers seeking to publish their papers to provide details of their plans for disseminating their findings to participants and other relevant communities, or to declare that they have no such plans. The British Journal of Learning Disabilities requires an accessible summary to be submitted with all article submissions for ease of all readers when scanning the content and with the explicit aim of making research findings more accessible to people with learning disabilities.

Post-trial access

Informing people with dementia who contributed to research about the findings is not the only way to show respect and appreciation for their time and efforts. In the context of biomedical research, post-trial access arrangements are becoming more important. Based on a justice argument, people who had the risk, burden and benefit of participation, shouldn't be worse off after participation. Therefore, since 2013 the Declaration of Helsinki sees it as a moral obligation towards research participants to make post trial arrangements.

> *"In advance of a clinical trial, sponsors, researchers and host country governments should make provisions for post-trial access for all participants who still need an intervention identified as beneficial in the trial. This information must also be disclosed to participants during the informed consent process" (Declarations of Helsinki 2013, article 34).*

In practice, this entails, giving participants the opportunity to continue taking the experimental drug before it obtains marketing approval. One possible way to do this is through open label extension studies which seem quite fair as they give each participant the opportunity to freely consent to continue with the study in the full knowledge that s/he will receive the experimental drug.

Open label extension studies are often carried out immediately after a double blind randomised clinical trial of an unlicensed drug. The aim of the extended study is to determine the safety and tolerability of the experimental drug over a longer period of time, which is generally longer than the initial trial and may extend up until the drug is licensed. Participants all receive the experimental drug irrespective of which arm of the previous trial they were in. Consequently, the study is no longer blind in that everybody knows that each participant is receiving the experimental drug but the participants and researchers still do not know which group participants were in during the initial trial.

There are a few relevant ethical issues related to this practice. For example, people may base their decision on continued participation on whether they had a positive or negative experience of the trial. However, they would not know whether their experience was linked to having taken the experimental drug, another drug or having received a placebo. For those who were not taking the experimental drug, their experience in the follow-up trial may turn out to be very different (Taylor and Wainwright 2005).

Also, if they are told about the possibility of the open label extension trial when deciding whether or not to take part in the initial trial (i.e. with the implication that whatever group they are ascribed to, in the follow-up study they will be guaranteed the experimental drug), this may induce some people to consent to the initial study. Taylor and Wainwright (2005) suggest that this might even amount to a form of coercion. However, by the time they reach the end of the study, of those who did not initially receive the experimental drug, some may no longer meet the inclusion criteria, may no longer be in a position to benefit from it (assuming that it is effective) or the drug might be counter-indicated for them for reasons not known at the outset of the study. The extension study, although planned, may not even be carried out if the data from the main study are not such as to justify the extension. Consequently, researchers should be very clear about the prospect of the open label extension trial and avoid it becoming a potentially misleading incentive to participation.

Taylor and Wainwright (2005) suggest that the open label trials may serve the purpose of prescribing an unlicensed drug on compassionate grounds, which whilst laudable, should not be camouflaged as scientific research. Rather governments should take responsibility and set up the appropriate legal mechanisms to make it possible for participants whose medical condition merits prolonged treatment with the experimental drug to have access to it. It would also be important to ensure that any claims made at the recruitment stage of the trial are clear and enable potential participants to make a fully informed decision. For the re-consent linked to continuing (or in some cases starting) to take the experimental drug, it should be emphasised that it has not yet received marketing authorisation and that the long-term benefits which might arise from the taking it, as well as the potential risks, are not yet known.

Recommendations for researchers

- Plan, budget for and write plain language summaries (for guidelines on writing plain English summaries for people having participated in a particular study and for relevant lay communities, see: https://www.invo.org.uk/resource-centre/plain-english-summaries/).
- Engage with people from the communities (including gatekeepers) about how and what to give back and in what format, recognising that this may not always be written.
- Include costs for dissemination, including accessible dissemination, in funding proposals and bids.
- Be sensitive to how the findings or any feedback are communicated (e.g. issues related to terminology, tone and how members of different communities are portrayed). Please see guidelines developed in collaboration with the members of the EWGPWD in 2013 on the portrayal of dementia and of people with dementia https://www.alzheimer-europe.org/Ethics/Ethical-issues-in-practice/2013-The-ethical-issues-linked-to-the-perceptions-and-portrayal-of-dementia-and-people-with-dementia/Guidelines
- Carefully plan how to bring the involvement of people with dementia in a particular study to an end so that people feel appreciated and valued rather than used and cast aside when no longer needed.
- Communicate findings to research participants and relevant lay communities using appropriate means of communication and dissemination (e.g. talks, pamphlets, reports on Internet, podcasts, web pages, newspapers, pictorial guides, graphic facilitation, professional/non-academic journals, social media etc.).
- Involve a broad range of people with dementia, people from relevant sub-groups and gatekeepers so as to ensure the best way to do this.
- Avoid sensationalistic reporting and unnecessarily complex explanations, without nevertheless this resulting in over-simplification and failure to communicate sufficient details for people to be able to understand the possible significance of the findings and to be able to use them to lobby for change.
- Explain how the findings fit into the ‘big picture’ (i.e. what is the next step, what now needs to be explored, what kind of changes will result from the findings?).
- If possible, discuss the results with people with dementia (i.e. those who were not involved in the study).

Recommendations for research ethics committees

- Check whether lay reporting has been envisaged and the appropriateness of plans for the dissemination and communication.
- At the end of the study, ask for information on how research results have been disseminated and communicated to the participants and communities.
- Review policies on the return of results and make sure they are described in the protocol and information materials.
- Review post-trial arrangements in the protocol and make sure they are ethically acceptable.

Recommendations for funders

- Clarify study dissemination plans and routes to impact with a range of audiences (e.g. to individuals, communities and policy makers etc.).
- Provide a dedicated budget for lay reporting (i.e. reporting of the findings to the general public in a form and in places which are accessible to them).
- Include costs for open access publications within funding call.

Summary

Just as research is not finished when the last piece of data has been analysed, neither is the involvement of people with dementia who contributed to a study through PI or as a research participant. Research findings need to be published in order to ensure that studies are not unnecessarily repeated and so that other researchers can verify the findings, challenge them and build on the knowledge that has been acquired. People with dementia and other members of the general public often don't have access to findings published in peer-reviewed journals (although open access publishing is becoming more common). They should be provided with lay summaries. To ensure that such summaries are understandable and disseminated in appropriate places, using appropriate means, a diverse range of people with dementia should be involved in the dissemination process. Out of courtesy, respect and acknowledgement of the considerable investment that people with dementia make to research and to society, researchers should consider whether there is something they could give in return. This might be a small token of gratitude, feedback on the results of the study (as just mentioned) or, in clinical trials, post-trial access to the experimental drug, subject to ethical approval.

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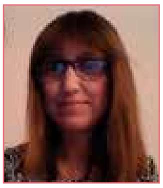
Appendix 1 – Co-authors and contributors to this paper

Alzheimer Europe would like to thank the members of the expert working group who graciously donated their time and expertise, and drafted this report. Please find below a brief overview of their backgrounds.



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Saloua Berdai Chaouni is a biomedical scientist and gerontologist. After years of field experience in developing projects of inclusion of ethnic minorities and consulting several organisations about inclusion and diversity management, she returned to research in 2015. As a PhD-student, affiliated to the Erasmus University College of Brussels and Vrije Universiteit Brussel, she conducts research about the experiences of dementia and care for older migrants with dementia in Belgium. Her other research work is situated at the intersection of ageing, migration and ethnic diversity; diversity management; disability and ethnic diversity; informal and formal care; qualitative research and decolonisation of research. She is also a lecturer at Karel De Grote University College in Antwerp.

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European Working Group of People with Dementia
https://www.alzheimer-europe.org/Alzheimer-Europe/Who-we-are/European-Working-Group-of-People-with-Dementia/Current-Members

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Appendix 2 – Glossary

The following list contains a brief explanation about some of the key terms used in this discussion paper.

Diversity

Diversity refers to the recognition, acceptance and respect of individual differences (e.g. in relation to gender, age, political beliefs, socio-economic status, appearance and dress-style, abilities and disabilities, sexual orientation, religious beliefs, language and so on) which are what make people unique. It amounts to more than a toleration of difference. It is about moving beyond a narrow focus on difference and from reducing people to a single defining characteristic. It is about embracing and celebrating the richness of individual and group differences.

Ethics/ethical

Ethics is a branch of philosophy which focuses on issues related to what is 'right' and 'wrong', 'good' and 'bad'. Over the centuries, philosophers, as well as researchers, healthcare professionals, politicians and members of the general public, to name but a few, have debated such issues. Guidelines, principles, values and theories abound. Determining what is ethically sound and what is not is a complex issue and a dynamic process. A clear overview of different ethical perspectives of relevance to the ethics of research can be found in Chapter 1 of the European Textbook on Ethics in Research (European Commission 2010).

Ethnicity

References to 'ethnicity' (or 'ethnic') are generally associated with the identification with a group of people or a community on the basis of a perceived shared culture. According to Smedley and Smedley (2005), "Ethnicity refers to clusters of people who have common culture traits that they distinguish from those of other people. People who share a common language, geographic locale or place of origin, religion, sense of history, traditions, values, beliefs, food habits, and so forth, are perceived, and view themselves as constituting an ethnic group" (2005, p.17).

Ethnic groups are not permanent, inflexible entities but rather open to change, with the possibility of people moving in and out of them. People define themselves as belonging to a particular ethnic group and are also identified by others as belonging to that group (as opposed to another group) (Barth 1998). They develop together the criteria for group membership, emphasising similarities between members of the group which are significant and ignoring those which are not (also with regard to intra-group differences).

An important aspect of ethnicity is that it is not something that is biologically determined, fixed or linked to nationality or place of birth, even though members of a group might share a common ancestral geographical origin and a tradition of common descent.

Intellectual disability

Intellectual disability is a disability that is characterised by significant limitations in both intellectual functioning (e.g. learning, problem solving and judgement) and in adaptive functioning (e.g. activities of daily life such as communication and independent living). It is not a disease and the disability starts before the age of 18. Down's syndrome is the most common identifiable cause. People with intellectual disability may still be able to learn new things and cope with various aspects of daily life but they may need more time, find it more difficult and need support.

Intersectionality

Intersectionality is about exploring the relationships between socio-cultural categories and identities. It emphasises multiple positioning, looks at differences between and within groups, at power relations between people and how the creation of the 'Other' serves to oppress, discriminate against and marginalise some groups of people, placing the blame for any societal problems they may experience on them personally whilst failing to acknowledge structural discrimination.

Paradigm

Kuhn, who coined the term 'paradigm', described it as a "constellation of beliefs, values, techniques and so on shared by members of a given community" (Kuhn 1970, p.175). In the context of research, paradigms are world views or belief systems which influence the way that researchers approach and carry out their studies (Guba and Lincoln 1994).

- The positivist paradigm emerged in the 19th century as a rejection of metaphysics. The validity of scientific theories was considered to rest on observable and empirical analytical facts. The focus was on what could be observed and measured with the ultimate aim of being able to predict and control it. Post-positivism was a reaction against the perceived limitations of the positivist paradigm and the

realisation that the positivist paradigm was not suited to the complexities of much social science research (e.g. including research into emotions and psychological factors which cannot be observed). It involves a critique of the positivist assumption that there is an absolute truth which with the right approach can be discovered.
- The social constructivist (or interpretivist) paradigm emphasises the social construction of reality based on shared and multiple meanings. This emphasises the social construction of reality, the importance of meaning, the impossibility of obtaining value-free knowledge, an emphasis on inductive logic and an awareness that methods of inquiry used in the natural sciences are not wholly appropriate for the study of social phenomena because the social world is mediated through meanings and human agency (Tashakkori and Teddlie 1998). Qualitative research methods are generally associated with the interpretivist paradigm.
- The paradigm of pragmatism favours the adoption of whichever methods, techniques or procedures are most relevant to providing the best understanding of the research problem and is based on the premise that no single method is perfect. By combining different methods, it is believed that certain weaknesses in one method may be cancelled out or balanced by strengths in the other and vice versa (Creswell 2009). Adopting a pragmatic approach involves inductive and deductive reasoning, as well as mixing qualitative and quantitative methods where appropriate (Tashakkori and Teddlie 1998, Creswell 2009).

Positionality

Posititionality is about where researchers stand in relation to the people with and on whom they are conducting research. A person's position in society, including their cultural background as well as a range of factors and characteristics which shape and reflect their identities, affects the way they make sense of the world. In the context of research, this may have an impact on how participants are treated, on decision making an on the conclusions drawn.

Public Involvement

The term Public Involvement (PI) is usually understood as meaning carrying out research and developing policies with or by members of the public and patients rather than on or for them. People who contribute towards PI are not considered as research participants. Rather, they are advisors or in some cases co-researchers who help improve research by sharing their personal experience and perspectives with researchers. Various levels of involvement are possible. All contributions are valuable.

Qualitative research

Qualitative research typically involves collecting, analysing and attempting to uncover the deeper meaning, significance and uniqueness of human behaviour and experience, including contradictions, ambiguities, behaviours, perspectives and emotions. The overall aim is to gain an in-depth understanding of people's experience and not to obtain information which can be generalised to the larger population. Possible ambiguities and contradictions in the data are considered as a reflection of social reality rather than a problem (Denscombe 2010). Researchers are often guided by a theoretical lens, a kind of overarching theory which provides a framework for their investigation.

The approach to data collection and analysis is methodical but allows for greater flexibility than in quantitative research. Data is collected by means of observation and interaction with participants (e.g. interviews, focus group discussions, immersion in the culture) or the review of documents. This is often an iterative process involving the methodical and simultaneous collection and analysis of data. With grounded theory, for example, researchers seek and determine hidden social and collective patterns and constructions from the data, avoiding theoretical preconceptions and adjusting research questions, if appropriate, as the theory starts to emerge.

Quantitative research

Quantitative research involves the systematic investigation of phenomena by means of the statistical analysis of numerical data in order to test hypotheses. The aim of such analysis (with the exception of descriptive statistics) is to determine whether it can be concluded that the findings were not due to chance alone. Researchers seeks patterns, trends, correlations and causal relationships between different variables. The focus is on whether and to what extent something is the case. Quantitative researchers use several different approaches.

Correlational research, for example, looks at non-causal relationships between variables using statistical analyses and is therefore mainly observational. With quasi-experimental approaches, researchers look for possible causal relationships between variables but without manipulating the variables. Cross-sectional studies compare different groups at a single point in time (almost like a snapshot of a particular variable), whereas longitudinal studies observe the same groups of people over an extended period of time which makes it possible to detect changes at the level of the individual and the group. The experimental approach seeks to establish a causal relationship between a group of variables. It is often considered as being the most rigorous approach and considered a kind of gold standard of (quantitative) scientific research. A classic example of an experimental design would be a randomised controlled trial. As suggested by Trochim (2006), if conducted properly, this design is the strongest with regard to internal validity.

Race

Race is a concept which categorises groups of people on the basis of biological differences (often including visible physical traits or characteristics), which it is claimed have been passed down from generation to generation (i.e. genetic differences). The concept of race is often associated with the belief that some races are inferior and even 'less human' than others, with devastating consequences for certain groups of people (e.g. discrimination, colonisation, slavery and genocide). According to Smedley and Smedley (2005), two main beliefs about race have persisted since the 20th century, namely race as consisting solely of human biogenetic variation (prevalent amongst scientists) and race consisting of a combination of physical and behavioural differences (a folk perception, also prevalent in some policies and laws). The concept of race has been challenged with opponents of this concept pointing out that so-called racial groups are not genetically discrete, measurable or scientifically meaningful and that there is more genetic diversity within 'races' than between them (Smedley and Smedley 2005, Mersha and Abebe 2015).

There may sometimes be valid reasons in the context of research for being aware of genetic differences between groups of people but the concept of 'race' (based on observed differences in biology, physical appearance and behaviour) is not useful in establishing such differences. Mersha and Abebe (2015) argue in favour of using 'ancestry informative markers' (AIMs). These are a "set of genetic variations for a particular DNA sequence that appear in different frequencies in populations from different regions of the world" (2015, p.4).

Reasonable accommodation

Reasonable accommodation is a term used in the United Nations Declaration of the Rights of Persons with Disabilities to describe reasonable adjustments or adaptations that should be made to ensure that people have the same opportunities (e.g. to use services, access buildings and play a role in society).

Reflexivity

It is not possible to rule out researcher reactivity (i.e. the possibility that the researcher might influence the research situation or the participants) or achieve objectivity (in the sense of freedom from bias) completely. It is therefore important to reflect on this and attempt to gain insight into it (a process which Kvale and Brinkmann call "reflexive objectivity") thereby enhancing sensitivity. This involves reflecting on one's own background, history and other relevant factors as well as regularly writing memos of one's thoughts and observations about the data collection and analysis.

Research

Research consists of a systematic, organised inquiry to find answers to worthwhile questions, using predefined methods or procedures which are clearly documented. The answers to such questions should contribute towards a body of knowledge or theory and it should be possible for other people to understand exactly what researchers did to arrive at their conclusions and any limitations there may have been to the study.

Sampling

Sampling strategies in quantitative research fall into two categories, namely probabilistic sampling and non-probabilistic sampling. The term probabilistic refers to the probability that everyone in the population has an equal chance of ending up in the study. Whilst non-probabilistic sampling is considered as permitting the lowest level of generalisability, its advantages in terms of reaching certain populations may sometimes need to be considered.

Qualitative research usually involves relatively small numbers of participants and contrary to the focus of much of quantitative research on random sampling, samples are often purposively selected. This is in keeping with one of the fundamental aims of qualitative research which is to gain an in-depth understanding of people's experience, for which diversity is necessary. Purposive sampling involves the selection of potential participants based on knowledge about the population and research topic, and on the research question. The number of people to be included in the sample is often not set in advance. Sampling evolves over the course of the study.

Note

Readers might also find the Jargon Buster document produced by INVOLVE (2007) helpful. It contains definitions of several terms which are often used in the context of Public Involvement: https://www.invo.org.uk/wp-content/uploads/2011/12/PIP44jargonbuster.pdf

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