D8.3 Brief on findings of ELSI focus groups for a RWE approach to AD

116020 - ROADMAP

Real world Outcomes across the AD spectrum for better care: Multi-modal data Access Platform

WP8 – Ethical, Legal and Social Implications

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Definitions

- Partners of the ROADMAP Consortium are referred to herein according to the following codes:
  - **UOXF**. The Chancellor, Masters and Scholars of the University of Oxford (United Kingdom) – **Coordinator**
  - **NICE**. National Institute for Health and Care Excellence (United Kingdom)
  - **EMC**. Erasmus University Rotterdam (Netherlands)
  - **UM**. Universiteit Maastricht (Netherlands)
  - **SYNAPSE**. Synapse Research Management Partners (Spain)
  - **IDIAP JORDI GOL**. Fundació Institut Universitari per a la Recerca a l'Atenció Primària de Salut Jordi Gol i Gurina (Spain)
  - **UCPH**. Københavns Universitet (Denmark)
  - **AE**. Alzheimer Europe (Luxembourg)
  - **UEDIN**. University of Edinburgh (United Kingdom)
  - **UGOT**. Goeteborgs Universitet (Sweden)
  - **AU**. Aarhus Universitet (Denmark)
  - **LSE**. London School of Economics and Political Science (United Kingdom)
  - **CBG/MEB**. Aagentschap College ter Beoordeling van Geneesmiddelen (Netherlands)
  - **IXICO**. IXICO Technologies Ltd (United Kingdom)
  - **RUG**. Rijksuniversiteit Groningen (Netherlands)
  - **Novartis**. Novartis Pharma AG (Switzerland) – **Project Leader**
  - **Eli Lilly**. Eli Lilly and Company Ltd (United Kingdom)
  - **BIOGEN**. Biogen Idec Limited (United Kingdom)
  - **ROCHE**. F. Hoffmann-La Roche Ltd (Switzerland)
  - **JPNV**. Janssen Pharmaceutica NV (Belgium)
  - **GE**. GE Healthcare Ltd (United Kingdom)
  - **AC Immune**. AC Immune SA (Switzerland)
  - **TAKEDA**. Takeda Development Centre Europe LTD (United Kingdom)
  - **HLU**. H. Lundbeck A/S (Denmark)
  - **LUMC.** Academisch Ziekenhuis Leiden – Leids Universitair Centrum (Netherlands)
  - **Memento**. CHU Bordeaux (France)
- **Grant Agreement.** The agreement signed between the beneficiaries and the IMI JU for the undertaking of the ROADMAP project (116020).
- **Project.** The sum of all activities carried out in the framework of the Grant Agreement.
- **Work plan.** Schedule of tasks, deliverables, efforts, dates and responsibilities corresponding to the work to be carried out, as specified in Annex I to the Grant Agreement.
- **Consortium.** The ROADMAP Consortium, comprising the above-mentioned legal entities.
- **Consortium Agreement.** Agreement concluded amongst ROADMAP participants for the implementation of the Grant Agreement. Such an agreement shall not affect the parties' obligations to the Community and/or to one another arising from the Grant Agreement.

Publishable Summary

This deliverable reports results from:

(1) An updated literature review on empirical research into patient and publics attitudes to the secondary use of data. (Initially was completed as part of deliverable 8.2.)

(2) Findings from two focus groups conducted with people with dementia and their supporters to understand patient and carer attitudes towards the ethical, legal and social implications of a real world data platform for Alzheimer's disease.

Existing studies suggest broadly supportive attitudes among patients and publics to the sharing of health data for both research and care. However, that support is almost always conditional on more specific views about the measures that need to be in place to secure acceptability. For example, sharing with universities and health services rather than commercial organisations; conducting research for public benefit; and ensuring that trustworthy and accountable governance mechanisms are in place to give individuals control over the way their data is shared and used. Against this background of existing knowledge, focus groups were designed and conducted in collaboration with ROADMAP partner, Alzheimer Europe (AE). In December 2017 the European Working Group of People with Dementia (EWGPWD) were consulted to explore their concerns and expectations about the re-use and combination of health data to create a real world evidence platform for AD research.

In line with the findings from previous qualitative studies, participants in the focus groups expressed conditional acceptance of many data sharing activities. A key finding from the focus groups was the view that greater engagement about the research would enable participants to deal with uncertainties around the risks that are seen to face the sharing of health data. Participants in the focus groups shared the desire to be more informed about the results of research, and ideally to have a stronger more engaged relationship with the research process. For them this meant closing the gap between study participants and researchers, who are further removed from each other in cases of secondary research, to establish stronger links between participants and the different ways their data are being used.

1. Introduction

Europe now faces a substantial healthcare challenge due to an ageing population, increasing cost pressures, and more specialised and costly treatments. Greater access for medical researchers and policy-makers to more and better quality data, enabled by biobanks and other medical data repositories, can help meet these challenges (Mostert et al., 2016). ROADMAP is a private-public partnership (PPP) of leading institutions and companies with an interest in improving the situation of people with AD. Its goal is to develop efficient uses of 'real world evidence' (RWE) for the benefit of AD patients and their caregivers. Approaches based on RWE are one promising area to deliver targeted and increasingly effective healthcare, while deriving additional value and knowledge from existing data sources.

RWE is the evidence derived from the analysis and/or synthesis of real-world data (RWD) collected outside of randomised trials. RWD may be primary research data, collected in contexts that reflect clinical practice, or secondary research data derived from routine clinical data itself. Using data that was collected for a different purpose is called data re-use, or the secondary use of data. Sources of such data include observational studies, Electronic Medical/Health Records, and disease and patient registries. The kinds of data these sources may include ranges from clinical and patient reported outcomes to economic outcomes and quality of life indicators (Goettsch and Makady, 2017). Platforms that support RWE may better inform regulators (efficacy & safety), healthcare providers and payers (cost effectiveness and budget impact), industry (pricing & manufacturing), and scientists (mechanisms & pathways) to accelerate decision-making on re-purposing current treatments and developing new treatments. In practice, ROADMAP brings together data from 6 European countries (Denmark, France, Netherlands, Spain, Sweden, UK), 75 national databases and clinical registries, more than 40 cohorts, several studies using wearables and smart devices, and 5 dementia relevant trials.

Using RWE raises ethical, legal, and social issues (ELSI) as increasing interconnectivity between datasets gathered within and beyond traditional medical institutions can challenge accepted ethical, social, and legal norms and frameworks (Mittelstadt and Floridi, 2016). The secondary use of data can raise concerns among people because it is a use of data that may not have been foreseen, or even conceived, when participants or patients were asked permission for their data to be collected. As part of ROADMAP's extensive stakeholder engagement work, to ensure patient interests and concerns are accounted for in an ELSI framework for a RWE platform for AD, focus groups were conducted to collect patient attitudes towards the specific platform and data integration proposed by ROADMAP.

An initial literature review on empirical research into patient and publics attitudes to the secondary use of data was completed as part of deliverable 8.2: this has been updated and reproduced here. This deliverable then provides a brief summary of findings from two focus groups conducted with people with dementia and their supporters to understand patient and carer attitudes towards the ethical, legal and social implications of a real world data platform for Alzheimer's disease. Specifically, the focus groups explored attitudes to the sensitivity of health data, the safeguards that should be in place to protect data, and how participants in studies can be kept informed and involved about the way their data is being use.

1.1. Objectives

- Update literature review on empirical research into patient and publics attitudes to the secondary use of data.
- Provide insight into concerns and expectations of AD patients and supporters, regarding the re-use and combination of health data to create a real world evidence platform for AD research.
- Provide recommendations to ROADMAP about how to address and meet those concerns and expectations.

2. Updated review of empirical studies of public and patient attitudes to sharing health data

In the following we have reviewed empirical studies of attitudes to data sharing in order to broadly survey the range of views held by various publics and patient groups. The attitudes of individuals potentially contributing their data to research is fundamental to understanding the salience of ethical issues identified in D8.1. Participants' understandings and expectations around these issues provides an important perspective not necessarily captured in ethical debate or policy development, for example, around how to strike a balance between privacy and openness, or what constitutes acceptable uses of data. This review was used to inform the design of the focus groups (see below) and has been updated to include new literature published between July 2017 and March 2018. This review and results from the focus groups will contribute to producing an empirically informed set of requirements for an ELSI framework for ROADMAP.

2.1. Methods

2.1.1. Search strategy

Two databases were searched (Web of Science, PubMed) to identify studies of public and patient attitudes to sharing health data. Preliminary search strategies to identify studies specific to AD and dementia returned very few results (<10), consequently these condition-specific elements of the search strategy were removed. A broader search enabled us to identify studies of attitudes to data sharing in a range of clinical populations, as well as the general public. Keywords were chosen to capture articles that reported qualitative or quantitative research into attitudes and preferences about sharing health data (See Table 3). No date restrictions were imposed and non-English language results were excluded.

The results returned from the searches were screened for relevance from their title and abstract. Only articles reporting empirical work, or systematic reviews of empirical work, were included. We only included studies seeking the views of patients (with any condition) or various publics (e.g. students, prospective biobank participants, representative samples of a national population). We did not include studies seeking the views of various other research stakeholders (e.g. researchers themselves, research and healthcare managers, healthcare professionals). While these research stakeholders clearly have important perspectives on the sharing and re-use of health data, we chose to focus here on those stakeholders who had, or may, consent for health data about themselves to be made available for research. From 586 results returned, 42 relevant articles were included. These are characterised in Annex I.

Database	Search String	Returned July 2017	Returned March 2018
Web of Science	TS=(attitud* OR preference* OR elicitation OR survey OR questionnaire* OR qualitative OR interview* OR "focus group") AND TS=("medical data" OR "health data" OR "health research data" OR "medical research data" OR "epidemiological data" OR "epidemiological research data" OR "clinical data" OR "clinical research data" OR "clinical trial data" OR "biomedical data" OR "biomedical research data" OR "cohort data" OR "cohort study data" OR "EHR" OR "individual level data" OR IPD OR "Individual patient data" OR microdata OR "administrative data" OR "research data" OR "real world evidence" OR RWE OR "medical big data" OR "biomedical big data") AND TS=("data sharing" OR "sharing data" OR repurpos* OR "responsible sharing" OR "secondary use" OR re-use OR "reuse" OR "open data")	177	204
PubMed	((attitud* OR preference* OR elicitation OR survey OR questionnaire* OR qualitative OR interview* OR "focus group")) AND (("medical data" OR "health data" OR "health research data" OR "medical research data" OR "epidemiological data" OR "epidemiological research data" OR "clinical data" OR "clinical research data" OR "clinical trial data" OR "biomedical data" OR "biomedical research data" OR "cohort data" OR "cohort study data" OR "EHR" OR "individual level data" OR IPD OR "Individual patient data" OR microdata OR "administrative data" OR "research data" OR "real world evidence" OR RWE OR "medical big data" OR "biomedical big data"))) AND ((((("data sharing" OR "sharing data" OR repurpos* OR "responsible sharing" OR "secondary use" OR re-use OR "reuse" OR "open data")))	324	382

2.1.1.1. Analysis

The content of the final corpus of documents was analysed thematically. Thematic analysis can be described as “a method for identifying, analysing and reporting patterns (themes) within data” (Braun and Clarke 2006, 78). We used thematic analysis inductively so that themes were closely linked and representative of the data. All included articles were read in detail and segments of text were

highlighted and grouped into similar issues. Issues were in turn grouped into wider themes by iteratively comparing similar issues and going back to the text to verify the grouping. Where there were clear keywords that characterised an issue or theme (for example, “trust” or “commercial interests”) all articles were searched for these keywords to ensure full coverage. Analysis focused on what was common and novel in study findings, not simply on the frequency or prominence of certain viewpoints within the corpus of documents.

2.2. Results

The articles reviewed included studies of the attitudes from a variety of publics and patient groups. Publics studied in these articles included random samples of populations, but also healthy volunteers, students, early adopters of health tracking technologies, and community representatives. The more clinically focused study populations included individuals visiting both primary and secondary care, such as those attending GP appointments, emergency departments, and outpatient clinics or specialist services, with a range of conditions such as HIV, Parkinson’s disease, dementia, and diabetes. Additionally, other studies sought the attitudes of participants in epidemiological cohort studies, clinical trials and disease registries, as well as patient community groups.

While most of the studies were conducted in either the US or Europe, a small subset were conducted in low and middle income countries (For example, Kenya (Joa et al., 2015a, 2015b) and Vietnam (Merson et al., 2015)). Studies mostly employed either structured survey, focus group, or interview methods, but there were also many mixed-method studies using some combination of these methods. Our search also identified three literature reviews, which explored the factors that influence consumer preferences for sharing health information (Moon, 2017), public attitudes to data sharing and linkage of health data for research (Aitken et al., 2016), and public attitudes to the use of medical data for research and about consent for secondary use for research (Hill et al., 2013). With little exception studies reported favourable attitudes towards data sharing when certain conditions or expectations could be met (see below). Even among the few exceptions, the divergence was not characterised by negative attitudes to data sharing, but rather by confused or mixed opinion (see for example: Audrey et al., 2016).

Many of the quantitative surveys sought to examine the demographic characteristics of those supportive of data sharing (Moon, 2017). Typical characteristics examined include age, gender, ethnicity, level of education, and health status, however, the relationships are mixed. Some studies find that older individuals are less likely to consent to sharing medical data for research (Goodman et al., 2017) or that younger individuals are more likely (Padrez et al., 2016; Page et al., 2016). Although the age ranges of the participants in these studies is highly variable, for instance, Padrez et al (2016) found greater acceptability of data sharing among those aged 18-30; the same gradient of greater acceptability among younger individuals was found by Page et al (2016) and Mursaleen et al (2017a) but from participants, respectively, with a mean age of 58 (SD 17.3) and with the majority with an age between 55-74. Many other studies found no association between data sharing attitudes and age. Kim et al (H. Kim et al., 2017) and Sanderson et al (2017) both found less willingness to share data for research among black or ethnic minority participants, whereas Padrez et al (2016) found greater

willingness for black participants to consent to having their social media data linked to their health data, and further studies outside Europe and the US have found greater trust and willingness for patients to share clinical data (Kimura et al., 2014; Merson et al., 2015). Sanderson et al (2017) found lower education to be associated with less willingness to share data for research, whereas Page et al (2016) found the opposite. Riordan et al (2015) found that low education and less experience with computers was associated with greater expectations that explicit consent would be sought for sharing of anonymous health data, and Perera et al (2011) found higher education and frequent computer and internet use to be associated with less concern about the technological risks of sharing electronic data. Regarding the influence of one's health status on attitudes to data sharing, Wicks et al (2010) and Goodman et al (2017) both found that participants with serious illnesses or cancer, respectively, were also more willing to share their health data.

It seems that there is some limited evidence across studies showing that attitudes to data sharing are less positive among some minority ethnic groups, and more positive among younger, more educated individuals, and those with a serious medical condition. However, the studies identified here are heterogeneous and overall present a very mixed picture of the relationships between demographic factors and attitudes to data sharing. What seems more important is less who people are, but rather what data sharing means to them. This is explored in many of the qualitative studies identified. Below we describe some of the key findings of these studies, grouped into four themes: trust, data sharing practices, consent, and the benefits of data sharing.

2.2.1. Trust

The characteristics that promote trustworthiness, and the kinds of entities that participants express trust in, include the nature of the organisations involved (university, commercial, health service) (Mazor et al., 2017), the kind of oversight mechanisms that are in place and how well-understood these are (Spencer et al., 2016), as well as the role of individuals who are involved in the sharing of data (Aitken et al., 2016; Bell et al., 2014; Manhas et al., 2015; Moon, 2017). Studies also found that trust was eroded or undermined by involvement of the private sector, due to the perceived tension between commercial interests and the integrity of researchers, the privacy of participants, and public good of research results (Aitken et al., 2016). Lack of trust in research and hence less willingness to consent to data sharing for research was also found among studies of ethnic minority communities (Lucero et al., 2015). Moreover, one representative national survey of US citizens found a general lack of trust in clinical information sharing systems (Platt et al., 2017), and a related survey found concerns about privacy to be a key predictor of reduced trust (Platt and Kardia, 2015).

Sheikh and Hoeyer (2017) studied perceptions of trust in research participants in Denmark and Pakistan and use their results to challenge the easy deployment of the concept of 'trust' in policy documents without sufficient explanation or sensitivity to the situations in which it is invoked. They observe that "when participants discuss trust they are trying to shape their relationship(s) with researchers while simultaneously communicating important hopes and fears in light of their situation" (p. 8) and go on to argue that "'trust' should not be thought of as the name of a phenomenon characterising willingness to donate" (p. 2) but rather use it as the starting point to examine what and why participants invest in research.

2.2.2. Data sharing practices

The acceptability of data sharing was rarely seen as unconditional, instead it was most often premised on practices being controlled or governed appropriately, such as assurances that confidentiality and privacy would be maintained, that data would be handled securely, and that mechanisms to prevent misuse would be in place (Aitken et al., 2016; Darquy et al., 2016; Haga and O'Daniel, 2011; Manhas et al., 2015; Mazor et al., 2017; Weitzman et al., 2010; Zalin et al., 2016). For example, the acceptability of data exchanges was conditional on the data-flows being auditable and secure (Aitken et al., 2016; Moon, 2017), and only disclosing the minimum data necessary to those accessing it (Moon, 2017). However, there is also some evidence of scepticism about maintaining the security of electronic records (Perera et al., 2011). Many studies found that data sharing was more acceptable if data are anonymised (Aitken et al., 2016; Haga and O'Daniel, 2011; Hill et al., 2013; Mahlmann et al., 2018; Mursaleen et al., 2017b), and less acceptable if data are shared with pharmaceutical or insurance companies, or used for commercial gain (Hill et al., 2013; Mahlmann et al., 2018; Mazor et al., 2017; Perera et al., 2011; Zalin et al., 2016). One notable exception however is the study by Ostherr et al (2017) who explored views towards sharing personal health-related data, collected through apps or devices rather than research or healthcare systems. In this highly commercially-mediated context, individuals showed little concern about sharing health related data with private companies, due in part to the personal convenience of the services offered and the social features that encourage sharing. Ostherr et al note that: "researchers who are required to participate in ethics review procedures and follow explicit protocols for data privacy, security and storage are subject to considerably more suspicion by members of the general public than are the corporations that overtly participate in data profiling with far less ethical supervision" (p. 9).

Other uses of data that were seen as less acceptable include, use for political purposes, for surveillance or in ways that result in stigmatisation or discrimination of individuals and groups, or more generally when there was no restriction on possible use (Aitken et al., 2016). Beyond particular uses of data, certain types of data are also viewed as being particularly sensitive, requiring stricter mechanisms for sharing (if they are shared at all). Commonly, this includes mental and sexual health data (Aitken et al., 2016; Bell et al., 2014; Moon, 2017; Powell et al., 2006), as well as identifiable or individual-level data or richer qualitative data (Aitken et al., 2016), and genetic data (Bell et al., 2014; Mahlmann et al., 2018). Although interestingly, in a large survey of nearly 1500 patients presenting to an emergency department, many of those who were social media users (Facebook or Twitter) were willing to consent to their social media data being linked with their medical data (Padrez et al., 2016).

This picture is also coloured by lack of engagement with the patients and publics around the safeguards (such as anonymisation) (Hill et al., 2013; Mazor et al., 2017) and the governance mechanisms that apply to research (Aitken et al., 2016). An interview study of young adults participating in a birth cohort study found variable and inconsistent understandings of data sharing and data linkage (Audrey et al., 2016), and a survey of over 300 individuals with Parkinson's disease found lack of consensus about the ownership, access or use of their medical data for research, attributable to lack of communication about those issues (Mursaleen et al., 2017a).

There is also evidence for cultural differences in attitudes, for example, Kimura et al (2014) found, in a study of US and Japanese citizens, that US citizens preferred access to their medical records for clinical purposes to be limited to only those healthcare professionals managing their care directly, whereas Japanese citizens had less objection to hospital-wide sharing of medical records. In a study of data sharing practices in Vietnam, Merson et al (2015) also found high levels of trust placed in researchers by patient representatives, and few if any conditions on data sharing, in contrast to the many studies conducted in Europe or the US which find widespread but conditional support for data sharing (Aitken et al., 2016).

2.2.3. Consent

Many studies have found patients and publics express a preference for explicit opt-in consent as a sign of respect for participants (Aitken et al., 2016; Hill et al., 2013; H. Kim et al., 2017) even though it may not be required if data are de-identified. In fact, studies have also found that participants hold the expectation that they will be asked to consent to any data sharing whether it involves de-identified or identifiable data (Page et al., 2016; Riordan et al., 2015). However, there is also recognition and acceptance among some individuals that this can be in tension with the efficiency and effectiveness of research (Aitken et al., 2016; Sanderson et al., 2017; Smith et al., 2016; Weitzman et al., 2010).

Furthermore, such studies typically consider the initial consent to participate in research, rather than re-consent for further studies. In relation to re-consent, Kelly et al (2015) found that some participants in the TwinsUK registry would be comfortable with not going through a re-consent process. The proportion of participants that were comfortable without re-consent depended on (in order of most to least comfortable) whether the further research was carried out by the same researchers, whether it was carried out by the same researchers but investigating a new condition, or whether it was carried out by different researchers (investigating the same or a different condition). A key finding here being that consent preferences are shaped by the existing relationships and trust that participants have with particular kinds of institutions already.

Related to this, studies have also found preferences for flexible models of consent that allow greater control over what kinds of data are shared with whom (Aitken et al., 2016). For example, in studies of technologies that presented patients with information about the data they were sharing, designs that allowed patients to exercise greater choice and granularity of data sharing were preferred (Bell et al., 2014; Caine et al., 2015; Harle et al., 2018; Moon, 2017). Similarly, other studies have found desire for knowledge about the planned uses and users of data that is shared (Hill et al., 2013; Spencer et al., 2016), and notably, transparency about of use and governance of data, achieved though greater engagement with participants about data sharing activities (Aitken et al., 2016; Haga and O'Daniel, 2011; Manhas et al., 2015; Mursaleen et al., 2017b).

2.2.4. Benefits of data sharing

Positive attitudes to data sharing very often rest on expectations of benefit to individuals themselves as well as to the general population (Aitken et al., 2016; Goodman et al., 2017; Hill et al., 2013; K. K. Kim et al., 2017; Lemke et al., 2010; Mahlmann et al., 2018; Manhas et al., 2015; Mazor et al., 2017;

Sanderson et al., 2017; Weitzman et al., 2010), or individuals with a specific condition (Darquy et al., 2016; Moon, 2017). In particular, clinical populations expressed supportive views about the use of their data for research (Darquy et al., 2016; Mursaleen et al., 2017b; Page et al., 2016; Trinidad et al., 2010) and for their own care (Perera et al., 2011; Zalin et al., 2016). Furthermore in studies of low-resource settings, the protection and promotion of benefits to those communities was also emphasised as a key component of acceptability (Jao et al., 2015a, 2015b). Similarly in surveys of healthy users of self-tracking technologies that collect personal health information there was found to be widespread support for the anonymous sharing of such data, if used for research for the 'public good' (Bietz et al., 2016; Chen et al., 2016; but see also: Ostherr et al., 2017).

2.3. Summary of findings

In summary, the reviewed studies suggest broadly supportive attitudes among patients and publics to the sharing of health data for both research and care. However, that support is almost always premised on more nuanced views about the measures that need to be in place to secure acceptability. In terms of who data is shared with researchers and universities, and healthcare professionals and health services were often trusted to protect individual's interests and act for the public good, in contrast, there is scepticism about trustworthiness of commercial organisations to do likewise. In terms of how data is shared, proper governance is key and individuals naturally expect mechanisms to protect confidentiality and privacy of data, but also greater control over data being shared was also found to be valuable to individuals. Indeed, the common expectation that individuals would be asked for explicit consent to data sharing out of respect, even in cases where it was not necessary is particularly noteworthy and speaks to the importance of engagement to maintain trustworthiness, legitimacy and 'social license' (Carter et al., 2015). In terms of what data is shared, supportive attitudes were often premised on only certain uses of data being permitted, or only certain types of data being shared. For example, that there would be no commercial use, or that types of data viewed as particularly being sensitive would be more strictly control, such as data about genetics, and mental and sexual health. The range of permissible uses of data also feeds into the question of why data is shared, where again supportive attitudes often rest on the promise of some benefit to individuals themselves, to individuals similar to them (for example, those with the same condition), or to the general population.

It will be crucial to understand attitudes to these issues in the specific setting of ROADMAP and to explore the conditions that promote support for data sharing among patients with AD and other stakeholders. This review therefore lays the groundwork for the focus groups (below), and subsequently for the final requirements for an ELSI framework (deliverable 8.5) to ensure they are aligned with those attitudes and values.

3. Focus Groups

3.1. Methods

Focus groups were designed and conducted in collaboration with ROADMAP partner, Alzheimer Europe (AE). AE convene the European Working Group of People with Dementia (EWGPWD), which is comprised entirely of people with different forms of dementia who are nominated by their national Alzheimer associations. The EWGPWD works to ensure that the activities, projects and meetings of AE duly reflect the priorities and views of people with dementia. The focus groups therefore consulted the EWGPWD members and their carers/supporters to understand the concerns and expectations of AD patients and supporters regarding the re-use and combination of health data to create a real world evidence platform for AD research.

3.1.1. Focus group members

We recruited all members of the EWGPWD who attended an AE consultation event in December 2017. The focus groups took place alongside other AE activities (not related to ROADMAP) that made up the consultation event.

There are currently 10 members of the EWGPWD, each from a different country. (The working language is English.) Members of the EWGPWD all have mild to moderate dementia and every member has the right to be accompanied to the meeting by a person of his/her choice to ensure safe travel and/or provide support during the meeting. The carer/supporter is only expected to facilitate communication and not to speak on the person's behalf, however we separately sought the views of any carers/supporters in attendance as well. Hence two focus groups were conducted: the first made up of 11 EWGPWD members, and the second made up of 10 of their supporters.

Members of the EWGPWD have prior experience of similar consultations. Most consultations led by Alzheimer Europe have been in the format of focus group discussions. During the Working Group's first two terms of office (2012-2014, 2014-2016), the members actively participated in the Alzheimer Europe conferences and gave keynote presentations in the European Parliament. In addition, they contributed to several face-to-face consultations for different European projects in which AE is involved (e.g. PredictND, EPAD, MinD, INDUCT, SMART4MD and PACE) and to the work that AE develops (especially in the context of the European Dementia Ethics Network and various Yearbooks). Consultations have also been conducted involving the EWGPWD by and for JPND (on dementia-friendly communities) and INTERDEM (on outcome measures), as well as in connection with research on people with dementia as peer researchers (Di Lorito et al., 2017). Individual members of the group have attended various international dementia events and given numerous interviews, both in their countries and internationally.

3.1.2. Consent

Members of the EWGPWD were provided with information about the focus group in advance of the consultation event and again on the day, so that they could decide whether to participate. AE

consultation events are typically considered as Patients and Public Involvement (PPI); individuals joined the EWGPWD to provide guidance to AE, either directly or in the context of projects in which AE is involved. However, by participating in the focus groups, members of the EWGPWD and their carers/supporters were treated as research participants. As such both working group members and carers/supporters were asked to give informed consent to take part the focus groups. Copies of the consent forms and participant information sheets can be found in Annexes II and III.

Most members of the EWGPWD take part in the group for at least 2 years. Some stay on for a second term of office, whereas some leave and are replaced by others. Within any 2 year term of office, it can arise that a particular person experiences cognitive decline which interferes with his/her ability to contribute meaningfully to a particular consultation. When this has occurred in the past, it has been the result of a gradual realisation and on the basis of discussions with the person concerned, his/her carer/supporter and AE, the person has decided to withdraw from the group. All current members of the EWGPWD have capacity to give informed consent, and there was no test of capacity at the consultation event. Each EWGPWD member was free and able to decide not to participate in the proposed focus group.

3.1.3. Organisation of session

The focus groups took place during an AE consultation event in December 2017. During the half-day focus groups members of the EWGPWD and their supporters were asked to reflect and give their views about what kinds of data should be used for research, who it should be shared with, and crucially what measures should be in place to make such sharing acceptable. An initial brainstorming session collectively explored what is understood by health data and data sharing. This was then followed by two, two-hour, focus group discussions made up of one group of EWGPWD members (facilitated by DG and AD) and a second group of the supporters (facilitated by AT and SL), so that potentially different attitudes and concerns could be explored separately. Within each group vignettes describing different data sharing scenarios provided a basis for detailed discussion and ranged over topics such as consent, the protection of confidentiality and privacy, the benefits to patients and the public of data sharing activities, as well as the conduct of data sharing activities in ways that are trustworthy and transparent. See Annex IV for a copy of the focus group schedule.

3.1.4. Ethics approval

Research ethics approval for this study was granted by the University of Oxford Social Sciences and Humanities Interdivisional Research Ethics Committee (IDREC) (Ref No: R53988/RE001).

3.1.5. Analysis

Focus group were audio recorded and transcribed. Transcripts were anonymised by replacing participant names with pseudonyms that preserve participants' genders (name were taken from lists of the top 20 male and female names of the 1950's in the USA). Each member of the European Working Group comes from a different European country, so references to participants' home country was disguised as northern, eastern European etc. References to native languages were replaced by

'non-English language'. The membership of the EWGPWD is small and public, so anonymity cannot be guaranteed however these measures make it difficult to associate quotes with particular individuals and make the risk of reidentification low.

Transcripts were analysed thematically. We followed Braun and Clarke (2006) who describe thematic analysis as "a method for identifying, analysing and reporting patterns (themes) within data" (p. 78). Themes were derived inductively to ensure they were closely linked to the data. Transcripts were coded independently by all authors using NVivo qualitative analysis software. Focus groups conducted with EWGPWD members and with supporters were treated separately for the purpose of generating codes and themes. This allowed for differences in the responses of the two groups to be preserved during coding and allowed the groups to be compared after themes have been generated. Initially transcripts were coded according to their content, then differences in codes and interpretations were reviewed and refined to reach consensus on the overall themes. The transcripts were reanalysed once the final set of codes was agreed.

3.2. Results

The aim of the focus groups was to provide insight into the concerns and expectations of people with AD and their carers regarding the re-use of health data. In broad terms, both focus groups articulated similar views that were in line with the findings from other qualitative studies in different populations, as described above in Section 2. That is to say, participants in the focus groups expressed 'conditional acceptance' of many data sharing activities. The participants in our focus groups explored the circumstances and conditions necessary for acceptable sharing but did not express strong principled objections to the sharing and re-use of data.

In the results that follow we first summarise the outcome of the initial brainstorming session that preceded the focus groups. In this session EWGPWD members and their supporters collectively discussed what 'health data' and what 'data sharing' means. Second, we describe the analysis of the focus groups themselves, examining participants' understanding of the ways health data are made available and used for research, and the different kinds of concerns and risks they view as salient. Third, we describe the ways they thought those concerns should be addressed, which focused most strongly on the social and relational aspects of their engagement with research, rather than the implementation of technical protections.

3.2.1. What is health data and data sharing?

In the initial brainstorming session participants expressed nuanced views about what counts as health data. All participants characterised health data as being information that describes individuals' medical conditions and general state of health (even if not suffering from a particular condition). Most participants also saw this as a narrow concept of health data and in various ways sought to broaden its scope:

First, participants noted other kinds of information about individuals which, in their view was not strictly health data but was important and clearly impacted health. For example, many kinds of lifestyle, education and socioeconomic information were described, as well as online information from social

media. Second, participants broadened the notion of health data out beyond single individuals, noting that health data (including genetic data) was not just data about themselves but also contained information about their family and social group.

In addition to broadening the scope of what counts as health data, participants expressed clear views about how it should be shared and used. Some participants raised the view that health data are an important resource that should be preserved and compared between generations, but also that they are sensitive and must be protected, especially against commercial exploitation. Here again, participants emphasised the collective aspect of such data, and indeed all participants expressed the view that health data should be shared in order to help others.

3.2.2. Risks and concerns

Throughout the focus group discussion, participants were encouraged to reflect on their concerns and the kinds of scenario that posed the most risk when sharing health data. Among both people with dementia and their supporters, discussion touched on generalised concerns with privacy, as well as specific concerns about how data would be processed and whether it would be vulnerable to 'hacking'. Worries about personal information being disclosed shaped much of the discussion, but participants also reflected on the accuracy of data being collected.

3.2.2.1. Data quality

One type of concern that participants raised was around how their data was processed. This typically involved scenarios in which there was a risk of bias or error being introduced into data. For example:

> *"Nancy: And I also know from other people, you get in a certain clinic certain results. You go into another clinic, you get other results.*
>
> *Facilitator: Hmm mm.*
>
> *Nancy: And in the third one you get again different results.*
>
> *Facilitator: Yeah.*
>
> *Nancy: So, it's not possible because my state can't change in the time. They even change – they make mistakes, for example, you make a scan and you get, as a result, a scan of a lady who is 20 years older than you, because they [the doctor] are distressed, they are not – the technology has to be higher, I think, safer, because they are humans who are working there, and they make mistakes." (EWGPWD members)*

Nancy identifies human error as a factor in data handling by suggesting that there is sometimes little consistency between the results of test performed at different clinics because doctors are under pressure and make mistakes. Furthermore, the acknowledgement that humans always make mistakes and that 'technology has to be higher' to deal with this suggests that Nancy is invested in the veracity and value of her data, rather than being fundamentally sceptical that data can be handled properly. As well as the possible introduction of error, participants also suggested there was a risk that bias may be introduced by those involved in collecting and processing data:

> *"Susan: [… an individual who] doesn't get on with her GP for whatever reason, or her psychiatrist, then they [the GP] will make notes and their particular bias, unconscious bias, will influence how they write their notes.*
>
> *Barbara: Absolutely.*
>
> *Susan: And will that data then be used, because that has a bias? It has a personal bias for whatever reason. That, I would be uncomfortable, very uncomfortable, with, because it's not strict… You take a blood test, you're looking for things. There's usually no bias." (EWGPWD supporters)*

Susan considers a situation where an individual has a poor relationship with their GP, potentially leading to information recorded about her being biased. Here again human factors enter the discussion of data sharing and are highlighted as a risk to data quality. Moreover, the potential for such biases also affects Susan's willingness to share data, as she states, she would be 'very uncomfortable' with biased data being used for research. Like Nancy, this shows Susan's investment in the veracity of her data but also that data collection processes are *fair* and not influenced, for example, by a GP 'who doesn't get on with her'.

Although the risk of bias and error being introduced into data collection was highlighted in both focus groups these ideas played a relatively minor part in the discussion. In contrast the dominant concerns in the discussions were around the possibilities for the disclosure of personal information.

3.2.2.2. The availability of too much information

Various scenarios in which data sharing introduced risks of personal information being disclosed were discussed by participants. Many examples centred around an unnecessarily large amount of information being available to others, and to combat this the notion of privacy was invoked. Privacy was used to justify participants' discomfort and hesitancy about data sharing, even when that information was being used in well-understood ways:

> *"Facilitator: But even if somebody's been given information about how the data will be used, can you understand why some people are hesitant?*
>
> *James: Yeah. Entirely.*
>
> *Facilitator: Why do you think that –*
>
> *James: Some people are very private.*
>
> *Mary: Yeah.*
>
> *Facilitator: It's a question of privacy then?*
>
> *James: Yeah, privacy. And some people think that too much information's going to be shared" (EWGPWD members)*

James refers to some people's desire to be private as a reason for hesitancy around data sharing, and adds, as further justification for hesitancy, that this may be because of the amount of information

that could be shared. Limiting the amount of information shared, so that seemingly unnecessary information would not be available to others, was a point that occurred throughout the focus group discussions. This highlights an important principle of information governance that only data necessary for the task at hand (research or care) should be shared, (see for example: Caldicott, 2013). Participants used a series of examples from their own experiences of care to express this idea. For example, Mary states:

> *"Because everything is put down on computer now, I've been in my own GPs, I been to, psychiatrists, psychologists, hospitals, and they just tap in, and all my data's in that. Everything. I'm not sure... I could be at the hospital for something entirely different, but they can get everything about my life, and I think it's a [... little] bit [of an] intrusion of privacy" (Mary, EWGPWD members)*

Mary describes her unease about the amount of information about her that was available in the context of her own care. Her concern stems from the fact that she could be 'at the hospital for something entirely different' but that health care professionals can 'get everything about my life'. That is to say, too much information is available in circumstances where it would in fact be unnecessary. This idea was also developed further by Linda:

> *"You wonder about in a care home or in a hospital, where the patients, how much protection you have, because everybody can tap into your – can tap into your records. So I'd have a question about that, you know, that it's free for all – that it's free for all staff. So should that be right? I don't think it should be." (Linda, EWGPWD members)*

Linda, like Mary above, questions the unrestricted availability of data because it allows others – in both cases health care professionals with legitimate access to such data – to learn too much about people. Other participants drew on further examples of unintended, unavoidable, and inappropriate disclosures that could occur during the use of their health data in routine care. For example, concerning the visibility of personal health information that was 'all spread out' while a doctor answer a question from another member of staff who had just entered the room (EWGPWD supporters); concerning the disclosure of one's conditions in order to receive assistance while travelling (EWGPWD members); and concerning situations where staff handle the data they can legitimately access unprofessionally, as illustrated by Susan's statement that:

> *"I would say my health data is only... is protected only as good as the blinking doctor's receptionist because my niece works in a doctor's receptionists and tells me things that she shouldn't be telling me, you know, just gossiping" (Susan, EWGPWD supporters).*

All the examples above involved participants drawing on their experiences of healthcare, rather than any real or hypothetical interaction with researchers. Deborah in the EWGPWD supporters focus group however broadened the discussion beyond the care context and was keen to emphasise that these same issues around the amount of data available could arise from health insurance companies combining multiple datasets and linking together difference sources of data about the same individuals:

> *"[another problem is] – how to say – to mix data from different resources, you have to have insurance, no? They ask, "Are you smoking? Are you sport[y]?" as in your lifestyle. And then there's the results of your illnesses, so they mix up and they want to control our*

> *lives, so this is the point, I think. And then the data will be also used for financial interests of very big companies, so you give your information and they earn money. Is this fair? It is here. Your health insurance then pays the medicaments which are very expensive." (Deborah, EWGPWD supporters)*

Deborah describes the process of data linkage as posing a risk if health insurance companies use the rich information they can collate to 'control our lives' and make medicines 'very expensive'. In the case of research data however, Deborah also argued that data linkage posed a problem for maintaining anonymity:

> *"For the quality of the research you must have a lot of information to make the people comparable, so you must have the social status, you must have the retail data, so what is then... except the name, […] medical records always link to data, as in [northern European Country] all the data is on a card from the health insurance, so what is then anonymous? You... and every moment you are able to mix up the data, to match the data." (Deborah, EWGPWD supporters)*

Deborah's argument here is that as multiple data sources are linked together and the information available on a given individual becomes richer, then the simple absence of their name from the data becomes increasingly insufficient to protect their anonymity.

Participants used reference to privacy and anonymity to push back against the amount of information about them that should be available. Importantly, the examples given by participants above all refer to information available to individuals, mostly, health care professionals, who have at least some legitimate interest in accessing a person's heath data; albeit if participants' main concern was that they have access to too much of it.

In the following section, however, a second kind of scenario was also discussed, drawing on examples where health data is accessed by individuals without a legitimate interest in it, that is to say, examples of malicious disclosures of information.

3.2.2.3. Unauthorised access and harmful uses: Hacking

Beyond the risks of unintended or unavoidable disclosures of information, arising from too much information being available to health care professionals, participants in both focus groups spent time discussing more malicious scenarios that involved unauthorised access to data that could lead to the disclosure of information and other harms. Foremost among their concerns was the problem of 'hacking':

> *"Linda: A hack. To me I think we are so vulnerable in the world today, and everything about us is – whether you go to the supermarket and you give your form for rewards or your revenue, you pay your taxes, everything, all your data is already there. So, every time you buy something, every time you pay your taxes, every time you go to your doctor, all your data's already there. It's already online. You go to the hospital, or you go you have your X-ray, by the time you're home, an hour later, the results of your X-rays is sent by computer –*
>
> *Facilitator: And what's the risk in that? What would be the …?*
>
> *Linda: I think the risk is that when we know that it is, that we have some anonym…*

anonymity with our… with the information that we have provided […] but then to see that it's all, you know, that it can be hacked into as easily as we have seen fairly recently.

Facilitator: So, it's about hacking?

Linda: It's about hacking. For me it's about hacking." (EWGPWD members)

Linda explains why she sees hacking as a concern: she describes the ubiquity of data about them that is online, claiming that this makes people 'so vulnerable in the world today' even if the data has been anonymised in some way, because it can be hacked into 'easily'. Linda goes on to describe the kinds of information disclosure they believe may result from hacking, claiming that:

"Linda: [… Hacking is] of great consequence. You don't want your mental health records out in the open.

Facilitator: Yeah.

Linda: You know, they [mental health records] can have huge consequences for people that are still employed, for example." (EWGPWD members)

Here Linda describes the kind of circumstances where hacking of health data can cause problems for individuals by suggesting that if one's mental health history was public, then this could create employment problems. In fact, later in the discussion Linda returns to this idea to re-emphasise the problems that stigma about dementia can cause for people who have revealed their condition, saying that: "We know from some of our own members across Europe the difficulties that that can create for employment."

Hacking and its potential consequences were discussed in both focus groups. Among supporters, the risk of hacking was similarly focused on the disclosure of sensitive data, like mental health information above. As well as the possible harmful consequences however, their discussion also touched on the value of hacking health data:

"Susan: […] If they're going to hack into this broad-spectrum [meaning: anonymised] data, it's not going to be as useful as hacking into specific [meaning: personal] data, so basically finding out about generally her health condition, her medications, as opposed to generally an anonymous person's health conditions. So, I actually think this data is – it had better be protected properly…. But it's probably less useful to those that want to steal it because of the randomness of it." (EWGPWD Supporters)

Susan reflects on the value of health data to those who want to hack or steal it. She makes the point that anonymised data may be less valuable than personal data and then notes that personal data must be well-protected because of its greater value. However, this is qualified with the further claim that health data may be too 'random', that is, random in the sense of the information being odd and not very useful for anything other than health research.

The question of value is a crucial aspect of assessing the risk that hacking poses, because it introduces the idea that risk is constituted by the motivations to hack health data as well as the feasibility of doing so. That is to say, while the risk of hacking depends on the effectiveness technical protection measures, it also depends on whether the data itself would be valuable to potential hackers.

Risk decreases if hacking is technically feasible but not worthwhile. Susan grapples with various views on the value of health data and whether hacking is worthwhile, admitting that personal data is likely to be more valuable that anonymous data, but also that the kind of information contained in health data sets may not be particularly valuable in any case. Indeed the value of health data to potential hackers is a question that some authors in the Statistical Disclosure Control literature (a field that studies the assessment and control of disclosure risks) have noted is not well-examined (Elliot et al., 2010; Elliot and Dale, 1999; Turner et al., 2017).

Participants in both focus groups also took the view that it was not possible to eliminate the risk of hacking and that human factors will always be present, making hacking possible. As James put it:

> *"I think it's well protected and they do the best they can, that there's lots of rules around it, but they'll always make mistakes and there's always someone a bit cleverer… who can access this stuff." (James, EWGPWD members)*

James simultaneously expresses the belief that data is well protected and that organisations 'do the best they can' to protect data, but that those organisations are one step behind hackers who will 'always' be a 'bit cleverer'. Similarly, supporters also expressed some scepticism about the protection of data:

> *"Barbara: I don't think… it's not 100% secure.*
>
> *Facilitator: Okay.*
>
> *Barbara: You can have people hack into it. Alright, say they probably even sometimes if the system's down, you could get someone else's information.*
>
> *Donna: Of course, it happens, yes. Yes. It happens.*
>
> *Barbara: Yeah.*
>
> *Karen: It's a vulnerable system and some people aren't able to handle the technical side.*
>
> *Barbara: Not fool proof." (EWGPWD supporters)*

Here the participants explain that hacking is always a possibility, partly attributing this to the technical measures in place to protect data, as seen in the reference to a system being 'down' or 'vulnerable', but also attributing it to people who 'aren't able to handle the technical side'. This understanding that security fundamentally involves a human element was explained further later in the discussion:

> *"Susan: I think research institutions will probably have stricter standards based on all their ethics and getting it through all the committees that they have to get through.*
>
> *Donna: Yeah.*
>
> *Barbara: And paperwork. On paper they have it. But is it actually happening? That's … but you can have all the constraints in the world, but if you don't implement them or there's always going to be some – I just, this is my feeling, there is always somebody. I don't think everything, no matter what company, who you're dealing with.*
>
> *Facilitator: So, it's an idea of human factor?*

Barbara:Yeah.

Facilitator: Yeah. So, what will make, could influence human factor then? What will improve the …?

Barbara: I don't think it can ever change.

Facilitator: Yeah?

Barbara: That's human … somebody's always going to be there looking, the opportunist, isn't it?

Karen: Yeah, or somebody being pressured." (EWGPWD supporters)

Susan suggests that research institutions have strict standards for the handling of health data, however Barbara questions whether these really translate into practice. Barbara's concern here is that it is always possible that protections are not properly implemented or that people may circumvent them. Indeed, Barbara and Karen identify two separate kinds of individual who they suggest pose a threat to research data: 'opportunists', taking advantage of some vulnerability, but also people being 'pressured' to release data. Again, it is not simply the protection measures that participants challenge (in this case ethical and governance standards, rather than technical measures) but their translation into practice and the people who are part in the system.

Hacking was a key scenario that both focus groups imagined when considering the risks that health data face. Previous research with biomedical scientists has found similar reference to hacking scenarios when they were asked to consider privacy and security risks facing data sharing technologies. Murtagh et al (2012) found two distinct scenarios invoking different malicious actors: "independent hackers" who are external and perhaps opportunistically trying to take advantage of vulnerable systems, and "unscrupulous scientists" who "have legitimate access to the data but abuse that right" (Murtagh et al., 2012, p. 251). A similar distinction was made by participants in our focus groups who differentiated between the risks posed by those both with and without authorised access to data. The reference to these kinds of imaged scenarios, here and in the study by Murtagh et al is framed by wider public concern about privacy and security caused by the increasing incidence and awareness of data accidentally leaked or stolen from organisations. For example, the UK National Data Guardian for Health and Care's 2016 *Review of Data Security, Consent and Opt-Outs* makes a clear connection between these kinds of wider concerns about cybercrime and the governance of health data, stating that:

"[…] the main threat to the public and private sectors is from basic cyber-attacks, which use hacking tools that can be purchased readily and cheaply online and exploit publicly known vulnerabilities" (National Data Guardian for Health and Care, 2016, p. 14).

This background of concern highlights some of the possible scenarios that may result in the disclosure of information, however, there are many open questions about how this background shapes expectations and the understanding of risk. For example, Moraia and Kaye (2014) worry about the potential impact increasing awareness of such incidents may have on sharing health research data, noting that "the level of public concern and outrage risks triggering a political overreaction that could be detrimental to biomedical research that crucially relies on sharing data and samples" (2014, p.

200, see also O'Dowd, 2013; Robeznieks, 2005). A key question that is thrown into sharp relief by the issues raised in the focus groups is therefore which aspects of these concerns are salient to the risks that research data face, and what measures are necessary to reassure participants that risks are being managed appropriately.

In contrast to the discussion of hacking risks, one participant, Michael, who was a member of the EWGPWD sought to frame the issue differently. He challenged the relevance of considering possible but illegal activities:

> *"Michael: But if we're talking about hacking and that sort of thing, it brings up a totally different scenario, so I... when I was read this, I have not thought about hacking, oh, that's a problem, because hacking, of course, is a problem. Misuse of total databank is a problem and we ... whenever we add to our own databank, I assume that this is not being hacked.*
>
> *Facilitator: Well – that may be being optimistic, this is talking about people's fears.*
>
> *Michael: Yeah, yeah, okay, yeah, but I assume that that is – that's not possible.*
>
> *Facilitator: Okay.*
>
> *Michael: Otherwise I would stop living if I was going to be concerned about how they behaved, misuse." (EWGPWD members)*

Michael suggests that considering hacking scenarios undermines a working assumption of participation in the 'databanks' of research or healthcare institutions, namely, that hacking is not possible. For him, taking such risks seriously would make it impossible to engage with those institutions. Later, Michael explains this idea further:

> *"Michael: So, I have to disregard possibilities that are really unlawful – It's not lawful to misuse or to go into the, my personal record, that's not lawful in [Northern European country]. So, if somebody's doing that, of course they are able – I'm sure they could be able to do it.*
>
> *Facilitator: Yeah.*
>
> *Michael: But if they're doing it I can get them to jail." (EWGPWD members)*

Michael concedes that illegal hacking activity is indeed possible, but suggests that the relevant risks to consider when thinking about the protection of their health data is not whether the law could be broken. Rather, for Michael, what matters is that there is such a law and that anyone who broke it could be sanctioned. Michael re-frames the issue of risk as being about what activities the legal and governance rules in place to protect data in fact prohibit, rather than about the range of scenarios that may be possible.

3.2.3. Trust and the protection of participants' data

Participants also spent time discussing how their concerns, and what they viewed as the most salient risks facing the sharing of their health data, could be addressed. In both focus groups this discussion

exclusively focused on disclosure risks and not the data quality issues identified above. Participants touched on many of the existing measures that were in place to protect their data, such as anonymisation, usage restrictions and robust legal and ethical rules.

Participants identification and endorsement of particular measures was typically brief, however more extended discussions developed as participants deployed notions of faith or trust in order to defend the validity of the above-mentioned protection measures. This occurred, for example, in the supporter's focus group as they were talking about the consequences of the presence of high-profile individuals, such as politicians, in a dataset:

> *"Susan: This wouldn't … the researchers wouldn't know that it came from Politician A. They would just know that this person has an alcohol abuse problem and this, that, and the other problem; or this particular number, rather than person, if it's depersonalised. They wouldn't know that it was Politician A.*
>
> *Deborah: No, they'd know because where they get the money from for research, they get it from there, from the state or from the companies or this is a whole team.*
>
> *Susan: The way I understand this research is, it's going to be anonymised. They are not going to know where it came from. And they're not going to go and think, "Ooh, Politician A, let's go and take his blood for research," because that would be unethical and if somebody were going to do that, they're going to do it anyway." (EWGPWD supporters)*

Susan suggests anonymisation would provide adequate protection for an individual, because it is impossible to infer information about that person from an anonymised dataset. Deborah challenges this idea by suggesting that funder, state or organisational interests in learning information about a high-profile individual would provide an incentive to de-anonymise the data. This therefore draws on the scenario, noted above, of the 'unscrupulous scientist' rather than the hacker. In response, Susan defends the integrity of anonymisation as a robust technique: 'it's going to be anonymised. They are not going to know where it came from', but furthermore suggests that researchers working to undermine the anonymisation 'would be unethical'. However she immediately qualifies this by claiming that 'if somebody were going to do that they're going to do it anyway'. Susan therefore admits that radical mistrust is possible and that little can be done to prevent de-anonymisation if 'unscrupulous' researchers want to do so, but she also suggests that a working faith in the ethical conduct of researchers is, in fact, what underwrites the integrity of anonymisation techniques.

The idea that participants use their trust in institutions or individuals as a way of dealing with the uncertainty and difficulty of making decisions about whether it was appropriate to share their data was also expressed by the EWGPWD focus group:

> *"Michael: I know there are obvious reasons to not trust the system. But I have to, when we sit here talking, I have to believe that the system is operating correctly.*
>
> *Facilitator: Okay.*
>
> *Michael: Otherwise there wouldn't be any discussion at all around these tables.*
>
> *Facilitator: Hmm. So you have to be in faith?*

Michael: You have to have that faith in the system.

James: There's a lot to trust involved, yeah.

Sandra: I agree with that." (EWGPWD members)

Michael describes his faith that healthcare and research systems will protect him and his data. Later in the discussion he describes how this faith is a fundamental premise for interacting with such systems, saying that "to survive I need a certain trust". James above also stresses that trust is needed and later draws the analogy that "It's only like telling someone a secret. […] You've just got to hope that they don't share it". Similarly again, Sandra above agrees and subsequently expands by stating that "Yes, I … I think they protect it [health data]. Yes. I trust …" and then, tellingly responds to the direction question of whether they think their health data is protected by first saying "Yes, I think so" then immediately qualifying this with "I hope so".

Trust is invoked, either directly or via the language of 'faith' and 'hope', as being necessary because it helps address participants' uncertainty. Beyond this, it was also used as a way of raising and opening-up this uncertainty for discussion.

"Susan: [sensitive data is] anonymised or protected and with all the -- hopefully things that would be put in place.

Pamela: Yes, but are they really protected? That's the issue because trust – I don't know." (EWGPWD supporters)

Susan defends measures such as anonymisation with a statement of faith that 'hopefully' they will be put in place, which Pamela uses to open-up the issue of whether such faith is well placed by contrasting trusting and knowing whether they are 'really protected'. In fact, when pressed to explain their trust in the face of uncertainty, participants often turned to their relationships with institutions or individuals. For instance, Susan talked about data being "used for the greater good" and then reflected on her own uncertainty about what the 'greater good' might mean by asking "how [would] you establish that[?], I would have to trust your institutions", and later using the example of her own participation in the focus group to elaborate further:

"Susan: What I'm doing … where my trust comes from is Alzheimer Europe allowing you [the facilitators, to conduct the focus group].

Robert: Yeah.

Susan: If Alzheimer Europe are happy with you then I'm happy with you. I think that's what I'm trying to say. So, my trust is in them, and it also happens to be in you as well." (EWGPWD supporters)

Susan uses Alzheimer Europe's approval of the focus group and facilitators as a proxy for her own approval: the relationship between Susan and Alzheimer Europe supports a new relationship between her and the focus group researchers themselves.

Participants deploy the language of trust to deal with uncertainties about the protection of their interests and their data. Their relationships with particular individuals or institutions underwrites their trust and ability to participate without being paralysed by the risks and concerns they identified.

3.2.4. Engagement and Feedback

Participants in both focus groups shared the desire to be, at a minimum, informed about the results of research, and ideally to be involved in the research process beyond the point where they contribute data. However, this raised a problem in the discussions, as some participants considered that their anonymity would be a barrier to feedback:

> *"Facilitator: So, what does everybody else feel about being informed about the research that's being done with their anonymous data, because if you've given broad consent, you might not get much more information.*
>
> *Barbara: No.*
>
> *James: How can they inform you if it's anonymous?*
>
> *Mary: They don't know [who you are]" (EWGPWD members)*

Here three participants are all puzzled by how it can be possible to feedback the results of a study to individuals who have contributed truly anonymous data. The question of how anonymity could be maintained alongside keeping participants engaged and informed about the research was also raised in the supporters focus group. They discussed the idea of broad consent and the possibility of being informed about the different ways their data was being re-used:

> *"Deborah: Or you also can regularly ask, "Okay, you have this data," and then you say, "Okay, it now will be used for this and it was for that".*
>
> *Facilitator: Ah, that's a really interesting idea. Okay.*
>
> *Susan: Oh yeah. "We're looking at your blood in relation to this particular piece of research." That would take some doing though, if you're anonymising all the data because then it's not anonymised." (EWGPWD supporters)*

This tension between anonymity and the ongoing involvement of participants speaks to a distinction that was not made in either focus groups, between personalised feedback, that is, feedback specific to particular individuals, and summary level feedback. Personalised feedback can include reporting back 'incidental findings' relevant to participants health, as well as other individual-level information. (While the norm in much epidemiological research has been not to feedback incidental findings, this is increasingly being questioned, see for example: Minion et al, 2017). Summary level feedback on the other hand refers to the overall findings of a study and therefore would not typically include information specific to individuals. Instead summary level feedback can be restricted to the low-dimensional or aggregate data found in scientific publications or public reports. Even if it is not possible to give feedback that is specific to individuals, because data are anonymised, it is still very likely to be possible to identify the set of individuals who participated in a study and provide summary level feedback. Indeed, summary level feedback is precisely the kind of information that could be fed back to everyone who participated, even when it is not possible to make a link between specific participants and study data/results.

The view that maintaining any kind of relationship or engagement with study participants – either to feedback information, or to inform or re-consent for further research – is impossible if data are

anonymised, could be detrimental to participants' expectations of participation in research. Such a view could discourage participation, if it was thought that no feedback could be provided; or equally if it was thought that receiving feedback meant that data were not properly protected. It is crucial therefore that different kinds of feedback, and kinds of relationship that can be maintained alongside robust anonymisation measures, are made clear to study participants.

Despite the puzzle of how feedback from research with anonymised data is possible, participants did nevertheless describe a strong preference for feedback. Moreover, while feedback at the end of the study was acceptable, participants also expressed a greater preference for regular updates and information as a study proceeded. As Linda suggested: "We'd [like to] get updates every so often". James expanded on this, stating they wanted information "Not just on – at the end of the research, the whole process", and they wanted "the same [information] as everyone else that's involved in the research programme". James suggests that this more extensive regular information should be similar in content to that received by other stakeholders in the research, firstly because "It would be nice to see what value your contribution was" and secondly out of respect to study participants.

At the same time, participants also lamented the fact that even receiving information at the end of a study is something that rarely happens:

> *"James: You don't usually get any [feedback].*
>
> *Linda: […] that's one of the greatest complaints we have, we never get the feedback" (EWGPWD members)*

One of the supporters, Barbara, who had some experience of research participation expressed this dissatisfaction in the strongest terms:

> *"Yeah, I think the PPI at home's one of the things I was involved in, a few pieces of research, and it was about getting the feedback and when it's complete, the research, and this has happened in [Western European City] that a paper has been published but the people that were involved in the research like myself, and we weren't … I don't know how many times I've emailed asking has it been published. I know it has now. I still haven't got a copy. I wasn't invited to the launch. Well, hello! Who gave you your information!? So, they forget the importance of us participating and working with them. Once they've got the information they're gone. And of course I've signed my consent. So, what do you do then?" (Barbara, EWGPWD supporters)*

Barbara does not want information about the study simply because she is interested in the results. She sees it, additionally, as a crucial way in which her participation can be recognised, emphasising this point with the exasperated rhetorical question 'Well hello! Who gave you your information!?'. But further explaining a sense of exploitation when researchers seem to 'forget the importance of us' and are 'gone' once participants have contributed data. Later Susan also expressed the same view, saying that:

> *"you see, you gave your permission, you were involved, and they haven't had the courtesy to say, 'Look, this is what we've produced'. So, it's very bad form." (Susan, EWGPWD supporters).*

Susan illustrates again how feedback about the results of a study is seen as recognising participants' contributions and maintaining a respectful relationship, as much as it is about providing information.

As described above the notion of trust was deployed by participants to deal with uncertainty in the research process. In discussion of what generated trust, participants also drew on the ways in which they were engaged in the research process itself:

> *"Facilitator: What else do you think helps foster trust …?*
>
> *Karen: Well, that's all the results are being openly discussed.*
>
> *All: Hmm, hmm.*
>
> *Karen: That there is the transparency in the results along the way, because you have stages, don't you, before the end result. And that you have a complete transparency all the way through, and the people involved – well, you are using anonymous data, so you can't go back to each person who might then withdraw or withdraw his consent. But I think to know what's happening on the end result, who benefits, yeah, and what else might the research have divulged on the way that might be useful, because sometimes, not just in what you hoped for, you get a lot more." (EWGPWD supporters)*

'Complete transparency', at each stage of research, was described by Karen as a mechanism for fostering trust, that is, reducing uncertainty about the research process, by creating and maintaining a relationship between researchers and participants. Later in the discussion, Barbara and Karen expanded on this idea further:

> *"Barbara: And the more open I think you are about research and explaining to people, the more people would buy in as well.*
>
> *Facilitator: Hmm, okay.*
>
> *Barbara: Because if we don't know about it and we don't understand the importance of it, we're all – well, there's always fear, isn't there, because that's …*
>
> *Karen: And when the researchers get to know about this, they would have to be even more careful that they are operating properly, so to speak. They have new ethical dilemmas proposed to them, they know all the doubts that the public may have about this, so they have to raise the level of security and whatever in their research, so us being critical and asking lots of questions will – or should – improve the quality of research and the way they will answer." (EWGPWD supporters)*

Barbara suggests that engagement with research will foster greater buy-in from participants and also that knowing about and understanding the research in question is an antidote to 'fear' and uncertainty. Karen follows this up by noting that transparency with respect to participants is not only useful for encouraging participants but also that, by opening-up the research to the input of a wider range of stakeholders, this increases the accountability of the researchers who must 'raise the level of security' and 'improve the quality of research'. In fact, these ideas were subsequently built upon by other participants when discussing information they would like to receive:

> *"Susan: Put it in the media. Let people know. But also let us know where something may*

not have worked as well as you thought.

Barbara:Yeah.

Susan:How you've changed that. So, to build up the trust, show us where you've gone wrong. You know, don't –

Barbara:Yeah, don't be afraid to say, "Hmm, we got this wrong, but we've corrected it".

Karen: That's the transparency of it." (EWGPWD supporters)

Here Susan and Barbara suggest a more radical kind of transparency and relationship between participants and researchers, one where study participants are informed about the details of scientific decision-making rather than just the findings that emerge at the end of a piece of research. In this sense then, they echo the view of James in the EWGPWD focus group who advocated for receiving 'the same [information] as everyone else that's involved in the research programme' putting participants on a par with other research stakeholders in terms of knowledge about decision-making, so that, as Deborah expresses it, "[participants are informed] about every step, you inform us what happened with our data." (EWGPWD supporters). In a similar way to the above calls from participants for recognition of their contributions, Susan noted that, "if you send it [information about a study] to us, we can read it or not read it. You don't send it to us, we don't have a choice". Thus the act of giving information about the results is seen as respecting participants' autonomy and their ability to engage with the research process.

3.3. Summary of findings

Participants in the focus groups highlighted concerns about data sharing and re-use that focused partly on data quality issues but most notably on risks associated with the disclosure of personal information. Participants drew on examples from their own care to describe scenarios in which health care professionals with legitimate access to their data, in various inadvertent or inappropriate ways, could learn information about patients. Participants used discourses of privacy and anonymity to challenge situations where seemingly more information than necessary about them was available to health care professionals. In addition, participants also described scenarios where personal information became available to individuals without legitimate access to their data, most prominently 'hackers'. The risk of personal information being disclosed by hacking was a key concern among many participants. Wider awareness of hacking clearly informs participants' understandings of data sharing activities, however, there was also reflection on the salience of such risks and the value of health data to potential hackers.

In response to the risk of personal information being disclosed, participants deployed the language of trust to deal with uncertainties about the protection of their data. They held the view that strong relationships with research and healthcare institutions underwrote their ability to participate in research without being paralysed by the risks and concerns they identified. Participants in both focus groups shared the desire to be, at a minimum, informed about the results of research, and ideally to have a stronger more engaged relationship with the research process: to them this meant participation beyond the point where they contribute data and greater recognition of their contribution. If

participants' relationship with institutions and individuals grounded their acceptance of some degree of uncertainty about how their data are used and protected and was therefore constitutive of trust, then their desire for a stronger more engaged relationship can be seen as seeking to reduce their uncertainties and build trust.

4. Conclusion and next steps

4.1. Recommendations: Feedback mechanisms for the secondary use of data

A key finding from the focus groups was the view that greater engagement about the research enabled participants to deal with uncertainties around the disclosure risks that are seen as facing the sharing of health data. However, study participants and researchers are removed from each other in cases of secondary research, which is primarily mediated by the data providers (for example, cohort studies). While engagement strategies are managed by data providers, findings from these focus groups suggest that there may be more work that consortia can do, in collaboration with data providers, to establish stronger links between participants and the different ways their data are being used.

We recommend three consortium-level activities that can validate and improve ROADMAP's approach to participant engagement:

(1) Audit ROADMAP's compliance with reporting and feedback requirements to ROADMAP data providers.

(2) Collect from ROADMAP data providers their best practices for engaging and informing participants about secondary research.

(3) Ensure these are adopted in all ROADMAP analyses.

4.2. Next steps

A session to discuss the results and recommendations from this study with members of the EWGPWD and their supporters is likely to be schedule for June 2018. Additionally, the EWGPWD will have opportunity to provide feedback and comments on subsequent publications to ensure that their contribution is properly acknowledged.

The findings from the focus groups will then be integrated into an ethical framework that will guide data integration practices in ROADMAP and provide recommendations for an EU-wide real-world evidence platform for AD: to be reported in Deliverable 8.5.

Along with a review of other empirical studies of public and patient attitudes to data sharing (Section 2), these findings will be written up for publication in an appropriate social science journal.

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ANNEXES

Annex I. Characterisation of reviewed articles

Article	Aim	Setting	Sample	Method	Basic finding	Results suggest attitudes towards data sharing are:
Aitken, Mhairi, Jenna de St Jorre, Claudia Pagliari, Ruth Jepson, and Sarah Cunningham-Burley. 2016. 'Public Responses to the Sharing and Linkage of Health Data for Research Purposes: A Systematic Review and Thematic Synthesis of Qualitative Studies'. Bmc Medical Ethics 17 (November): 73. doi:10.1186/s12910-016-0153-x.	Review and synthesise qualitative research examining public attitudes to data sharing/linkage of health data for research			Systematic Review	"widespread general—though conditional—support for data linkage and data sharing for research purposes. It has found that whilst a variety of concerns are raised (e.g. relating to confidentiality, individuals' control over their data, uses and abuses of data and potential harms arising) where members of the public perceive there to be actual or potential public benefits arising from re- search and where they have trust in the individuals or organisations conducting and/or overseeing data link- age/sharing they are generally supportive. However, the thematic synthesis has also highlighted current low levels of awareness about existing practices and uses of Data,"	Positive, but conditional
Audrey, Suzanne, Lindsey Brown, Rona Campbell, Andy Boyd, and John Macleod. 2016. 'Young People's Views about Consenting to Data Linkage: Findings from the PEARL Qualitative Study'. Bmc Medical Research Methodology 16 (March): 34. doi:10.1186/s12874-016-0132-4.	To examine ALSPAC participants' views about data linkage	Bristol, UK	55 ALSPAC participants (age 17-19)	Interviews	"Findings from this study question the validity of 'informed consent' as a cornerstone of good governance, and the extent to which potential research participants understand different types of consent and what they are consenting, or not consenting, to"	Inconsistent / Issues not well understood

Bell, Elizabeth A., Lucila Ohno-Machado, and M. Adela Grando. 2014. 'Sharing My Health Data: A Survey of Data Sharing Preferences of Healthy Individuals.' AMIA ... Annual Symposium Proceedings. AMIA Symposium 2014: 1699–1708.	To understand choices individuals would make about sharing data from their health record for research	UCSD, USA	70 healthy volunteers	Survey and interview	"Having choices available did make participants more willing to share their data, and they expressed interest in keeping specific categories of information private. […] participants appear to be more willing to share when given granular choices over what categories of information to share, as well as when they are given information about who is accessing their information."	Positive, but conditional
Bietz, Matthew J., Cinnamon S. Bloss, Scout Calvert, Job G. Godino, Judith Gregory, Michael P. Claffey, Jerry Sheehan, and Kevin Patrick. 2016. 'Opportunities and Challenges in the Use of Personal Health Data for Health Research'. Journal of the American Medical Informatics Association 23 (E1): E42–48. doi:10.1093/jamia/ocv118.	To understand barriers to using personal health data for research among individuals who self-track.	USA	465 individuals, early-adopters of self-tracking technologies	Survey and interview	"Individuals reported willingness to anonymously share PHD if it would be used to advance research for the good of the public."	Positive, but conditional
Caine, Kelly, Spencer Kohn, Carrie Lawrence, Rima Hanania, Eric M. Meslin, and William M. Tierney. 2015. 'Designing a Patient-Centered User Interface for Access Decisions about EHR Data: Implications from Patient Interviews'. Journal of General Internal Medicine	To understand patient views towards the design of a user interface for an EHR system that allows them to make choices about who can access their health data	USA	30 adults receiving healthcare in Indiana, with EHRs containing sensitive information.	Survey, tasks and interview	"Patients overwhelmingly expressed a desire to have access to their own medical records, as well as a desire to control who views their health care data."	Positive, but conditional

30 (January): S7–16. doi:10.1007/s11606-014-3049-9.

Chen, Juliana, Adrian Bauman, and Margaret Allman-Farinelli. 2016. 'A Study to Determine the Most Popular Lifestyle Smartphone Applications and Willingness of the Public to Share Their Personal Data for Health Research'. Telemedicine and E-Health 22 (8): 655–65. doi:10.1089/tmj.2015.0159.	To understand use of lifestyle-apps for information tracking and attitudes to sharing such data for research	Sydney, Australia	101 adults (staff and students) University of Sydney	Survey	"While anonymity and privacy were recurring concerns raised by survey respondents, individuals were generally open to sharing their personal health data with researchers to improve health and to advance understanding of group behaviors."	Positive, but conditional
Goodman, Deborah, Catherine O. Johnson, Deborah Bowen, Megan Smith, Lari Wenzel, and Karen Edwards. 2017. 'De-Identified Genomic Data Sharing: The Research Participant Perspective'. Journal of Community Genetics, April, 1–9. doi:10.1007/s12687-017-0300-1.	To understand the views of study participants towards the sharing of their de-identified genetic data for research	USA	450 participants in the Northwest Cancer Genetics Registry (cases, controls and relatives)	Survey	"while it was important that their privacy and information be protected, having their information available to many research studies is important to them. […] the decision to make personal data available to a research repository is influenced by the participants' desire to acquire health information about themselves."	Positive, but conditional

Haga, S. B., and J. O'Daniel. 2011. 'Public Perspectives Regarding Data-Sharing Practices in Genomics Research'. Public Health Genomics 14 (6): 319–24. doi:10.1159/000324705.	To explore public attitudes to genomic data sharing for research	North Carolina, USA	100 individuals, mostly Female, mostly African-Americans.	Focus groups	"while focus group discussants recognized the importance of data-sharing, they desired to be informed of how the data would be shared due to concerns about privacy and confidentiality."	Positive, but conditional
Hill, Elizabeth M., Emma L. Turner, Richard M. Martin, and Jenny L. Donovan. 2013. '"Let's Get the Best Quality Research We Can": Public Awareness and Acceptance of Consent to Use Existing Data in Health Research: A Systematic Review and Qualitative Study'. Bmc Medical Research Methodology 13 (June): 72. doi:10.1186/1471-2288-13-72.	To explore public views on the use of medical data for research and about consent for secondary use for research	UK	19 men (aged 50-59) eligible for on-going randomised trial of test for prostate cancer	Systematic Review and Focus group	"All participants were keen to contribute to NHS-related research but some were concerned about data-sharing for commercial gain and the potential misuse of information"	Positive, but conditional
Jao, Irene, Francis Kombe, Salim Mwalukore, Susan Bull, Michael Parker, Dorcas Kamuya, Sassy Molyneux, and Vicki Marsh. 2015a. 'Involving Research Stakeholders in Developing Policy on Sharing Public Health Research Data in Kenya Views on Fair Process for Informed Consent, Access Oversight, and Community Engagement'. Journal of	To understand views on fair data-sharing practices	Kenya	30 community members with relatively low research experience. And 40 research experienced individuals from a variety of	Focus groups	"A majority of stakeholders were generally supportive of data sharing in principle, but important conditions were proposed by all"	Positive, but conditional

Empirical Research on Human Research Ethics 10 (3): 264–77. doi:10.1177/1556264615592385.			stakeholder groups			
Kim, Hyeoneui, Elizabeth Bell, Jihoon Kim, Amy Sitapati, Joe Ramsdell, Claudiu Farcas, Dexter Friedman, Stephanie Feudjio Feupe, and Lucila Ohno-Machado. 2017. 'ICONCUR: Informed Consent for Clinical Data and Bio-Sample Use for Research'. Journal of the American Medical Informatics Association 24 (2): 380–87. doi:10.1093/jamia/ocw115.	To evaluate a web based consent tool that elicits patients preferences for data sharing	USA	126 outpatients attending an internal medicine or HIV clinic	Interviews	"Obtaining consent for de-identified data is legally not necessary, but our results suggest that it is not only feasible but also confers a higher level of trust in research and has no negative impact on participation."	Positive
Kim, Katherine K., Pamela Sankar, Machelle D. Wilson, and Sarah C. Haynes. 2017. 'Factors Affecting Willingness to Share Electronic Health Data among California Consumers'. Bmc Medical Ethics 18 (April): 25. doi:10.1186/s12910-017-0185-x.	To explore consumers' willingness to share electronic health information for healthcare and research	California, USA	800 randomly dialled Californian adults	Survey	"For healthcare, those who believe that sharing healthcare data through HIEs improves privacy and safety are more likely to consent to share data for healthcare purposes. Those who believe EHR positively impacts healthcare quality and research quality are more likely to consent to electronic data sharing for both research and healthcare."	?

Kimura, Michio, Jun Nakaya, Hiroshi Watanabe, Toshiro Shimizu, and Kazuyuki Nakayasu. 2014. 'A Survey Aimed at General Citizens of the US and Japan about Their Attitudes toward Electronic Medical Data Handling'. International Journal of Environmental Research and Public Health 11 (5): 4572–88. doi:10.3390/ijerph110504572.	To explore attitudes towards handling of electronic medical data	USA, Japan	200 US individuals, 457 Japanese individuals	Survey	"Participants from the US think that the extent of the sharing their identifiable medical records should be limited to the doctors-in-charge and specified doctors referred to by their own doctors. On the other hand, Japanese people find it acceptable for doctors of the same hospital to share their medical records."	?
Lemke, A A, W A Wolf, J Hebert-Beirne, and M E Smith. 2010. 'Public and Biobank Participant Attitudes toward Genetic Research Participation and Data Sharing'. Public Health Genomics 13 (6): 368–77. doi:10.1159/000276767.	To understand attitudes towards the collecting and sharing genetic research Data	Chicago, USA	21 participants in the Nugene biorepository ; 28 members of the public	Focus groups	"Although focus group participants discussed both positive and negative reasons for participating in genetic research, overall, most indicated they would consider participation to benefit individuals and society."	Positive, but conditional
Lucero, Robert J., Joan Kearney, Yamnia Cortes, Adriana Arcia, Paul Appelbaum, Roberto Lewis Fernandez, and Jose Luchsinger. 2015. 'Benefits and Risks in Secondary Use of Digitized Clinical Data: Views of Community Members Living in a Predominantly Ethnic Minority Urban	To explore community members' views on secondary use of clinical data for study recruitment and studies linking data to biosamples	Columbia , USA	30 adults, mostly Latino from ethnic/racial minority neighbourhoods	Focus groups	"They were concerned that secondary use of their personal health information for research recruitment constituted a privacy violation. This sentiment reflected participants' fear, uncertainty, and lack of trust regarding research."	Mixed

Neighborhood.' AJOB Empirical Bioethics 6 (2): 12–22. doi:10.1080/23294515.2014.949906.						
Manhas, Kiran P., Stacey Page, Shawn X. Dodd, Nicole Letourneau, Aleta Ambrose, Xinjie Cui, and Suzanne C. Tough. 2015. 'Parent Perspectives on Privacy and Governance for a Pediatric Repository of Non-Biological, Research Data'. Journal of Empirical Research on Human Research Ethics 10 (1): 88–99. doi:10.1177/1556264614564970.	To examine parent perspectives on pediatric research data (not sample) repositories	Canada	19 interviewees, 18 focus group participants; adult participants from two birth cohorts.	Focus groups and interviews	"The parent participants in this study valued sharing non-biological research data derived from themselves and their children, when clear and effective governance strategies are in place."	Positive, but conditional
Merson, Laura, Tran Viet Phong, Le Nguyen Thanh Nhan, Nguyen Thanh Dung, Ta Thi Dieu Ngan, Nguyen Van Kinh, Michael Parker, and Susan Bull. 2015. 'Trust, Respect, and Reciprocity: Informing Culturally Appropriate Data-Sharing Practice in Vietnam'. Journal of Empirical Research on Human Research Ethics 10	to explore attitudes to sharing clinical research data for research	Vietnam	15 patient representatives (patients and family members). Plus other research stakeholders	Focus groups	"Patient representatives expressed willingness to entrust researchers with all decisions regarding the use of their data. They showed a low level of interest and lack of concern for personal risk with respect to data sharing."	Positive

(3): 251–63. doi:10.1177/1556264615592387.

Moon, Lisa A. 2017. 'Factors Influencing Health Data Sharing Preferences of Consumers: A Critical Review'. Health Policy and Technology 6 (2): 169–87. doi:10.1016/j.hlpt.2017.01.001.	To identify factors influencing consumer preferences for sharing electronic protected health information			Systematic Review	"this critical review shows that, (1) Trust relationship, (2) Harm Threshold, (3) Balance Risk and Benefits, (4) Transparency of Data Exchange and (5) Access and Control of Data are important when considering how to best include the consumer voice in the development of legal / public policies related to the privacy, security and consent management of ePHI."	Mixed
Mursaleen, Leah R., Jon A. Stamford, David A. Jones, Richard Windle, and Tom Isaacs. 2017. 'Attitudes Towards Data Collection, Ownership and Sharing Among Parkinson's Disease Patients.' Journal of Parkinson's Disease, June. doi:10.3233/JPD-161045.	To explore patient attitudes to ownership and sharing of their medical data	UK	310 individuals with Parkinson's disease (aged 55-74)	Survey	"The lack of consensus on data ownership and general absence of clear demographic predictors of data sharing implies impaired communication pathways."	Positive, but issues not understood
Mursaleen, Leah R., Jon A. Stamford, Peter Schmidt, John M. Dean, Richard Windle, David A. Jones, and Helen Matthews. 2017. 'Choices on Selective	To explore patient attitudes to clinical data sharing	UK	43 individuals with Parkinson's disease, participating	Focus groups	"PwP are more likely to share their information if there is assured anonymity and transparency about the use of their data.	Positive, but conditional

Clinical Data Sharing by People with Parkinson's Disease'. Journal of Parkinsonism and Restless Legs Syndrome 7. doi:10.2147/JPRLS.S133922.			in a charity organised conference			
Padrez, Kevin A., Lyle Ungar, Hansen Andrew Schwartz, Robert J. Smith, Shawndra Hill, Tadas Antanavicius, Dana M. Brown, Patrick Crutchley, David A. Asch, and Raina M. Merchant. 2016. 'Linking Social Media and Medical Record Data: A Study of Adults Presenting to an Academic, Urban Emergency Department'. Bmj Quality & Safety 25 (6): 414–23. doi:10.1136/bmjqs-2015-004489.	To determine the acceptability of linking patients' social media content with their electronic medical record data.	USA	1432 individuals who presented at an emergency department, who were also facebook or twitter users	Survey	"Many patients are willing to share and link their social media data with EMR data. Sharing patients have several demographic and clinical differences compared with non-sharers."	Positive, depending on demographic characteristics
Page, Stacey A., Kiran Pohar Manhas, and Daniel A. Muruve. 2016. 'A Survey of Patient Perspectives on the Research Use of Health Information and Biospecimens'. Bmc Medical Ethics 17 (August): 48. doi:10.1186/s12910-016-0130-4.	To explore patient attitudes towards the use of their medical data and biosamples for research	Canada	211 outpatients attending a renal clinic	Survey	"In general, these patient participants were supportive of medical research and very trusting of medical researchers. Most believed that consent should be sought for use of health information or biospecimens and most indicated they would always give consent for any medical research."	Positive

Perera, Gihan, Anne Holbrook, Lehana Thabane, Gary Foster, and Donald J. Willison. 2011. 'Views on Health Information Sharing and Privacy from Primary Care Practices Using Electronic Medical Records'. International Journal of Medical Informatics 80 (2): 94–101. doi:10.1016/j.ijmedinf.2010.11.005.	To explore patient and physician attitudes to sharing electronic health data for healthcare and research	Ontario, Canada	490 patients with diabetes already enrolled in a randomized trial of a web-based diabetes tracker	Survey	"patients generally embraced the potential benefits that computers can bring in terms of sharing, integrating and evaluating information when used for their direct care. [...] they were concerned about the potential for dissemination of their private information over the Internet, especially to certain groups not involved in their health management, […] There was less concern with university or hospital-based researchers using de-identified information about them."	Positive, but conditional
Platt, Jodyn E., Peter D. Jacobson, and Sharon L. R. Kardia. 2017. 'Public Trust in Health Information Sharing: A Measure of System Trust.' Health Services Research, January. doi:10.1111/1475-6773.12654.	To measure public trust in a health information sharing	USA	Nationally representative sample of 1011 adults	Survey	"a majority of the U.S. public does not trust an integrated health information sharing system in at least one or more dimensions. […] We also found that the public is more inclined to feel the system is competent and has their best interests in mind (i.e., fidelity), but it is less confident in the system's integrity and overall trustworthiness."	n/a
Platt, Jodyn, and Sharon Kardia. 2015. 'Public Trust in Health Information Sharing: Implications for Biobanking and Electronic Health Record Systems.' Journal of Personalized Medicine 5 (1): 3–21. doi:10.3390/jpm5010003.	To examine characteristics of the general public that predict trust in a health system	USA	447 individuals who participate in a crowd source marketplace that is "younger, less diverse, and more educated	Survey	"Knowledge and concerns about privacy were found to be the key factors in predicting lower levels of Trust. […] One of the strongest predictors of system trust was having a positive view of data sharing."	n/a

			than the US population."			
Powell, John, Richard Fitton, and Caroline Fitton. 2006. 'Sharing Electronic Health Records: The Patient View.' Informatics in Primary Care 14 (1): 55–57.	to explore accuracy and willingness to share electronic record data on a national database.	UK	31 patients attending (consecutively) a GP.	Preference elicitation task	"Eighty-four percent of the patients in this study were happy to have their whole record shared. Of the five patients who felt that there was at least some information in their primary care record that they would not want to be shared, the items they identified related almost entirely to matters of pregnancy, contraception, sexual health and mental health."	Positive, but conditional
Riordan, Fiona, Chrysanthi Papoutsi, Julie E. Reed, Cicely Marston, Derek Bell, and Azeem Majeed. 2015. 'Patient and Public Attitudes towards Informed Consent Models and Levels of Awareness of Electronic Health Records in the UK'. International Journal of Medical Informatics 84 (4): 237–47. doi:10.1016/j.ijmedinf.2015.01.008.	to examine attitudes towards sharing identifiable and de-identified data for healthcare, research and planning.	London, UK	3157 patients or members of the public attending primary and secondary care clinics	Survey	"This study indicates that most members of the public expect to be asked for explicit consent before their identifiable data stored within integrated EHRs is shared for health provision, research and policy. Even for de-identified health records, however, half of the respondents expect their explicit consent to be sought."	Positive, with more knowledge of EHRs

S, Darquy, Moutel G, Lapointe As, D'Audiffret D, Champagnat J, Guerroui S, Vendeville Ml, Boespflug-Tanguy O, and Duchange N. 2016. 'Patient/Family Views on Data Sharing in Rare Diseases: Study in the European LeukoTreat Project., Patient/Family Views on Data Sharing in Rare Diseases: Study in the European LeukoTreat Project'. European Journal of Human Genetics : EJHG, European Journal of Human Genetics 24, 24 (3, 3): 338, 338–43. doi:10.1038/ejhg.2015.115, 10.1038/ejhg.2015.115.	to explore patient and family views on the sharing of their medical data in the context of compiling a European leukodystrophies database	France, Italy, Belgium, Spain, Germany	46 patients with leukodystrophies, and 149 close relatives of patients	Survey	"A major result is that patients/families are strongly driven to participate in any research that collects data. Patient registries and databases are widely recognized as highly vital in the context of rare diseases"	Very positive, but conditional
Sanderson, Saskia C., Kyle B. Brothers, Nathaniel D. Mercaldo, Ellen Wright Clayton, Armand H. Matheny Antommaria, Sharon A. Aufox, Murray H. Brilliant, et al. 2017. 'Public Attitudes toward Consent and Data Sharing in Biobank Research: A Large Multi-Site Experimental Survey in the US.' American Journal of Human Genetics 100 (3): 414–27. doi:10.1016/j.ajhg.2017.01.021.	To examine attitudes to consent and data sharing for biobanking research	USA	13000 patients who had attended an Electronic Medical Records and Genomics (eMERGE) Network medical site	Survey	"the results from this study suggest that bio-banks using broad consent may not be less successful in recruiting participants than if they use more specific consent approaches. Open data sharing may be almost as acceptable to participants as controlled data sharing"	Positive, but conditional

Spencer, Karen, Caroline Sanders, Edgar A. Whitley, David Lund, Jane Kaye, and William Gregory Dixon. 2016. 'Patient Perspectives on Sharing Anonymised Personal Health Data Using a Digital System for Dynamic Consent and Research Feedback: A Qualitative Study'. Journal of Medical Internet Research 18 (4): e66.	To explore patient perspectives on the use of anonymized health care data for research	UK	35 patients attending a rheumatology outpatient clinic. 5 individuals from a patient and public involvement health research network.	Interviews and focus groups	"Patients were supportive of sharing their anonymized electronic patient record for research, but noted a lack of transparency and awareness around the use of data, making it difficult to secure public trust."	Positive
Trinidad, Susan Brown, Stephanie M. Fullerton, Julie M. Bares, Gail P. Jarvik, Eric B. Larson, and Wylie Burke. 2010. 'Genomic Research and Wide Data Sharing: Views of Prospective Participants'. Genetics in Medicine 12 (8): 486–95. doi:10.1097/GIM.0b013e3181e38f9e.	to explore attitudes of current and potential research participants to genome-wide association studies and repository-based research.	USA	34 individuals with dementia or their carers participating in the Adult Changes in Thought cohort, and 45 patients of the Group Health research institute	Focus groups	"Overall, participants endorsed the value of data sharing and, while they recognized some risks, most considered the potential benefit of high-throughput genomic research to outweigh the possible harms."	Positive, but conditional

Weitzman, Elissa R, Liljana Kaci, and Kenneth D Mandl. 2010. 'Sharing Medical Data for Health Research: The Early Personal Health Record Experience'. Journal of Medical Internet Research 12 (2). doi:10.2196/jmir.1356.	To explore attitudes toward sharing personally controlled health record information for health research	USA	151 early adopters of a PCHR platform for survey. 13 PCHR users and 17 community members for focus groups and interview	Survey, interview and focus groups	"Across subject groups, regardless of level of exposure to personally controlled health record technology, sex, age, and social role (student or employee), we found high levels of willingness to share personal health information from a PCHR with public health agencies for purposes of disease monitoring, evaluation, and needs assessment."	Positive, but conditional
Wicks, Paul, Michael Massagli, Jeana Frost, Catherine Brownstein, Sally Okun, Timothy Vaughan, Richard Bradley, and James Heywood. 2010. 'Sharing Health Data for Better Outcomes on PatientsLikeMe'. Journal of Medical Internet Research 12 (2): e19. doi:10.2196/jmir.1549.	to understand patient attitudes to sharing their health data online (among other study aims)	Online community	1323 members of the PatientsLike Me website, with either MS, PD, ALS, fibromyalgia, HIV or mood disorders	Survey	"Patients who opt to join the site are, by and large, already comfortable with the notion of sharing their health data when they join […] Those patients with the most serious illnesses were most comfortable with sharing, suggesting that patients are making risk/benefit analyses about sharing their health data and taking prognosis into account."	Positive
Zalin, A., C. Papoutsi, K. Shotliff, A. Majeed, C. Marston, and J. Reed. 2016. 'The Use of Information for Diabetes Research and Care: Patient Views in West London'. Practical Diabetes 33 (3): 81–86A. doi:10.1002/pdi.2008.	To understand the views of people with diabetes regarding access to electronic health records for healthcare and research.	UK	404 patients with diabetes from recruited from outpatient clinics or GP surgeries. (Separate recuitment of patients for focus	Survey and focus groups	"a large majority (79,4%) of people with diabetes would allow their EHRs to be used for research"	Positive, but conditional

			groups, unclear N)			
Mahlmann, L., Schee Gen Halfmann, S., von Wyl, A., Brand, A., 2018. Attitudes towards Personal Genomics and Sharing of Genetic Data among Older Swiss Adults: A Qualitative Study. Public Health Genomics. https://doi.org/10.1159/000486588	To assess the willingness of older Swiss adults to share genetic data for research purposes and to investigate factors that might impact their willingness to share data	Switzerland	40 members of a continuing education programme for seniors	Interviews	"older citizens are willing to share their data for research purposes. However, most of them will only contribute if their data is appropriately protected and if they trust the research institution to use the shared data responsibly."	Positive, but conditional
Sheikh, Z., Hoeyer, K., 2017. "That is why I have trust": unpacking what 'trust' means to participants in international genetic research in Pakistan and Denmark. Med Health Care and Philos 1–11. https://doi.org/10.1007/s11019-017-9795-9	To understand how participants in collaborative international genetic research think about trust and the relationships in which they provide biomaterial and health data	Pakistan and Denmark	42 research participants having biological material collected by a single laboratory (Denmark: 23 people with balanced chromosomal rearrangements identified through public registers.	Interviews	"when participants discuss trust they are trying to shape their relationship to researchers while simultaneously communicating important hopes, fears and expectations"	n/a

			Pakistan: 19 people with autosomal recessive disorders identified primarily through researchers' personal contacts and snowball sampling.)			
Harle, C.A., Golembiewski, E.H., Rahmanian, K.P., Krieger, J.L., Hagmajer, D., Mainous, A.G., Moseley, R.E., 2018. Patient preferences toward an interactive e-consent application for research using electronic health records. J Am Med Inform Assoc 25, 360–368. https://doi.org/10.1093/jamia/ocx145	To assess patient views towards an interactive electronic consent application, when giving broad consent for research using identifiable EHR data	Florida, USA	32 community members who had previously agreed to be approached for future research studies	'Think aloud' Interviews - "Think-aloud interviews simulate a task while asking users to constantly verbalize their thoughts and decisions"	"this study provides preliminary support for the value of electronic applications with interactive features that allow patients to customize their consent experience. At the same time, this study suggests a need to support people who have reservations about electronic consent platforms as well as the importance of communicating information about administrative processes and safeguards that protect personal health information when used in research."	Positive

Mazor, K.M., Richards, A., Gallagher, M., Arterburn, D.E., Raebel, M.A., Nowell, W.B., Curtis, J.R., Paolino, A.R., Toh, S., 2017. Stakeholders' views on data sharing in multicenter studies. Journal of Comparative Effectiveness Research 6, 537–547. https://doi.org/10.2217/cer-2017-0009	To understand stakeholders' views on data sharing in multicenter comparative effectiveness research studies and the value of privacy-protecting methods.	USA	34 stakeholders (15 patients, 19 other organisational stakeholders)	Individual and group interviews	"stakeholders are open to data sharing in multicenter studies if the research offers benefits and value to patient care, minimizes data security risks, and can be done at reasonable cost."	Positive, but conditional
Ostherr, K., Borodina, S., Bracken, R.C., Lotterman, C., Storer, E., Williams, B., 2017. Trust and privacy in the context of user-generated health data. Big Data & Society 4, 2053951717704673. https://doi.org/10.1177/2053951717704673	To explore trust and privacy in the context of user-generated health data	USA	32 adults, (9 researchers, 6 start-up employees, 17 members of general public approached in a park)	Interviews	"Members of the general public expressed little concern about sharing health data with the companies that sold the devices or apps they used, and indicated that they rarely read the ''terms and conditions'' detailing how their data may be exploited by the company or third-party affiliates before consenting to them"	Positive

Annex II. Focus group schedule

Schedule for the ROADMAP consultation involving people with dementia and carers/supporters

09.00 – 09.10 Welcome to the consultation, special welcome to Andrew (Helen).

09.10 – 09.25 How the consultation today will be organised (Dianne)

The aim of the consultation today is to explore your opinions and feelings towards the sharing of health data in the context of the creation of a real world platform for Alzheimer's disease. It has been organised by the work package on the ethical, social and legal implications of the ROADMAP project, which is led by Dr Andrew Turner. He will give you some background information about this whole topic and answer any questions you may have about this consultation before asking you to sign the consent form and starting the actual discussion.

The discussion will be structured around the presentation of vignettes and questions that probe opinions about the hypothetical sharing of health data. With your permission, we will make a sound recording to ensure that we don't miss anything that is said. [check everyone OK about that]

The use of short stories (vignettes) to describe possible situations involving the sharing or re-use of health data will hopefully enable you to focus on the topics of discussion and get you thinking about what the key issues might be and how you feel about them. Don't worry about whether you will have something to say. No prior knowledge is needed and once the discussion starts, you may have some ideas. You may find that you agree or disagree with some of the things that other people say. This is equally important for us to know. Different views and opinions don't mean that one person is right and the other is wrong. It's important to capture different perspectives. We are interested in all ideas and suggestions.

We will be splitting up into two groups: a group of carers/supporters and a group of people with dementia (including Nelida to provide translation for Idalina). For each group will have two moderators Andrew and Sébastien for the carers/supporters group and Ana and Dianne for the people with dementia group.

We hope you enjoy taking part in this consultation but if at any time you wish to leave, you are free to do so and do not have to explain to anyone why.

09.25 – 09.55 Information about data sharing, questions and signing of the informed consent form (Andrew)

A brief overview of the ROADMAP project, highlighting the re-use of existing data from multiple sources.

09.55 – 10.20 Warm-up exercise (whole group together) (Dianne and Ana)
Before reading the vignettes and starting the discussions, we would like you to think about (5 minutes):

- what "health data" means to you.
- what "data sharing" means to you.

Now, we would like each of you to discuss (with the person who is sitting next to you) what health data and data sharing means to each of you. You have 10 minutes to discuss this, (remind the group that there is no right or wrong responses) and write on the yellow post-its a few words about what each of the terms means to you both. If it helps, you could perhaps also include examples of what you consider "health data" to be and what you would not consider as "health data". If you do not share the same understanding, use the pink post-it to explain the differences in understanding.
Sebastien to collect all the post-its and paste them on the flipchart.
Dianne and Ana to make some comments about the post-its and let people talk about whether they found it difficult to describe / define what these concepts mean and what they found most challenging about them.
10.20-10.40 –Coffee/tea break
10.40– 12.30 (if necessary the moderators could suggest a 10-minute break)
10.40-11.10 Discussion 1: Sensitivity of data

Explain that this first discussion is about the re-use of data, in particular about how people feel about information obtained (e.g. during their individual care) being passed on to researchers.
Remind them that we are asking this because ROADMAP is about the secondary use of data and how 'real world' data will be combined in the ROADMAP project (e.g. the anonymous combination of health records (data from individual care), clinical trials and epidemiological studies (data from research studies).

Vignette 1: Mavis and Cynthia are both in their eighties and have gone to their local doctor's surgery for their annual blood pressure and blood tests. They go together now because Cynthia has mild dementia and has difficulty finding her way around. When she comes back from giving blood, Mavis complains to Cynthia that the nurse wanted her to sign a document agreeing to the results of her blood test being used for research. Cynthia says she wouldn't mind, adding, "Anything for a quiet life!" but Mavis feels uneasy about giving people access to her test results. Cynthia is very much in favour of research and doesn't really understand why Mavis is concerned. The two ladies start discussing the whole issue of whether it is OK for researchers to obtain samples and test results in this way.

What do you think about Mavis and Cynthia's reactions to the issue of data sharing?

- Questions and prompts
    - *Should this kind of information be shared? Why/why not?*
    - *Are there any kinds of data that are particularly sensitive? If so, which ones?*
    - *What, if anything, do you worry about happening when your data is shared?*

- *Why? How would this affect you or others?*
- *What do you think the main benefits are to data sharing?*
- *What do you think about the combination of data to build a more detailed picture about the 'real world'?*

11.10-11.40 Discussion 2: Oversight of data sharing

Explain that this discussion is about the kinds of safeguards and mechanisms that are or should be in place to control who can access data and for what purposes it should be possible to use it.

Vignette 2: The nurse overhears their discussion and reminds Mavis that she is not in any way obliged to consent to sharing her test results but that if she does, her test results would in any case be anonymised. So it would not be possible for the researcher to know that they are hers. Later, Mavis discusses this with her son, Bob. He points out that all kinds of data about people's lives are shared nowadays (e.g. where you shop, what you buy, how often you use public transport etc.). He points out that anonymising health data is one safeguard to protect the interests of the people providing it. Mavis and Bob start to discuss whether this is sufficient and what else, if anything, might be needed.

So, the question we'd now like to ask you is:

- *How well do you think/feel your health data is protected?*
- *What, if anything, could be done to make data sharing more acceptable/secure?*

  (Make a list on the flip chart)
- *And should there be any limits on who or what kind of organisation should be able to access such data? Prompt: Why?*
- *Should there be limits on the ways that data can be used by other people or organisations? Prompt: Why?*

11.40 -12.10 Discussion 3 – Consent and engagement

Explain that this discussion is about whether and if so in what circumstances it should be possible to re-use patient data without the prior consent of the patient concerned.

"Durham General" is a teaching hospital which also has links to external research institutes. The Board of the hospital has just adopted a data sharing policy which will soon be implemented. It establishes the conditions (including the application of necessary safeguards) under which information, collected in the course of individual treatment of patients, can be shared with researchers. It also establishes the conditions which must be fulfilled for such data to be shared without the prior consent of the patient concerned and encourages transparency about how data collected in this way is used.

- How would you feel about new research being proposed that wanted to re-use Mavis's test results for an additional study (without asking for additional consent)?

- If the safeguards we agreed on before were in place (show the flip chart to remind everyone), would you approve of your anonymous data being shared without prior consent?
    - Why?
    - Are there any exceptions (e.g. linked to situations or types of data) where you would nevertheless expect to be asked to consent to the re-use of your health data?
    - Are there any circumstances where you would consider it unnecessary to ask for consent to sharing of your health data (i.e. even if not anonymised)? Or are we always assuming some degree of anonymisation? If so, this question would already have been answered by the first/main question above.

- Would you like to be informed about the research that is done using your anonymous data?
    - Is this reasonable to expect?
    - What would you like to know?
    - How would you like to be informed?
    - How regularly?

12.10 -12.30 Discussion 4 – Public interest/benefit to society

The deal between the NHS Hospital “Royal Free” and DeepMind (a company owned by Google) first became public in February 2016 and caused controversy over the amount of patient information being shared without public consultation.

- Information from 1.6 million patients in the Hospital was used to develop an alert, diagnosis and detection system that can spot when patients are at risk of developing acute kidney injury.
- DeepMind gained access to sensitive patient information such as HIV status, mental health history and abortions.
- The Royal Free did not tell patients that Google's DeepMind would have access to such information. DeepMind insists that it has never shared patient data with parent company Google.

The result was an app called Stream and looks for acute kidney injury, which affects up to 18 per cent of those admitted to hospital.

- ***Question: What do you think about what happened here?***
- ***We then read the following text and ask if this makes any difference?***

Afia Ahmed, 38, suffered complications following the delivery of her daughter by emergency Caesarean at the Royal Free hospital.She developed the whole-body infection sepsis during labour, which led to acute kidney injury. This was detected by the Streams app, which then sent text alerts to specialist kidney doctors, allowing them to intervene quickly. This saved her life.

12.30-13.00 Brief feedback from the 2 groups, main conclusions from the day and how this relates to the ROADMAP project (Andrew)

13.00 Lunch at the hotel

Annex III. Consent forms

EWGPWD MEMBER CONSENT FORM

Ethics Approval Reference: R53988/RE001

ROADMAP
Real world Outcomes across the AD spectrum for better care: Multi-modal data Access Platform

Purpose of Study: What concerns are raised by re-using and combining data to create a real world evidence platform for AD research?

		Please initial each box
1	I confirm that I have read and understand the information sheet for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.	☐
2	I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason, and without any adverse consequences.	☐
3	I understand that research data collected during the study may be looked at by designated individuals from the University of Oxford where it is relevant to my taking part in this study. I give permission for these individuals to access my data.	☐
4	I understand that this project has been reviewed by, and received ethics clearance through, the University of Oxford Central University Research Ethics Committee.	☐
5	I understand who will have access to the recording and transcript of the focus group, how this data will be stored and what will happen to the data at the end of the project.	☐
6	I understand how this research will be written up and published.	☐
7	I understand how to raise a concern or make a complaint.	☐
8	I consent to being audio recorded.	☐
9	I understand how audio recordings will be used in any reports resulting from the research.	☐
10	I agree to take part in the above study.	☐
Optional:	I agree for research data collected in this study to be given to researchers, including those working outside of the EU, to be used in other research studies. I understand that any data that leave the research group will be anonymised so that the risk of being identified is as low as possible.	☐

Name of Participant Date Signature

Name of person taking consent Date Signature

SUPPORTER/CARER CONSENT FORM

Ethics Approval Reference: R53988/RE001

ROADMAP

Real world Outcomes across the AD spectrum for better care: Multi-modal data Access Platform

Purpose of Study: What concerns are raised by re-using and combining data to create a real world evidence platform for AD research?

		Please initial each box
1	I confirm that I have read and understand the information sheet for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.	
2	I understand that my participation is voluntary and that I am free to withdraw at any time, without giving any reason, and without any adverse consequences.	
3	I understand that research data collected during the study may be looked at by designated individuals from the University of Oxford where it is relevant to my taking part in this study. I give permission for these individuals to access my data.	
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Optional:	I agree for research data collected in this study to be given to researchers, including those working outside of the EU, to be used in other research studies. I understand that any data that leave the research group will be anonymised so that the risk of being identified is as low as possible.	

Name of Supporter/Carer Date Signature

Name of person taking consent Date Signature

Annex IV. Participant Information Sheets

ROADMAP
Real world Outcomes across the AD spectrum for better care: Multi-modal data Access Platform

INFORMATION SHEET FOR EWGPWD MEMBERS

Ethics Approval Reference: R53988/RE001

Thank you for taking the time to read this information sheet. It provides information about the scope of the study, and what your participation would involve.

You are being invited to take part in a research study conducted by the University of Oxford and Alzheimer Europe. The study involves a focus group where we will ask you about your views on re-using medical data for research into Alzheimer's disease.

Participation in the study is entirely voluntary.

1. *Background and aims of the study*

This study hopes to answer the following question: What concerns are raised by re-using and combining data to create a real world evidence platform for AD research?

Real world evidence is a term for information about the effects of medical treatments that is collected outside of medical trials. For example, it includes information collected from people's electronic health records or from studies that observe people's health over extended periods of time.

This study is funded by the Innovative Medicine's Initiative, which is a partnership between the European Union and the pharmaceutical industry.

2. *Why have I been invited to take part?*

You have been invited because you are a member of the European Working Group of People with Dementia.

3. *Do I have to take part?*

No. You can ask questions about the study before deciding whether or not to participate. If you do agree to participate, **you may withdraw from the study at any time**, without giving a reason and without penalty. To withdraw, please advise the researchers of this decision.

4. *What will happen in the study?*

If you agree to take part in the study, you will be asked to participate in a focus group discussion. In the focus group, you will be invited to share concerns you may have about re-using and combining medical data on a digital platform to help with research into Alzheimer's disease.

The focus group will take place at the Alzheimer Europe consultation event on December 7, 2017. It should take approximately 3 hours and there will be regular breaks. There will not be any follow-up, but you will be informed about the outcomes of the study.

Please treat the focus group with the same level of confidentiality as other EWGPWD consultation events. You are free to discuss information from the focus group elsewhere, but please do not disclose views or comments made by other participants.

The discussion will be audio recorded.

5. Are there any potential risks in taking part?

If you feel uncomfortable with any of the topics discussed or you have had enough of the discussion, then you can take a break or withdraw at any time. You can withdraw without giving a reason and without penalty, by advising the researchers of this decision.
Every effort will be made to ensure the risk of you being identifiable in any published reports is very low, however the EWGPWD is a small group so this cannot be guaranteed.

6. Are there any benefits in taking part?

There will be no direct benefit to you from taking part in this research.

7. What happens to the data provided?

The audio recording will be transcribed. The recording and transcript are sensitive data. This sensitive data will be:

- Securely shared with, and accessed by, the transcriber and the research team at the University of Oxford and Alzheimer Europe.
- Encrypted and stored confidentially using secure file storage at the University of Oxford and Alzheimer Europe.

From the transcript, your responses will be anonymised. The anonymised transcript will have directly identifying information removed or replaced with pseudonyms (e.g. names and places). The content of the transcript will be reviewed for sensitive information and edited appropriately. The original transcript and audio recording will be retained by the University of Oxford and Alzheimer Europe. The anonymised transcript:

- Will be securely shared with the research team at the University of Oxford and Alzheimer Europe.
- May be shared with and made accessible to other project collaborators.
- Will be used for analysis and anonymous quotes in publications.
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All sensitive data and research data will be stored by the University of Oxford and retained for at least 3 years after publication or public release of work resulting from this research. Alzheimer Europe will also keep a copy of the data for the same period of time and will act as the contact point for any future questions or requests about it.

If you have any questions about data stored at Alzheimer Europe, please contact Dr Dianne Gove. If you have any questions about data stored at the University of Oxford, please contact Dr Andrew Turner. Their details are provided at the end of this document.

8. Will the research be published?

The research will be published in reports for the ROADMAP project and also in articles for scientific journals. A summary report will be produced for everyone who took part in the focus group.

Only anonymised quotes will appear in publications. Every effort will be made to ensure the risk of you being identifiable is very low, however the EWGPWD is a small group so this cannot be guaranteed.

9. *Who has reviewed this study?*

This study has been reviewed by, and received ethics clearance through, the University of Oxford Central University Research Ethics Committee (reference number: R53988/RE001).

10. *Who do I contact if I have a concern about the study or I wish to complain?*

If you have a concern about any aspect of this study, please speak to Dr Andrew Turner (andrew.turner@oii.ox.ac.uk, 01865 212329) or Professor Luciano Floridi (luciano.floridi@oii.ox.ac.uk, 01865 287202) at the University of Oxford, or Dr Dianne Gove (dianne.gove@alzheimer-europe.org) at Alzheimer Europe, who will do their best to answer your query.

They should acknowledge your concern within 10 working days and give you an indication of how they intend to deal with it. If you remain unhappy or wish to make a formal complaint, please contact the relevant chair of the Research Ethics Committee at the University of Oxford who will seek to resolve the matter in a reasonably expeditious manner:

Chair, **Social Sciences & Humanities Inter-Divisional Research Ethics Committee**; Email: ethics@socsci.ox.ac.uk; Address: Research Services, University of Oxford, Wellington Square, Oxford OX1 2JD

11. *Further Information and Contact Details*

If you would like to discuss the research with someone beforehand (or if you have questions afterwards), please contact:

Andrew Turner
Digital Ethics Lab
Oxford Internet Institute
41 St Giles
Oxford
OX1 3LW
Tel: 01865 212329
Email: andrew.turner@oii.ox.ac.uk

Dianne Gove
Alzheimer Europe
14, rue Dicks
L-1417 Luxembourg
Email: dianne.gove@alzheimer-europe.org

ROADMAP

Real world Outcomes across the AD spectrum for better care: Multi-modal data Access Platform

INFORMATION SHEET FOR EWGPWD SUPPORTERS/CARERS

Ethics Approval Reference: R53988/RE001

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Participation in the study is entirely voluntary.

12. Background and aims of the study

This study hopes to answer the following question: What concerns are raised by re-using and combining data to create a real world evidence platform for AD research?

Real world evidence is a term for information about the effects of medical treatments that is collected outside of medical trials. For example, it includes information collected from people's electronic health records or from studies that observe people's health over extended periods of time.

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13. Why have I been invited to take part?

You have been invited because you are a member of the European Working Group of People with Dementia.

14. Do I have to take part?

No. You can ask questions about the study before deciding whether or not to participate. If you do agree to participate, **you may withdraw from the study at any time**, without giving a reason and without penalty. To withdraw, please advise the researchers of this decision.

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The focus group will take place at the Alzheimer Europe consultation event on December 7, 2017. It should take approximately 3 hours and there will be regular breaks. There will not be any follow-up, but you will be informed about the outcomes of the study.

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The discussion will be audio recorded.

16. *Are there any potential risks in taking part?*

If you feel uncomfortable with any of the topics discussed or you have had enough of the discussion, then you can take a break or withdraw at any time. You can withdraw without giving a reason and without penalty, by advising the researchers of this decision.
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