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Dynamic Consent: a mechanism for engagement.

A thesis submitted for the degree of

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by

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Thesis abstract

This research emphasises that the burden of communication lies with data-collectors rather than the people who are asked to provide personal information. The research questions addressed in the thesis concern a service review of one study's implementation of a dynamic form of consent over time, rather than presenting research participants with a one-off decision. This thesis argues, based on a critical review of previous work, that the value of engagement is underdeveloped and underexplored. Three studies are carried out in collaboration with an online study collecting survey data on rare diseases (RUDY). Findings suggest that developing engaging data-practices offers a return on investment. In other words, participant engagement enriches data collection in the study under test and there are further steps that could be taken.

Study 1 consisted of interviews with participants, and a focus group with researchers. While researchers report user experience and engagement as design priorities, most participants could not recall any information about the project. Participants indicate that the type of feedback they want should be interesting, provide knowledge and allow further opportunities for active participation. Study 2 involved a series of focus groups with RUDY participants, exploring how an intervention study could enhance the feedback provided. This provided a specification for 'enhanced feedback': participants wanted to know project aims, what data was collected, how that data was used, the value of their contributions, and research outcomes. Participants wanted this information to be communicated in a way they could understand, free of technical jargon. The resulting specification was used to make changes to the project website, online portal and reminder emails.

A pre-test/post-test intervention study [n=1,936] showed two benefits to making information about a research study more accessible and relevant to members of the public: increased retention and a reduced drop-off in participation over time. From 2019 to 2020, questionnaire completions increased by 5% and questionnaire drop-off fell from 32% to 8.9%. Those who collect, control, use and share personal data have responsibilities that they must, but do not often, demonstrate publicly. Public demonstration of responsibility is important it is known to build trust, as illustrated in the literature review. A tool-kit is provided to help researchers build engagement into their own data-practices, tools which also serve as design prompts to stimulate conversations around how to demonstrate trustworthiness in research projects.

The thesis makes several contributions: theoretical, methodological, empirical and practical. This work demonstrates a need for researchers to describe how they will engage with participants over time about the importance of their data-contribution. Co-design is used to validate the research direction of this work, ensuring its relevance and impact. The importance of engagement is indicated, and justified empirically. This work demonstrates that engaging data-practices are just as valuable to those who collect data as they are to those providing it.

Declaration of Authorship

I declare that this thesis is entirely my own work, and except where otherwise stated, describes my own research.

Research papers published over the course of this DPhil relating to this thesis:

I was the primary author of each output listed below.

The conceptual leaps in "**Dynamic Consent in Cybersecurity…**", which have since been heavily refined, are evident in the literature review where responsibility for data protection is accorded to experts rather than patients, participants and other laypeople. I tried to add 'persistence' as a third aspect of Dynamic Consent (in addition to 'revocation' and 'engagement'), but this idea actually matured into "**Wider Research Applications…**". This output reports the results of a workshop I used to validate the findings from the focus group and interviews reported in chapter 4, and feeds into the discussion section for that chapter. "**Why We Trust Dynamic Consent…**" is of particular note, as this was the first time I had consolidated Dynamic Consent literature into a specification. This was an early version of the tool-kit presented in chapter 7. Conceptually, this output also tries to develop a practical approach to privacy, leading to the 'modelling privacy requirements' section in chapter 2.

- ⇨ Schuler Scott, Arianna & Goldsmith, Michael & Teare, Harriet & Webb, Helena & Creese, Sadie. (2019). Why We Trust Dynamic Consent to Deliver on Privacy. 10.1007/978-3-030-33716-2_3.

Overview: Dynamic Consent offers data controllers a way to show compliance with individual privacy preferences, and data subjects with control as and when they require it. We must re-think how consent is implemented to develop useful privacy controls.

- ⇨ Schuler Scott, Arianna & Goldsmith, Michael & Teare, Harriet. (2019). Wider Research Applications of Dynamic Consent. 10.1007/978-3-030-16744-8_8.

Overview: as research processes change due to technological developments in how data is collected, stored and used, so must consent methods. We explore wider research problems that Dynamic Consent could help address: research design, expectation management and trust.

- ⇨ Schuler Scott, A., Goldsmith, M., Teare, H., Creese, S., & Kaye, J. (2018). Dynamic Consent in Cybersecurity for Health. International Conference on Health Informatics and Medical Systems, CSREA Press.

Overview: patient care is moving away from traditional models of medical care but patient-centred approaches cannot put the responsibility for data protection directly onto patients. Those who control and process information are better placed to leverage the appropriate risk controls.

Workshops and talks developed and delivered over the course of this DPhil relating to this thesis:

- ⇨ "Protecting Data as a Software Developer".

Overview: a two-hour online workshop introducing secondary school students to concepts of social, ethical, and legal software requirements. This workshop was designed to be fun and to develop critical thinking and reflection skills, running several times due to popular demand.

- ⇨ "RE: I've been forced to sign this and I am not happy".

Overview: a 20-minute talk addressing the recent upheaval in European data-handling and its impact on privacy awareness.

- ⇨ "Protecting Ourselves Online: Why We Shouldn't Take Cookies from Strangers".

Overview: a 20-minute public engagement exercise focusing on how data-use has developed and institutional responsibility.

- ⇨ "Designing Responsible, Participatory Research".

Overview: a 15-minute talk to postgraduate students providing an overview of methods used in this thesis, suggestions as to how others could start doing so.

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Grateful thanks to the tax-paying British public, without whose support this work would never have been possible.

☙ ✝ ❧

In memory of Elizabeth Scott.
Friend, Gran and all-round excellent example.

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Chapter 1

Thesis introduction

"Therefore, while dynamic consent is currently a biobanking project, it is also an approach or concept that is being applied more broadly, not only within health but also in other fields", (Kaye *et al*, 2015).

1.1 Introduction

Dynamic Consent was originally conceived to empower individual's choices (Kaye, 2011; Casassa Mont *et al*, 2012; Whitley *et al*, 2012; Kaye *et al*, 2015); rethinking most biomedical research processes, but especially informed consent (Manson & O'Neill, 2007). In designing a methodology allowing participants to re-revisit choices made during the informed consent process, researchers can re-use and share data with consent and confidence, maintain a record of data-use for auditing purposes, and recruit for further studies.

There is a deep literature, of many threads, voicing the nature of consent to support patients to volunteer to participate in biomedical research, why it is sought, its implications for privacy, and the safeguarding role of experts as facilitators. This work focuses on how consent processes are implemented. A typical research situation, as we will discuss, is that a participant is presented with pages of information and then asked to tick 'I agree' or 'I do not agree' to the use of data. This is usually a one-time decision, and participants do not see the impact of this data-sharing agreement. As we will explore,

there are benefits for those in 'expert' roles (e.g., researcher and doctor) when they seek information from so-called 'non-experts' (e.g., participants and patients). There is information that can be gathered by offering options other than simply 'yes' or 'no'. Indeed, a key finding of this work is that those often relegated to 'non-expert' roles when seeking support, advice or services, are sources of considerable expertise. Dynamic Consent offers one way to leverage this expertise.

Ensuring Consent and Revocation (EnCoRe) was a project that aimed to give individuals more control over personal data, using consent as a way to do so. EnCoRe sought to explore data control in terms of a person's ability to take back (i.e., 'revoke') permission; providing an approach to consent in which the power dynamic between those seeking data and those providing it, was more balanced (Whitley, 2009). Researchers wanted people who used technology to be able to say yes or no to their personal data being used, as easily as turning a tap on or off. They constructed data-sharing options to highlight, and accommodate, more nuanced data-sharing choices (Agrafiotis *et al*, 2010a). The theory of Dynamic Consent was developed in response to initial discussions within EnCoRe (EnCoRe, 2008; Casassa Mont *et al*, 2012) which identified privacy requirements within three use cases: an employer needing to protect personal data, identity assurance and biobanking (Casassa Mont *et al*, 2010; Casassa Mont, Sharma, *et al*, 2011; Bramhall *et al*, 2011).

The subsequent literature on Dynamic Consent tackled problems with the general approach to consent employed in biobanking (Curren & Kaye, 2010; Kaye, 2011; Casassa Mont *et al*, 2012). This work offers a fourth use case for Dynamic Consent; that of an online longitudinal research study. Continuing EnCoRe's premise, that consent must be able to be re-visited over time ('dynamic'), if it is to be fit for purpose within online research, this work focuses on how choice is communicated. Rather than focus on a technical architecture, as EnCoRe showcased, I investigate the cultural change required for implementing Dynamic Consent (Kaye *et al*, 2015); a change that requires technology developers to seek feedback from users as well as their personal data. After EnCoRe, Dynamic Consent theory detailed "a new approach for engaging individuals about the use of their personal information" (Kaye *et al*, 2015) that built public trust. Biobanks (research institutions collecting biological samples) offered no information to donors and delegated decisions to unseen experts. Dynamic Consent researchers (namely Kaye *et al*) interviewed biobank stakeholders to identify specific problems posed by these practices: participants wanted to learn about the research process, researchers wanted to be able to (re)use samples more easily, and policy-makers wanted a more innovative approach to secondary and future uses of data.

While proponents of Dynamic Consent posit that the theory offers a solution to these problems and their ilk, there is a distinct lack of service evaluation work and tools to support Dynamic Consent's development. As we will discuss, research papers reporting the use of Dynamic Consent often overlook engagement, and those who mention it do not prioritise it. This is a significant oversight; engagement is one of Dynamic Consent's two key tenets, the other being 'revocation (of consent)'. We will also see that existing evaluation tools are unwieldy and difficult to translate into practical research requirements. This work describes 'engagement' in practice and equips other researchers to build and report engagement as part of their own longitudinal research studies online. In addition, it provides insight as to why participants trust researchers, and some of the assumptions regarding data-use on which this trust rests.

1.2 Problem statement

Research participants have more to offer researchers than their data alone, but invaluable input is often left unsought; such expertise can make research more relevant and useful, help define what is ethically acceptable, and improve the participant experience of a study. The informed consent process is an opportunity to start a (two-way) conversation.

While medical research is the context for this work, there is a wider application – where a relationship can be described by an 'expert' seeking information from a 'non-expert'. The problem in this case is similar to the above. Without building processes to seek input from non-experts, experts may never have the opportunity to re-think or re-shape existing practices.

A university researcher hoping to collect data from human participants must subject a plan of their work for ethical approval. Ethical review is important in ensuring public trust and support, but may be restrictive to researchers. One response to such constraints would be for the researcher to draft a broad participant consent agreement (Whitley *et al*, 2012):

> "…one coping strategy was to 'make our ethics as broad as possible because we are trying to give ourselves a bit more room to manoeuvre without actually having to go and fill in another form… they cover themselves absolutely for everything and it doesn't necessarily help the patient to give informed consent'"

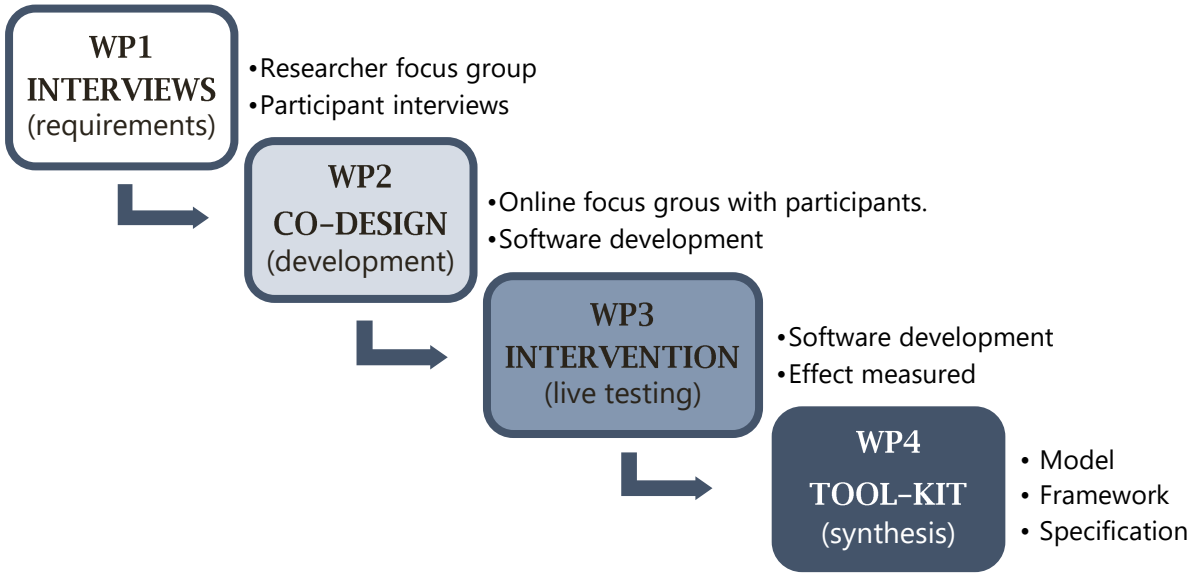
Default consent options must offer a basic level of protection because research participants place themselves in a position of vulnerability when taking part in research. Researchers are responsible for how consent options are designed and presented, acting as "choice architects" (Thaler & Sunstein, 2008).

As research moves online, researchers must be aware of fundamental issues with informed consent that have become magnified in these spaces: online consent options tend to overload readers with information (Obar & Oeldorf-Hirsch, 2020), and are designed to coerce the reader into making immediate judgements without understanding the information provided (Millett *et al*, 2001). Researchers must communicate information about a study to potential participants in a way they understand, so that consent is voluntary informed and un-coerced. If these issues are ignored, there is a risk that informed consent, a process put in place to protect citizens from harm, is instead protecting the institutions with the capacity to cause these harms. Without researchers advocating for, and prioritising, the participant experience, then informed consent practices are unfit for purpose. Rather than resign ourselves to this as fact, the problem statement of this thesis is that we must address the problem of one-time consent in online spaces head on, and work towards re-designing these interactions.

1.3 Research overview

Figure 1 illustrates the work packages carried out over the course of this project; each work package is a study described in its own thesis chapter. Work package 1 (WP1) describes interviews with RUDY participants and a focus group with the research team, to understand Dynamic Consent looked like in practice. This informed WP2, a series of online focus groups where participants were asked what they wanted to know about the study. I returned to the RUDY team with these findings and, with permission from the project lead, I worked with the software developers to implement, test and refine changes to the website, online research portal, and email notifications. The impact of these changes was measured using a pre/post-intervention service evaluation, described in WP3. Questionnaire responses were compared at time points before and after the changes were made, to observe any change in participation. WP4 describes a suite of tools developed to support researchers in developing or enhancing feedback processes within their own work.

Figure 1: Overview of research work packages.



1.4 Research aims

This thesis investigates the theory of Dynamic Consent in practice to discover limitations, provide a more realistic assessment of what Dynamic Consent can be used to achieve, and provide empirical evidence to justify its application. Research participants can be unaware of precisely how their data is collected, used and shared for research purposes while still being eager to contribute (Simon *et al*, 2011). The 'informed' consent process can involve vague descriptions of the study ahead, and it is at this point that participants are asked to absolve researchers of any potential negative consequences that their work may have(Manson & O'Neill, 2007). Research participants may agree to requests under the 'therapeutic misconception' that researchers will act in their best interests (Appelbaum *et al*, 1987; Corrigan, 2003), without requiring any evidence.

Dynamic Consent encourages researchers to design their methodologies in such a way as to inform participants over time, equipping them with information or access to resources they can later rely on to participate on their own terms. This work extends the existing body of knowledge by providing a richer account of Dynamic Consent within an online research platform. A key focus is to investigate how technology can be, and is, used to support Dynamic Consent. The position of this thesis is that consent is normative, individuals have different preferences and may choose to share personal data in different ways. Rather than making a single recommendation for the 'best' consent practices, emphasis is placed on a need to accommodate genuine consent that is informed over time.

The three research aims of this thesis are:

- ➔ RA1: to observe and evaluate an existing implementation of Dynamic Consent, identifying conflicts and similarities with existing academic literature.
- ➔ RA2: to extend the theory of Dynamic Consent: provide empirical evidence to strengthen a research requirement for engagement.
- ➔ RA3: to develop practical guidelines for researchers who want to implement a dynamic form of consent in their own work.

Each aim has corresponding research questions, as illustrated in Table 1 below.

Table 1: Research aims (RA) and corresponding research questions (RQ).

RA1: to observe and evaluate an existing implementation of Dynamic Consent, identifying conflicts and similarities with existing academic literature.
⇨ RQ1a: what similarities does RUDY's Dynamic Consent implementation bear to the Dynamic Consent literature?
⇨ RQ1b: how does RUDY's Dynamic Consent implementation differ from the Dynamic Consent literature?
RA2: to extend the theory of Dynamic Consent: provide empirical evidence to strengthen a research requirement for engagement.
⇨ RQ2a: is research participation affected by how information is communicated to research participants?
⇨ RQ2b: what is the relation between participant engagement and the richness of longitudinal research data?
RA3: to develop practical guidelines for researchers who want to implement a dynamic form of consent in their own work.
⇨ RQ3a: how can Dynamic Consent be described, implemented, measured and reported in research?
⇨ RQ3b: what kind of tools can support these processes and accommodate knowledge-sharing among researchers implementing, or trying to implement, Dynamic Consent?

1.5 Collaborators

The Rare and Undiagnosed Diseases studY (RUDY) launched in 2014, is an online study in rare genetic conditions such as vasculitis (inflammation of blood vessels), fibrous dysplasia (where scar-like tissue grows instead of bone), myeloma (cancer of the plasma cells), hypermobility and brittle bones. These conditions often restrict physical or mental capacity, as well as life expectancy (Schieppati *et al*, 2008), and are under-researched due to the scarcity of data from which to draw meaningful conclusions. RUDY is an example of best practice due to the way the research process is amended to reflect participant suggestions (Watts *et al*, 2016).

RUDY has recruited over 3,400 participants to their online platform as of January 2022[1], and participants are asked to submit questionnaires every six months. The questionnaires received provide data regarding quality of life and daily function, as well as clinical information (Ponte *et al*, 2015). This data is vital in understanding the lived experiences of people with rare conditions (Orlando *et al*, 2020). RUDY is an online research platform, with three traits that differentiates it from other databases and registries: an online self-registration process, an implementation of Dynamic Consent (Teare *et al*, 2017), and participant data-entry. This form of data-entry saves in-clinic and research time and has resulted in improved recruitment and retention overall (Javaid *et al*, 2016).

There is a risk, in recruiting people with rare conditions, that only the most severely affected individuals are included, since they are recruited as soon as they are diagnosed. RUDY presents an opportunity to collect a range of experiences providing insight into the quality of life for people with rare conditions (Zhang *et al*, 2016). A patient forum was created to involve participants in the research design process (Watts *et al*, 2016). RUDY uses the term 'patient' rather than participant because all participants either have, or are the primary caregiver (e.g., a parent) of someone with, a rare condition. I will use 'participant' in this thesis because research participation, rather than healthcare status, is the subject of study. For many RUDY participants, these terms are interchangeable.

The reason that Dynamic Consent has been implemented as part of RUDY is that the research team prioritises participant engagement. Their aim is to develop an on-going two-way dialogue between participants and researchers as standard practice, where participants can securely view their consent options online at any time (Javaid *et al*, 2016). RUDY offers a rare opportunity – a project implementing Dynamic Consent that has been running for many years, with the same research team.

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[1] As indicated on the project website: https://research.ndorms.ox.ac.uk/rudy.

1.6 Key findings

Each of the studies described in this thesis is presented as a separate chapter. Key findings from each of these are as follows:

Chapter 4 describes interviews with RUDY participants, and a focus group with the research team who had developed RUDY. Researchers reported that the study had been designed with patient engagement at its heart. Most participants did not report any level of engagement with the project, which demonstrated a need to explore the disparity between researcher and participant experiences. Participant interviews were also analysed to provide a model of engagement referenced throughout the project:

Engagement = Participant interest + Transfer of knowledge + Active participation

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Chapter 5 describes a series of focus groups with participants where the criteria for enhancing the feedback available to participants is drafted. This provides requirements for a software development cycle with RUDY's development team to make changes to the project website, online portal and reminder emails. Participants were unaware of the importance of their contribution to the study. A desire to record positive experiences was thwarted by only being offered space to report negative experiences. Participants reported that they were less likely to prioritise the study when deciding where to invest time and energy when they received no updates. Focus group data was analysed to provide a model of enhanced feedback that would drive development: participants wanted to know project aims, what data was collected, how that data was used, the value of their contributions, and research outcomes. Participants wanted this information to be communicated in a way they could understand, free of technical terms.

Enhanced feedback = Project aims + What data are you collecting? + How are you using this data? + Value of data-contributions + Research outcomes

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Chapter 6 describes an intervention study measuring the effect of enhanced feedback on research participation. Research participation was monitored before and after changes were made to the study website, online portal and reminder email. From 2019 to 2020, questionnaire completions increased by 5%. As a long-term study, participant retention is a key metric for the RUDY research team. In 2019 the number of participants who joined the study and remained active dropped by 32% from July-September to October-December. In 2020 this drop was 8.9%, suggesting that the intervention, implemented in late September 2020, reduced drop-off in participation over time.

1.7 Thesis contributions

In addition to the findings described above, this thesis offers theoretical, practical and methodological contributions. The theory of Dynamic Consent is developed, adding to a considerable body of literature, and a critical analysis of current work is offered that shows engagement is underserved in existing discussions of Dynamic Consent. The practical contribution of this work is a tool-kit consisting of a model, framework and specification. These tools were created as design prompts for incorporating Dynamic Consent into research methods.

This research also offers a novel, mixed-methods methodological approach. Interviews identified that participants had needs that were being left unmet. Online focus groups were used to explore those needs, and a software development cycle implemented changes designed to fill those needs. An intervention study was used to validate whether fulfilling those needs may have affected participation.

This work is also interdisciplinary: I am a Computer Scientist leveraging a Design Science methodology, drawing from Social Science and Legal theory. As far as I have uncovered, no similar methodology has been applied to the study of Dynamic Consent. This thesis demonstrates originality in its conceptual and methodological approaches. The established literature surrounding Dynamic Consent has not grappled with the nature of the theory as a proxy for wider discussions around a need for engaging data-practices. This work establishes a fundamental responsibility for safeguarding information, drawing from consent and privacy literatures, before aligning this responsibility with ideas and examples from professional practice. The studies supporting these ideas were designed to illustrate that seeking feedback from research participants enriched data-collection. The research approach is novel as findings relied on methods that were inclusive of, and responsive to, participant feedback. This improved research quality and the relevance of findings. This work will also be useful to anyone with an interest in an interdisciplinary perspective; participants, clinicians, software developers, computer scientists, lawyers, social scientists and biomedical researchers all contributed to the development and delivery of this thesis.

1.8 Overview of chapters

Chapter 2 offers a review of relevant literature in two parts: first, a brief history and theoretical overview of Consent and Privacy literatures to draw out key concepts, and a positioning of this thesis regarding the 'Social Contract' between Science and Society. Second, a deeper exploration of these elements in practice. A discussion of consent and privacy gives way to a critical review of data protection failures, and the development of a responsibility owed by those who build technologies that seek user data, to better architect data-sharing options. Here, research gaps are highlighted, as is the institutional lack of guidance offered to researchers regarding how they can build 'engagement' into their work. **Chapter 3** describes the overall research methodology and justifies the decision to apply a mixed-methods approach combining qualitative and quantitative techniques. This chapter also describes the ontological and epistemological positions of this research, its development process, and overarching ethical considerations.

Chapters 4, 5 and **6** describe the substantive work supporting the 'tool-kit for engagement' developed in **Chapter 7**. **Chapter 4** describes interviews with RUDY participants, and a focus group with the research team, evaluating the similarities and differences between Dynamic Consent theory and RUDY's implementation. While researchers designed their study to be participant-centric, participants are unable to recall basic research aims. **Chapter 5** describes the co-design and software development processes behind the intervention study. Feedback on findings from chapter 4 is sought from participants via online focus groups, and during these calls a suggestion for enhancing the feedback available to participants is developed into a more rigorous set of features that are then passed to RUDY's development team to implement. **Chapter 6** describes the intervention study testing the effect of this 'enhanced feedback' on study participation. **Chapter 7** presents a tool-kit for engagement based on the definition of engagement from chapter 4, criteria for enhanced feedback in chapter 5, and initial justification demonstrated in chapter 6. Expert opinion is sought on the tool-kit as a validation step.

Chapter 8 offers a reflection on research aims and a summary of theoretical, empirical, methodological and practical contributions. Notes on the development of Dynamic Consent theory and the value of participant engagement in research practice are offered, as are: a critical review, and directions for further work.

Chapter 2

Literature review

"Consent is a propositional attitude, so intransitive: complete, wholly specific consent is an illusion", (O'Neill, 2003).

2.1 Introduction

What follows is a literature review in two parts. The first concerns key themes of consent and privacy. These ideas underpin and drive this work. The nature of consent is described, including the purpose for which it was created. We will see how this purpose, protecting individuals from institutional harms, while noble-minded, is eclipsed by stubbornly paternalistic approaches. Privacy is then introduced as a form of control; over information and knowledge about a person. The concept of privacy can be seen as a private space that can grow or shrink; it is a pre-requisite for someone to be able to express themselves. I describe two ways in which privacy requirements can be modelled, so that they can be built into new technologies (Nissenbaum, 2004; Solove, 2006). These ideas highlight a general responsibility for expert practitioners to safeguard personal information and architect data-sharing choices in an accessible way.

The second part of this review introduces the idea of a social contract (Gibbons, 1999a; Meslin & Cho, 2010) between science and society. This contract is used to illustrate how conversations should occur between scientists and the public – they should be two-way. Examples where communication errors caused a breakdown of trust are offered, and a re-framing of trust is described; rather than seek to build trust, we should prioritise demonstrations of trustworthiness (Aitken *et al*, 2016) through honesty, competency and reliability (O'Neill, 2013). Dynamic Consent is described in further detail, as a digital mechanism to demonstrate trustworthiness and support the fulfilment of researcher data-responsibilities (Kaye *et al*, 2015; Teare *et al*, 2015). Of particular interest are two knowledge gaps; Dynamic Consent literature fails to report on participant engagement, and institutional 'Responsible Innovation' resources cannot describe what 'engagement' looks like in practice.

Finally, reflections are offered on the findings of this review as well as three lines of thought that will be revisited throughout the thesis. These are ideas that germinated as part of the literature review and developed through the experimentation reported in chapters 4, 5 and 6: responsibility for data protection lies with those who design information systems rather than those who use them, feedback must be sought from users to inform and improve practice, and there is great need for consent over time in digital spaces as data-uses change, new technologies are developed and levels of trust fluctuate.

2.2 Part 1: Consent and Privacy

2.2.1 Consent

Informed consent was created to protect people from harm, in response to atrocities committed in the name of medical research (see Appendix 1). There are ethical problems arising in biomedical research that are reverberating across other fields, particularly where researchers are operating more and more in digital spaces – we will come to these issues later in this review. Consent methods provide an opportunity. They can be used to raise awareness by designing forms and information sheets to clarify what is expected of participants and what is expected of researchers (Riordan *et al*, 2015). There are different ways to ask for consent, which I describe as consent 'modes': blanket, broad, tiered, opt-in, opt-out, meta and specific. Each mode has its own advantages and obstacles to user choice, and some modes are given different names in the literature. For example, consent given for a specific purpose might be referred to as 'specific' or 'explicit' consent.

Table 2: Modes of consent.

Mode of consent	Also known as	Sources
Blanket	General	(Hansson, Dillner, Bartram, Carlson, *et al*, 2006; Caulfield, 2007; Simon *et al*, 2011)
Broad	Blanket; general	(Sheehan, 2011)
Tiered	Categorical	(Ram, 2008; Simon *et al*, 2011)
Opt-in		(Junghans *et al*, 2005; Simon *et al*, 2011)
Opt-out		(Simon *et al*, 2011)
Meta		(Ploug & Holm, 2016; Manson, 2019)
Specific	Study-specific; narrow	(Árnason, 2004; Simon *et al*, 2011)
Genuine	Valid	(Taylor, 1949; Boulton & Parker, 2007)

Blanket (or 'general') consent places few restrictions on data-use (Hansson *et al*, 2006). Biobanks are research frameworks used to collect biological samples from people, that have historically used blanket consent. This is because it is difficult to re-contact tissue donors, and there is support from the public in delegating research decisions to researchers (Caulfield, 2007). One criticism is that blanket consent does not provide enough information to inform people about future data-uses (Simon *et al*, 2011). **Broad** consent is a term sometimes used interchangeably with 'blanket' or 'general' consent. I use broad consent to mean that participants consent to general research purposes. This does not include all possible purposes, some of which may not be research-related. As with blanket consent, key criticisms of broad consent are that: the method fails to keep participants informed over time (Sheehan, 2011) about how biobank samples are used, vague statements are used at the point of consent to prompt agreement to undefined future data-uses, and decisions about what 'appropriate use' of data entails is delegated to a review board.

Tiered consent (also referred to as 'categorical' (Simon *et al*, 2011)) presents "a menu of research categories" to which a participant may consent (Ram, 2008). This includes keeping biobank samples for researching a specific condition, widening the use of samples to any research purpose, or requesting to be contacted before the sample is used at all. **Opting-in** to participation is when someone actively signals that they are willing to take part (Junghans *et al*, 2005). This approach more accurately reflects

individual autonomy and choice but is often more resource-intensive in practice (Simon *et al*, 2011). **Opting-out** of participation includes patients who might be willing to participate and allows researchers to identify and attempt to communicate with non-responsive individuals. Opt-out is preferable to opt-in in terms of a lower self-selection bias and larger sample sizes (Junghans *et al*, 2005), but automatic enrolment is problematic as individuals may not know they are taking part. Situations where individuals do not know their information is being shared for a particular purpose are to be avoided, as this may violate the criteria of voluntary informed consent.

Meta-consent has been used to describe an approach where participants express a preference for how and when to provide consent, essentially designing their own consent process (Ploug & Holm, 2016). Neil Manson claimed that there was no ethical imperative to employ meta-consent (Manson, 2019), due to the fact that:

> "Nobody is wronged by making a non-coercive, truthful proposal, whilst denying the addressee the opportunity to "design" the scope of consent or to shape the nature of an ongoing consent relationship"

He then describes practical reasons for employing meta-consent, e.g., as a recruitment aid, before immediately weighing these ill-defined benefits against the vague notion that such an implementation would be incredibly costly[2]. More work is needed to describe the benefits of more flexible forms of consent, and to measure the cost such methods entail. **Specific** consent is sought for a particular reason, as "the more general the consent is the less informed it becomes" (Árnason, 2004) and can be referred to as 'study-specific', or 'narrow' (Simon *et al*, 2011). In a research study, for example, a potential participant will be told about what they will need to take part in and what that participation involves and this will not apply to participation in any other study.

2.2.1.1 Consent in research

Some experts are of the opinion that informed consent need not be compulsory, and is one item in a tool-kit of many, to be wielded by researchers as they see fit. Indeed, Ingelfinger suggests that the only real protection for participants lies in a compassionate researcher (Ingelfinger, 1972); informed consent is simply one way to articulate such compassion. Without values or ethics driving research purpose, informed consent has become an empty exercise where participants skate over pages of documentation, offloading any substantial decision-making to researchers. In discussing the legal environment surrounding informed consent practices, Katz (1977) describes:

[2] A deeper exploration of the Manson/Ploug and Holm debate on meta-consent lies outside the scope of this work, for further reading: (Ploug & Holm, 2020); (Manson, 2020).

> "… a fairy tale-like optimism about human capacities for "intelligent" choice and for being respectful of other persons' choices; yet in its implementation… pessimism of human capacities to be choice-makers"

So while Ingelfinger's compassionate researcher and Katz's pessimistic clinician are both using informed consent processes for different purposes, there is a shared frustration regarding the 'ritualisation' of such processes. In fact, informed consent provides opportunities[3]; to teach participants about the realities of research (e.g., its flexible nature) and its 'unknowns', and to listen to any questions a participant might have. Beecher argues that consent will always be improper, and informed consent always unattainable because it is "exceedingly difficult or impossible to obtain in any complete sense" (Beecher, 1966). Beecher takes exception (as do I) to the simplistic assumption that in research, our ends justify our means – that data collection does not need oversight. Accidents happen, and researchers must be held accountable for the mistakes they make; they must report data breaches, for example.

Rather than a one-off decision made at the start of a study and never re-visited, informed consent must persist over time for two reasons: one-time consent means coerced or accidental decisions cannot be corrected (O'Neill, 2003; Whitley & Kanellopoulou, 2010), and unreadable terms and conditions disempower "a raft of the population" (Luger *et al*, 2013). The former of these delegitimises consent, and the second normalises a sense of ennui. I use 'informed nature' and 'informedness' interchangeably to describe a participant's understanding of a study, which may differ from the point of consent to the end of a study. Data may be processed differently that initially proposed within any study, which is why researchers must create ways to inform participants throughout a project.

2.2.1.2 Digital consent

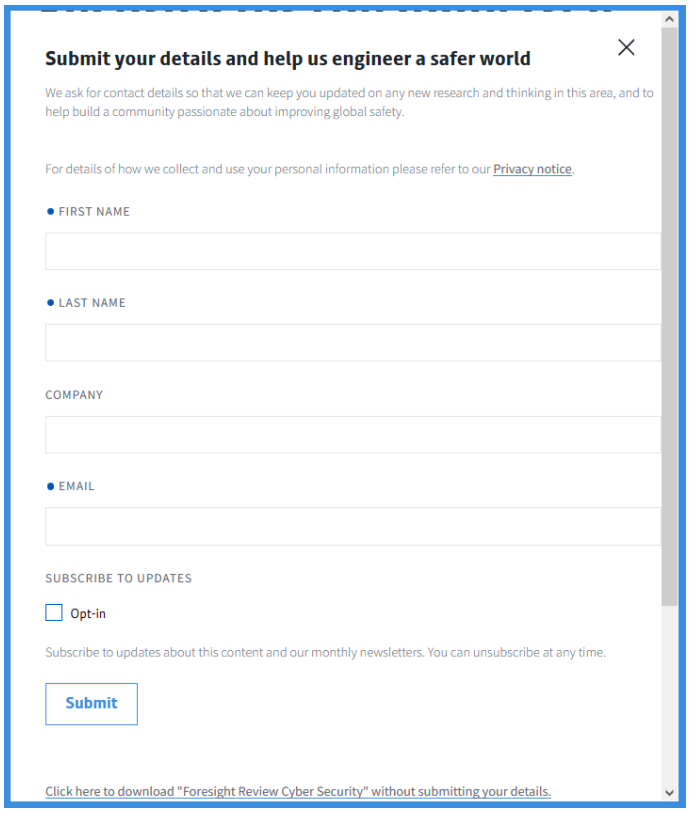
Consent procedures are in place to limit the deception and coercion of individuals (O'Neill, 2003). Unfortunately, the procedures used to collect consent online, such as cookie banners and consent notifications, afford no such protection. In online environments, people provide personal data without needing to demonstrate any understanding of why they have been asked, or in some cases required, to do so. Perhaps this is because if there was a need to demonstrate this understanding, we would observe a distinct lack of it. The exchange of personal data for services is ingrained, and legal efforts to formalise requirements and standardise practice have sowed dissent and confusion (Hayes, 2012).

[3] Opportunities identified in previous work (Teare *et al*., 2015), and leveraged in this project.

> "[H]ad browsers in 1995 (or even in 1997) implemented "decline all cookies" as the default setting… [most sites] would have responded with designs that were functional without cookies", (Millett *et al*, 2001).

Cookie banners ask for permission to allow 'local' information (such as location and IP address) and internet-browsing habits to be collected and stored as a text file. This file, referred to as a 'cookie', is stored on the user's machine or device for the website to bookmark that user's behaviours, preferences and any other, relevant, information. This is used to target the individual for advertising. While it can be convenient for users to have their preferences stored; cookies are collected for a data-processor's benefit. For some services, a user who does not give consent to their data being shared will be cut off from that service - coming up against a 'data paywall' (see Figure 2). A user who might want to share some, rather than all, of their data is usually directed towards complex wordings and highly detailed privacy policies. This user will find themselves prompted at every step to take the easy route and simply agree with what is being asked of them. The figure below, for example, asks for personal information and only at the bottom of the page, after the submit button, is there an option for accessing the website without providing information. As we will explore, "dark patterns" built to obstruct, sneak, interfere and coerce (Soe *et al*, 2020) encourage, rather than limit, the deception and coercion of users.

Figure 2: A 'data-paywall' for access to an industry report.



Historically, consent processes are established to reduce harm, but Fenech *et al* (2018) claim that consent also inhibits innovation. The authors describe this in relation to the use of data for AI:

> "The sheer size of datasets used to train AI [artificial intelligence] for medical… use [makes] consent a potentially impractical framework to operate under - it may be impossible to get specific informed consent from each and every patient"

While this example is AI-specific, the authors' outlook appears fairly limited. Rather than abandon harm-reduction measures, a more innovative and non-traditional approach to these processes is required. Organisations such as the Algorithmic Justice League, who offer a structured approach to algorithm auditing, and the fledgling Distributed AI Research Institute (DAIR), looking to 'slow down' AI research, leverage diverse expertise, and a value-driven approach. This work aligns with the Responsible Innovation approach previously discussed. EnCoRe demonstrated that technical architectures protecting user interests are feasible; responsibility lies with practitioners to prioritise human interests during development when building systems.

2.2.1.3 A form of data-protection

The historic use of informed consent as a way to physically protect patients has paved a route towards using such processes as a data protection mechanism, as reflected in recent legal trends and policy direction. The GDPR provides a specific description of consent when used to fulfil a data protection requirement (European Parliament, 2016):

> "Consent should be given by a clear affirmative act establishing a freely given, specific, informed and unambiguous indication of the data subject's agreement to the processing of personal data [by written, electronic or oral means] … Silence, pre-ticked boxes or inactivity should not therefore constitute consent… When [data] processing has multiple purposes, consent should be given for all of them… [a request by electronic means] must be clear, concise and not unnecessarily disruptive to the use of the service for which it is provided."

There are three key criteria for consent: it must be active, informed and explicit. Note the parallels this draws with a Dynamic Consent approach; revocation ("it shall be as easy to withdraw as to give consent") and, loosely, engagement. While two-way communication is not a focus in the GDPR, communication between data-controllers and data-subjects should provide clarity as to what someone's personal data will be used for. The criteria for informed consent within GDPR demonstrates the importance of communication in informed-consent practices.

The need for such guidelines comes from an increase in data's use and importance. While GDPR was not intended for researchers, research institutions must still ensure that data processing practices are lawful, fair and transparent, and that, where personal data is collected and used, information is provided about these practices (UKRI, 2020). Where consent was originally established to protect medical research participants from physical harm (United Nations, 1948; Taylor, 1949; DHEW, 1979; Shultz, 1985), individuals are now exposed to data-harms, especially as researchers increase their use of online spaces. Ethical review committees, bastions of quality and peer review in the approval of research studies, are unable to accurately assess such harms (Hibbin *et al*, 2018)[4]. This suggests that the governance infrastructures supporting ethical research practices need to be considered in the digital era. Examples of data-harm include unauthorised access and sharing of data, discrimination and identity theft. These harms are less tangible than physical harms, and so more difficult to understand. This means it is incumbent on researchers, as subject matter experts, to be able to effectively communicate such risk, and informed consent process offer an opportunity to start these discussions, with a proportionate degree of nuance (Agrafiotis *et al*, 2010b):

> Consent itself is not a simple yes/no ("store this data" or "do not store this data"), but has many subtly different gradations, depending on the type of data it refers to.

Manson and O'Neill describe an informational obligation owed by researchers to participants (Manson & O'Neill, 2007), which is violated by 'bad' or unethical behaviour. This obligation relates to how information is used rather than an exhaustive list of specific types of data that ought to be collected. For example, a research participant sharing symptoms and treatment history for research purposes is unlikely to find third-party, for-profit pharmaceutical access to their record acceptable. If a research project shared that person's data with a newly on-boarded pharmaceutical partner, this would violate their informational obligation. A different individual may find such an arrangement acceptable – a flexible mode of consent would allow such preferences to be recorded.

2.2.1.4 Problems with consent

There are two potential problems with informed consent: the first is information overload, the second is consent fatigue. Moving from static to more dynamic consent practices that better reflect user preferences offers one way to address these issues.

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[4] Hibbin *et al* identified that committee members who had experience carrying out research drawing data from social media were more likely to have a nuanced view of the requirements of such work. Guidance then, is required for researchers who lack such experience.

When providing consent, individuals put themselves in a vulnerable position. They make a trade-off between their personal interest and the risk that a service might cause them harm (Manson & O'Neill, 2007). Let us take Manson and O'Neill's example of a surgeon and patient. A surgeon recommends a surgical intervention to a patient, who consents to the procedure as part of treatment. The patient's agreement means that they have made short-term sacrifice for long-term benefit. They will be in pain and need time to recover, which may have tangible effects such as a reduced capacity to earn money. The patient shoulders some of the responsibility for these restrictions as they agreed to the surgery, but our surgeon retains the burden of responsibility for her patient. She must have the relevant surgical expertise; she is responsible for the patient being under anaesthetic (delegating the finer details to her anaesthesiologist); and her patient must receive the appropriate level of post-operative care.

Manson and O'Neill use the example above to illustrate how the burden of responsibility is distributed between the expert surgeon and non-expert patient. Online, non-expert users shoulder a disproportionate level of responsibility when making decisions to share data (or not). They are overloaded with information with no expert to support them in their decision-making process; they are asked to scan, in seconds, terms and conditions with advanced reading level requirements (Luger *et al*, 2013). These tasks are imposed, and users are unlikely to revisit the choices they have made. I have declined to identify who the fulfils the role of an expert in online spaces in part due to another important problem. Identifying who the 'expert' is, is difficult. In the surgery example, clearly the surgeon is the expert. Online, the responsible expert is unclear; e.g., the organisation asking for data, the project manager who delivered the choice architecture, or the software developer who implemented it. A research study using online recruitment methods will likely have an expert investigator with overall responsibility for the project, and non-expert participants. Sharing responsibility across these parties may not be possible to do initially, but methods such as inviting a participant representative to the project's oversight committee provide one way to ensure an ongoing conversation.

Information overload

Receiving too much information can cause people to disengage from the decision-making process and decline to make any choice at all (Ram, 2008). In the process of informed consent, information is given and an individual demonstrates that they understand this information by giving, or declining, their consent (Miller & Wertheimer, 2010). Miller and Wertheimer are perhaps overly eager when they say that providing consent shows understanding. Research participants have been shown to consent to a study and not know basic information about the research (Ormond *et al*, 2009), online users tend not to read the information provided before agreeing to requests for data

(Bashir *et al*, 2015), and some patients may agree to intervention under the therapeutic misconception that experts have only their best interests at heart (Appelbaum *et al*, 1987; Corrigan, 2003). In the example of a surgeon and patient, the surgeon provides information about the procedure and asks the patient to sign a consent form. This prompts a conversation where the patient asks questions and clarifies details they are unsure of. In the example of a user being asked to share information via cookies, the user is flooded with information and there is no opportunity to ask questions. Where a researcher uses online methods to recruit participants, there is an opportunity to set up the 'data-transaction' between researcher and participant using informed consent by providing information, and ways to communicate over time. Where there is no interaction between participant and researcher, this transaction offers a low return on investment, and a reason to disengage from the decision-making process.

While people may undervalue their data:

> "… our genes are not that important, maybe we can do our little bit to share", (Lemke *et al*, 2010).

They also feel that they should have a say in how that information is used and shared:

> "Participants… want to know that their DNA data are going to be shared [and] think that it is important to have some control over who accesses and uses their information", (McGuire *et al*, 2008).

Researchers are experts who have knowledge about research methods and practices that most participants do not:

> "We may disclose information that we do not understand, or do not see as relevant. Such unwitting disclosures may gain significance only for an audience that knows certain further facts" (Manson & O'Neill, 2007).

Two-way communication is not necessarily a pre-requisite for informed consent decisions to be truly informed. Perhaps if research participants had all the information they needed to make their consent decision, they would not need an ongoing dialogue with researchers. A conversation is required, however, as a mechanism to support participants in deciding on trade-offs that taking part in a study might entail. Such processes could help identify the information needed by particular participants – e.g., the time commitment required.

Those who build consent options should know what information is needed, and why. This puts them in a position to communicate their objectives in a way that is easily understandable. Placing the burden of comprehension on users means that, rather than

translating technical language into accessible terms, there is a risk that experts obfuscate and individuals are forced to take on the work of translating technical information for themselves.

User rights initiatives such as Terms of Service; Didn't Read (tos;dr, see Figure 3) serve as an example of how experts can translate information for non-experts to absorb more easily. The example below relieves the burden on the reader through interface design; short sentences and colour, size and font are used to highlight key information. Figures Figure 4, Figure 5 and Figure 6 show the initial data-access (cookie) requests, and optional information from each of the tos;dr examples (Amazon, Reddit and Facebook). Notice how the eye is immediately drawn to the 'accept all' options, while alternative (read: undesirable) actions likely to be muted in colour. The way that these screen captures have been laid out also highlights the amount of information users have the option of exploring if they want to exercise control over the data they share. In Figure 4 users can either immediately configure their data-sharing preferences or read more information, being redirected back to the preference list. Figure 5 shows specific options, with users eventually being able to configure their preferences. Figure 6 shows configuration options and 'More Options'.

Figure 3: Examples taken from "Terms of Service; Didn't Read" website.

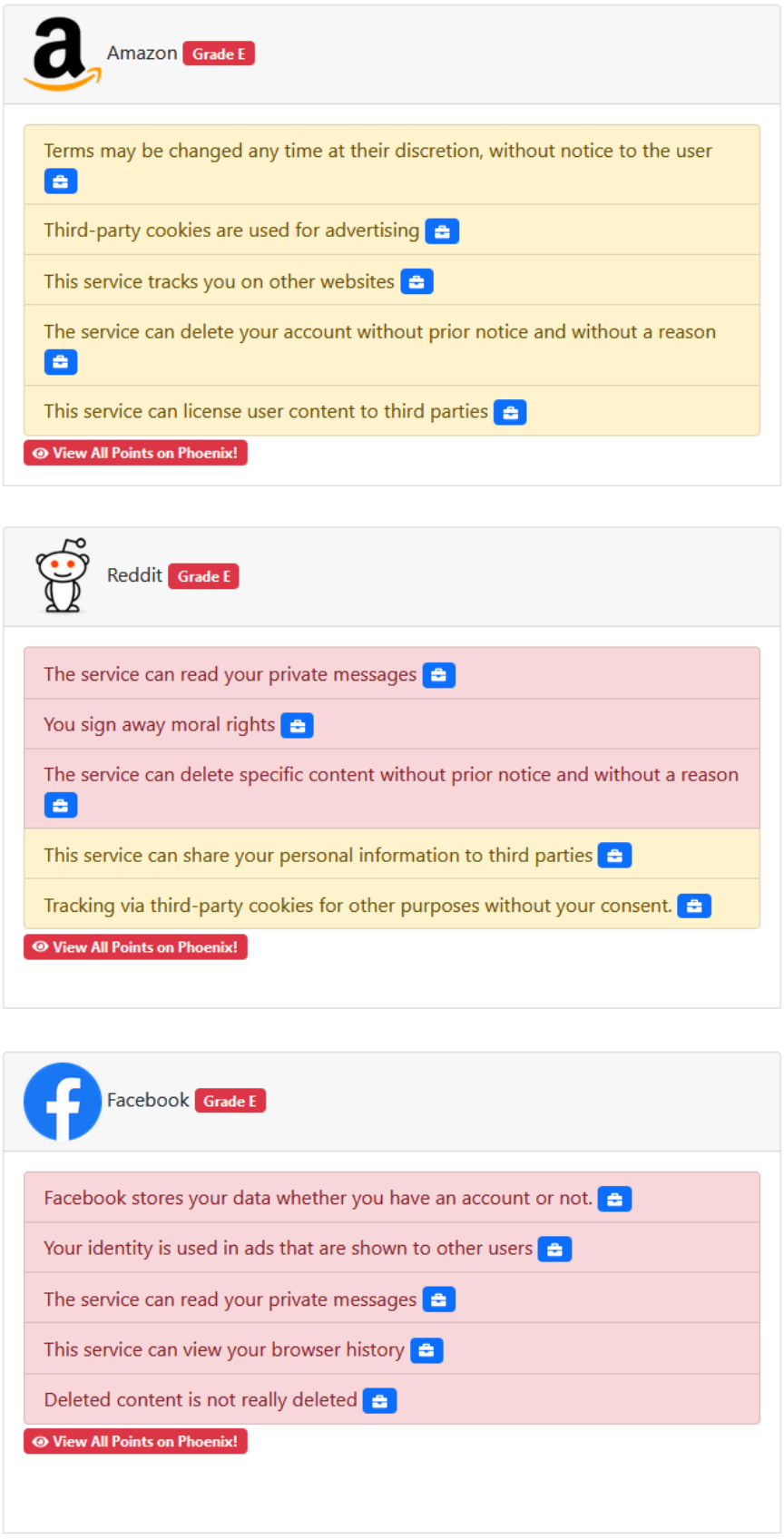


Figure 4: Data-access (cookie) request from amazon.com

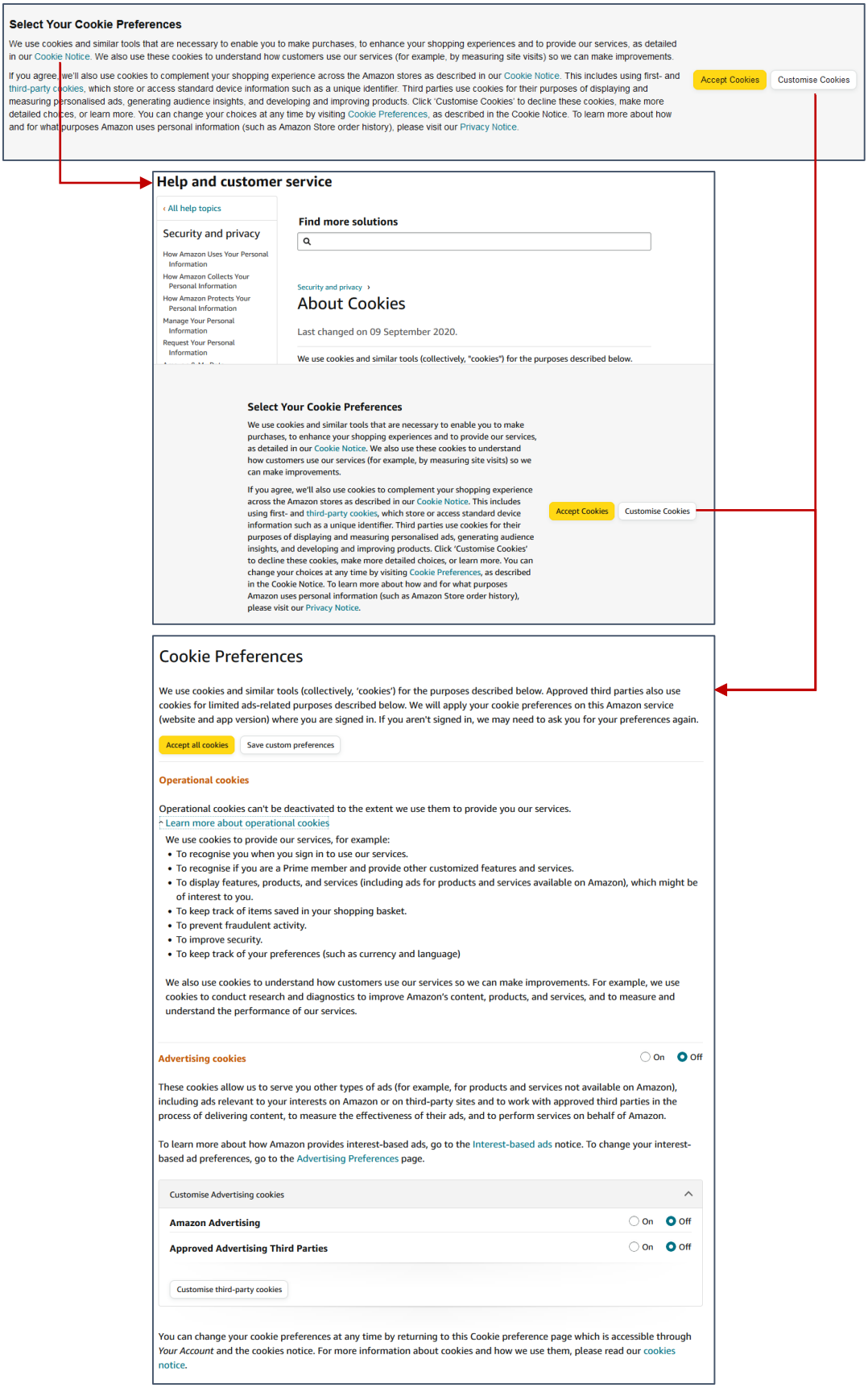


Figure 5: Data-access (cookie) request from reddit.com

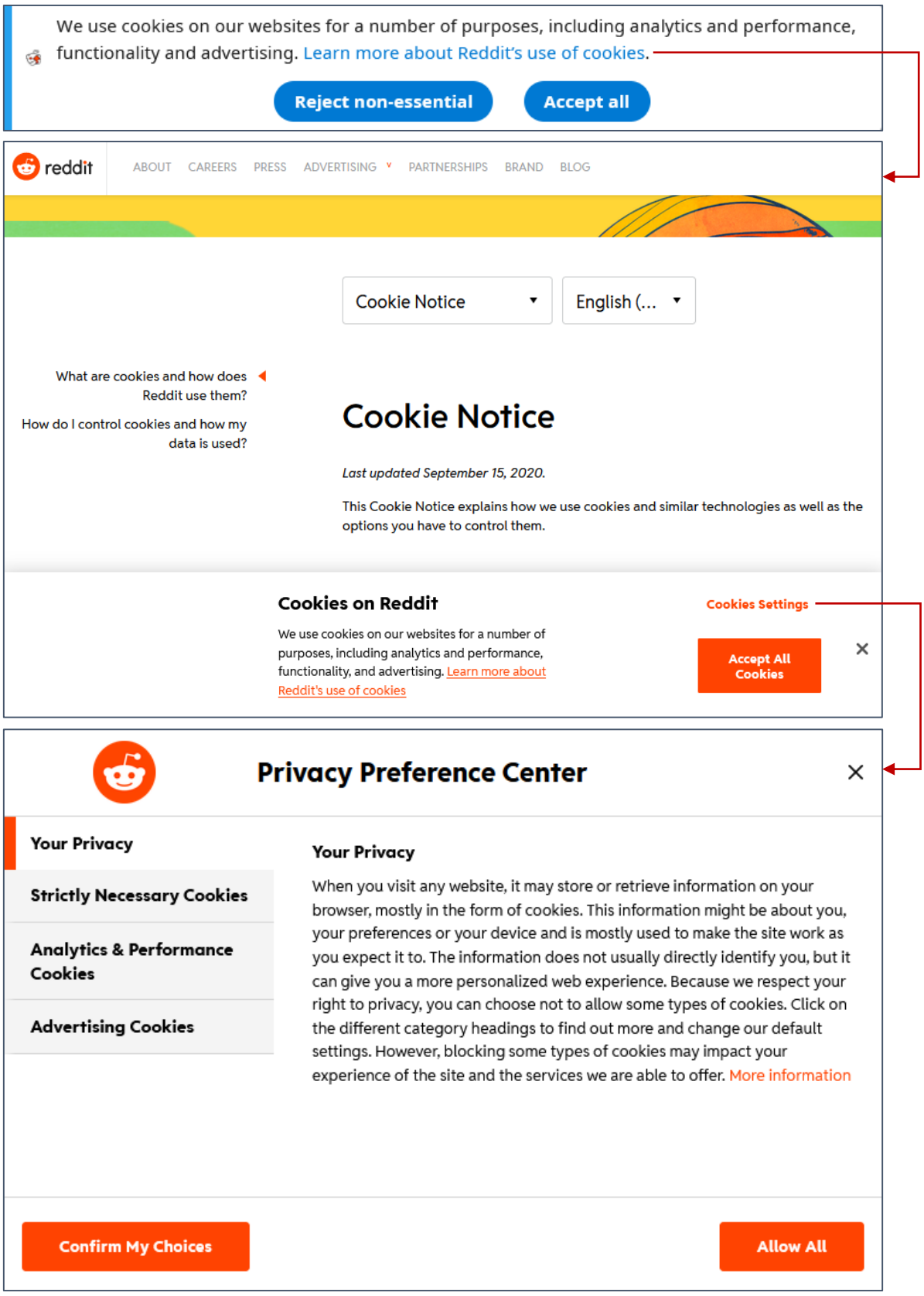
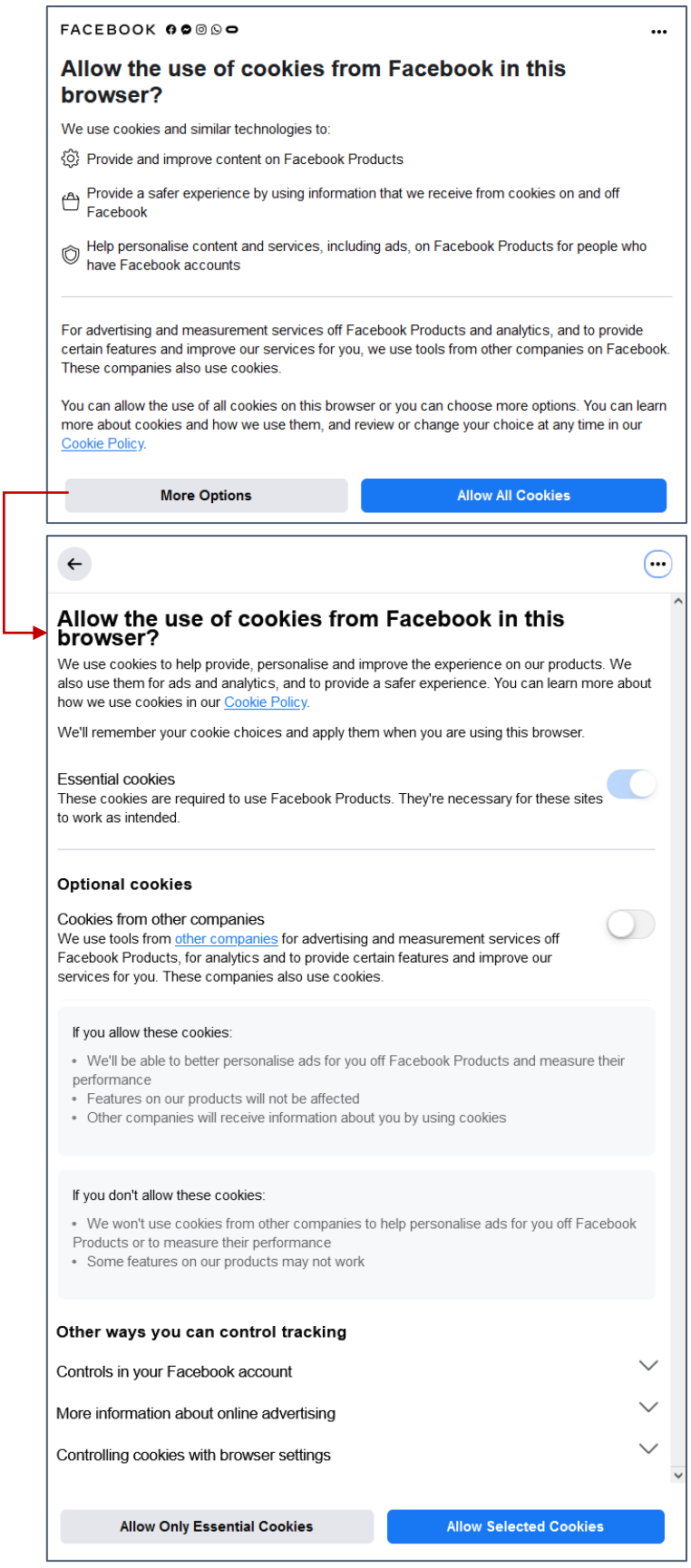


Figure 6: Data-access (cookie) request from facebook.com



Consent fatigue

'Security fatigue' has been described as a symptom of exhaustion caused by the over-zealous application of security processes and procedure – especially where these practices get in the way of day-to-day operation (Bada *et al*, 2015). Security fatigue occurs because it can be "stressful to remain at a high level of vigilance and security awareness… [and users could] become lax". In other words, where users operate in a reactive mode of operation over a long period of time, they become desensitised to security issues. 'Consent fatigue' describes an inability to make choices that reflect actual preferences. This inability comes from being forced to interact with consent notices frequently and on a regular basis. People are presented with notices that coerce an acceptance, so they fail to invest the time and energy in these decisions that they ought to have done. In cases such as this, consent is a concession rather than an active (or informed) choice. The term consent fatigue is not new (Kaye *et al*, 2015; Prictor *et al*, 2018), but is somewhat under-developed.

The purpose of informed consent is to prevent harm. If users are being over-exposed to notices they do not care to understand, they are unlikely to keep engaging with data-sharing decisions and make the fastest choice possible as a form of avoidant coping. There is evidence that 'privacy fatigue' results in an avoidant coping (Choi *et al*, 2018) – those showing signs of such fatigue are more likely to disengage with privacy decisions. Importantly, Choi *et al*'s work indicated that privacy fatigue had a stronger influence on behaviour than privacy concern. This raises a concern that consent fatigue may cause a user to rely on cognitive shortcuts, even if messages are communicated in an accessible way. This 'wearing down' presents harm on a wide scale.

Information overload happens when someone is presented with more information than they can process. The brain defaults to shortcuts, or heuristics, that are not representative of someone's informed preferences (Ram, 2008). Where someone has to make a choice about their privacy for example, they might use heuristic cues such as expert recommendations or the advice of friends and family. They might also be motivated by general principles, like a need to be accurate, to keep their data safe, or to make a good impression (James *et al*, 2021). Consent dialogues impose a time cost on privacy-aware users (Hils *et al*, 2020); I use consent fatigue to refer specifically to a result of over-exposure to prompts presenting users with choices they do not want to make. McDonald and Cranor (2008) calculated that users were presented with 1,462 privacy policies a year, each taking 10 minutes to read. In one experiment, measuring how long people spend reading privacy policies on social media, Obar and Oeldorf-Hirsch (2020) found that users invested 73 seconds in documentation that would have, in other circumstances, required 30 minutes to complete.

2.2.2 Privacy

Historically, 'individual' privacy concerns the protection of personal data and communication channels for such data (Warren & Brandeis, 1890; Prosser, 1960; Bloustein, 1964; Westin, 1968; Thomson, 1975; Kupfer, 1987; Cooke, 1999). The sources referenced in this section demonstrate a general responsibility to protect information, owed by those collecting and consolidating personal data, to those they have collected data from. This is especially the case as information is often gathered, based on an agreement that it is used for a particular purpose. Legal provision, such as the General Data Protection Regulation (GDPR) (General Data Protection Regulation: Regulation (EU) 2016/679 of the European Parliament and of the Council of 27 April 2016 on the Protection of Natural Persons with Regard to the Processing of Personal Data and on the Free Movement of Such Data, and Repealing Directive 95/46/EC, 2016), also directs data-controllers towards the responsibilities they hold with regards to their data-subjects[5]. I will explore limitations of individual empowerment (Curren & Kaye, 2010), and a need for mechanisms that allow individuals to shape data-sharing relationships (Agrafiotis, Creese, Goldsmith, Papanikolaou, *et al*, 2010). These issues highlight the way that privacy controls are used as evidence of compliance with data-protection responsibilities (e.g., section A.18.1.4 of ISO/IEC, 2013). Such controls could, if well implemented, demonstrate individual preferences and indicate the degree to which an organisation has used someone's information in line with their preferences (Casassa Mont *et al*, 2011a).

2.2.2.1 A right to privacy

At the end of the nineteenth century, American lawyers Samuel Warren and Louis Brandeis argued that individuals have a right to be left alone and their work grounded privacy as a legal right. Members of a wealthy elite uncomfortable with a tabloid press, they sought protection from the "idle… curiosity" of the general public on the grounds of inviolate personality, asserting that "personal writings and all other personal productions" should not be published (Warren & Brandeis, 1890). It took another fifty years and a World War for the United Nations to enshrine privacy as an individual, legally-protected right (United Nations, 1948), where: "no one shall be subjected to arbitrary interference with his privacy… nor to attacks upon his honour and reputation". A key criticism of privacy as an individual right describes harms caused when one person's privacy veils the abuse of another (Cooke, 1999):

> "…the function of privacy laws cannot be the demarcation of… the family or household – in which everything that happens is held to be a

[5] "Controller" and "subject" are legal terms, as we will discuss. Here they illustrate the relationship between those who collect information and those from whom that information is collected.

> matter for private concern, not subject to public thematization or to criticism by strangers"

Cooke suggested that privacy is valuable in its own right, as a precondition for autonomy. Privacy fosters an individual's personal development (Cooke, 1999) by providing a personal space in which they can exercise independence and self-determination (see Kupfer, 1987). Privacy has also been described in terms of human dignity (Bloustein, 1964); a construct in its own right rather than a composite interest (Prosser, 1960). In fact, Bloustein goes so far as to describe privacy intrusions as an imposition and affront to one's liberty – similar to (physical) battery, but through different means. Prosser described privacy as a product of reputation, emotional tranquillity and intangible property. A more pragmatic approach to this idea is suggested by Thomson, who seconds Prosser's thinking that a 'right' to privacy derives from a cluster of other rights. Rather than reputation and emotional tranquillity, Thomson proposes that privacy derives from ownership rights as well as the right not to be harmed (Thomson, 1975):

> "The wrongness of every violation of the right to privacy can be explained without ever once mentioning it"

DeCew (1997) rejects reductionist and derivative approaches; that privacy can be reduced to other claims or derived from property and physical rights. Privacy "should be guaranteed in a wide variety of contexts" because it protects information control, access control and freedom of choice. For DeCew, context is key, privacy is pinned firmly in its place by the social context of how data is used. Along a similar line of suggestion, Kanellopoulou advises that a right to privacy exists, and that conceptualisations of privacy must be nuanced to allow for contextual complexity. Privacy is "inextricably linked with one's essence as a human being… an expression of respect for an… autonomous being" (Kanellopoulou, 2009).

In sum, an individual's right to privacy exists but the characterisation of this right in terms of information-sharing is unclear. The position of this thesis is that privacy requirements provide a way to consider user autonomy within technical systems. While these systems may not be private spaces, they must allow a level of control over the information that is shared about someone. This is one reason why privacy preferences are important to record and abide by.

2.2.2.2 Legal enforcement

Organisations have privacy requirements that operate at 'high' and 'low' levels: high-level requirements include legal obligations, and low-level requirements are more operational, usually technical (Papanikolaou *et al*, 2010). Regulations such as Europe's GDPR (General Data Protection Regulation: Regulation (EU) 2016/679 of the European Parliament and

of the Council of 27 April 2016 on the Protection of Natural Persons with Regard to the Processing of Personal Data and on the Free Movement of Such Data, and Repealing Directive 95/46/EC, 2016), the California Consumer Privacy Act (SoCA, 2018), Japan's Act on the Protection of Personal Information (PIPC, 2016) and New Zealand's Intelligence Act (Parliamentary Council Office, 2017) demonstrate how organisations are obliged to protect data. Such data protection requirements can cause conflict within organisations seeking to repurpose data without consulting users[6].

The table below provides definitions of specific legal terms from the GDPR. I use these terms throughout this thesis as a broader representation of what they portray in legislation.

Table 3: Terms defined in the GDPR.

Personal data	Any information relating to a data-subject.
Data-subject	An identified or identifiable natural person. This means someone who can be identified, directly or indirectly, by name, identification number, location data or online identifier. This also includes physical, physiological, genetic, mental, economic, cultural or social factors of that person's identity.
Data-controller	The natural or legal person, public authority, agency or other body which, alone or jointly with others, determines the purposes and means of the processing of personal data.
Data-processor	A natural or legal person, public authority, agency or other body which processes personal data on behalf of the controller.

Data-subjects have a right to see what a data-controller holds about them for two key reasons. The first is that specific rights are enshrined in data-protection legislation around the use of personal data, such as the right to be forgotten (data-erasure) and the ability to move data from one controller to another (data-portability). This recognises the importance of such data-rights. Second, insights can be gained from personal data that may not be what data-subjects gave permission for a controller to know, access or use. This could affect how someone is treated, posing risks of profiling and discrimination. Further, if information is inferred about someone and proves to be incorrect, a data-

[6] When GDPR came into effect in Europe for example, American news sources came under fire for restricting users' freedom of choice by blocking audiences from GDPR-compliant nations (Connolly, 2018; Hill, 2018).

subject should be able to correct what is known about them. These highlight the importance of regulatory and practical considerations in this area; there are no simple explanations and enforcing privacy usually involves trade-off of some kind.

Existing online privacy controls are unfit for purpose as they do not protect privacy. Privacy policies are offered, and user agreement to share data sought, to demonstrate that organisations are in compliance with user preferences. However, McDonald and Cranor (2008) describe how they think responsibility lies squarely with data-controllers rather than data-subjects:

> "… some corporations take the view that their users should read privacy policies and if they fail to do so, it is evidence of lack of concern about privacy. Instead, we counter that websites need to do a better job of conveying their practices in useable ways, which includes reducing the time it takes to read policies. If corporations cannot do so, regulation may be necessary to provide basic privacy protections"

2.2.2.3 Data-sharing choices

Creating new regulation takes time. In the meantime, efforts must be made to improve practice. The example of a potential research-study participant is used to illustrate the following example. Embedded within research-consent processes are choices to be made around who personal data is to be shared with. I refer to these as data-sharing choices. Researchers must be careful that options are phrased in such a way that participants can clearly understand them (Ram, 2008), and the informed consent process is used to record privacy preferences and demonstrate compliance with these choices. While the 'privacy paradox' (see Appendix 2) suggests that people do not care about the realities of data-sharing, we know that people express privacy preferences when they understand what their choices are (Agrafiotis *et al*, 2010a).

Individuals value autonomy in decision-making, while also seeking expert guidance (Hammami *et al*, 2014). They are also more likely to exercise autonomy when provided with information that they understand (Agrafiotis *et al*, 2010a). 'Choice architecture' refers to the way in which options are presented, with presentation influencing the choices people make. In their foundational work on behaviour nudges, Thaler and Sunstein describe an approach to architecting choice; libertarian paternalism: "Libertarian paternalists want to make it easy for people to go their own way" (Thaler & Sunstein, 2008). I will simply use the term 'choice architect' to describe data-controllers who are

responsible for providing non-coercive data-sharing options[7]. The libertarian paternalism movement seeks to preserve an individual's right to choose, while nudging them towards a decision that is in their best interests. Organ donation and public hygiene provide examples of experts architecting choice in a way that positions people to make decisions in the public interest (Oullier *et al*, 2010). E.g., public health (Thaler & Sunstein, 2008):

> "Libertarian [parentalists] see countless opportunities for improving people's health… Framing matters: people are more likely to engage in self-examinations for skin and breast cancer if they are told… about the increased risk if they fail to do so"

There are two criticisms that I will level at libertarian paternalism and 'nudge' theory. First, their focus lies with the provision of multiple options rather than whether or not the framing of such options is coercive. I level this criticism because informed consent cannot be coercive. To adapt this theory for use in informed-consent practice, non-coercive framings must be prioritised. My second criticism is that user involvement is not a design consideration in choice architecture discussions, meaning that a wealth of experience is left untapped. User input can be invaluable – the role of an expert can be filled by users themselves, due to their own areas of expertise and understanding. Choice architectures must be designed with feedback from the people who are going to be asked to make choices, and this feedback can come in the form of consultation before, during and after architecture development, or as user-testing throughout.

Data-sharing choices can be complex; choice architecture can simplify consent choices and reduce consent fatigue. In what is still some of the most fascinating work in this arena, EnCoRe found that users will make a choice where they have the options available for them to do so. In this study, participants were asked how they thought they could revoke personal data they had provided, in hypothetical scenarios (social networking, medical, legal, public data-use and private data-use). Participants reported that they would choose to delete all data, or left intact. Researchers then spent time describing different ways participants could revoke data, and participant preferences began to reflect those nuances. Table 4 shows the revocation mechanisms participants initially thought should be available ay default. Table 5 shows the revocation mechanisms that participants thought should be available after researcher intervention (Agrafiotis, Creese, Goldsmith, & Papanikolaou, 2010).

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[7] "Libertarian paternalism" is layered with sentiment that I would like to simplify. Not least, to remove the gendered term "paternal", which also indicates coercion and control (the antithesis of informed consent). Choice architects are responsible for presenting people with options, this thesis claims simply that researchers are choice architects.

Table 4: Revocation mechanisms users expect to see, by default.

	Social networking	Medical environment	Public data controller	Private data controller	Legal environment
Deletion	☑	☑		☑	☑
No revocation		☑	☑	☑	☑

Table 5: Revocation mechanisms users expected to see when told about different options.

	Social networking	Medical environment	Public data controller	Private data controller	Legal environment
Deletion	☑	☑		☑	
Anonymisation		☑			
Cascading revocation				☑	
Revoke permission to process		☑	☑	☑	
No revocation	☑	☑	☑	☑	☑
Revoke permission to disseminate		☑		☑	
Consentless revocation		☑	☑	☑	☑
Delegated revocation				☑	☑

One example of consent choice architecture comes from a recent review of data security, consent and opt-outs in the NHS. "Information profiles" are suggested, that patients of the National Health Service might choose (NDG, 2016):

Table 6: Example information profiles.

My health and social care information profile		
☐ **Standard setting** Information about me can be used to run the NHS, social care and to support public research.	☐ **Limited setting** Information about me can be used to run the NHS and social care. Not public research.	☐ **Restricted setting** Information about me can only be used by the people directly providing my care.

2.2.2.4 Modelling privacy requirements

The importance of modelling privacy requirements as part of the technology-development process cannot be understated. In 1996, Smith *et al* constructed a framework to describe concerns about organisational information privacy practices (Smith *et al*, 1996), identifying the following areas of concern: data collection, unauthorised secondary use, errors and improper access (see Table 7). Researchers are making the same recommendations 25 years later, shadowing Smith *et al's* recommendations concerning the use of access controls (Qiu *et al*, 2020), tracking internal use of personal data (Legg *et al*, 2013), and creating databases designed to use information sparingly (Datta *et al*, 2011).

Table 7: Privacy concerns in organisational information practices (Smith *et al*, 1996).

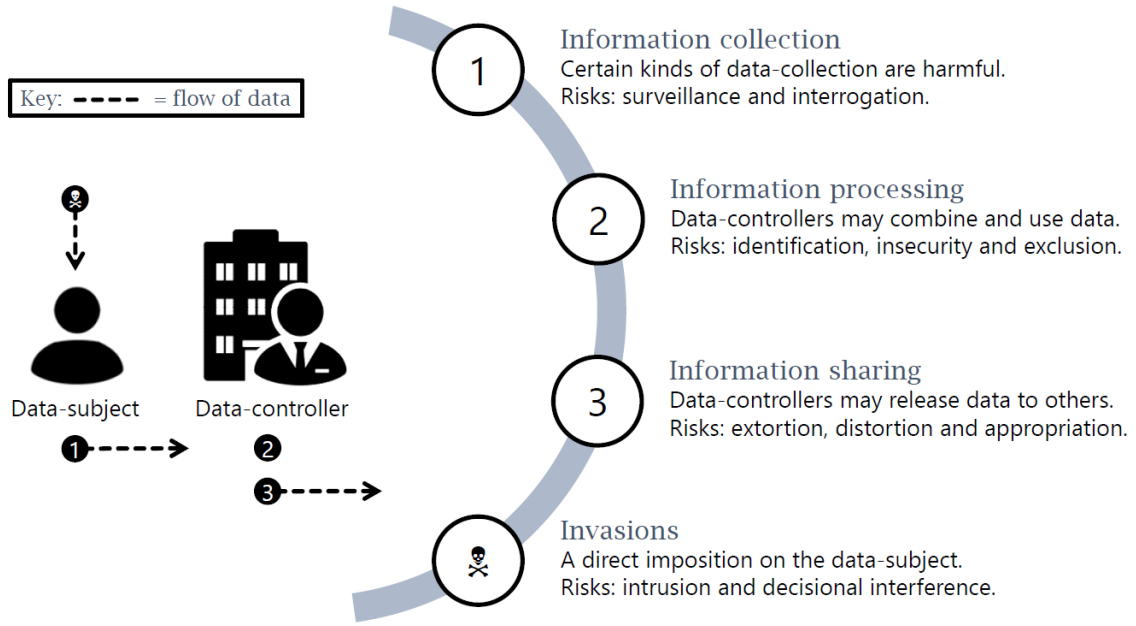
Area of concern	Recommended actions	Recent source
Improper access	Implement technological controls on access to systems.	(Qiu *et al*, 2020)
Unauthorised secondary use	Ensure that all internal uses of personal data can be tracked.	(Legg *et al*, 2013)
Collection	Practice parsimonious database design.	(Datta *et al*, 2011)

To identify what may reasonably be required of information privacy practices, we turn to two frameworks: Helen Nissenbaum's theory of contextual integrity, and Daniel Solove's privacy taxonomy.

Both frameworks were designed to identify privacy challenges during design stages of technology development. The first of these, contextual integrity, describes privacy violations as a lapse in norms of 'appropriateness' and 'distribution' around information. A "foundation for policy and law… [is grounded] in terms of moral, political, and social values", and the theory works from "a right to privacy as it applies to information about people"(Nissenbaum, 2004). Contextual integrity is built on the idea of informational transactions happening in a particular context, or 'sphere' of life. These can be sector-specific (e.g., education or politics) or situational (e.g., a job interview, or visiting the dentist) (Nissenbaum, 2009). These contexts are governed by their own norms, and there are two types: norms of appropriateness which dictate information-type and data-sharing restrictions, and norms of distribution which address data-transfer (or 'flow'). The 'contexts' of contextual integrity are structured social settings that can be defined by internal values, roles, relationships and norms. Nissenbaum's theoretical framework of privacy models how context-relevant norms can be violated. If expected (and 'correct') behaviour can be defined then unexpected behaviour can be identified, although it is important to note that unexpected behaviours are not necessarily privacy violations.

In a similar vein, Solove reiterates that "the question of when and how the law should regulate can only be answered in each specific context in which the question arises". The taxonomy of privacy (see Figure 7) focuses on privacy-infringing actions, grouped under four categories of harmful activities: information collection, information processing, information dissemination and invasions (Solove, 2006). A lawyer, the author wanted to move away from vague definitions of privacy in favour of a clearer understanding of actions and consequences - if a criterion for privacy violation was that a user had not consented to data processing, then secondary data–use for a purpose that someone had not agreed to would constitute a privacy violation. Solove argues that violations can occur at different levels of severity. For example, a local newspaper may cause annoyance when reporting a burglary victim's name, but a company selling medical equipment being found selling customer data to a health insurer.

Figure 7: Categories of privacy-harming activity (adapted from Solove, 2006).



Nissenbaum and Solove provide specific understandings of privacy that also allow for the nuance of social context, by providing methods to identify privacy expectations (norms) and behaviour (actions). Online tools can be used in a number of different social contexts, so identifying expected norms and appropriate actions will help those seeking to develop consent-specific tools. Online privacy is a significant element of any digital consenting tool and must be considered so that research participation can be supported, without undermining the privacy (or group privacy, see Bélanger & James, 2020) of participants.

2.3 Part 2: fulfilling the social contract.

2.3.1 Social contract

A "social contract" exists between science and society where public institutions fund science that operates in the public's interest (Gibbons, 1999b). In return for science being carried out on behalf of the public, members of the public need to be involved; providing data and participating in research studies, for science to be successful, and persist. This contract requires a two-way relationship where public awareness is raised about the importance of research, and outcomes demonstrate value. Philosopher Onora O'Neill argues that research participants place themselves in a position of vulnerability in consenting to take part in research, demonstrating their trust in researchers (O'Neill, 2013). As we will discuss shortly, public engagement efforts help to demonstrate that

researchers are trustworthy (Aitken *et al*, 2016), which in turn may help to secure public trust.

Meslin and Cho (2010) suggest that education "on both sides" is key to the successful implementation of a social contract between science and society. Communication must go both ways.

Table 8: A proposed social contract between science and society (Meslin & Cho, 2010).

Scientific community commits to:	In return, society provides:
Articulating goals and visions of what brings benefit.	Participation in research.
Achieving goals over pursuing individual interests.	Sustained, reliable funding.
Involving the public in the scientific process, demonstrating trustworthiness.	Academic freedom from political manipulation.
Greater transparency.	

The existence of a social contract between science and society means that researchers must challenge the status quo of consent processes that fail to inform participants over time. In this research project, I uphold the social contract by producing relevant, reproducible and actionable findings, consulting members of the public as part of the design process, and creating useful tools to support other researchers in their own work.

2.3.2 Controversies

There are several historic instances where public trust has been severely diminished by poor practices. Two examples follow, the first is an account of Alder Hey Children's Hospital where children's organs were retained for research purposes without parents' knowledge. The second describes the impact of a lack of public engagement within a government campaign to digitise medical records in the UK; the initiative garnered very little support with policy-makers having to concede, that "legal authority does not necessarily command social legitimacy" (Carter *et al*., 2015).

Alder Hey Children's Hospital

Before collecting tissue from deceased children, Alder Hey Children's Hospital failed to seek appropriate consent from the parents of those children. Parents were coerced into agreeing to a post-mortem, and multiple organs were retained without notice from approximately 850 post-mortem examinations carried out between 1988 and 1995. Between one and three containers held multiple organs and many pieces and fragments of tissue from each child (The Stationary Office, 2001). While medical professionals may have been guided throughout their training to protect patients from difficult decisions, this case offers a damning indictment of paternalistic practice (Bauchner & Vinci, 2001). As a result of these failures, an oversight committee was enacted: the Human Tissue Authority (Hall, 2001), and the Human Tissue Act of 1961 was re-written in 2004 to mandate how organs were to be used for research purposes (The Stationary Office, 2004).

Care.data

In 2013, care.data was going to make primary care records available for others uses (Carter *et al*, 2015):

Figure 8: Project aims of Care.data.

Box 1 Aims of *care.data*

NHS England has described the *care.data* service as: '…a new, modern data system for the NHS in England. Known as *care.data*, its purpose will be to provide timely, accurate information to citizens, clinicians and commissioners about the treatments and care provided by the NHS' (http://www.england.nhs.uk/wp-content/uploads/2013/05/ces-tech-spec-gp-extract.pdf).
The aims of the *care.data* programme are sixfold:

1. To support patients' choice
2. To advance customer services
3. To promote greater transparency
4. To improve outcomes
5. To increase accountability
6. To drive economic growth by making England the default location for world-class health services research.

This project offers a cautionary tale; the scheme was opt-out by default which made people feel as if a decision had already been made for them, and there were no public-facing doctors or experts to field questions. Public trust in NHS England plummeted and the development of potentially valuable services was dropped. Expert reviews called for better security provisions and new patient opt-outs for the use of personal information (Limb, 2016), and a statement from the British Medical Journal identified a specific need for "dynamic consent" processes (Godlee, 2016). On the advice of the National Data Guardian, and Care Quality Commission, care.data was closed down (Freeman, 2016); the project promised data-sharing benefits without offering patients any form of control.

2.3.3 From trust to trustworthiness

While there is no shortcut to developing trust relations, it is entirely possible for trust to be misplaced. To avoid this, researchers must provide evidence of trustworthiness: competency ("can they get the job done?"), honesty ("will they tell me the truth?") and reliability ("are they consistent?") (O'Neill, 2013). Instrumental in building trust are a researcher's assurances of competence and accountability, through research process and procedure. However, researchers must also demonstrate trustworthiness and credit members of the public with the intelligence required to make a decision about whether or not to trust them with their data (O'Neill, 2014). This sentiment is echoed by Aitken *et al* (2016) who interviewed members of the public to explore attitudes to sharing electronic patient records. Opinions relied on a range of factors, such as process transparency and institutional accountability. The authors concluded that consistent demonstration of trustworthiness would be more effective in building and fostering relationships with the public over time, rather than a focus on building trust.

Aitken *et al* (2016) also concluded that engagement can be used to demonstrate trustworthiness. Rather than, rather paternalistically, deciding what the public needs to know, public engagement can be used to identify concerns and design processes that address them. Community engagement has been known to also uncover barriers to adoption (Voss *et al*, 2007). Indeed, a rather helpful conceptualisation of engagement describes a process of four potential (and interchangeable) parts: a point of engagement, engagement over time, a point of disengagement, and re-engagement (O'Brien & Toms, 2008). Experts may not know how to engage with non-experts however, and may struggle to know where to start the process[8]. Seeking feedback is the first step in this process, a response to such input is required. This demonstrates the two-way communication required to fulfil the social contract.

Effective engagement requires expert architecture. In workplaces, for example, employees need opportunities to be proactive (Knight *et al*, 2019). Carolan and de Visser describe an engaging online environment (Carolan & de Visser, 2018):

> "The perfect digital mental health intervention was described as… a short interactive course [with] time-unlimited information and advice that was regularly updated and could be dipped in and out of. Participants also wanted access to e-coaching support"

[8] This is why informed consent is identified as an excellent first-step in engagement efforts. Researchers are familiar with this process, and may be persuaded to expand their approach.

2.3.4 Dynamic Consent

In theory, Dynamic Consent provides avenues for engagement such as an online interface that provides project updates and allows participants to revisit data-sharing choices (Whitley *et al*, 2012). Dynamic Consent was developed to address problems with the broad consent approach (Curren & Kaye, 2010; Kaye, 2011; Casassa Mont *et al*, 2012), and lack of participant oversight (Kaye *et al*, 2015) in biobanking. While individuals are comfortable delegating study-specific data-sharing decisions to a biobank's ethical review process (Simon *et al*, 2011), broad consent to non-specific use removes participants from future decisions.

Fundamentally, Dynamic Consent was "a new approach for engaging individuals about the use of their personal information" (Kaye *et al*, 2015), relying on two principles: revocation and engagement. Revocation is described in terms of "an interactive personalised interface that allows participants to engage as much or as little as they choose and to alter their consent choices in real time". Engagement is described as "a personalised, communication interface to enable greater participant engagement in clinical and research activities". In focus groups with biobank participants, Dynamic Consent researchers found that participants were keen to learn about their role within research processes:

> "I suppose you'd you want an email alert to say 'Just used your sample' and then you can log in and see what it's being used for", biobank participant (Teare *et al*, 2015).

Engagement 2.0 underpinned early development of Dynamic Consent, re-evaluating how researchers could incorporate interactive and user-led technologies. Engagement 2.0 consisted of five TRACKs: timing, research participation, autonomy, consent and knowledge (see

Table 9).

Table 9: The TRACKs of Engagement 2.0.

Feature	Engagement 1.0	Engagement 2.0
Timing	• Linear flow. • One-time decisions.	• Iterative processes supported • Can review and revise decisions.
Research participation	• Flyers and leaflets. • Conversation from consultant to participant.	• Find participation opportunities. • Dialogue with researchers. • Notification of opportunities.
Autonomy	• Passive.	• Active, empowered, ongoing.
Consent	• Paper-based, broad.	• Dynamic.

Knowledge	• Biobank-specific leaflets and resources.	• Opportunities to share with community of participants.

Prictor *et al* describe Dynamic Consent as an online tool:

> "developed [to] improve engagement, communication and participation with a range of communities [allowing] greater control over how their data and samples are used" (Prictor *et al*, 2018).

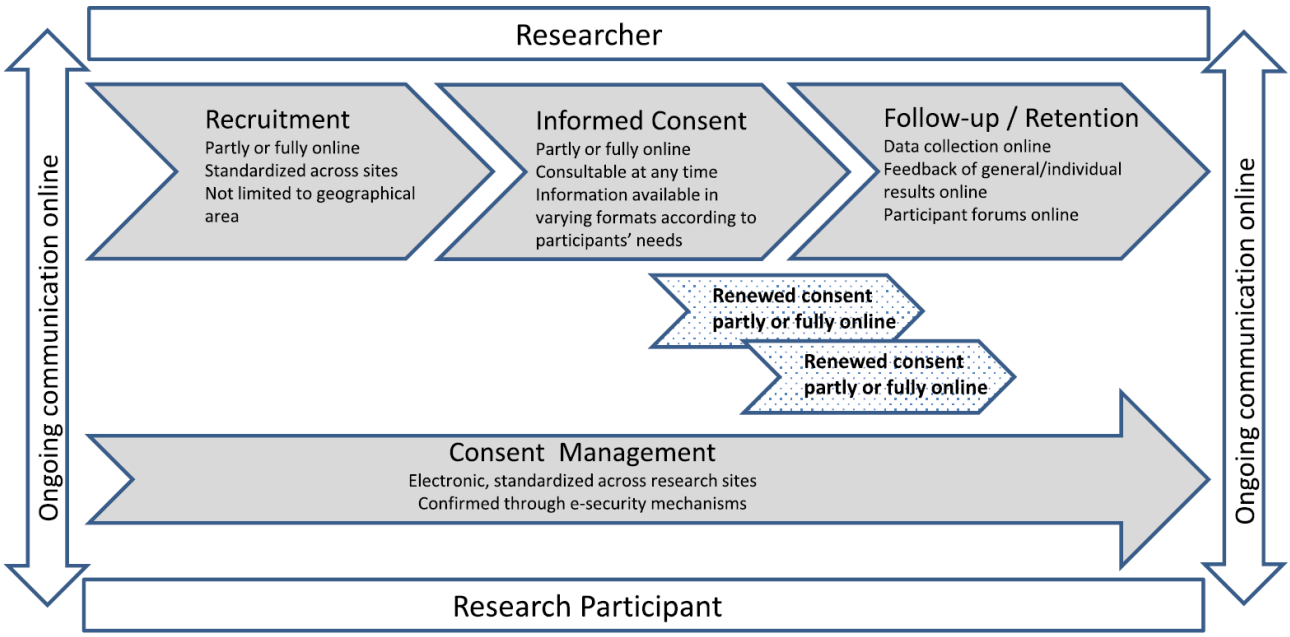
The benefit of this tool lies in its capacity to support informed consent by prompting potential participants to "think through important aspects" when they are introduced to a study. In their survey of existing literature, the authors describe Dynamic Consent as being administered via a "secure, personal profile", allowing participants to "receive information about the research… news and updates". Through this interface, researchers can manage "communication and engagement strategies… and keep a record of interactions". The authors assert that a dynamic form of consent may overcome barriers to participation in clinical studies almost exclusively dominated by white, middle-class participants, due to its flexibility. Ultimately, these claims are baseless due to a lack of empirical evidence. Prictor *et al* identified two key research areas regarding Dynamic Consent: general project evaluation, and information overload. The authors provided an evaluation framework for Dynamic Consent, recommending that future work investigates whether multiple consent options might overwhelm participants, cause consent fatigue, or lead to disengagement.

Limits of existing literature

EnCoRe demonstrated that Dynamic Consent could be built into technical architectures. One issue with the literature that has followed is that the ideas can be difficult to operationalise. Budin-Ljøsne *et al* (2017) created a process flowchart showing where Dynamic Consent could benefit biomedical researchers (see Figure 9). Use of Dynamic Consent, the authors argue, solves problems in participant recruitment, collection of informed consent, participant retention and consent management. The model is built on qualitative findings from a workshop seeking clinical, academic, ethical, legal, participant and patient expertise on the use of Dynamic Consent in research. This means that the work offered theoretical rather than practical insights, and there are two points of order to address in this regard. The authors make two bold claims that are difficult to substantiate: that Dynamic Consent is "future-proof" and "relatively easy to set up and maintain". Future-proof is a dangerous term to use because this implies that Dynamic Consent offers a complete solution that is resilient to change. While the authors claim

that Dynamic Consent is easy to set up and maintain, they provide no implementation guidance to support this assertion. These claims are somewhat inappropriate because the authors do not have any evidence for them. They did not find issues with, or 'troubleshoot', a live project. This may seem an unfair point, as Budin-Ljøsne *et al* are not carrying out a service evaluation of Dynamic Consent. My contention, and overarching point, is that without practical examples of Dynamic Consent in practice it is difficult to develop methodologies or evaluation metrics with any degree of certainty. More work is needed in this area.

Figure 9: Applying Dynamic Consent to research processes (Budin-Ljøsne *et al*, 2017).



Prictor *et al* created an evaluation and reporting framework for Dynamic Consent (see Figure 10) in the form of a "logic model framework" (Prictor *et al*, 2020b):

> "A logic model of DC should provide a framework for describing (a) the relationships between the resources needed for a DC approach, (b) the activities required for this approach, and (c) the results as they relate to the goals of DC. It provides a means by which to examine the assumptions that underlie a DC approach by making them explicit"

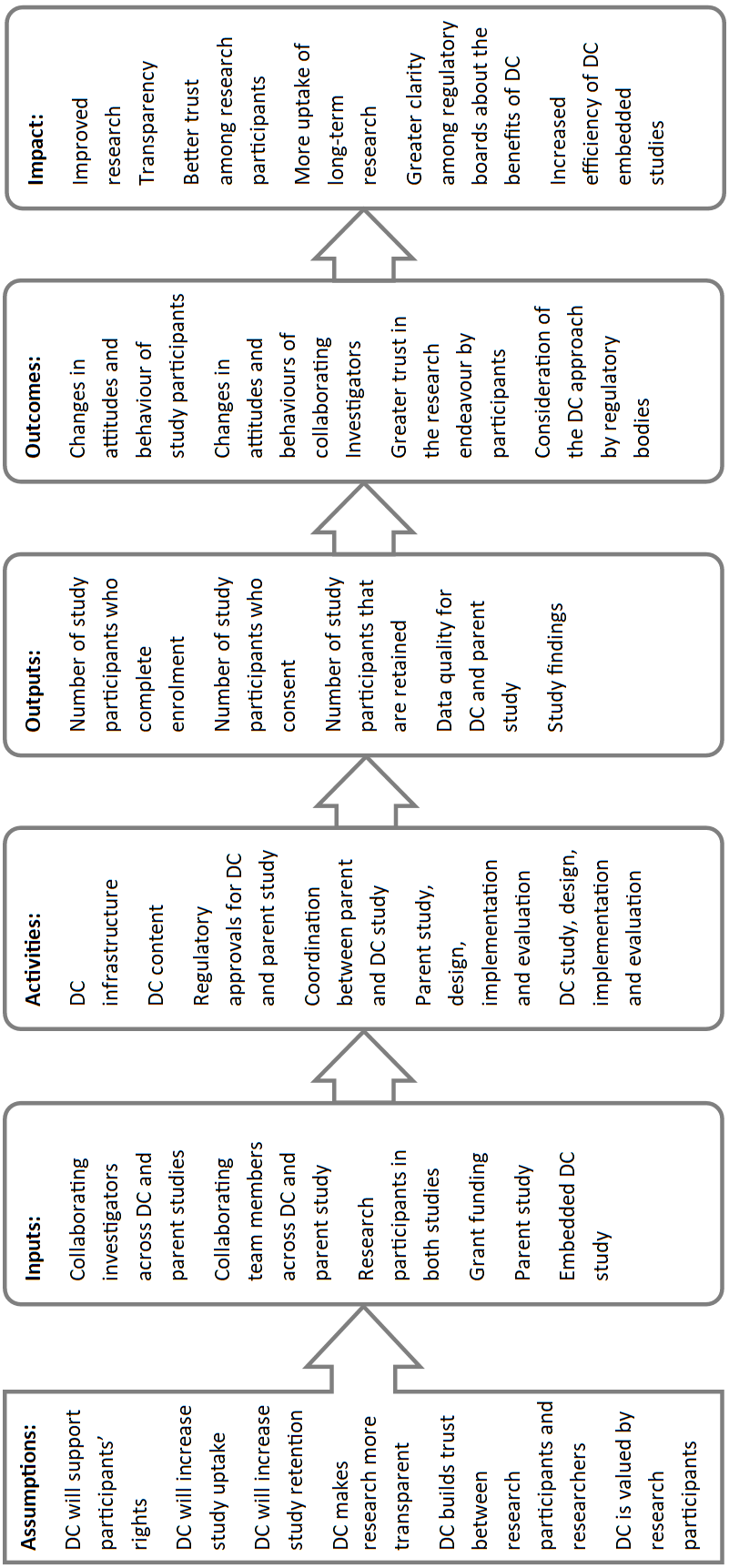
There are two issues with the approach used in this paper. The first is that the authors develop a logic model framework for Dynamic Consent which seems to depart from the supporting methodological literature. The second is that engagement does not appear in the logic model framework, despite being one of the two original criteria for Dynamic Consent. As to the first point, the authors describe logic models as "proven tools to help in evaluation", citing Frechtling (2007) and Knowlton & Phillips (2013). With reference to

those sources, while there is overlap between logic models and frameworks, the two constructs are different. Logic models can aid evaluation – Frechtling describes how logic models can be used to create frameworks. These frameworks create an outline of shared criteria within a group, or 'family', of projects. One example of such a family is projects that implement Dynamic Consent, which would include RUDY (Javaid *et al*, 2016), CHRIS (Pattaro *et al*, 2015; Motta *et al*, 2019) and My Health My Data (Panetta & Cristofaro, 2017).

Prictor *et al*'s paper considered how Dynamic Consent could be evaluated within projects, which is why I turned to it for insights regarding Dynamic Consent requirements. Frechtling (2007) explains that logic models can be used as frameworks to compare projects that share overarching goals, but differ in focus and implementation. Logic models can also help generate new frameworks. Frechtling also describes logic models as decision-making tools, as they help illustrate specific use-cases. Knowlton and Phillips describe logic models as providing a common vocabulary and shared understanding among stakeholders, a reference point (or anchor) for implementation, evaluation and dissemination, and helpful in identifying a project's next steps (Knowlton & Phillips, 2012). Both sources emphasise the practical applications of both logic models and frameworks – a good quality model, these authors argue, is both plausible and feasible. While Prictor *et al*'s paper is a useful review of the available literature on Dynamic Consent, their logic model framework is neither plausible nor feasible. As to the former, when attempting to use the framework I was unable to deliver the framework, largely due to overly abstract prompts such as "Dynamic Consent will support participants' rights". As to the latter, criteria for successful evaluation is not clear in Prictor *et al*'s paper and I found it difficult to understand how this tool was to be used in practice.

Does Dynamic Consent offer a way for researchers to take a longer-term view of their data-practices? The intuitive answer, from the authors, is 'yes'. It is surprising that the notion of Dynamic Consent's application in existing projects is not raised, as evaluation tools would be helpful in managing a migration from static to dynamic consent processes. Prictor *et al* advise researchers to consider cost and maintenance, the degree of collaboration with research ethics committees that will be required, platform accessibility and personal responsibility, before implementation. Note that neither revocation or engagement are included in this list, which is symptomatic of the tendency for literature citing Dynamic Consent to move away from its basic principles.

Figure 10: Dynamic Consent evaluation framework (Prictor *et al*, 2020).



2.3.5 Knowledge gaps

Dynamic Consent does not seem to have been implemented in many research projects, so there is a lack of work reporting its implementation. Service evaluations are required from projects implementing Dynamic Consent, to test existing evaluation tools, to develop new tools, and to further understand the challenges of putting Dynamic Consent theory (i.e., principles of revocation and engagement) into practice.

There is also a lack of guidance surrounding engagement within research practice more widely – Responsible Innovation is a key part of the methodological approach within this work, discussed in more detail in the next chapter (section 3.2). There are glaring omissions in the guidance available regarding the design and reporting of participant engagement.

2.4 Reflection

This literature review has consisted of five lines of enquiry:

- First, the importance of consent as an established, but admittedly flawed, form of data protection. Consent process offer an opportunity to start a conversation between experts and non-experts but can face limitations when applied to new technologies and emerging data-uses.
- Second, the importance of a right to privacy in allowing people to self-determine and control what is known about them. To design privacy-protecting technologies, one must create technical design requirements that can describe privacy. One way of doing this is to define context, which will dictate norms which can be used to define expected behaviours.
- Third, there is a social contract between science and society that means rather than simply 'educating' the public, lessons can, and should be expected to be, learned on both sides.
- Fourth, demonstration of trustworthiness should be a design goal of any public-facing process, rather than building trust. This can be done through transparency of process and using 'engagement' to inform projects, making findings more relevant.
- Finally, Dynamic Consent was designed to disrupt the status quo in biomedical research, but its underlying theory does not provide a solution by itself and needs testing.

Three lines of thought have also surfaced in the exploration of literature described above. First, that responsibility for data protection lies with those who design information systems rather than those who use them. Consent is not currently working as it should,

and there is a need for work that shifts the focus of informed consent from data-subject autonomy to data-controller practices (O'Neill, 2003). Quite simply, data-controllers are not fulfilling their obligations to data-subjects. Second, feedback must be sought from users to inform and improve practice. This review has provided some insight into the practicalities of 'responsible innovation', and how to design with social context in mind. All a 'responsible' researcher may need to do is communicate the lessons they have learned. We simply do not know, due to a dearth of research-implementations that could serve as examples. Third, there is great need for consent over time in digital spaces as data-uses change, new technologies are developed and levels of trust fluctuate. The literature indicated that people know they are left out of decision-making processes that use their data, but are not equipped to do anything about it. Research participants, for example, often receive no indication that their contribution has had any impact at all.

Chapter 3

Methodology

"Innovation is about transforming futures" (RRI Tools, 2015).

3.1 Introduction

The work underpinning this thesis consists of four studies: the description of a live Dynamic Consent use-case based on qualitative data; identification of where implementation aligned and departed from Dynamic Consent literature, using qualitative data to justify suggestions for improvement; an empirical justification for the importance of engagement within Dynamic Consent architectures, grounded in quantitative findings; development of a tool-kit to provide implementation guidance.

Chapter 2 demonstrated the importance of mixed methods in the development of Dynamic Consent. EnCoRe described technical architectures that used "robust consent and revocation methods" built in (Bramhall *et al*, 2011), and uncovered issues with institutional oversight and day-to-day research practices through focus groups with medical professionals (Whitley *et al*, 2012). Following this work, researchers explored a need for Dynamic Consent in clinical research. A workshop was run with members of the public to understand why people shared data for research purposes (Sanders *et al*, 2014), and Teare *et al* (2015) ran focus groups with biobank participants who expressed interest

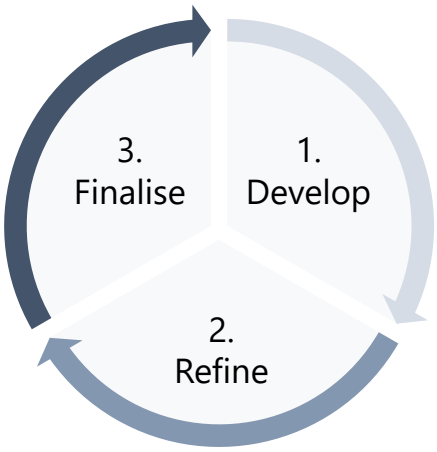
in a more interactive relationship. As the literature surrounding Dynamic Consent has developed, broader imperatives have been identified. An interdisciplinary workshop showed that researcher/participant relationship dynamics needed to be reconsidered if communication methods were to change at all (Budin-Ljøsne *et al*, 2017), and Prictor *et al* (2020b) have also highlighted a lack of empirical work in testing Dynamic Consent theory in practice.

This chapter describes the research methods used in this work, in defence of a mixed-methods approach. This approach was taken to provide qualitative insight and quantitative nuance (Toomey *et al*, 2017). Research philosophy discusses the ontology and epistemological positioning of this thesis, using 'Responsible Innovation' as a framework to illustrate how this philosophy has been put into practice. Project management describes the requirements engineering approach taken in an effort to combine qualitative research findings and suggested actions, with the software development and evaluation operations. Research methods simply lists, and describes in general terms, the methods used over the course of this work. These include participatory methods, e.g., co-design, qualitative methods, quantitative methods and tool-kit synthesis. Chapters 4, 5, 6 and 7 each contain a short, detailed account of how specific research methods were implemented. Ethical considerations are also provided, which offer an account of the ethical approvals sought and how their development informed the series of studies (e.g., applying for approval from the RUDY review committee gave me clarity on the type of data the study could provide). This chapter concludes with a short summary of why a mixed-methods approach has been adopted, the benefits this provides, and a final reflection on the value of including input from key stakeholders[9]. Namely, the value of a mixed-methods approach.

This research was carried out in stages. Knowledge gained from preceding work was used in the development of each study, and findings were used to inform next steps. Each stage consisted of three steps: development, refinement and finalisation (see Figure 11). The development step collated information from pre-existing sources as well as the data-collection described in this work. The refinement step reflected on feedback gathered through validation work, and the finalisation step was used to complete and add polish to research contributions.

[9] RUDY participants as well as the RUDY research team.

Figure 11: Stages of research.



Study 1	Study 2	Study 3	Study 4
Design interviews	Participant validation	Gather insights	Draft framework
Pilot and adjust	Feasibility check	Intervention study	Expert validation
Peer validation	Revision of intervention design	Develop framework	Re-draft framework

3.2 Research philosophy

A relativist ontology

This thesis serves as the significant contribution towards a degree in cybersecurity; my work is ontologically novel as I seek to understand different stakeholder framings, and it is inherently social. A relativist ontology posits that there is no shared social reality; only a series of different, individual constructions (Ritchie *et al.*, 2013). A system is designed to function in a particular way; this is its proposed reality. Systems are composed of many different components, e.g., hardware, software and the human processes supporting them. Such systems are a human construction, resting on the cultural assumptions of their creators. A user's 'reality' of such a system will be shaped by how they interact with it, and this may be shaped by the user's role in relation to that system (e.g., participant, researcher or external collaborator), their expertise or personal experiences. This work of this thesis rejects a realist framing, an absolute notion of reality, in favour of relativism. In asking both researchers and participants to take part, this work collated different, individual constructions of RUDY in terms of experience and understanding. The literature surrounding Dynamic Consent is growing, so an incomplete modelling of RUDY as a Dynamic Consent implementation is acceptable in this case as a description of the study in-situ (Hughes & Sharrock, 2016, p114):

> "This leads on to another general property of descriptions, namely, that they are always, in principle, incomplete"

A partial, illustrative model is helpful as a development aid, rather than seeking explanations of behaviour and 'universal' laws. I sought academic, clinical and participant understandings of RUDY as it is one of only a few implementations of Dynamic Consent in practice. A combination of understandings would, I posited, allow me to model the study in terms of criteria for Dynamic Consent found in academic literature. The different framings would allow such a model to be as complete and honest as possible. This has certainly been the case, as we will explore further in Chapter 4. RUDY researchers illustrated how they had meant Dynamic Consent to operate in practice, while participant experiences offered frank insights. Academic literature provided a theoretical construction of Dynamic Consent against which to compare these views. RUDY provided a specific context in which different (and shared) constructions and understandings of Dynamic Consent were rooted.

A constructivist epistemology

How then, did this work create knowledge, given its relativist nature? Knowledge has been produced by exploring and attempting to understand the social world of those with a stake in RUDY (Ritchie *et al*., 2013). Meaning has been constructed, not discovered (Crotty, 1998):

> "What, then, is constructionism? *It is the view that all knowledge, and therefore all meaningful reality as such, is contingent upon human practices, being constructed in and out of interaction between human beings and their world, and developed and transmitted within an essentially social context*"

This epistemology has been chosen due to the intangible nature of software, and the need to collect accounts and experiences of it to understand how it is used and whether that use correlates with its intended function. Note, that my intention is not to use Crotty's idea of constructionism to make grander claims about the nature of reality in its entirety. In other words, I have derived an understanding of what RUDY means to people, and constructed my own interpretation of the study, based on the experiences reported to me by my participants.

To analyse this data, an abductive approach was used, a cyclical strategy (Blaikie, 2007):

> "This logic involves moving back from observations to a creation of a possible explanation… Abductive logic is also a creative process in which social scientific concepts and theories of social life are derived from social actors' everyday conceptualizations and understandings"

An entirely inductive approach beginning with data collection instead of a literature survey would have neglected the wealth of knowledge available. Instead, I developed research aims based on insights from academic literature, and was receptive to new and relevant areas of interest that might emerge during empirical work. RUDY participants were interviewed about their research participation to discover their experience of research in their own terms. I expected to uncover shared understandings among participants instead of 'universal' facts. I used these interviews to define terminology and concepts for use in further studies. Namely, a specific definition of engagement, and the components of 'enhanced feedback' used to structure the intervention study.

This work draws from interpretivism; specifically, that human knowledge of the world around them is constructed and shaped by experience and understanding (Dudovskiy, 2012; Walsham, 1995). This interpretation is heavily grounded in, and supported by, the data collected and reported in each chapter (Ritchie *et al*., 2013). There are three characteristics of this work owed to an interpretivist influence: first, I regarded participants as people to interact with and be influenced by, rather than objects to distance myself from; second, I explored the nature of participant realities as subjective and dependent on individual experience and social construction, rather than immediately quantifiable; third, I wanted to understand the meaning RUDY held for participants (Scauso, 2019) to identify needs that were not being met.

Responsible Innovation (RI)

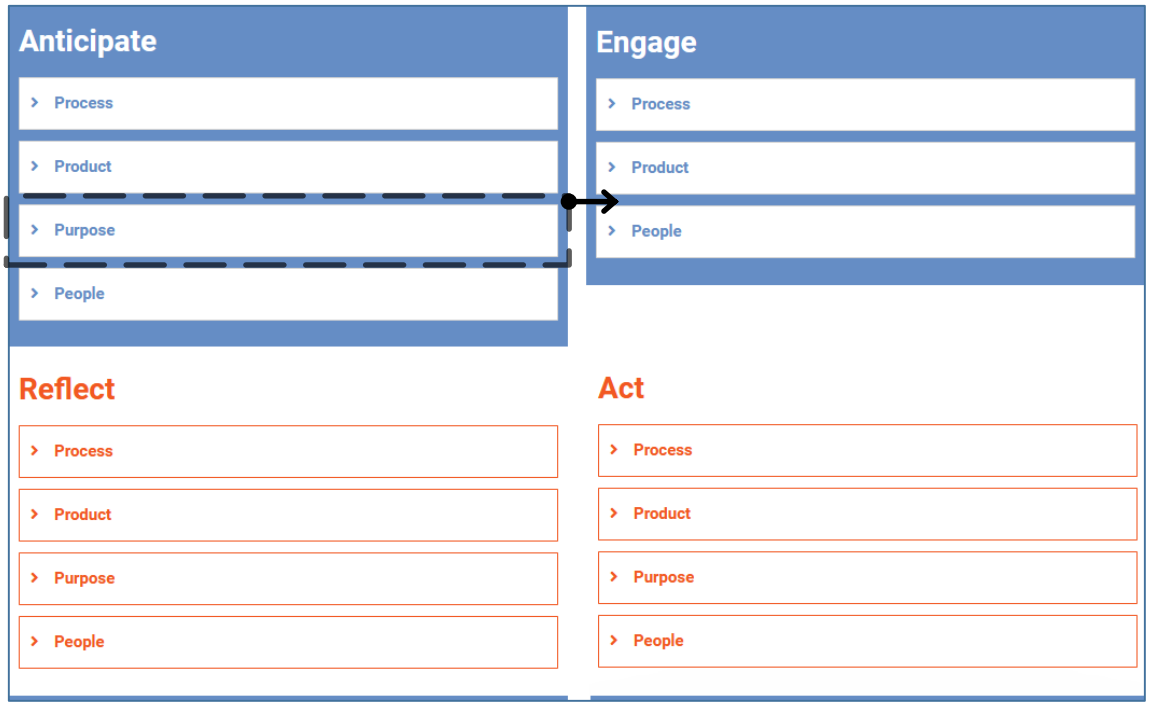
The phrase Responsible Research and Innovation (RRI) is commonly used to describe anticipatory, adaptive approaches to research that promote a safe introduction of new technologies into society (European Commission, 2012; IPCEU, 2014; EPSRC, 2018, 2020). Research commitment to this enables responsible research under conditions of uncertainty: researchers may revisit the riskiness of their project as it unfolds. On a semantic note, RRI's foundational work (Owen & Goldberg, 2010) originally used "responsible innovation" to describe basic principles which was formalised as RRI over time (Owen *et al*., 2012; RRI Tools, 2015). I use Responsible Innovation to indicate a wider concept; using science in a responsible way to develop technologies that improve, or challenge society to improve in some capacity[10]. I also use the term to avoid subscribing to the RRI school of practice – there are issues with public engagement guidelines that, given the subject of this thesis, seem sensible to distance from.

Responsible Innovation is a key part of the methodological approach within this work, but there are glaring omissions in the guidance available regarding the design and

[10] In fulfilment of the wider social contract between science and society.

reporting of participant engagement. The Anticipate, Reflect, Engage and Act (AREA) framework describes how researchers should approach RI and has the backing of the UK's Engineering and Physical Sciences Research Council (EPSRC, 2018). The Observatory for Responsible Research and Innovation in ICT (ORBIT, active as of January 2020) mapped the AREA framework to '4Ps'; core aspects of Responsible Innovation: Process, Product, Purpose and People (ORBIT, 2018). ORBIT's strategic aim is to promote and apply Responsible Innovation practices within information and communication technology (ICT) communities, but the teams who developed the AREA and 4P AREA frameworks were different groups of people which means that the current, institutional, approach to Responsible Innovation in practice has been disjointed. A lack of oversight is perhaps at its most obvious in the lack of information on ORBIT's website regarding why engagement is necessary for ICT research and innovation projects (i.e., the Purpose (PP**P**P) of Engage (AR**E**A) is blank, see Figure 12):

Figure 12: A visual indication of the lack of RI guidance for the "Purpose" of "Engage".



This is, perhaps, an oversight. Fortunately, this work provides guidance regarding the purpose of engagement which fills this methodological gap. Another point to note regarding ORBIT's construction of the 4P AREA framework is that "public engagement mechanisms" are identified as a way to structure engagement but no further guidance is offered (see Table 10):

Table 10: Insufficient and confusing 4P AREA guidance in how to "Engage".

ENGAGE	**How to engage a wide group of stakeholders?**	Identify stakeholders. Participatory processes. Process evaluation.
	What are viewpoints of a wide group of stakeholders?	Public engagement mechanisms. Prototype/ demonstrator evaluation (public).
	Is the research agenda acceptable?	Public engagement mechanisms.
	Who prioritises research?	Public engagement mechanisms.
	For whom is the research done?	Public engagement mechanisms.

The 4P AREA framework is aimed at ICT communities who operate on the bleeding edge of technological development. Available guidance is limited as to how ICT researchers and innovators can implement engagement, or the value in them doing so. If these communities lack guidance as to the purpose of engagement in research, then they will develop technologies that do not seek participant input or feed information back in an accessible way. Such work will lack inclusive and responsive practices, meaning that this innovation will be somewhat irresponsible.

In practice, Responsible Innovation proposals include researcher commitment, reflective methods as standard practice, and a diverse approach (Owen & Goldberg, 2010). Richard Owen described the wider philosophy of RI in a 2015 interview (RRI Tools, 2015):

> "[Responsible Innovation] recognises the transformative power of Science… that innovation is socially, politically and ethically entangled"

Responsible Innovation, or the on-going process of aligning research and innovation to the values, needs and expectations of society (IPCEU, 2014), addresses the challenge of unexpected problems being unearthed by science (EPSRC, 2020). Owen describes four qualities: aims and project trajectory should be inclusive, recognising the importance of diverse thinking and experiences; researchers should be able to anticipate risk and reflect on their work as part of standard practice; researchers must commit to research integrity and open access; responsiveness is key at individual and institutional levels (RRI Tools,

2015). Responsible Innovation equips researchers with tools, such as the anticipate, reflect, engage and act (AREA) framework, to directly address controversial issues. Standardising Responsible Innovation across the research community is important in developing public trust and confidence in research (Sutcliffe, 2011).

This thesis describes research that has been carried out to be socially desirable and act in the public interest, using Responsible Innovation as a framework to do so. This work uses inclusive and responsive data-practices to seek input from, and to offer feedback to, participants. Participatory research practices were used to do this; participatory research uses non-research input to improve the relevance and validity of research, making it more easily actionable (Barreteau *et al*, 2010). Participants have been known to contribute towards the research agenda, prompting discussions that researchers had not previously explored (Black *et al*, 2019). In this work, non-research input refers to feedback received from RUDY participants, the clinical project lead, the study and patient and public involvement co-ordinator, and the technology development team. In further chapters we will explore how RUDY participants helped to refined the research agenda of this thesis.

3.3 Project management

This section describes how I managed my research because this work required the development of a bespoke research approach. Over the course of study, Dynamic Consent and Responsible Innovation literatures did not provide sufficient implementation reporting, neither did they offer methodological guidance. This prompted me to design my own processes and procedures, which are reported here in the hope that it offers reproducible research and a novel methodological contribution. I used Requirements Engineering as a management approach (J. A. Hughes *et al*., 1993):

> A key objective of requirements capture is maintaining, by negotiation if necessary, consistency throughout the various stages to avoid incompatibilities arising in services the system is expected to deliver.

Requirements Engineering is a form of engineering where a specification for technology is from human needs being elicited, analysed, specified and validated (Jirotka and Goguen, 1994; Randall *et al*, 1994; Westfall, 2005). The type of requirements I sought were privacy requirements, which can be under-represented due to their ambiguity (Ambreen *et al*, 2018). As the literature review highlighted, I structured my thinking around privacy in terms of appropriate use. This framing was then used in interviews with participants to explore how data was collected, used and shared within RUDY. The reason that I consulted the RUDY team as well as RUDY participants was that users do not always

know what their requirements are (Maiden, 2008). So, asking RUDY participants for input without seeking information from those who had designed the platform would have ignored a valuable source of data. Engaging with multiple stakeholder groups is one way to establish different needs (Goguen & Linde, 1993; Westfall, 2014) that technology must serve.

Prototyping helps developers learn what their users need (Davis, 1993). Throwaway prototypes are low quality and useful for scoping, evolutionary prototyping develops a product over time, and operational prototyping develops a workable idea into a viable product. I used operational prototyping as a way to discuss the changes under development, with the RUDY team. I held regular meetings to share findings with the RUDY team, to suggest tasks, to seek approval and assignment of those tasks by RUDY's project lead. Throughout the process, I solved issues with insight and approval from RUDY. Where my findings identified issues, RUDY team members were better placed to advise on feasible solutions[11].

I had the opportunity to work directly with a software development team on a system currently in operation. The intervention study proposal (see Appendix 3), commentary on proposal (see Appendix 4), user testing document (see Appendix 5) and edits agreed with RUDY (see Appendix 6) demonstrate the evolution of this process. I produced an initial proposal based on interview and focus group findings, to which the RUDY team responded. This discussion helped to refine the proposal and prioritise certain tasks over others. Once the intervention had been implemented, I carried out user tests, stepping through the redesigned website with RUDY participants. From these tests, I drafted a list of final edits, which the RUDY team again commented on and helped to prioritise. RUDY's project lead officially accepted the redesign and those changes were published.

3.4 Research methods

Chapter 1 outlined the research aims of this this work, derived from a literature review. Specific research questions were provided for each research area, and these questions have been answered based on findings from both qualitative and quantitative data. Findings based on qualitative data can be problematic because problems arise due to data-subjectivity and researcher bias. To mitigate this, validation steps were used throughout this work to maintain as objective an analysis as possible, and to ensure the results were meaningful. Chapter 4 will describe in further detail how I consulted colleagues throughout my analysis, and I presented findings at a workshop, gathered

[11] Style of project management reporting taken from (Haas *et al*, 2021).

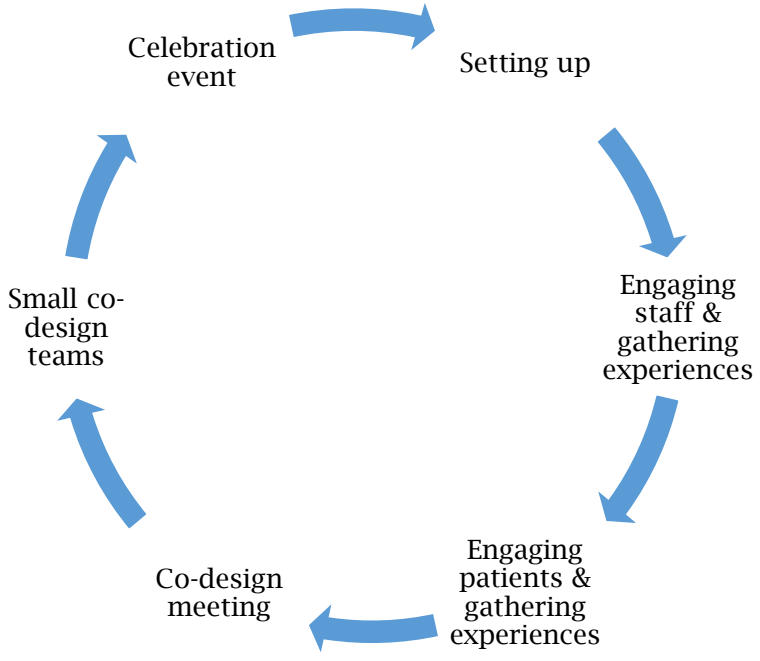
feedback and wrote a peer-reviewed paper. Similarly, the Chapter 5 will illustrate how I worked closely with a supervisor during data collection and analysis to reflect on emerging findings. At every stage of development, I presented findings to RUDY researchers and participants for review.

This chapter provides an overview of co-design, the inclusive research approach chosen for this work. Qualitative and quantitative data collection and analysis methods are described. A note on tool-kit synthesis provides an account of how the practical output of this thesis, the Dynamic Consent tool-kit, was developed from my findings.

3.4.1 Co-design

The academic literature surrounding experience-based co-design (EBCD) is largely clinical and relates to improving healthcare quality (Dimopoulos-Bick *et al*, 2019). Figure 13 shows Donetto *et al*'s (2015) model of EBCD as six stages: set-up, gather staff experiences, gather patient experiences, meet, break into teams, and celebrate. I had to reject parts of Donetto *et al*'s model, such as having larger and smaller co-design meetings, in favour of a process that was easier to manage and has less organisational overhead. However, the model provided direction for developing the enhanced feedback intervention. I engaged with staff by running a focus group with the RUDY research team, and engaged with participants by carrying out interviews. The online focus groups and software development cycle served as small co-design teams exploring and then implementing a solution. The celebration event is to be a short video sent to participants via RUDY and posted on the project's social media page.

Figure 13: The six stages of experience-based co-design (from Donetto *et al*, 2015).



To engineer requirements, user input is needed. A well-defined and structured software development process is crucial to delivering a good-quality solution on time and within budget (Sadraei *et al*, 2007). Co-design was used to draw out user need by encouraging target users to reflect on their experiences. Palmer *et al* describe co-design as a zeitgeist, illustrating a shift towards consulting participants for solutions that are tailored to their needs (Palmer *et al*, 2019). Palmer *et al* previously used co-design to develop an intervention improving psychosocial recovery from severe mental illness (Palmer *et al*, 2015). The authors used interview and focus-group data to inform the development of an intervention, an approach also adopted in this work. User-testing was used as part of the co-design process as a point of reference, to reflect on whether user requirements had been met by the solution under development.

In another example, researchers worked with 48 young people to develop an eHealth intervention. These young people had long-term physical conditions and helped the researchers to design, develop and refine ideas. The researchers then recruited a further 20 participants to take part in a pilot trial to observe whether the implementation was acceptable and usable (Thabrew *et al*, 2017). Another health-related intervention was a study that gamified cognitive behavioural therapy on smartphones (Christie *et al*, 2019). In this example, software development cycles were combined with "think-out-loud" user tests. In this work, user tests allowed problems to be fixed before the intervention was published. I distributed a call guide ahead of time to allow participants to familiarise themselves with the tasks. Some participants took the initiative to explore changes and provide feedback of their own accord. This feedback highlighted issues that neither I nor my RUDY collaborators had identified.

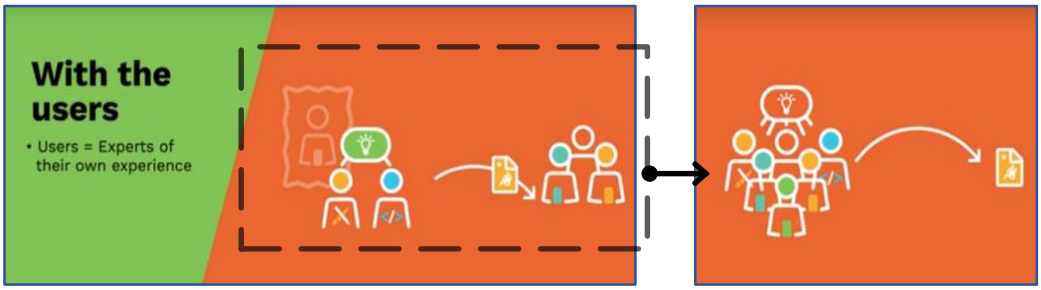
Co-creation design (hereafter referred to as "co-design") is a form of project management and development that involves a key, but often overlooked stakeholder: users (Goodyear-Smith *et al*, 2015):

> With co-creation design… [a] participant is not someone on whom research is "done", but is actively engaged in designing and implementing the research process"

Some co-design processes take years (Hjelmfors *et al*, 2018) and others host "world cafés" (Gahan *et al*, 2019). This project used co-design for scoping work (Fleming *et al*, 2019): to understand user experiences of RUDY. Participants, researchers and software developers were key stakeholders in the development of this work. During co-design "a participant is actively engaged in designing & implementing the research process" (Goodyear-Smith *et al*, 2015).

This work used co-design as an overarching methodology rather than a prescriptive method due to its flexibility. Co-design is adaptable; examples of its use include the development of anxiety and mental health treatments for young people (Thabrew *et al.*, 2017; Christie *et al.*, 2019; Fleming *et al.*, 2019) and screening for elder abuse (Gahan *et al*, 2019). As a method, it is well-suited to serving vulnerable populations, making it an excellent choice of method for collaborating with RUDY. Co-design is grounded in academic and clinical literature, but this work also draws from resources published by the National Co-ordinating Centre for Public Engagement (NCCPE) and the Co-Create Project. NCCPE and Co-Create offer practical guidelines for implementation, for example Figure 14 is from Co-Create's website and illustrates how users should be consulted as experts during design processes.

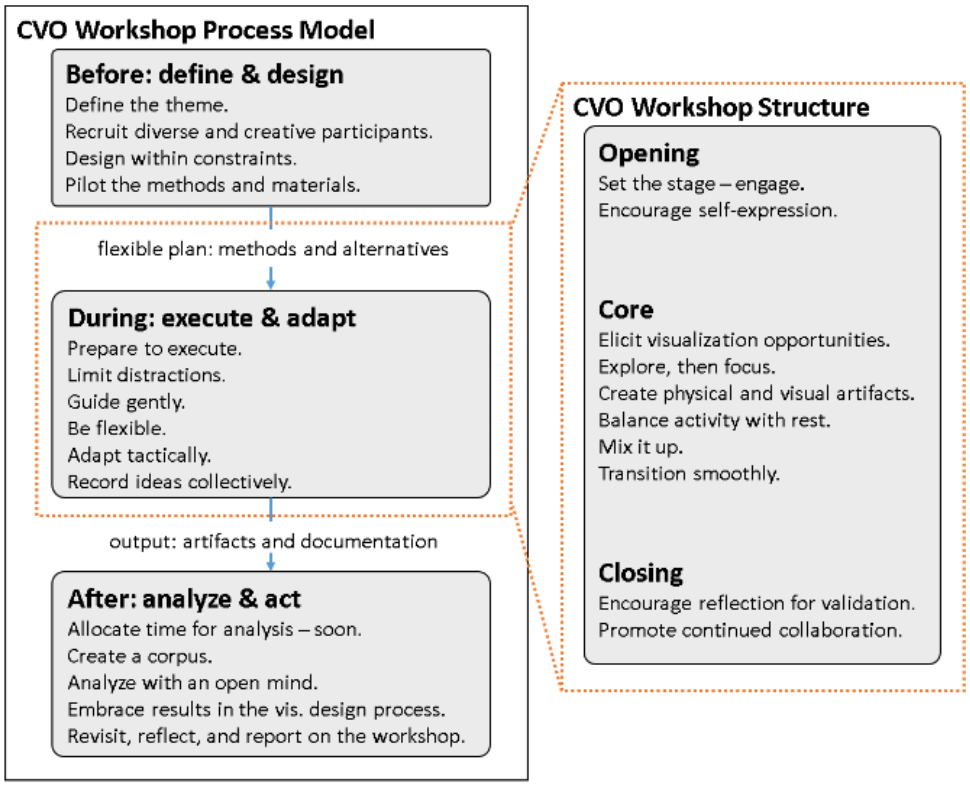
Figure 14: An illustration of co-design (Co.Create.Training, 2019).



This research collected insights from two types of people who used the RUDY platform: the research team who created it, and research participants. These groups have different types of expertise, each of which was invaluable during the intervention design process. Seeking user input as that meant misunderstandings could be identified (Chapter 4 outlines conflicting accounts of engagement within RUDY), and design flaws smoothed earlier (Chapter 5 identifies website usability issues and overly-technical wording).

The principle of co-design is to consult those who will use a product during the design of that product. This is a form of risk mitigation, avoiding as many problems in advance as possible. Co-design involved online focus groups with participants and a software development cycle with the RUDY development team. To develop the online focus groups, I drew from Kerzner *et al* (2018), who designed a workshop process model and structure. The authors prioritise participant wellbeing, open communication channels and flexible methods.

Figure 15: Workshop process model and structure (Kerzner *et al.*, 2018).



This is a research approach that I incorporated these considerations into my own research approach by ensuring breaks, providing contact details and offering different communication channels (telephone, in-person, video-calling and email). Figure 15 shows how a workshop can be situated within a wider research process, with pre- and post-workshop activities. The first step in developing my co-design work was to draft information flows, thinking about the information that would be received from and communicated to participants before, during and after the focus group. Where Kerzner *et al* "recruit diverse and creative participants" and "record ideas collectively", My recruitment pool was pre-determined due to the fact-finding nature of the session, and I retained the administrative burden of synthesising outputs. The aim of the focus group was to develop the enhanced feedback intervention with input, agreement and validation from stakeholders. The collective recording of ideas would have been difficult and placed an administrative burden on participants[12], most of whom took part via a mobile phone that would have made digital notetaking untenable.

I chose to run online focus groups because this was the only way to gather RUDY participants in one place. Many participants cannot travel, and are located across the UK.

[12] E.g., by asking them to learn new tools (online whiteboards, collaborative note-taking, etc.).

Members of the patient forum who took part were already familiar with Skype, as this was how they had taken part in the forum before. I asked other participants which tools they were comfortable using, and Skype was the most popular option. Virtual focus groups are not ideal however – while there are similarities between online and face-to-face interactions, online environments can be distracting (Abrams *et al*, 2015). People can find it harder to focus, concentrate for extended period of time, and can be easily distracted by technical issues (e.g., repeatedly asking "can you hear me?").

"Before: design & define", refers to initial preparation and call guide design. "During: execute and adapt", refers to the focus group execution, and "After: analyse & act" refers to the collation and analysis of the notes gathered. I used the workshop structure to create an agenda for the sessions, with "opening", "core" and "closing" sections. Where, under "Workshop structure: Core", Kerzner *et al* "Elicit visualization opportunities", the focus of this work was on eliciting opportunities for engagement. I provided information ahead of time, to allow participants to read and reflect on content ahead of time. Participants were also encouraged to communicate questions, comments or concerns beforehand, but no such feedback was received. During the session, timings were flexible but no amendments were necessary. I checked participant understanding throughout the call by asking for opinions and summaries of key points. This was done in case repetition was needed – it was not. After the call, I provided feedback to participants. Kerzner *et al* "promote continued collaboration", and I chose to notify participants of next steps and provide a point of contact. One participant sent me some suggested reading.

3.4.2 Qualitative methods

Qualitative methods, interviews and focus groups, were used in this work to explore user perceptions of data-use in research. The resulting data was detailed, insightful and demonstrated nuances within different people's experiences. Qualitative data corroborates quantitative results, increasing confidence in findings and quality of research (Sofaer, 2002; Toomey *et al*, 2017). The methods used in the work supporting this thesis were telephone, online and in-person one-to-one interviews, and in-person and online focus groups.

During in-depth interviews a researcher asks particular questions to get an understanding of a particular subject, e.g., patient-centred care (Fix *et al*, 2018). Interviews generate descriptions and interpretations of people's social worlds (Ritchie *et al*., 2013), and can be useful in eliciting technological requirements (Ferrari *et al*, 2016). Kvale describes a "traveller-interviewer" as someone who "asks questions that lead the subjects to tell their own stories of their lived world" (Kvale, 1996), a philosophy used in this work. Chapter 4 describes how scenarios were used to encourage participants to share their lived experiences. I wanted to explore the motivations behind individual

attitudes to data-use for research purposes, and to identify any evidence of the key principles described in the academic literature behind Dynamic Consent: revocation and engagement. Interviews were used by I to gather shared understandings and definitions, identifying issues for further investigation.

Core Dynamic Consent literature describes how focus groups have been used to describe the accounts of expert and participant perspectives. This work follows suit, collecting data from both researchers and participants. Focus groups provide rich data (McLafferty, 2004), drawing upon participant attitudes, beliefs, experiences and reactions in a way that other methods such as observation, one-to-one interviews and surveys do not (Gibbs, 1997). Group interaction is important because this allows a researcher to observe group dynamics as well as individual accounts (Holloway, 2005; Kitzinger, 1995). For focus groups to be successful, an interviewer will need to recruit a group of people who share something in common. Homogeneity within a group is important because of an (originally therapeutic) assumption that people who share a problem are more willing to discuss it in the company of others with the same problem (Lederman, 1990). Lederman also sets out practical steps for focus-group sessions as articulation of purpose, definition of recruitment criteria, interview guide creation, focus group interview, data analysis and presentation of findings. Analysis of the content, linguistics and word count for different focus group methods (face-to-face, online audio-visual, and online text-only) suggest that face-to-face is the best way of running focus groups (Abrams *et al*, 2015) because interactions between participants are easier to observe. Section 4.1.2.1 describes a face-to-face focus group with the RUDY research team, and Section 5.1.1 gives an account of online focus groups.

Qualitative data lends itself to interpretation and analysis through thematic analysis and grounded theory. Thematic analysis (Braun & Clarke, 2006) and grounded theory (Charmaz, 2006), two different approaches that complement one another. Both provide structure and are flexible enough to allow undefined topics to emerge:

> "Grounded theory coding consists of at least two phases: initial and focused coding. During initial coding we study fragments… for their analytic import. From time to time, we may adopt our participants' telling terms as in vivo codes. While engaging in focused coding, we select what seem to be the most useful initial codes and test them against extensive data. Throughout the process, we compare data with data and then data with codes" (Charmaz, 2006).

Data analysis consisted of reading interview transcripts and "codifying" them with an initial, and two focused cycles.

> "to codify is to arrange things in a systematic order, to make something part of a system or classification, to categorize. When codes are applied and reapplied to qualitative data, you are codifying" (Saldaña, 2013).

The analytic approach was substantive rather than structural – I was concerned with what the data described, i.e., reported experiences, rather than specific wording.

3.4.3 Quantitative methods

Quantitative methods, statistics from preliminary data samples and an intervention study, were used in this work to observe participation in RUDY via questionnaire completion rates. Data provided by RUDY indicated that questionnaire completion dropped less for a questionnaire that allowed participants to report positive experiences like enjoyment, cheerfulness and relaxation (Zigmond & Snaith, 1983). This heuristic was compounded by qualitative co-design data (Section 5.2.1) and provided direction for the intervention study under development. Data collection pre- and post-intervention indicated an overall change in participation rates, showing that the intervention had an effect. The initial data provided served to justify the intervention study, which was mission-critical in supporting this thesis. This work does not use complex statistical methods because they were not needed. Comparative statistics were used to investigate data pre- and post-intervention, and graphs were generated to illustrate these comparisons. This research is scoping work that has identified barriers to Dynamic Consent's implementation, so the methods used have been designed to be "minimally sufficient" in setting up this problem and providing reporting criteria. More rigorous qualitative testing of these ideas lies in store for other researchers whose interest has been piqued by this problem, and whose interrogation of this work has opened up further avenues of investigation:

> "Our questions should dictate our methods and analyses, not vice versa. We should not do research in a particular way simply because we can. We should not use a statistical technique simply because we can understand the software", (Peterson, 2009).

Intervention studies are used to measure cause and effect. Rather than cross-section studies that show a single state at any given time, interventions follow behaviour over some period. Researchers have used intervention studies to investigate the role of communication within informed-consent practices for decades. "Extended discussion" has described the creation of more involved conversation between participants and researchers (Flory & Emmanuel, 2005) and shown to improve research understanding (Nishimura *et al*, 2013). Interventions have also been used to measure engagement in the workplace (Knight *et al*, 2019). Of particular interest to this work is that co-design has been employed by other researchers to scope the requirements for an intervention study

(Knight *et al*, 2017). This particular example used participatory action research, which asked domain-experts (nurses) to describe the problems they faced. The authors also concluded that implementation evaluation is an essential, and often overlooked part of the research process. This thesis addresses this particular concern by including a section on research quality (Section 6.1.3), drawing from frameworks evaluating methods and outcomes (Mullen *et al*, 1985; Proctor *et al*, 2011).

Reporting research findings after they have been discovered without describing the initial design work required is a barrier to good practice. While emergent findings do occur, normalising the description of post-hoc analysis as part of the original design approach is a limitation that makes findings harder to replicate (Curran-Everett & Milgrom, 2013). "Post hoc" means "after this". Curran-Everett and Milgrom advise caution in post-hoc data analysis in biomedical research, offering the example of a researcher who has carried out their planned analysis to no avail and encountered unexpected results which they report as part of their intended analysis. Unplanned post-hoc analysis can be useful in providing a means to generate scientific hypotheses for further study. The work described in Chapter 4 describes an analysis plan, as well as how presenting findings to RUDY collaborators resulted in a post-hoc analysis data stratifying initial and follow-up participation.

3.4.4 Tool-kit synthesis

Chapter 7 describes a tool-kit created to address a lack of participant engagement in research. This tool-kit is a series of design prompts: a model, a framework and a specification. Each of these draws from the original work supporting this thesis and from academic literature. I identified problems with existing tools, namely that they were difficult to understand and use in practice. I developed the tool-kit in response to these problems by using illustration, focusing on key information and developing tools that could be used in isolation as well as with the available guidance.

3.5 Ethical considerations

I submitted two applications to the Central University Research Ethics Committee (CUREC) for this work. The first covered the interviews and focus group (Chapter 4[13]), and the second supported the co-design research reported in Chapter 5. Ethical approval was also sought from RUDY's data-access committee so that the project had oversight on the

[13] The participant information sheet was used in an Introduction-to-ethics training session for the Social Sciences and Humanities, cited as "a great example of using common sense to deviate from the 'official' text while still getting the right information across" *(personal correspondence)*.

work that was taking place. The intervention study did not require university approval, on the advice of a Senior Clinical Research Support Manager, from the clinical trials and research governance (CTRG) department. A before and after service evaluation approach was taken because a randomised trial would have required a substantial amendment. This would have delayed the work past my expected DPhil end date.

During the first study, key areas of ethical concern were confidentiality and data-protection. Participants details were stored in a password-protected file on a university laptop. Interview and focus group transcriptions were kept in secure university data storage. In addition, I was conscious that that interview discussions about online data-use might increase levels of paranoia, and made sure to use non-emotive and neutral language. Given the nature of her research as an exploration of consent in healthcare-related fields, I was also aware that discussion topics could touch on sensitive and personal areas. Part of the introduction scripts communicated to the participants that they could stop the interview of take a break at any time.

Ethical approval sought for the co-design work described confidentiality and data-protection as issues, also highlighting risks to me and the people taking part in my study. During the previous study, I underestimated the level of resilience required to interview over fifty people suffering with rare-diseases would. To address this, I created a "care plan" making time for regular reflection and use of support networks such as her supervisory team and resources provided by the Department. A key concern was participant fatigue. Rare conditions impact communication, stamina and other factors affecting social and intellectual interaction. To mitigate this, and given the success of email as a form of asynchronous communication, I recruited via email. Call guides were sent to participants ahead of time so that they could take part at their own pace.

A short note on integrity and data collection: I decided early on that I wanted to focus on scoping issues that RUDY had, and that the output of this would serve the RUDY community. This meant that, rather than demand RUDY's entire dataset from 2014 to 2020[14], I would work with specific data-samples that RUDY could more easily provide. This is why I decided to use periodic intervals to glean insight into participant behaviour over time.

Consent is often a trade-off where the consenting party takes on a degree of liability. This has had two practical implications: I abided wholeheartedly by existing legal protections, and made decisions that would impact RUDY's live operation with the

[14] The focus-group indicated that different consent versions were in operation due to the longitudinal nature of the project. I decided to use data provided by participants with the most up-to-date consent provision.

agreement of key stakeholder groups. As to the first point, it is pertinent to note that institutional ethical approvals do not provide any guarantees to participants as to what they can expect from the research process. This was my responsibility as lead investigator for this project. Secondly, technologies are created to fit human needs. Rather than simply drawing conclusions from academic literature, I consulted RUDY participants and researchers throughout the project to ensure the work was relevant. This consultation also served to validate data collection. E.g., as part of the co-design process described in Chapter 5, focus group findings were emailed to participants. Their responses confirmed some findings and asked for clarification about others, which informed the software development process. This research procedure has compensated for one-time consent's failure to inform over time by seeking participant input and oversight.

3.6 Reflection

This chapter has outlined the overarching methodology of the work supporting this thesis, described fundamental research principles and described the mixed-methods approach. This methodology was developed to address research questions drawn from a literature review on consent, privacy and social contract between science and society. A mixed-methods approach draws from both qualitative and quantitative research methods, increasing the confidence with which the conclusions of this work are drawn.

Gathering qualitative data through interviews and focus groups has allowed a deeper and richer exploration of an implementation of Dynamic Consent. The results from interviews and focus groups are not generalizable but they provide a rich illustration of the RUDY project, allowing the reflection needed to refine the intervention study's hypothesis: enhancing feedback has a positive effect on research participation. Quantitative data was gathered via an intervention study – data was collected pre- and post-intervention to empirically test this idea. The quantitative data collection validated qualitative findings. These conclusions were developed into a set of materials designed to support researchers in designing engaging data-practices.

The studies that follow have been designed to be rigorous and replicable. Validation steps include seeking participant feedback; findings are presented in a lay-summary and feedback is sought from RUDY participants, researchers, professional colleagues and other experts. Suggestions are offered for others seeking to develop similar practices. Chapters 4 – 7 describe the data-collection, analysis and findings of four different studies, linked by an overall thread of investigation: a service evaluation of Dynamic Consent. Each chapter has a methods section that offers more comprehensive reporting of how research methods described in this chapter have been put into practice. In this work, I

will highlight the importance of two-way communication; the importance of Responsible Innovation research data-practices that are both inclusive and responsive.

Chapter 4

Study 1: rules of engagement.

"Your consent preferences allow a system to be personalised to the person's level of risk sharing", RUDY project lead.

4.1 Introduction

This chapter describes an empirical study that outlines challenges faced by RUDY researchers trying to implement inclusive and responsive data-practices: namely, participants reported a lack of engagement. RUDY is an epidemiological study collecting data over time, meaning that a long-term approach to consent is important. Dynamic Consent offers one way to design consent practices that inform over time, and while RUDY implements Dynamic Consent, there is little empirical evidence to justify its use.

Revocation of consent was not reported at all, but participants reported conditions under which they might revoke their consent: if they lost confidence in an institution, or the institution started to behave in an unethical manner. Researchers had provided ways in which participants could change their data, or control access to it. While participants agreed that they should have ultimate control, or "the final say", there was no evidence that this kind of control was actually exercised; participants showed little interest in having granular control over research processes, favouring delegation to

researchers as experts. "Feedback" refers to how information is communicated and how interactions are designed (Coathup *et al*, 2016). Participants showed interest in receiving feedback and were able to describe the kind of information they would like to receive, but the majority reported a distinct lack of feedback from the study. The majority of participants interviewed had no idea of RUDY's overall goals. This lack of engagement was problematic because user experience and involvement were reported as two key design considerations by the research team. Where experiences of engagement were reported, this was by members of RUDY's patient focus group. These participants reported feeling involved with the study whereas other participants reported that they felt uninvolved.

Analysis of the data collected over the course of this study corroborates previous reporting of how informed consent has failed: participants do not understand research, even at an abstract level, trusting researchers entirely to work in their best interests (Ludman *et al*, 2010; Ormond *et al*, 2009; Simon *et al*, 2011). However, the insights provided show how RUDY researchers have had to balance competing and changing priorities, often with positive results. I also describe nuanced expectations that participants have of researchers, regarding data-use, often influenced by positive or negative interactions with medical professionals.

4.2 Methods

Section 3.4.2 outlined the importance of qualitative research methods to this work, in exploring user perceptions of RUDY. Interview and focus-group data was collected to investigate the first research aim of this thesis: to observe and evaluate an existing implementation of Dynamic Consent. I ran a focus group with the RUDY research team on-site: the project lead (an epidemiologist), the study coordinator, the technical lead and network programmer, and the database and web development programmer. I also carried out interviews with RUDY participants via telephone and in-person. When a participant requested an email interview, I added this option to my research protocol. The interviews and focus group were structured around simplified scenarios (vignettes) to encourage participants to share lived experiences. Thematic analysis and grounded theory were combined in the analysis of this data to draw out overarching themes. I ran a data-session with colleagues in the Computer Science Department to test ideas as analysis was ongoing, and these findings were validated as part of a workshop presented at a prestigious summer school. This workshop was written into a conference paper.

4.2.1 Vignette development

A vignette is an abstraction of a real-life scenario that can be used to explore sensitive topics because they distance a participant from the situation (Torres, 2009). As a researcher who recognises that social context plays a role in understanding how people behave, vignettes allowed me to conduct an in-depth exploration of participant responses (Hughes, 1998). Hughes *et al* also used a story-telling method, where one vignette develops from another, which I initially modelled my work on but later had to re-visit. Rose *et al* (2015) used vignettes to present a scenario with choices for how a clinician would react to a patient presenting with certain symptoms. These vignettes were constructed to reflect clinical practice and maintain consistency across the 2,795 participants in 11 different jurisdictions. When asking people from different cultures about aging, Torres wanted to avoid imposition of culture because the issue laden with cultural context (Torres, 2009). Vignettes could not on their own convey real-life complexities or guarantee that a participant's response reflected their actual behaviours, but they provided prompts for discussion. They allowed participants to talk about their understanding of a situation and voice an opinion without having experience to draw from. In this study I investigate the social context around research data-use, and wanted to collect data from different stakeholder groups: researchers and participants. Rather than ask participants about decisions that they may have made in the past, I wanted to draw out individual values and attitudes.

I developed vignettes to encourage participants to provide answers that reflected their own experiences rather than what they thought I wanted to hear. Two vignettes were used during data collection, which were reviewed and modified after several interviews, to include a familiar example of where data needs securing: banking. This change was inspired by participants using their experiences with banks as an example of "good behaviour", without any prompting. In the first iteration, vignette 1 involved a fictional participant who has enrolled in a study "that allows them to revoke access". The character hears of a data breach in the news and the interview questions explore whether or not the participant thought that the character should remove themselves from the study, and why. Vignette 2 involved two characters, both signed up to a fictitious study. One character receives emails regularly and the other receives both emails and social media updates. This was designed to explore whether a demand for Dynamic Consent might exist outside of research context. I wanted to know whether or not participants thought that their interactions with RUDY served as an example for their relationships with other organisations collecting and using their data.

I used vignettes to discover conditions under which a participant might revoke their consent from a study. I wanted to explore any data-protection responsibilities that the

participant thought that researchers had. Vignettes 1A and 1B are the first iterations of vignettes used in interviews. There were two significant problems with them. First, I assumed that participants would equate the fictitious study to RUDY, and second, that the participant understood specialised terms such as "revocation" and "data breach". Vignette 1A was simplistic and distracted participants with undefined variables. The language used in 1B was dense – participants were unable to follow the vignette's story.

Rather than an overarching story with two parts, vignette 1B was given a new story with a security focus, resulting in vignette 2B. This is because after 3 interviews, participants were reporting data-security concerns without any prompting at all. They used experiences of their bank's reaction to credit-card fraud as a positive example of institutional accountability. If the event of a breach, they argued, they wanted a trusted party to notify them and take action. In response to these initial interviews the vignettes were refined and simplified and this structure made them easier to follow. Vignette 2B explores Anna's experience of credit card fraud while 2A explores Manisha's experience of a study "where she can provide contact details and log symptoms and information about diagnoses". Note the clarity between this description and a study that "allows them to revoke access" which was used in 1A. The changes made to the vignettes allowed participants to engage in discussion more easily. I observed that participants drew parallels between the vignettes and their own experiences and asked clarifying questions which suggested that they were more engaged with the task; the new vignettes had clarity. Another indication of this was that participants were more forthcoming with their opinions, and seemed more confident in giving them.

Vignette 1A

The participant has enrolled in a study that allows them to revoke access.

In the news there is a data breach at the University responsible.

- What should the participant do?
- Is there any risk here?

The participant revokes the study's access to all of their information.

- What might happen?
- Why do you think they might have done this?
- Should they give that access back?
- Would anything be needed from the university to encourage this?

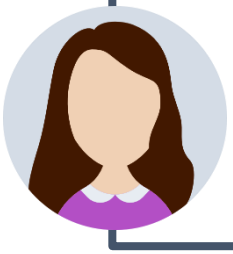
Nothing seems to happen.

Vignette 1B

Person A is signed up to [one] ~~a~~ study [and] Person B to another. Both of these are long-term studies over a year or so.

Study A sends emails every month to arrange a meeting, the participant confirms a time that suits them and if they cannot make it they must call 24 hours ahead of time. There is no penalty for cancellations but if you miss three consecutive studies the scheduling stops.

Study B also sends an email to participants every month to schedule a meeting. In addition to this there is a Twitter feed that participants are encouraged to sign up to. Related articles are sent out, and participants can use this to engage with the project.



[Open answer]

Point of discussion: Is Dynamic Consent something that should be happening in other parts of the participant's life?

Vignette 2A

Manisha has a rare condition and has enrolled in a study at a local hospital where she can provide contact details, log symptoms and information about diagnoses.

She is largely housebound so a researcher enrols her over the phone. Once they have hung up, Manisha realises that she did not take the researchers name.

- How could she get back in touch?

6 months after joining the study, Manisha wonders whether the researcher she had been talking to had collected enough people's information for them to do their work.

- What might she have to do to find out?
- What kind of information might she discover (what might you want to know of a study you took part in)?

Manisha is sharing her mobile number, email and symptom log and is happy to be contacted about other research trials. On the news, a 'data breach' at the hospital is announced.

- What does 'data breach' mean to you?
- Is a 'data breach' a problem?
- What do you think the patient has a right to know?
- What responsibilities do researchers have in this area, if any?
- What is personal information and who does it belong to?

Manisha decides to withdraw consent for the study to access to her mobile phone number and share her information with other studies. She continues using the symptom log and after a couple of months allows the researchers access to her mobile number again.

- Is control of your information and decisions made about you important to the delivery of your healthcare?

Vignette 2B

Anna has signed up for a store credit card. Every time she shops at that store she collects points that add up to vouchers that she can spend in store.

Anna notices that her credit limit has increased without her permission.

- How could this have happened?
- What should she do?

She decides to call the shop the next day but does not get round to it. Three days later she receives a call saying that a purchase had been made in another county that reached her (new) credit limit. The shop refunds her the amount.

- Is this a problem?

Anna worries that her card details were taken via transactions [that] she had made online or that her computer is infected with a virus that collects her information.

- Do you think she is right?
- Do you worry about providing data or interacting with people online?
- What kind of information is important to be kept private or secure?

4.2.2 Qualitative methods

Interviewees were based across the UK and unable to travel, had different levels of communication difficulties, and were at different stages of their condition and care. Focus-group participants were interviewed at their workplace. My analytic approach focused on the substance of what was said, rather than terminology or sentence structure; I found that there were sentiments shared across both groups.

Focus group

People who share a problem are more willing to discuss it in each other's company (Lederman, 1990). Lederman's paper provides practical steps for carrying out focus-group sessions. Table 11 shows how these steps were used to design this study. The focus group was run with the aforementioned members of the RUDY team: the project lead, study coordinator, technical lead and web developer. Vignettes 1A and 1B were used as prompts for the focus group and the session lasted 90 minutes. The initial intention was to interview each member individually, but the subjects suggested that a focus group setting would spark discussion, offer group consensus on key points and provide an insight into team dynamics. During the interview, team members reminded each other of past events, and their rapport helped draw out shared understandings, such as what the project aims of RUDY were. This rapport also meant that individuals did not hesitate to talk about both positive and negative experiences. One limitation, however, was that dominant voices took over the conversation. This was helpful at times, e.g., when the project lead would summarise and other members would confirm this and provide other insights. Elsewhere however, discussion veered off-course and quieter voices were less forthcoming than their more vocal counterparts which means that insights may have been missed.

Table 11: Designing a focus group session (following Lederman, 1990).

Attribute	Implementation in this study
Articulation of purpose	The purpose of the interview is to collect supplementary data on the RUDY research team's perspectives of the RUDY project. The overall research question I am answering with this focus-group data is RQ1: to observe and evaluate an existing implementation of Dynamic Consent to identify conflicts and similarities with existing literature. I break this down into the following: 1. Why did the RUDY team consider Dynamic Consent appropriate for their project? 2. How did the RUDY team design Dynamic Consent into their research study? 3. What are the RUDY team's perceptions of Dynamic Consent at present? In particular, the issues I hope to cover are the following: how RUDY participant data is used, perceptions of data-risk and ownership, the role of consent, and the perceived importance of communication.
Definition of recruitment criteria	The recruitment criteria for this was quite simply, that the participants were part of the research team who had developed the RUDY study.
Interview guide creation	An interview guide was developed (Appendix 7), as a prompt for individual interviews. I used this as a guide for the focus group, but I did not include time prompts, such as "allow focus group members to respond one by one [2 – 3 minutes]". This meant that some participants spoke more than others. I address this in study 2 by adding structure to the interactions – participants are called on one by one in order. This continues, until participants are comfortable interacting freely.
Focus group interview	In this study of a research team's perceptions of implementing Dynamic Consent, I was the only interviewer. When the focus group was carried out I was an experienced group leader, drawing on my experience within a research context. When responses to questions

	had been exhausted, I asked whether there was anything else anyone wanted to say. During the focus group, when I required clarification I provided a summary and asked for corrections to minimise potential misinterpretation. I did not provide regular summaries which I think would have minimised unwarranted inferences and assumptions during the focus group. One positive example was that a participant asked a question and gave a summary unprompted, which led to further discussion about a related and relevant point I had not thought to ask about. I recorded the focus group and took brief observational notes. A transcription of the audio recording served as the data I later analysed.
Data analysis	I used thematic analysis to explore data gathered from this focus group. Please see Section 4.1.3 for a more in-depth description.
Presentation of findings	The findings from this study have been presented in this chapter in Section 4.2, contributing to this body of doctoral work.

Interviews

The inclusion criteria for these interviews was simple: an individual had to be taking part in RUDY, and their participant record needed to indicate that they had given consent to be contacted for other (i.e., non-RUDY) research purposes. RUDY's study coordinator sent emails out to the appropriate participants, who were asked to contact me independently. Any responses accidentally directed to the coordinator were forwarded on. When participants expressed interest, I sent participant information forms (Appendix 8) and asked the individual to indicate their availability. Initially, telephone and in-person interviews were offered. Email interviews (Carlsson *et al*, 2007) were made available when one participant asked whether the interview could be done asynchronously. This was added to the research protocol, and used by later participants.

54 RUDY participants were interviewed, and two accounts excluded: one of these participants did not want to contribute to non-clinical research at the end of their interview, and I failed to collect appropriate consent for the other. Of these 52 interviews, this study's findings are based on 22 accounts due to a hard-drive failure combined with

a software-backup failure. It is perhaps of some good fortune that findings converged at 20 interviews.

Of the 52 interviews, 2 were in person, 46 were carried out over the phone and 4 were done via email. Of the 22 final transcripts, 66% of testimonies were from women and 95% were from white people. 66% of testimonies were from participants educated to college, vocational or secondary level and a minority of participants held postgraduate qualifications. The majority of participants were over 55 years of age, with 35% being over 65. These trends are consistent with the larger set of interviews. Unfortunately, demographic information was lost as well as interview data.

4.2.3 Analysis and validation of interview data

"Pragmatic eclecticism" is when a researcher decides to finalise their analysis methods after initial data collection and review (Saldaña, 2013):

> "Others, like me, believe in the necessity and payoff of coding for selected qualitative studies, yet wish to keep themselves open during initial data collection and review before determining which coding method(s) – if any – will be most appropriate and most likely to yield a substantive analysis. I label this personal stance 'pragmatic eclecticism'"

This was the approach I employed, to ensure that the findings drawn from this series of interviews represented what participants were saying, as much as possible. The 22 surviving transcriptions were imported into Nvivo software. Analysis was broken down into three parts: initial code development, first coding cycle and second coding cycle. The initial analysis plan consisted of three parts: transcribing, focused-coding, and applying themes to interview and focus-group data. The only substantial change made to this plan was that, as I collected and reviewed data, a validation step was added. This was done as a "data-session" with peers in the Computer Science Department before analysis was finalised. I did not have resources for two analysts to work through and code the data, so I used the data-session to provide a brief overview of the study alongside anonymised interview samples, and ask for feedback on the analysis in progress (Appendix 9). This feedback was incorporated as part of a final coding cycle.

During initial coding, I read through the focus-group transcript and chose extracts that I thought described Dynamic Consent in practice (i.e., I codified the data). I sorted these extracts into two broad categories: researcher (160 extracts) and participant (90 extracts). Nvivo has a word cloud function, which I used to provide an overview of key words within the two categories: participant-related and researcher-related. Figure 16 indicates that within the focus group, the RUDY team understood that researchers and participants

have different information requirements. Where they talked about themselves and their work they used more specific, technical terms like "data" and "patients". Where they discussed RUDY participants they tended to use broader terms like "information" and "people". These word clouds provided a cursory look at the data and were not used in any further analysis.

Figure 16: An overview of key words from focus-group data.

Researcher-related focus group extracts Participant-related focus group extracts

For a general overview of the interview and focus-group data combined, I initially grouped responses by interview question, looking for similarities and differences. Both types of session had used the vignette structure, so shared common prompts. Table 12 shows topics that emerged after initial analysis of both the focus-group and interview data. These topics were of interest because they appeared repeatedly: they were revisited over the course of the focus group, or appeared across different participant accounts. The focus group data helped to shape my thinking, but the thematic framework created to analyse the full dataset was based on interview data. After reading through all 22 interview transcripts, I highlighted extracts of interest from five of them. Table 13 shows how I analysed each extract by grouping directly relevant quotes, and identifying groupings where connections were less obvious and required further investigation (Rabiee, 2004).

Table 12: Overview of topics of interest arising from initial analysis.

Focus group	Interviews

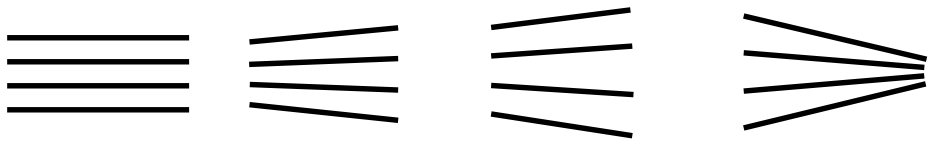
The RUDY project:
• Is a longitudinal study. They need patients to stay even if not sharing data with other researchers.
• Has relationships with patient groups (charities) → reputation and recruitment benefits.
• Has design principles: "don't be boring", "be relevant" and "report lived experiences".
 	
RUDY is a patient-facing project:
• RUDY collects data from patients directly, other projects collect data from clinicians.
• A significant priority is how to effectively communicate medical information to non-medical people.
• Save money on data entry.
 	
Dynamic Consent
• "Consent preferences allow a system to be personalised to individual levels of risk".
• When designing consent options, concerns were raised over:
  ○ Control is lost if there is not enough choice
  ○ Choice paralysis due to a patient being overwhelmed by too many choices.
• Dynamic Consent is an online consent form and patient engagement via a patient forum.
• The online consent form has consent controls:
  ○ Patients can edit information at any time.
  ○ Patients can withdraw data from research use entirely and still log data with RUDY (but this is rarely used).
  ○ Patients can specify who can access to data.
• The patient forum is a self-selecting group of patients who are consulted on development and changes.
  ○ Patients feed back into research design processes.
  ○ Patients teach researchers about engagement.
	Reasons for joining RUDY:
	• Individual has a rare condition.
	• RUDY focuses on rare diseases.
	• RUDY focuses on wellbeing.
	• So that others may benefit.
	
	Experience of the study
	• Just an email.
	• Patients feel their participation is needed but this relies on individual motivation.
	• Frustrated at inability to contribute.
	
	Withdrawal of consent
	• No one would withdraw consent.
	• Potential reasons:
	  ○ Unethical behaviour.
	  ○ Unauthorised data-sharing.
	  ○ Unauthorised commercial venture.
	  ○ The process is of no benefit to patients.
	
	Suggested changes
	• RUDY could feed information back to participants – "check-ins" for example.
	• Patients are unsure of how they can take part.
	• Patients do not feel that tick boxes represent their experiences – a conversation might.

Table 13: A systematic approach to qualitative analysis (from Krueger & Casey, 2000).

1. Did the participant answer the question that was asked?
    a. If yes, go to question 3.
    b. If no, go to question 2.
    c. If don't know, set it aside and review it later.
2. Does the comment answer a different question in the focus group?
    a. If yes, move it to the appropriate question.
    b. If no, go to question 3.
3. Does the comment say something of importance about the topic?
    a. If yes, put it under the appropriate question.
    b. If no, set it aside.
4. Is it something that has been said earlier?
    a. If yes, start grouping like quotes together.
    b. If no, start a separate grouping.

Table 14 shows the four work cycles that I found adequate to create a rigorous thematic framework that could be used to analyse data from both the interviews and focus group. My initial analyses consisted of three divergent cycles to explore emergent ideas. This divergence was important because it meant that the final thematic framework was not limited by a narrow pool of code choices. The final analysis narrowed in focus. Each cycle of thematic development is described in further detail below.

Table 14: Overview of thematic development.



Work cycle	1	2	3	4
	Extracts coded under common themes.	Extracts coded under motivation, assumption, experience, demand and trust.	Revisit research questions to explore ideas.	All interviews coded using categories from framework 3.
Themes	Reasons for study participation	Reasons for study participation	Research contribution	Communication
	Attitudes to healthcare	Attitudes to healthcare	Researcher responsibility	Data-controller
	Attitudes to RUDY	Attitudes to RUDY	Data subject entitlement	Data-subject
	Researcher responsibility (data-use)	Researcher responsibility (data-use)	RUDY participation	Data-use
	RUDY data-use	RUDY data-use	RUDY improvements	RUDY
	RUDY consent revocation	RUDY consent	Consent withdrawal	Security
	RUDY engagement	Revocation	Motivation	Trust
	Experience of the RUDY study	RUDY engagement	Background	
		Experience of the RUDY study	Attitude to data	

Work cycle 1

To start, I grouped responses by interview question. I identified repeating ideas or phrases and collected those extracts under a code. For example, "as long as it helps the kids" was coded under "reasons for study participation". When I finished this cycle, I had eight codes. As shown in Table 15 below, these lent themselves to sub-codes. Under "reasons for study participation" for example, I sorted extracts into medical, research and social reasons for taking part. "Emergent themes" was also included to allow for unexpected topics.

Table 15: Codes used in work cycle 1.

1. Reasons for taking part in the study
   - 1.1. Medical-related reasons
   - 1.2. Research-related reasons
   - 1.3. Social reasons
   - 1.4. [Emergent themes]
2. RUDY's data-use
   - 2.1. Understanding of how information is used
   - 2.2. Patient's own use of their record
   - 2.3. [Emergent themes]
3. Attitudes to healthcare
   - 3.1. Hospitals
   - 3.2. Medical research
   - 3.3. [Emergent themes]
4. RUDY: revocation of consent
   - 4.1. [Emergent themes]
5. Attitudes to RUDY in particular
   - 5.1. Responsibilities
   - 5.2. Communication
   - 5.3. Impact
   - 5.4. [Emergent themes]
6. RUDY: engagement
   - 6.1. [Emergent themes]
7. Views on researcher responsibility regarding data-use
   - 7.1. A researcher's duty
   - 7.2. Organisational responsibility
   - 7.3. Data-sharing
   - 7.4. Participant's role
   - 7.5. Research limitations
   - 7.6. [Emergent themes]
8. Experience of the RUDY study
   - 8.1. What is the study doing?
   - 8.2. Satisfaction with study currently
   - 8.3. Suggestion: improving feedback
   - 8.4. [Emergent themes]

Work cycle 2

I indexed the five interviews using the above codes and found that the resulting groupings were similar to simply grouping the transcripts by interview question. I wanted to find out whether I could find overarching themes rather than follow the structure of the interviews. I grouped extracts of interest based on whether the extract described motivation, assumption, experience, demand, or expectation. These themes required further clarification:

Table 16: Codes used in work cycle 2.

1. Motivation
    1.1. Internal
    1.2. External
2. Personal relevance
3. Others
4. Experience
5. Help
6. Assumption
7. Perception
8. Data-value
9. Behaviour
10. Knowledge deficit
11. Medical
12. Individual
13. Trust
14. Impact
    14.1. Practical application
    14.2. Future impact
15. Belief
16. Research

Work cycle 3

Work cycle 2 did not prove insightful, so I read the interviews again and identified key quotes. I discovered overarching themes such as "research contribution":

Table 17: Codes used in work cycle 3.

1. Research contribution
2. RUDY participation
3. Motivation
4. Researcher responsibility
5. RUDY improvements
6. Background
7. Data-subject entitlement
8. Withdrawal of consent
9. Attitude to data

Work cycle 4

I felt that these codes represented the topics that had emerged during interviews. Interviews were hand-coded and entered into Nvivo. The following codes emerged:

1. Communication
2. Data-controller
3. Data-subject
4. Data-use
5. RUDY
6. Security
7. Trust

The five interviews were re-coded and different themes converged (e.g., extracts coded under "Data Controller" were also relevant to the "Communication" theme). At this point I identified and hand-coded key quotes. Table 18 shows the themes identified, which were then used to thematically analyse all interviews.

Table 18: Themes emerging from qualitative data.

1. Trust
    1.1. How trust is built
    1.2. What trust builds
    1.3. Conflict
    1.4. Trusted individuals
    1.5. Trusted values
    1.6. Other
2. Security
    2.1. Protected information
    2.2. Security by design
    2.3. Harm
    2.4. Lack of individual control
    2.5. Trade-offs in research
    2.6. Role of technology
    2.7. Other
3. Communication
    3.1. Individual empowerment
    3.2. Providing richer data
    3.3. Two-way nature of research
    3.4. A preventative measure
    3.5. Engagement
    3.6. Other
4. Consent
    4.1. Purpose
    4.2. Data ownership
    4.3. Conditions for revocation
    4.4. Implementation details
    4.5. Other
5. RUDY
    5.1. Intrinsic motivation to take part
    5.2. Extrinsic motivation to take part
    5.3. Understanding of RUDY
    5.4. Experience of RUDY
    5.5. Participant feedback
    5.6. Other
6. Data Subject
    6.1. Experience as a data subject
    6.2. Duty to contribute
    6.3. What "data processing" means
    6.4. Attitude to participation
    6.5. Other
7. Data Controller
    7.1. Experience of data controller
    7.2. Duty to protect information
    7.3. Duty to use data responsibly
    7.4. Value-driven processing
    7.5. Institutional expectation
    7.6. Other

Once codes had been developed and refined for use across the dataset, one way to ensure reliability would have been to test this with another coder (Joffe, 2012). Since I did not have access to another independent coder, I organised a data-session. For a

comprehensive overview of the materials provided to colleagues and the resulting feedback, see Appendix 9. The themes that I proposed did not need substantial changes. The next section provides an overview of the final thematic framework, and results from the data collection.

4.2.4 Validation of findings: a return to the literature

Interviews provided me with examples of information that participants wanted to know about RUDY: project aims, what data is used, how that data is used, the value of these data-contributions, and outcomes of data-use. This is reported in further detail in the results section (4.3) below. In summary, reporting this kind of information enhances the feedback provided to participants. I used the enhanced feedback structure to model engagement in an analysis of three existing reports of Dynamic Consent in practice.

While there are indications that my contemporaries think about engagement as part of their work (Haas *et al*, 2021), this is not under the auspices of Dynamic Consent**.** Recent work in partnership with the National Centre for Indigenous Genomics in Australia indicated that a model for engagement and a Dynamic Consent platform were developed separately (Elsum *et al*, 2019). This is, perhaps, an indication of the future of this research field. 'Dynamic Consent' will describe technical data-controls and engagement is acknowledged, prioritised and developed separately throughout the project.

This trend is impacting the work citing Dynamic Consent; implementation studies are divorcing engagement from Dynamic Consent despite its position as one of the two main requirements. Three recent papers (Dankar *et al*, 2020; Mamo *et al*, 2020; Norval and Henderson, 2019) laying claim to Dynamic Consent have demonstrated that a participant's ability to control their data (the revocation arm of Dynamic Consent) has overridden engagement as a design priority. This veers from Dynamic Consent's original purpose, and the critical analysis below is meant in a constructive manner rather than to detract from the excellent academic work in this area.

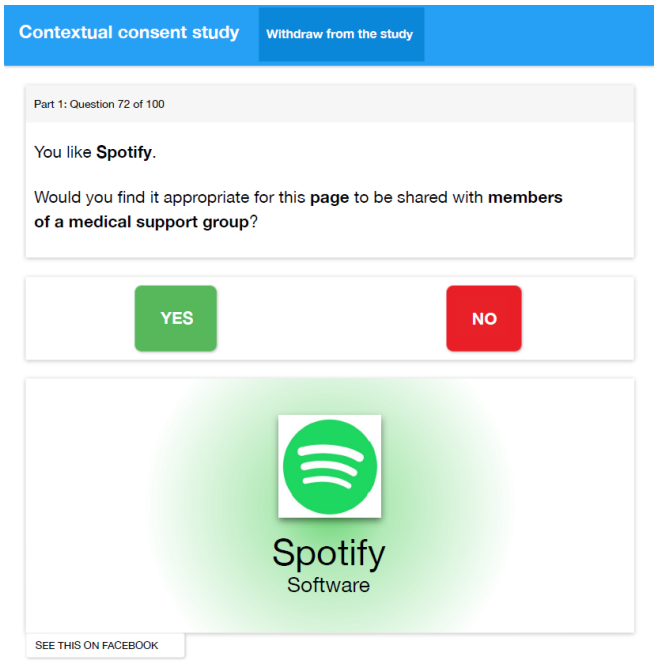
Automating consent choices (Norval & Henderson, 2019)

Revocation	Project aims	Data used	Data-uses	Contribution value	Outcomes
☑	☒	☒	☒	☒	☒

The authors cited Dynamic Consent as a potential solution to automating consent decisions for online users. Participants were recruited from Facebook patient groups, shown items of social media information, and asked whether they would consent to share this in a medical context. The answers to these questions were used to train an algorithm that would predict whether a user would give this consent for other data-items (termed "appropriate data-flows"). Recruiting from patient groups proved difficult, as the group

administrators (or "gatekeepers") were reluctant to allow access to their communities. The authors even received negative feedback from some gatekeepers, who emphasised their duty as protectors and cited privacy concerns. This issue alone suggests that early on, the authors had failed to consider how to engage with participants.

Figure 17: Example notification.



Once the data had been collected, algorithm trained and data-preferences automatically set, the authors reported that they wanted to lower false positives. In this case, a false positive consisted of an algorithmic assignation of "would share" whereas in reality the individual would not. When running their models, the authors chose to minimise leaks, resulting in a small number of automated choices to share data being incorrect. Such false positives are entirely unacceptable here and should be eliminated, which the authors try to address by suggesting that any algorithmic uncertainty should pass choices to the user for approval. This solution is neither elegant nor intelligent, overcomplicates the consent process unnecessarily, and fails to address the fundamental issues of consent fatigue and information overload:

> "Data Leaks can be eliminated at the cost of burdening the participant. If false positives are seen as entirely unacceptable, the participant could simply be consulted every time a 'Share' outcome is predicted… More conservative models would result in lower burden, but also lower sensitivity… In the worst case (i.e., all data is predicted as 'Share'), this approach would match the burden associated with non-automated

> Dynamic Consent (the participant is asked whether each item should be shared)."

Norval and Henderson argue that privacy-aware technologies need not perform worse than their counterparts, but they do not develop this line of argument. Rather than make a case for privacy-awareness by default (or indeed, 'privacy-by-design') and place responsibility for this on technology developers, the authors leave participants to indicate a preference for secure data-use. This work provides a promising use-case for automating Dynamic Consent, in the automation of Dynamic Consent within a research data-bank that uses social media data. This is a contrived example of course, but the authors discover that participants have privacy preferences that extend to publicly-available data, such as information that can be derived from social media. A third of participants reported that they thought that public data should not be 'fair game' for researchers, and 31% failed to share data with researchers as they thought it was uninteresting. If anything, this makes a case for engagement as part of research practice – communicating the type of data being collected, how that information will be used, and emphasising the importance of this contribution are all criteria for enhancing feedback.

The authors suggest that Dynamic Consent should be active and ongoing, seeking confirmation from participants throughout a study, but their work is not longitudinal. Data is taken at one time-point, and there is no further discussion on this point, which is important, because in doing so they ignore a basic tenet of Dynamic Consent as addressing data collection over time. As a final point, while their resolution that participants should be involved in "discussion prior to any predictions or automation taking place" is commendable, the authors fail to address how they might include this as part of their own study.

New Dynamic Consent requirements for genomics research (Dankar *et al*, 2020)

Revocation	Project aims	Data used	Data-uses	Contribution value	Outcomes
☑	☒	☒	☒	☒	☒

This author describes Dynamic Consent as a personalised consent and communication platform, and this paper offers an excellent resource as a Dynamic Consent literature review. Dankar describes projects claiming to have implemented Dynamic Consent in terms of three pillars of informed consent, developing a checklist of requirements applying these pillars in terms of Dynamic Consent (termed "dynamic-informed consent", see Figure 18). Unfortunately, these requirements are difficult to understand, and as this is a theoretical paper it draws from published work rather than conversations with the

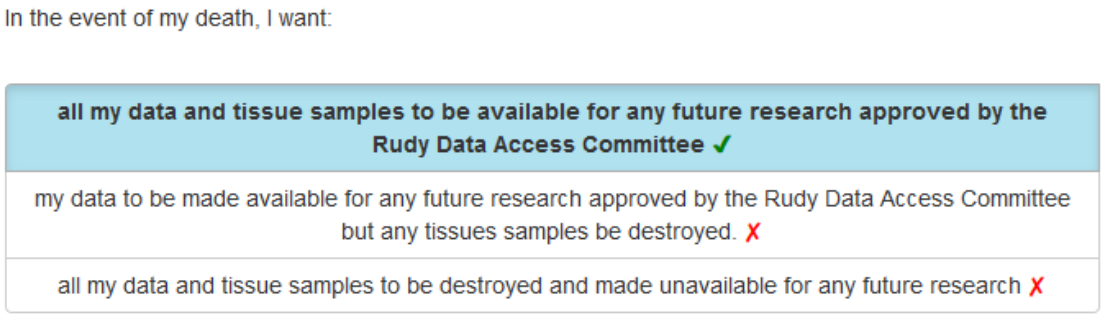
project teams. RUDY is described in this work, but the description is inaccurate. This suggests that the descriptions of other projects may also be inaccurate.

Figure 18: RUDY v. dynamic-informed consent requirements (Dankar *et al.*, 2020).

		RUDY
Dynamic Permissions	Online Portal	✓
	Per-Study Consent	✓
	Interpreted Results	×
Dynamic Education	Dynamic Education	×
	Dynamic Assessment	×
	Timely Research Information	×
	Up-to-Date Research Progress	✓
Dynamic Preferences	Select Consent Level	×
	Tailored Information	✓
	Choose to Receive Individual Results	×
	Select Specific Individual Results	×
	Share Individual Results	×
	Ability to Withdraw	✓

Interestingly, the author asks "What happens in Dynamic Consent after death?" which is an issue that RUDY has already addressed. Rather than researchers pre-empting the issue, members of the patient forum raised concerns about post-mortem data-use. To accommodate these concerns, the research team added options to the consent process (see Figure 19 below).

Figure 19: RUDY options for post-mortem data-use.



The requirements Dankar outlines in this paper for dynamic-informed consent do not refer to engagement. The author's recommendation for RUDY however, highlights a need to improve communication, a need corroborated by Chapters 4 and 5 of this thesis and empirically validated in Chapter 6. Dankar further outlines a requirement for dynamic-informed consent to "produc[e] participants that are ready to make autonomous and informed choices". This suggests that potential research participants are not ready to make such choices outside of a research context, which somewhat infantilises members of the public. This also implies that there is a need to prime people, without providing any suggestions as to how researchers could measure participant readiness to make such

choices. Designing engagement into research practice offers one way for researchers to plan how and when to check a participant's understanding. A forum, for example, provides an opportunity to simply ask participants questions.

Dankar does not discuss the opportunity that Dynamic Consent provides to continuously inform participants and support their decision-making process – they make their claim and leave it unsupported. Similarly, they claim to identify a need to examine "moral issues arising from… the open timelines of DC", briefly describing potential privacy issues surround the use of Dynamic Consent in blockchain. They fail to describe this examination in any detail, despite the opportunity that RI provides in this area. Finally, Dankar describes how participant competence reduces over time (with age, they infer) which, in combination with increasingly complex research processes, erodes informed consent. My first criticism of this point is that it is ageist – many technologies are simply not designed with the needs of older users in mind (Knowles & Hanson, 2018) and this presents a barrier to adoption. My other criticisms of this are that the argument is simplistic, and the claim entirely unsupported. If Dankar had considered engagement, they could have used it to mitigate such risks to valid consent. All in all, while an admirable goal, "building an integrative dynamic-informed consent process [that] connects people, solves all stakeholder problems and lets participants control data-flow" is an oversimplified and ultimately baseless claim.

Blockchain application (Mamo *et al*, 2020)

Revocation	Project aims	Data used	Data-uses	Contribution value	Outcomes
☑	☑	☒	☒	☒	☑

Mamo *et al* claim that Dynamic Consent increases trustworthiness, attributing this to Kaye *et al*, with no empirical basis. The authors apply Dynamic Consent to a blockchain implementation, Dwarna, designed to inform research partners and allow them to identify how to involve themselves in genomic research.

The Malta biobank website contains an online portal, hosts governance documents and videos, and contains a web form to allow expressions of interest in taking part. These expressions lead to an on-site visit, followed by participants being assigned a pseudonym and login credentials.

Figure 20: The Malta biobank's website.

Overwhelmingly positive work, these authors claim that data minimisation is important and want to demonstrate that data-protection compliance does not need to impede implementation. Despite their aim being to enhance trustworthiness and transparency, the authors focus on total control over data. Their failure to consider how to build engagement into their research practices means that they lose sight of Dynamic Consent's original purpose, to keep biobank participants informed over time. On further investigation, the biobank's website is difficult to navigate and while videos are available they are not immediately accessible.

4.3 Results

This study collected qualitative data describing participant and researcher experiences of an implementation of Dynamic Consent. The theoretical work grounding Dynamic Consent focuses on an individual's ability to revoke consent once given, and engagement with participants to inform them about a study over time. An analysis of the data collected, in combination with a brief return to the academic literature, suggests that the discourse surrounding engagement is almost entirely lacking; there is considerable work ahead for Dynamic Consent to be implemented in its entirety.

A working definition of 'engagement' was created over the course of the data analysis carried out supporting this study:

Engagement = Interest + Knowledge transfer + Active participation

To better describe the concept of 'knowledge transfer' I identified specific types of information that participants expressed an interest in receiving. These were grouped under the term 'enhanced feedback', an idea which is further validated in Chapter 5 before being empirically tested in Chapter 6:

Enhanced feedback = Project aims + Type of data used + How data is used + Value of data-contribution + Research outcomes

The following section describes themes arising from all qualitative data collects; a deeper exploration of data collected from the researcher focus group framed in terms of how responsible data-practice can drive research; a similar exploration of participant interview data, which focuses on the expectations people had of themselves as well as research and medical institutions; finally the validation steps outlined previously are summarised and a gap in Dynamic Consent literature is identified. Quite simply, of Dynamic Consent's core components (revocation and engagement), reporting often records how revocation is managed but fails to describe engagement in any way.

4.3.1 Themes

Table 19 provides an overview of the themes and subthemes that emerged from the qualitative data. These themes are described in further detail below.

Table 19: Final thematic framework.

1. Communication
   - 1.1. Engagement
   - 1.2. Two-way
2. Consent
   - 2.1. Revocation conditions
   - 2.2. Data-ownership
   - 2.3. Implementation
   - 2.4. Purpose
3. Data-controller
   - 3.1. Duty: responsible use
   - 3.2. Duty to protect
   - 3.3. Experience of…
   - 3.4. Institutional expectation
   - 3.5. Value-driven processing
4. Data-subject
   - 4.1. Attitude to participation
   - 4.2. Duty to contribute
   - 4.3. Experience as…
   - 4.4. Me v. Other
   - 4.5. Definition of processing
5. RUDY
   - 5.1. Experience
   - 5.2. Feedback
   - 5.3. Motivation
   - 5.4. Understanding
6. Security
   - 6.1. Control
   - 6.2. Data breaches
   - 6.3. Harm
   - 6.4. Protected information
   - 6.5. Role of technology
7. Trust
   - 7.1. Conflict
   - 7.2. Building trust
   - 7.3. Trusted entity
   - 7.4. Trusted values

4.3.1.1 Communication

Two-way research: the onus falls on both researchers and participants ensure that research is accurate and makes a positive contribution towards society. Researchers are responsible for designing protocols that offer shared decision-making and seek input from participants (this includes advocacy groups).

Engagement: analysis of data provided a working definition of engagement. Engagement was reported as a combination of interest, knowledge and level of active participation. Isolation was a key motivation for individuals to engage with research. A lack of peer[15] and specialist[16] support meant that individuals were actively seeking out relevant information. Newspapers and other media outlets were the primary and most easily understood source of information.

4.3.1.2 Consent

Purpose: consent showed that a participant had agreed that their data would be used for particular reasons and established "rules" for researchers to follow. For example, researchers are "not allowed to give it [data] out", and participants must be "telling the truth".

Data ownership: participants unequivocally agreed that they owned their personal data, and that ultimate control remained with them. Participants wanted to be asked their permission before data was shared.

Revocation conditions: no participant had withdrawn or changed their consent as part of RUDY and the overwhelming response was that they would not. There was evidence of conditions under which participants might revoke data, but they were not immediately offered and required some reflection. When prompted, participants said they would choose to revoke data-access for two main reasons: loss of confidence and a moral breach. Faltering confidence, presented as a lack of research contribution or "I feel the whole process is of no benefit", was seen to be a waste of the participant's time and effort, which are already limited resources. A moral breach was described as unethical behaviour on the part of the institution, such as personal data being sold to third parties.

Implementation: participants showed no desire to learn about the reasons for processing data, but there were opinions about how consent choices should be offered, and how their permission should be sought. Participants wanted to control how they were communicated with, as well as what about them could be communicated to others.

---

[15] Other people in similar situations.

[16] Research studies and other information about the participant's condition.

Any policies and procedures that exist around the consent process (e.g., "if you withdraw consent, you are still entitled to…") should be communicated at the first point of consent, and revisited throughout participation.

4.3.1.3 Data-controller

Duty to use data responsibly: participants agreed that controllers had an obligation to use personal data in a way that would contribute to a greater good. There should be no unauthorised sharing and if data was to be released, "the person should be told… and whose hands it's going into" despite whether or not they had originally consented to data-sharing. Responsibilities to follow ("data protection policies", for example) and to lead ("use it in the best way you see fit") fall to data controllers. Perceived as a figure of authority they must maintain oversight of policy and procedures, prioritise the anonymity of subjects and then update them as and when required.

Duty to protect: participants also agreed on the role of controllers as protectors of data, with principles of safety (being emotionally and physically protected from harm) and security (how the state of "safe" happens) often intertwined, as demonstrated by "not patient safety… patient confidentiality *and* safety". Confidentiality is a term traditionally used in information security literature for hiding or protecting data. Information security (confidentiality and anonymity of data) is important to participants. The "consideration and protection" of data includes putting security measures in places such as access control and updating keeping technology up to date. Participants believed that the data controller should make them feel safe.

Experience of data controllers: there were two significant negative experiences of data controllers in research, and no positive experiences. Participants who were themselves responsible for being data controllers (usually at work) had an in-depth understanding of standards such as GDPR and what was required in practice. Their data controller experiences were with the NHS rather than in research. Incorrect data being recorded on medical records is a significant problem as this has a direct impact on healthcare, "I made up something and they put it in". Lack of information-sharing across the NHS meant that many participants carried their medical records with them when they went to hospital, to consultations, and while travelling abroad. "It really is very frustrating".

Institutional expectation: participants had particular ideas about what they expected from medical ("I don't think hospitals are very good at security are they?") and research ("it's all safe") institutions. There were also indications of general expectations for data controllers. Medical data-controllers focus on provision of care and must work with vulnerable populations but do not check in regularly, leaving patients to "fall off the grid". Information is not shared. Research data-controllers focus on progress and must "push

the envelope". This is regulated in the form of ethical review boards. General expectations of data controllers focus on expertise and transparency. Processing purposes, data lifespan details and data protection policies must be communicated (how this information should be communicated is not mentioned, and is worth exploring[17]). In the case of a data-breach, details of the incident and further actions to prevent a similar incident should be communicated.

Value–driven processing: participants suggested that there were overarching values driving data-processing. Whether animal testing is bad, for example, can be argued based on the end goal of that research, "if there's intensive animal testing then [it's unethical] … though I know that the drugs that I have been given have undergone rigorous animal and human testing, so… [it is a means to an end]". A code of conduct, marked in the case of research by the used of an ethics committee to approve work. This can mark commitment to values such as respect for the participant, ethical data-use and privacy.

4.3.1.4 Data-subject

Experience as a data subject: striking commonalities suggest that most participants experience communication breakdown, misinformation and a lack of agency in a system responsible for providing care. Communication breakdown in health services was a commonly reported experience. Some participants reported that a lack of data-sharing between their consultants put pressure on them to centralise their own records "just in case things have gone wrong". There were also examples, however, of cases where the participant and their consultant worked together to ensure data was communicated to the appropriate experts. Mistakes made on medical records ("they wrote that he had Dementia on his record even though he doesn't have it") meant that future care decisions would be based on incorrect information. Participants reported experiencing a lack of agency – despite general agreement that "it is vital that [health] information is shared" people reported experiences of delayed diagnoses ("my GP said… 'you can't have that") and a lack of centralised data ("it [condition-related detail] doesn't go in my case history"). This meant that participants often had to judge what types of information were relevant and compile them.

Duty to contribute: participants often felt it was important for them to contribute and to encourage others to do the same ("I've got a duty to do") as providing correct information would mean useful research findings, more relevant to participants.

[17] This is the basis for Chapter 5.

Processing definition: participants were clear that data they provide "should not be used outside of research purposes" but within this broad purpose there was significant flexibility, such as data access for "anybody that might be looking into these things" because "you don't know which road you're going down". Security and anonymity were not synonymous, but it was expected that information to be processed would be kept secure.

Attitude to participation: RUDY participation held different meanings to different participants. Raising the profile of their condition was a common objective, with participants giving their data and time for no immediate return. Some participants explicitly stated that, as their participation was voluntary, it should not be leveraged from profit. Similarly, if data from another study or research activity was shared and re-used. Active participation was an example of how participants took initiative – "people should make a fuss" and "I'm quite capable of contacting them [researchers]". Participants also recognised that their data-contribution held value. They thought that the information they provided held value, believing it should be put to use ("it would be pretty silly not to take part"). Some participants identified a participant responsibility to report correct data – to "be honest and tell the truth".

Me v. other: participants identified a need to protect data, but generally had lower data security requirements for themselves than unnamed "other" people. Some participants reported that data-sharing should be restricted by default, allowing for individual risk tolerances ("I don't have an issue with sharing it however some people might have problems"). Participants expressed a desire for information, to keep on top of "the situation", whether that situation is talking with a consultant ("for many the doctor is always right"), or being informed about a data breach ("I don't know if it's the same for everybody"). Participants seemed confident in their ability to gather information but indicated that there should be more provision for others.

4.3.1.5 RUDY

Motivation: there were many reasons participants decided to take part in the RUDY study. Those who did not have a rare condition were caregivers to those who did and across the board, participants wanted to contribute to the literature on their condition. "I just think you can help somebody": other people were cited as a key motivation to take part in the study, from unknown strangers to family members ("my brother's got it, my sister's got it, my nephew's got it, my nephew's baby's got it, my son's got it so anything that can help them"). The long-term benefit of research contribution was important despite participants being "past help" themselves. RUDY focuses on wellbeing differentiated it from other studies on genetic conditions looking at "just symptoms and

outcomes". Lack of awareness and available information on the specific condition was also a significant issue.

Understanding: what became clear was that participants were not certain about RUDY's ultimate research objectives, neither did they feel that they needed to know. "I don't have a clue" was by far the most common response. There were ideals participants hoped that RUDY was working towards such as feeding back into clinical practice and raising the profile of their condition, but when asked they did not know what the study was actually doing. RUDY's position as an interdisciplinary project consistently emerged – that researchers, scientists and medical staff were working together or using participant's data to work towards a common, greater good. Other assumptions were that whatever RUDY is doing, data is safe with them, "they probably pick their participants and stick with them", and the project supports others that work along similar veins.

Experience: participants were all familiar with the surveys sent out by the RUDY study as they completed those regularly, but reports on overall experience differed significantly. "It's just an email isn't it?" represents a common response. Other than filling out the survey, the participant had no reason to interact with the project. This presented as both a positive (in that the participant did not feel "pestered") and a negative ("no one has ever come back to me"). Participants reported that the patient forum made them feel their time was not being wasted, and "quite involved". The symptom log (a survey on symptoms and wellbeing sent out every few months) is cited as largely positive ("I… monitor myself more closely… maybe not such a good thing because of obsession…!") but some participants said they felt that they could not communicate their pain via checkboxes or mark context ("if I had a bad day"). There was a perceived lack of consequence to participation, in that few participants had been recommended for other studies or taken part in physical tests. The study was seen as reflective rather than proactive.

Feedback: participants began to suggest information they would like to receive from the RUDY project. An annual check-in was repeatedly suggested. The suggestions were largely to have it in person but the underlying goal would be to give participants an idea of where they fit in the research population and what they (and others like them) have been contributing towards such as results and "what it [research results] mean for the future". Clear language is needed – those who had read, or tried to engage with technical research outputs could not understand the language used ("a brief update"). Participants want to be involved but are not sure how to engage with the subject matter. This may not be in the researcher's remit, but RUDY has connections with patient groups who are more likely to be able to provide that kind of information.

4.3.1.6 Security

Control: individuals should be at the centre of decision-making as far as use of their personal data is concerned and for this to happen they may need information from the data controller and they may need to check-up on what is happening with the project. Participants want to control who accesses their data ("I would like to maintain a degree of control"), with many having their own controls in place like checking that sites were secure online and limiting information disclosure on social media. A perceived lack of control was evident across participants. In the face of an overwhelming amount of data and "clever people out there" … "I would probably trust the good fortune".

Data breach understanding: participants identified data breaches to be leaks of information. They described malicious and non-malicious causes, the impact of such breaches and their own reaction to such an event happening. Unauthorised access was the perceived cause of data breaches, either "a hacker getting into the system" or a mistake because "no one does it on purpose". Breached are inevitable: while data breaches were described as illegal and a threat ("potentially critical information… could be used by other people"), participants also disclosed that they felt these events were "a fact of life" and were powerless to stop them. Reaction 1: "happens from time to time". This neutral response stems from data being given to researchers. "You're not giving this to Joe Public" so the personal data has been entrusted. Reaction 2: "contravenes the Data Protection Act". This negative reaction comes from "they've [the controller] broken that trust", despite the fact that it is "probably through no fault of their own". Participants did not raise attacks, hackers or advanced persistent threats. Data breaches were due to incompetence.

Harm: participants shared common concepts of harm that might be caused by data misuse, this discussion was largely theoretical because most experiences of "harm" were simply described as annoyances (e.g., spam emails or credit card fraud). The use of anonymised information was not problematic, but human-linked information needed protecting ("when it's linked to your name is the problem or you know people would be aware of it"). Participants had difficulty conceptualising harm or how medical records could be used in a harmful way ("I can't see what use my medical data can be to anybody"). Participants identified potential harms as: workplaces finding out about their condition and treating them differently, being embarrassed, and receiving spam calls/emails. The most common concern was actions being done under the participant's name without their knowledge or authorisation.

Protected information: banking information was cited as the most important information to be kept safe by most participants. In cases where they were compared,

the value of medical data was medical data always lower than financial details. Individuals had differing views on how they prioritised contact details and other personal information. Medical information seemed to have a sliding scale of confidentiality. Pregnancy history or AIDS diagnoses, things that "affect your day to day" were specific examples of information that needed protecting whereas a medical history was not as there was "nothing to hide". Individuals had different stances on whether their personal information was something to protect. In general, identity related data such as address, date of birth and "anything to do with me".

Role of technology: across participants, technology use varies. Most participants could identify where technology such as storage systems, data-sharing and online interfaces could make their lives easier but agreed that this was not currently the case. Participants with a positive view of technology assumed research data was shared securely, so no extra controls would be necessary. Online interfaces were associated with secure transactions like banking or a "secure research agency". Many participants were inhibited by technology. Online interfaces put off people unfamiliar with their use, Wi-Fi signal and connection times holding up hospital care, and information loss or incomplete records on file. Technology is an enabler – participants identified that data should be shared for the purpose of delivering care and underlying technologies could streamline how data was used.

4.3.1.7 Trust

Conflict: if trust is a belief ("one has to put one's faith somewhere") then deciding to entrust data and take part in research is a choice that potential participants need to make. Trade-offs like those participants felt they had to make between unethical research and research results, and insecurity and research value, suggest that participants are sacrificing individual goods (like personal ethics and individual data security) for an undefined research benefit. Participants feel that they must trust research, even if they are trusting "with caution". This judgement call on their part is rarely reciprocated and never justified by the party they are trusting – trustworthiness is not shown, yet this trust judgement is not expected to change accordingly.

How trust is built: participant's trust in the RUDY study was influenced by a number of factors. Data being shared without consent was perceived to be actively "going against that consent… a breach of trust". Positive experiences of the National Health Service, both working for and being treated by, encouraged participation. Expert recommendation from a consultant, the requirement for institutional ethical approval ("ethics checking") and associations with patient groups. Some participants were also prompted by a lack of any negative reports, with "no reason, so far in my life, ever not to trust".

Trusted entity: participants showed levels of trust in different people or groups. The three recurring motifs were the RUDY study and the participant's consultant. While RUDY was generally a force for good, reports differed on patient-doctor relationships. Some participants reported that if they did not trust the study they would not be taking part. Reports were that this trust was built largely through associations (with a university and various patient groups) and on the recommendation of doctors and patient group members. Trusted doctors were the consultants who helped participants coordinate with other doctors, answered questions and acted in the participant's interest (taking a participant-centric approach). Untrusted doctors were not untrusted due to initial misdiagnoses. Negative experiences such as the participant feeling that they were not listened to; trying to communicate "was a waste of time", and persistent misdiagnosis in the form of treating symptoms rather than the participant as a whole, significantly impacted participant attitudes to their consultant.

Trusted research values: participants allow a study to use their information for some "valid reason" which they might have some idea of, but largely did not. There were values that they hoped would shape the research they were participating in: honesty, compassion, and that data-use is driven by confidentiality, privacy ("share my data securely and appropriately") and the law. Finally, developing a "healthier, better quality of life" for people with rare conditions, and although individuals did not expect direct benefits they expected participation "not to worsen my condition".

4.3.2 Focus group: responsible data-practices drive development

RUDY was founded on strong design principles – the team described their role as collectors and curators of personal data, with data-ownership lying solely with the participant. The quote below demonstrates the importance of data minimisation:

> "PROJECTLEAD: We ask for them to send, we contact their lead clinician for a confirmation of the letter and we ask them to send us back a photo of their letter as well… so that way we can support that.
>
> INTERVIEWER: Okay so let's take this strand of being a data controller, you are in control of data. What do you understand by the term 'data minimisation'? What does it mean to you?
>
> PROJECTLEAD: It means you collect only the data you need to do your research, to answer your question."

The study is also patient-facing, meaning that data is collected from patients. The research team said that this was a novel position for an epidemiological study, most studies collect data from clinicians. As seen here, the project and development leads shared an understanding of consent, describing it as a mechanism to facilitate individual decisions around data-risk:

> "PROJECTLEAD: Uh I suppose they are important here because if you don't have them you assume everyone has the same level of morality. Risk sharing. Your consent preferences allow a system to be personalised to the person's level of risk sharing and that can be between people or the same person over time.
>
> DEVLEAD: it's also it's explicit you know, you're, there's very little grey area in terms of those consent options in terms of whether you can read or misread it. I'm thinking compared to iTunes you know three-thousand-line document that you have to say yes to where you don't know what you've agreed to… whereas I think because we've got essentially a tick box on every line essentially…"

In creating a "Facebook for rare disease" [DL] the project builds rapport with their participants to collects longitudinal data about rare genetic conditions. When designing the RUDY study, the team needed to communicate medical information to non-medical people and were constrained by their resources which made that online data-entry appropriate. Three key design principles emerged: to be of interest to patients, to be relevant, and to report patients' lived experiences. Designing an attractive, intuitive

interface was prioritised at the initial project design stage to encourage use of the platform. Below is a description of the importance of having relationships with patients over time:

> "INTERVIEWER: Why is a long-term relationship important?
>
> DEVLEAD: Because ongoing longitudinal data's the whole point of RUDY really, so collecting stuff over time where we need to maintain a good relationship or you know a good experience for those participants where they feel both the work they are putting in is benefitting or going towards something, going to be useful, but also that it's not too onerous or too much of a burden. I think generally with rare diseases they like the idea they are being asked questions at all you know because very little research has been done on them in the past so having something like RUDY where they feel like they're getting asked relevant questions or 'there is space for me to put down what I consider important factors', I think that's valuable…
>
> PROJECTLEAD: They feel listened to.
>
> DEVLEAD: Yeah
>
> PROJECTLEAD: It's about the lived experience and they get feedback on what they've done. We have it so that everything they enter they can see so they can track their healthcare over time and that may make people more involved with research."

As a longitudinal study, simply having participants on record is important because RUDY is a study in rare genetic diseases. This means that if participants do not want to share their records with (vetted) third parties, they can be enrolled without actively participating (filling in surveys). From the focus group arose a concern around "consent fatigue" in the form of choice paralysis or the "danger of overburdening them with too many options" [DL]. In the focus group, participant choice was equated with participant control and this was an overriding priority so to enable participant control was to enable choice and empower participants. The team tried to design consent choices with patient requirements in mind, such as low energy thresholds. Consent fatigue was a concern which the team tried to mitigate in two ways: grouping consent options, and trying not to overburden users with information they were required to understand. These points are noted in the discussion below:

> "INTERVIEWER: Okay, great. I am aware of the time but we have two more questions and then that's it. What changes would you make to the consent process? What about consent fatigue?

DEVLEAD: Yep. Well I've thought about that before, you know I suppose it depends on the individual patients and their kind of level of interest in their own data and how there is a danger in all of this stuff in overburdening them with choice paralysis – you're giving them too many choices, too many options. You've got to marry that with-

PROJECTLEAD: You've got to group them up.

DEVLEAD: If you don't give them enough choices then they don't have that control.

PROJECTLEAD: Give them a choice about the number of choices… [Laughter]"

Researchers had expectations of Dynamic Consent as a research tool. In this project, the team described "Dynamic Consent" as an online consent form, and participant engagement. These were implemented via consent controls accessible through an online portal, and a patient forum and website. With the controls, participants could log on and edit their information at any time. They were able to withdraw their data from research use while still logging information (they might use the portal to track their condition rather than take part in research), and could specify who was able to access their record. The patient forum was a group of RUDY participants who were consulted about the study. They provide requirement that feed into research design – one example of this was that the consent form did not originally have an option for what happened to data in the event that a participant died. This was a valid concern for participants, they communicated this to the research team and the consent form has since been amended. Researchers also described that the forum had educated them about how to engage with participants.

4.3.3 Interviews: personal and institutional expectations

Many participants believed that data-use should be driven by principles of confidentiality, privacy and compliance (with the law), as demonstrated below:

> "I'm confident in the NHS, I think it's one of the best institutions we have created and I would have – nothing is going to ever be totally secure but I would put my trust in them to share my data securely and appropriately", P13.

This indicates that participants are aware that protecting information is an institutional concern. Focusing on medical research as an institution, interviewees described the overarching goal of research as improving the quality of life for people with rare conditions, as demonstrated in the following account:

"Do you trust medical research in general? Yes.

With what? The good effect that it will have on people in general – by good I mean they'll be healthier, better quality of life.

Was participating the right decision? It will eventually, hopefully lead to understanding of the management of certain conditions. Or strange, rare conditions. Each condition is different but there are certain things that you can find over multiple conditions or questions you can apply to multiple conditions", P10.

This participant did not know how researchers achieved a "good effect… on people in general", but believed that this was a key goal. This account was representative of a wider belief that participating in RUDY would lead to better care and insight into rare conditions. This belief in scientific progress was not based on personal experience, as few participants reported any feedback from RUDY at all. In general, participants did not expect direct positive benefits from research they took part in. Some individuals, as this participant describes, reported that while they did not expect an immediate benefit, they also expected not to be harmed over the course of their participation:

"[I trust research] Not to kill me. Not to worsen my condition. If it improves, it improves and if it doesn't then I'm no worse off but I don't want to come out of it with my condition worse than it was", P11.

P11 also demonstrated a general expectation to be kept safe from all harm, physical and digital. Responsible data-use was described as important by many participants. In the quote below, the participant describes responsible data-use as an exercise in safeguarding data:

"[Researchers have a responsibility] To protect everybody's data and only release if a person's data is going to be released the person should be told what is going to be released and whose hands it's going into. Because well, you can't trust anyone these days? You know like you said, you're not part of the RUDY study but I don't know you could be a criminal at night. I don't know I can only presume that you're genuine", P44.

Researchers must share data in a responsible way, with people who can be trusted to use it in a suitable manner. This participant expressed a desire to know how their data was used over time, and who that information was shared with. This, and other accounts, support a need for data-practices that account for this and include designing data-

flows[18] early on in research development. Elsewhere in the data, responsible data-use did not rely on researchers knowing every implementation step ahead of time. Rather, tools like data-protection policies offer institutional oversight and research itself is directed by professionals with experience who make the "right" choices:

> "Well yes I'm just a patient, I mean I don't know where you're trying to go, I don't think it's going to find a cure… but it might. Doing something else, because for example I mean they found when I was first diagnosed, they pumped me full of anti-cancer drugs because they found somehow it helped. So you know if my reaction to talk to whoever health… that way then go with it, you set the ball running don't you, with investigations and developments you don't know which road you're going to go down, it's 'yes here's my information, use it in the best way you see fit and if it goes in a way that you haven't foreseen or could not have foreseen but you think might be useful then do it as well", P20.

Participants agreed that researchers have a duty to protect participant data; compassionate researchers demonstrate their care by showing respect for personal data and respecting participant preferences. Despite this, participants did not report any experience of such compassion or demonstration of respect despite describing them as essential requirements. Participants assumed that researchers played an active role in keeping data safe, as illustrated by the following quotes:

> "I think you do have a responsibility. I think you have the responsibility to exercise due care. [Please tell me what you mean by "due care"?] Due care can have many different meanings, but the consideration and protection of that data. And utilising it for a general positive advance… of whatever. New drugs, new treatments… I think you've got to have reasonable protections in place to ensure that not just anybody can access that data, and by anybody I mean they must be a certain level of authority. Whoever is the lead clinician of the research program and it spills down from there to whatever level is appropriate. I'm assuming automatically. People such as yourself would be required to sign some kind of contract and commitment to privacy", P11.
>
> "Well my personal information – who I give it to. If I'm allowing people to have that information, then they've got the rights. [Who do you think owns

[18] Data-flows: identify the information to be communicated, who it will be sent to, and how it will get there. For more information, see Nissenbaum's thinking on Contextual Integrity in section 2.2.2.4.

> your information at the moment?] Myself at the moment and someone I divulge it to – which means I've allowed you access to it. [Just to clarify, your info is yours and you divulge it. What does that do?] I'm giving you the authority to use what I give you. [So what comes with that?] That you keep it safe, what I give to you", P14.

The first participant discusses access control. Their idea of a "reasonable" protection is one where different levels of access are given to different levels of authority. They do not talk about hierarchy of an institution, rather the hierarchy within a project. The lead clinician of a program dictates who can access data. Research is a sector where not only are such controls commonplace, they are mandated. RUDY has a data-access committee that must approve potential collaborators. This ethical approval is, perhaps, similar to P11's notion of a "contract and commitment to privacy". The second participant crystallises this discussion when they say "keep it safe, what I give you". This simple statement reverberated across all 54 interviews and outlines a key principle that can be implemented differently from one study to another; data must be protected.

There were two reasons that participants decided to take part in RUDY. Those who did not have a rare condition were caregivers to those who did. Every participant wanted to help build the research literature on rare conditions. The long-term benefits of research to the rare-disease community were described as important, despite participants being "past help" themselves. There was no evidence that participants wanted to revoke consent once they had given it; none of the participants interviewed had revoked their data or withdrawn access for research use. There was overwhelming evidence that participants believed that they had the right to change their mind, as one participant argues below:

> "Well because if people don't give consent – at the end of the day if you're going to join a research study there's usually a reason. Usually a high percentage have a rare disease themselves or know someone who does. So the first thing when you come across this kind of thing is: it's all about consent because you're taking part in this research because you know you're not going to see a medical professional or get personal, or it's not a fast track way to see a surgeon or you know, yes you can learn by it in the research you participate in but you know that… it defeats the object if you're not going to give consent but if you're not then how are these researchers meant to research and progress if you're not going to give your consent? So it's a Catch-22. So basically what these research should say is that if you are not prepared to share… or do a separate consent form when you first sign up to say 'what data are you not prepared to share?

> What data do you not want revealed to third parties, e.g., phone number email…' therefore it would only be RUDY that would access that information. [Should you be able to change that?] Of course, freedom – it's a person's data. It's like anything. If you give your consent to something you have 24 hours and the right to change your mind!", P44.

In saying that "it defeats the object if you're not going to give consent" P44 outlines the social contract between the society and science and emphasises the importance of free and non-coerced participation. They describe, in no uncertain terms, that data can be shared with different parties and that there are different kinds of information to be guarded. Participant data-sharing preferences, P44 says, can be established during the initial informed consent process of a study and should be accessible throughout.

There were two conditions under which participants thought it would be acceptable to withdraw permission for a research project to use personal data. The first condition was a breach of morality - described as unauthorised use, or the sharing of personal data with an unapproved external party. The second condition related to participant perceptions of the value of research:

> "[Participant would revoke if] I feel the whole process is of no benefit", P63.

All participants interviewed reported that they were unlikely to ever revoke consent, as they believed that the benefits of research outweighed personal risk. One participant used a phrase often associated with privacy and information control:

> "I have nothing to hide", P21.

This suggests that while participants do not expect research to benefit them personally, taking part is a reflection of their character. There was also evidence that participants were making security trade-offs. The quotes below suggest that participants had different ideas about the level of data-protection they could expect from medical and research institutions, and were judging these risks as part of their routine participation:

> "[What do the words 'data breach' mean to you?] Yeah usually, it's when average security - they've got that security and somebody has hacked into their system and personal details have emerged or been compromised, but if it's only mobile numbers I mean it's not like bank account details and stuff like that is it. I don't think hospitals are very good at security are they? So yeah, that's what it means to me, it's sort of a fact of life these days isn't it?", P20.

> "[Who do you think personal information belongs to?] Well exactly what the word says, the person. It's like their money and they can give it to who

> they like. No matter how many times it passes around you must get permission from the first individual unless of course like me it's giving permission for the university to use, for the medical professionals to use… because it's all safe. You know, they're sending a parcel to each other it's all there online so they access the same data", P44.

Participants were confident that they would be able to withdraw from RUDY should they wish to, stating the assumption that researchers have a duty to safeguard data while participants have a duty to provide truthful information. Interestingly in the workshop I used to validate these findings, researchers and research students it was reported that boredom is one reason participants revoke access (Schuler Scott *et al*, 2019a). This is not necessarily the case – if the research was of no value, or a moral breach were reported. There was no mention of boredom from the participants I interviewed.

There was evidence of engagement within the RUDY study – "engagement" emerged as "interest", "knowledge" and "active participation". For the former, many participants were interested in RUDY because they felt isolated. A lack of peer and professional support around their condition meant they needed to actively seek information. "Knowledge" was not communicated by academic literature – participants reported reading news articles to learn about the world around them, particularly data breaches. Participants who actively took part in the patient focus group felt that RUDY was hands-on. They were a significant minority of my interview population. Few participants reported anywhere near this level of involvement as the only interaction they had with RUDY was filling in surveys. Participants did not expect to engage with the study at all, which may be because they do not currently engage with RUDY. Participants expressed a desire to take part in research and receive findings, continuing taking part despite these needs being unmet.

> "It's just an email isn't it?", P02.

Interestingly, many participants did not feel they had control over their personal data in daily life and expressed a preference for control within the context of research participation. Control was described as important by many participants and in the quote below the participant describes the value of control in the face of an overwhelming amount of day-to-day data-access requests online:

> "[What do the words 'data breach' mean to you?] It could be that someone has left a file somewhere or it could be that a computer has been hacked. [Do you think that presents a problem?] I suppose that I am in an age group [55 – 65] where it may be more of a concern but quite honestly it seems to be happening in so many places - I personally don't sign up to social media and things like that to try and keep a bit of a handle on it but

> people have so many ways of finding that sort of thing out nowadays. I would probably trust the good fortune. It depends how big the data breach was; how serious it was. What can anyone do with them (medical records)? I know it may end up with me having spam calls from companies that make equipment and stuff...", P03.

Elsewhere in the data, participants also referred to their own data-protection processes such as checking website security and limiting information disclosure on social media. The majority of participants however, did not give details about how they might exercise such control themselves.

4.3.4 Dynamic Consent and a lack of engagement

This review identified that there is a need to test live implementations of Dynamic Consent, to justify its use in practice. Table 20 below brings three reports of Dynamic Consent in practice (Norval & Henderson, 2019; Dankar *et al.*, 2020; Mamo *et al.*, 2020) together to indicate the scarcity of engagement as part of the discussions around Dynamic Consent, where practice and implementation are discussed.

Table 20: An indication of how three studies focusing on Dynamic Consent have considered engagement.

	Revocation	Project aims	Data used	Data-uses	Contribution value	Outcomes
Automating consent	☑	☒	☒	☒	☒	☒
Solving ethical dilemmas	☑	☒	☒	☒	☒	☒
Blockchain	☑	☑	☒	☒	☒	☑

Despite reaching its first decade, literature on Dynamic Consent tends to lack practical application which means that the theory remains largely untested. Because the field lacks guidance, tools and empirical measures, we cannot reliably justify the claims being made because they have not yet been measured or proven widely applicable. For digital, dynamic forms of consent to exist, researchers must collaborate – in talking with RUDY participants and researchers I have found that researchers, administrative staff, technology developers, clinicians and participants are all vital to the research process. The project has been designed to seek input from these different stakeholder groups. While technically, Dynamic Consent relies on a software development team to implement

the abstract ideas of consent revocation and engagement, these abstract principles need to be prioritised by researchers as they design their methodologies.

4.4 Reflection

Findings from this study contribute to the Dynamic Consent literature by identifying how the use of Dynamic Consent has been fundamental to RUDY's ongoing success. Dynamic Consent provides evidence of institutional compliance with data-protection[19], helps to accommodate participant autonomy (Kaye *et al*, 2012; Budin-Ljøsne *et al*, 2017; Schuler Scott *et al*, 2019a) and develops a 'research partnership' approach (Javaid *et al*, 2016; Teare *et al*, 2017). RUDY has focused on building rapport, in addition to carrying out rare-disease research. The team has done this by developing an online interface, building connections with advocacy groups (see Aymé *et al*, 2008), and maintaining a group of volunteers acting as project consultants. Participants trusted researchers to use data appropriately, with little to no expectation of any practical research outcomes - findings also suggested that enhancing the feedback available to participants may result in more effective longitudinal engagement. Enhanced feedback is offered as a way to structure practical steps for engagement within Dynamic Consent.

The Dynamic Consent literature to date is more theoretical than practical, which means there is a lack of empirical evidence supporting, or negating, its use in practice. Current use of Dynamic Consent fails to build engagement as part of a Dynamic Consent practice, which is unacceptable. This means, of course, that this work is timely and relevant in observing a practical example of Dynamic Consent and providing evidence to strengthen the position of engagement as a founding principle of Dynamic Consent. In this reflection, I will explore key findings, focusing on insights particularly relevant for the longitudinal engagement of study participants. I then offer a brief reflection on methodology; specifically, the use of emails as an interview tool and what that might mean for empirical research with patients in the future. Finally, a note on the validity of the core Dynamic Consent assumptions and what these findings suggest regarding consent choice architectures within research.

RUDY participants had their own expectations around data-protection and the use of data for research purposes. They did not want granular control of data, and were happy to delegate technical details to RUDY researchers. Participants expected researchers to protect their data, and to make them feel safe. They also wanted changes in how data was used to prompt a re-consent process. Borup *et al* describe expectations as the key

[19] In the focus group, researchers describe how versions of consent have improved over time.

to science and technology due to the way they "mediate boundaries" (Borup *et al*, 2006). When describing participation in RUDY, participants often blurred the lines between the medical and research domain; in one, they occupied a non-expert role as patient, while in the other they occupied a non-expert role as a research participant. Even though participants recognised their own wealth of experience as someone living with a rare condition, they relegated themselves to a non-expert role. RUDY researchers, on the other hand, described participants as experts whose feedback was invaluable to the project. Researchers should reiterate the importance of participant input, as expertise.

Participants described an "obligation to participate" (John, 2009), citing participation as a choice - choosing not to contribute was regarded as selfish. Some participants said that they felt a need to encourage others to contribute where they could, exerting influence within families and at support groups. These "peer pressures" to contribute to a study like RUDY reflects existing work highlighting the importance of social circles on an individual's data-sharing decisions, and the degree to which people are willing to delegate these decisions (Nissen *et al*, 2019). This work adds to a body of literature demanding that data-sharing options better represent user concerns (Culnan & Armstrong, 1999), and are introduced via informed consent processes at the start of a project (Helweg-Larsen & Shepperd, 2001).

RUDY's clinical lead, project manager and software developers were heavily focused on creating a positive participant experience. The online interface was designed to be easy to use in order to collect data directly from participants and retain users[20]. When the project started, RUDY created a patient forum to gather feedback regarding participant experiences and to communicate project updates. Through this, participants have made changes to the research process, such as introducing new consent options around the use of post-mortem data. Researchers said that they were concerned about 'consent fatigue'; they wanted to avoid presenting participants with an overwhelming number of data-sharing choices which could result in choice paralysis and disengagement with the decision-making process overall.

Members of the public can highlight concerns that are not immediately obvious to experts (Norberg *et al*, 2007). One such concern was a participant who was unable to take part in a telephone interview and wanted to know whether they could answer my questions at their own pace via email. This had not been a consideration, but email is a useful method for reaching overlooked, or difficult to access groups as they are permitted to respond on their own terms and in their own time (McCoyd & Kerson, 2006). Emails

[20] For further work on the importance of Dynamic Consent interfaces and ease of use online, see Thiel *et al*, 2015.

can also provide richer data as participants have time to reflect on questions (Stacey & Vincent, 2011). I concluded that emails could be leveraged to enrich the array of tools available for qualitative data collection (Burns, 2010), and as soon as I started offering this option participants started to request it.

These findings are likely to be relevant for institutions such as the UK Biobank[21], and centres such as Genomics England[22] and Australian Genomics[23]. These biobanks operate online, and are in a position to consider how to inform participants over time, and to provide information that is likely to encourage participation. These findings may also be of use to any of the number of cohort studies supported by the UK's Medical Research Council, such as the Avon Longitudinal Study of Parents and Children[24]. The findings reported suggest that core Dynamic Consent assumptions are valid; namely, that participants want a greater understanding of how their data is used, and they would prefer a degree of continuing control over that usage. Flying in direct opposition to proponents of the privacy paradox, people care about how their data is used. Unfortunately, this study showed that, even where researchers prioritised the participant experience, participants did not feel as empowered, informed or engaged as researchers hoped they would be. Hope then, is not the tool we require in building Dynamic Consent into research, which the next chapter will explore further.

RUDY offers an example of inclusive, but not responsive data-practice. Including participant input has led to positive interactions, high recruitment numbers from a small population, and research processes tailored to that participant population. The research team highlighted a need for participant engagement but had not focused on it due to other research priorities. There are three areas to be explored further. First, that engaging with RUDY participants may increase general awareness and participation. Second, that there is a need to empirically measure the research value of engagement: this data hints at that value, such as the enrichment of consent options based on participant feedback, but does not provide an empirical measure. Third, a more detailed description is needed of how relevant information can be communicated to participants, to guide implementation steps.

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[21] https://www.ukbiobank.ac.uk

[22] https://www.genomicsengland.co.uk

[23] https://www.australiangenomics.org.au

[24] https://www.bristol.ac.uk/alspac

Chapter 5

Study 2: ask the audience.

Enhanced feedback: information will be communicated to RUDY participants that makes the importance of their contribution to the project more immediately obvious.

5.1 Introduction

Chapter 4 highlighted an opportunity to focus on participant engagement within RUDY. Engagement is one of two fundamental tenets of Dynamic Consent, a theory that RUDY is putting into practice. When asking participants what they wanted to know about RUDY, they wanted interesting information, they wanted to learn, and they wanted to be told how they might be able to take part. Simply informing participants does not mean that they are engaged – a more effective informed consent process, that actually informs over time, may need to signpost participant forums, and showcase the monthly project newsletter. Longer-term studies might host annual workshops or publish annual reports. A hypothesis that this chapter explores is that to truly inform participants over time, feedback must be relevant and easy to understand. To do this, I asked RUDY participants how they wanted to receive information about the study. I then worked with the RUDY team to implement the most feasible changes (most of the suggestions).

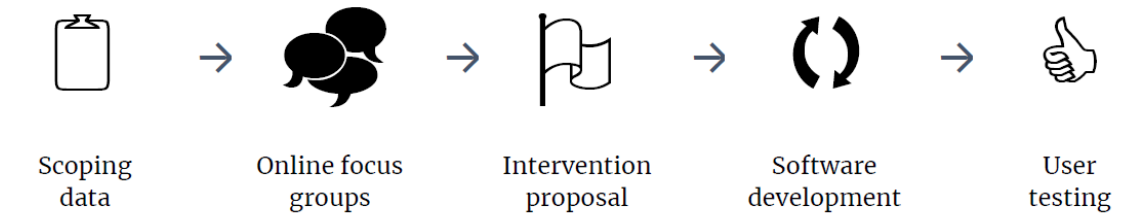
Accessibility and convenience were key design considerations during interview development in the last chapter. The co-design process described in this chapter was designed to examine the nuances of these requirements and identify opportunities for further development. I asked participants to describe what they wanted to know about RUDY, and worked with the research team to enhance the way information was fed back to participants about the project. These enhancements involved making information on the project website easier to find and understand, and identifying which key pieces of information to re-iterate at points of contact: the participation reminder email and online portal. The reminder email was a regular contact point controlled by RUDY, as participants received an email every six months to remind them to fill in questionnaires. Participants could visit the portal whenever they wanted to, this lay outside of RUDY's control. This chapter describes a co-design process with two objectives: to validate the findings from Chapter 4, and to develop a study providing an empirical measure of the value of participant engagement in research.

5.2 Methods

The co-design work described in this chapter was designed to elicit responses from RUDY participants and researchers. Participant feedback was gathered through online focus groups and researcher feedback was gathered throughout a software development cycle. This work supports the second research aim of this thesis[25], designing an intervention study measuring the effect of feedback on research participation. This study was co-designed with participants and researchers. Figure 21 illustrates the co-design process. "Scoping data" refers to findings from Chapter 4 that provided initial direction. I created a list of proposed changes to RUDY and invited participants to comment on this intervention proposal in online focus groups. Amendments were made and the proposal was presented to the RUDY research team, who worked with me to prioritise these changes and decide what was possible and necessary to implement. The proposal was developed into a list of software requirements which the development team implemented. I recruited participants from the focus groups to carry out user tests, reviewing these changes and identifying any unforeseen issues. Feedback from five user tests was presented to the research team, final edits were made and a date was confirmed to commence data collection.

[25] Research aim 2: to extend the theory of Dynamic Consent by providing empirical evidence to strengthen a research requirement for engagement.

Figure 21: Development methods for intervention study co-design.



The inclusion, once again, of both RUDY participants and researchers increased the chance that the outcomes of this work would be useful to both parties. This work is also predicated on sample data (Section 6.2.1.1) showing low questionnaire completion rates; the study would test an outcome that was valuable to RUDY. On submission of a data access request, RUDY provided an anonymised dataset that contained information for all adult participants with valid consent in the period from the June 1st to December 12th 2019. Table 21 shows that each participant record consisted of a patient identification number, their date of consent and a record of the different questionnaires (see Table 22) they had completed between June and December 2019. For an example of the kind of data RUDY provided, see the table below. "Rich" data collection can simply mean more people provide information, but RUDY's particular concern is that the same individuals return to provide data over time. This is what provides insight into lived experiences of rare conditions. The initial data provided by RUDY allowed me to observe low completion rates as an issue (see Table 23), and validate with the research team that this was a problem in the study. Identifying this as a key problem for RUDY provided a metric for improvement: questionnaire submission. I calculated 'completion rate' from the number of questionnaire assigned and the number of questionnaires returned.

Table 21: Example records from RUDY data sample (adults, Jun 01-Dec31 2019).

		ADL questionnaire			
Patient ID	Date started	Assigned	Submitted	Not done	Started
2	2014-04-23	1	1	0	0
13	2014-04-23	0	0	0	0
11	2014-04-24	0	0	0	0
18	2014-04-24	1	0	1	0

Participants are asked to submit multiple questionnaires via the online portal, to measure different variables. I started gathering data on questionnaire-specific response rates in longitudinal medical studies to compare to RUDY's data, but this took an excessive amount of time and research effort, which is why such a comparison is not included.

Table 22: Questionnaires for which I received preliminary submission data.

ADL	Nottingham Activities of Daily Living assesses daily activities (Yohannes *et al*, 1998).
EQ5D	EQ5D-5L measures quality of life: mobility, self-care and discomfort (EuroQol, 2018).
PSQI	Pittsburgh Sleep Quality Index measures sleep quality (Buysse *et al*, 1989).
MCGILL	McGill pain questionnaire collects ratings, description and intensity (Melzack, 1975).
SF36	Measures function, limitation, mental health, pain and perception (Brazier *et al*, 1992).
PAINDETECT	PainDetect measures pain (Freynhagen *et al*, 2006).
FACIT	Functional Assessment of Chronic Illness Therapy – Fatigue measures physical, social, emotional and functional well-being, and fatigue (Muszalik *et al*, 2009).
HADS	Hospital Anxiety and Depression scale measures possibility and probability of anxiety disorders and depression (Bjelland *et al*, 2002).
EPWORTH	Epworth Sleepiness Scale measures daytime sleepiness (Johns, 1991).
FATIGUE SCALE	Modified Fatigue Severity Scale (Krupp *et al*, 1989) measures fatigue severity.
WALK 12	Walk 12 measures the effect of neurological disability on walking (Holland *et al*, 2006).
OTHER	An unknown questionnaire, used as an indication of participation.

Table 23: Average completion rate by questionnaire, for RUDY participant cohorts.

Completion rate	ADL	EQ5D	PSQI	HADS	WALK12
Cohort 2019, n = 84	13%	13%	13%	13%	13%
Cohort 2018, n = 77	24%	20%	16%	16%	24%
Cohorts 2014-19, n = 273	26%	23%	21%	21%	27%

5.2.1 Online focus groups

The online focus groups were designed to gather participant recommendations, and explore in more depth the apparent lack of engagement within RUDY. I sought ideas for specific changes that could be made to the project to improve participant understanding. Focus groups were run over Skype, nine interview participants returned to take part and six further participants were recruited. To allow for limited time and energy restrictions, I offered participants different options for active engagement: they could contribute during a Skype call, give feedback via email on the final intervention proposal, or both.

A call guide was created to provide focus group structure, and sent to participants a week ahead of time so that they could familiarise themselves with the literature and reflect on what they wanted to say. The focus group was piloted with colleagues in the Department of Computer Science and Faculty of Law, who provided feedback that helped to refine the call guide and session structure. I made themselves available online 15 minutes before the calls were due to start, in case technical support was needed or participants had any questions. This meant that calls started promptly. I conducted the focus groups, acting as facilitator. Supervisor HT took notes, debriefed I after each call, and took notes. This provided more material for analysis, and another perspective on the content of the calls. Consolidation of My and HT's notes produced ideas for an actionable intervention that I developed into an intervention proposal (Appendix 3). This proposal was validated by focus-group participants before being presented to the RUDY research team.

Call guide development

For the call guide in its entirety, see Appendix 10. Section 3.2 describes the Responsible Innovation principles guiding this work; aligning research to societal need, prioritising diverse thought and experiences, inclusive and socially desirable aims, responsiveness and acting in the public interest. Focus group implementation drew from NCCPE guidelines on creating virtual public engagement events (NCCPE, 2020). Table 24 provides a summarised account of how these prompts were the starting point for designing the focus groups.

Table 24: NCCPE virtual event guidelines used to design the focus group.

Questions to ask	My reflections
What are you trying to achieve?	Changes to RUDY that increase questionnaire submissions.
Accessibility requirements?	Participants: limited time and energy.
Is there a script or prompt?	I can make (and send out) a call guide.
Have you rehearsed?	I can run a pilot with colleagues.
The event	
Encourage people to arrive early.	I will be online ahead of time, and say so.
Plan breaks ahead of time.	I can create a schedule to prepare participants, and for me to follow in-call.
Keep to the timings you have set.	
Tell people how they can take part.	Mute microphones when not talking.
Facilitation	
Be clear and repeat instructions.	Bear these in mind during the pilot, and ask for feedback. As there are multiple calls, I can ask HT to provide feedback on facilitation technique.
Agree etiquette at the start.	
Use slides, and have a welcome slide.	
Consider providing cue cards.	
Ethical consideration	
I do not want participants to sign up to a new service, sharing personal data. All participants have Skype and are familiar with its operation.	

The call guide included a project brief to provide an overview of what the sessions were for, and descriptions of the group activities. Rather than having me speak the entire time, I designed activities to invite feedback throughout and encourage a two-way conversation (Riddle & Treder, 2015). Table 25 shows an initial layout for the focus group,

held online because RUDY participants are based across the UK[26]. The overall session was broken into activities, and data-flows were used to model three types of data-exchange I expected to occur: one-way (from candidate to participant), two-way (from candidate to participant and vice versa), and co-creation activity. Co-creation activities had an activity prompt, time restriction and room for discussion. Two-way communication invited participants to provide input, influencing the research process. For example, in "Overview of work" I briefly describe the research progress and expect no participant contribution, but in "Agenda" I welcome recommendations for changes to the proposed session structure.

Table 25: Initial session structure for online co-design focus group.

Pre-session	Session	Post-session
Research problem	Warm-up	Summary
Overview of work	Introduction	Thanks
Session brief	Creativity task	Results
Videos	Idea collection	Review
Session policy	Break 1	
Agenda	Idea collection	
Questions	Break 2	
	Prioritise	
	Converge	
	Summary	
	Actions	

Session length
90 minutes

Colour code
Exercises
One-way info
Two-way info

[26] Incidentally, these focus groups coincided with the 2020 COVID-19 pandemic. The UK was under lockdown, so meeting in-person would carry significant risk, especially for an already vulnerable population.

Pilot call

A call guide was created for the focus group structure above, and a pilot call was run with five colleagues. The feedback received allowed me to streamline the session[27]. Three colleagues were academics based in the Faculty of Law, one was a project manager in the same Faculty and the other was a DPhil student in Cybersecurity with experience in mixed-method approaches. There were three categories of feedback: relating to the overall process, relating to data-use, and call-guide edits. The overall process was overly prescriptive. Rather than stepping through every technical adjustment with attendees, it was suggested that I give participants basic rules and made it clear that they would be willing to stop at any point. One pilot participant with experience working with patient-participants said:

> "You sound cautious about the group – don't be. They won't be shy and will ask questions", PC1.

They suggested that participants would want to talk because the call offered a rare opportunity to talk about something important to them. They also pointed out that the participants had self-selected to take part and would likely choose to talk rather than need to be coaxed. Another comment led to my deciding to take charge as a facilitator rather than make notes as an observer:

> "[the] protocol needs a way for you to direct discussion", PC3.

> "Taking notes and facilitating is hard. Are you recording?", PC4.

Even in a small focus group, the effort needed to facilitate and take notes effectively would be too much for one person. I asked a supervisor to sit in on the calls and take notes. I was reluctant to record the calls, because this simply was not necessary. Recording also seemed inappropriate, as participants would be talking about sensitive subjects. I was advised to email participants a link to the call ahead of time. This made the call more easily accessible and also served as a reminder to take part. One of the most important comments helped I elucidate what they wanted to take from the focus group:

> "If there is something you want to know, ask it", PC3.

This point was frequently revisited as I developed the call schedule, to help focus her priorities and ensure that the focus groups made the most efficient use of participants'

[27] E.g., feedback from the pilot suggested that I focus the introductory warm-up on the task at hand, rather than treat it as an informal, unrelated task which could have been distracting.

time. The question that I decided to put to participants as a result of this feedback was: "if there is one thing we can do to improve communication in RUDY, what would it be?".

Comments relating to data-use highlighted a lack of clarity in my own communication with participants:

> "Make it clear that you are collecting answers and passing on aggregate information – i.e., there is no identification", PC5.
>
> "Reiterate on the call that answers are in confidence and not linked to participants in any way", PC3.

The call guide was changed to include a note on data-use and a GDPR notice provided by the university. Colleagues also highlighted in-text issues that they saw as either incorrect or worth discussing, such as an over-cautious approach to participants. Phrases such as "if you need anything…", pilot participants suggested, opened the participant up to more work that was necessary. Overzealous provisions like this would likely confuse participants and distract them from the point of the exercise.

A group vote had originally been included as an activity, but this was removed on the advice that this task would be complex in an online environment. Pilot participants asked why the warm-up exercises did not use prompts from the interview vignettes, and suggested that asking for positive and negative responses to the warm-up over-primed participants. Prompts drawing from the vignettes would be relevant to the focus group and would prime participants for the discussion ahead. If participants were asked to focus on a positive and a negative experience, they would not be focusing on their own priorities. Finally, there were two terminology edits:

> "Don't say 'fail' as there are no right or wrong ways to do things", PC1.
>
> "What do you want to hear [or see] from RUDY… [is not inclusive, people experience the world differently] … what do you want to know?", PC3.

The comments received from the pilot highlighted two key issues and addressed minor issues with grammar and readability. The key issues were that the content was overambitious for the allotted time, and I needed to pay further attention to the language they had used regarding data-use. Pilot participants took 20 minutes to arrive at a shared understanding of what the session was for and just as ideas were forthcoming, time ran out. I increased exercise length from 20 to 30 minutes and added a 10-minute break halfway through the session. The entire focus group was lengthened from 90 minutes to 2 hours to accommodate these changes. The agenda was redeveloped to be more flexible, allowing time to explore ideas that emerged during the call. Storyboarding and

sketching activities would have worked well in person but did not translate into an online environment. This is especially a concern with participants who have accessibility issues and different levels of technical ability. "Session policy" was changed to "call etiquette" and prompts such as "please mute your microphone when you are not talking, to reduce background noise" were made more obvious. Table 26 shows the new structure:

Table 26: Refined session structure for online co-design focus group.

Pre-session	Session	Post-session	
Research problem	Introduction	Thanks	Session length
Overview of work	Warm-up	Invitation to comment	2 hours
Session brief	Fill-in-the-gaps	Results	Colour code
Call etiquette	Break		Exercises
Agenda	Discussion		One-way info
Exercises	Summary		Two-way info

The refined structure used three activities, described in Table 27. The first was a warm-up reflecting on the reason for the focus group, completing the phrase "I am a part of RUDY because…". Exercise two provided more fill-in-the-gap prompts, but these statements were more substantial. The third exercise was discussion-based. Prompts were provided and participants drew from and expanded upon the ideas that had been raised emerged in the previous exercise. Ahead of the calls, participants were invited to provide suggestions on their preferred call etiquette. No such suggestions were received.

Table 27: Exercises used in online focus groups.

Exercise 1	Exercise 2	Exercise 3
Warm-up	Fill-in-the-gaps	Discussion
1. I am a part of RUDY because…	1. When I think of RUDY I think of _____. 2. On the RUDY website I _____. 3. I would like the RUDY website to have _____ because _____. 4. When filling out the RUDY surveys I use _____. 5. I would like RUDY to make _____ available.	1. What do you want to know from RUDY? 2. What do you think RUDY wants to know from you?
Purpose of exercise:	Purpose of exercise:	Purpose of exercise:
Participants reflect on the above sentence, and read out their completed version.	Participants reflect on the above statements, and read out their completed versions.	These statements act as prompts for participant discussion.

Online focus groups with participants

Two focus groups were conducted, following the call guide as it was written. I facilitated the session, taking notes where possible. HT supported me by taking notes and clarifying key points that participants raised (e.g., "I liked your point about over-information rather than under-information. What does that look like?"). At the end of each call HT and I compared notes and discussed the call. The first focus group had four participants from RUDY's patient forum[28]. The second focus group consisted of four participants who were part of the wider RUDY population.

My early arrival to the call, and invitation that participants do the same, meant that calls started on time because there was time to troubleshoot technical problems. Participants

[28] A particularly engaged sub-section (and minority) of RUDY's participant population.

helped each other with such issues, which seemed to build rapport ahead of the call. Each call ran to time. Notes taken during the calls were drawn together to create a list of suggested changes that the RUDY development team could incorporate into the website, online portal and the email reminder sent to participants. This list of changes was sent to all eight focus-group participants, as well as three people who had expressed interest in being part of the co-design process but could not attend Skype calls. Feedback was received from eight people, and these responses validated findings from the focus groups (see Table 28).

Table 28: Participant feedback on report collating online focus group findings.

"The lack of contact on RUDY is a disincentive not only to participate but as part of my clinical experience. So in a small way, it's damaging to my health. Oh Dear! But doctors are so busy! With what? Ignoring patients?", P1.
"I think we all see it as one-way traffic, (from us to RUDY) which is great, but means a lot of the potential use is untapped. I must admit, even though I'm aware of these uses, I don't use them! Wonder why???", P4.
"I feel some information needs to be prior to log in to entice more participants: [e.g.,] current projects – videos or quote", P6.
"I am happy filling in the survey so that it can be of use to someone. As far as I am concerned it is not the end of the world for me if I do not know how it is being used and feedback to the patient. Those things are of course nice to know, but I can live without them", P7.
"I think you are very correct in your thoughts that people feel as if they give quite a lot by filling in the questionnaires every six months but get very little back", P9.
"I have read your report and it is very good you have covered ever thing we asked you for", P11.
"I think the email sent out to participants asking to fill out surveys...should just re-emphasise the value of the data generated to the medical professionals involved and explain that whilst some of the questionnaires may seem repetitive", P13.

> "Thank you for your document – I agree we give all the details but have no idea how the research is progressing as stated in page 2", P14.

Participants were in broad agreement that their views had been captured:

> "I think you are very correct", P9.

> "I have read your report and it is very good", P11.

One participant summarised what they thought was important in the document, reiterating the perceived lack of control observed in Chapter 4:

> "I agree we give all the details but have no idea how the research is progressing as stated in page 2… Things are of course nice to know, but I can live without them", P7.

Also evident are stronger opinions and more confident suggestions from participants:

> "[more] information needs to be prior to log in to entice more participants", P6.

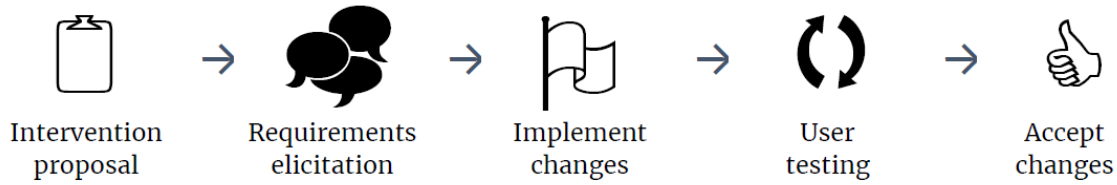
> "the email sent out to participants asking to fill out surveys…should just re-emphasise the value of the data", P13.

The participant-validated list of changes was presented to the RUDY research team as an intervention proposal (Appendix 3), and used as a reference document during software development.

5.2.2 Software development

The intervention proposal consisted of a list of proposed changes to RUDY's website, the online portal through which participants access questionnaires, and the email reminder. This proposal was used to start a discussion with the RUDY research team, and define requirements for a software development cycle implementing these changes. The development cycle consisted of five stages, illustrated in Figure 22. The first step was to meet with the RUDY research team, show them the focus group findings and discuss the intervention proposal. I worked with the research team to clarify overall development goals, set expectations and elicit software requirements. For each suggested change on the proposal, the project lead and development lead commented on how important that change was to them (priority), and the how realistic that change was to implement (feasibility).

Figure 22: Software development process for intervention study co-design.



Once the changes had been agreed, the RUDY development team implemented them. I prepared a user testing document, inviting co-design participants to take part. This document consisted of a list of tasks that had been defined in the original intervention proposal, and open questions such as "are these changes useful?" to identify any other issues that participants had. Once the project lead had agreed that user tests could be carried out, five user tests were carried out.

One user test was conducted via email, where the participant worked through the task list at their own pace and wrote down their feedback. Four tests were conducted over the telephone or via Skype, lasted approximately 30 minutes. Users were asked to describe what they thought a button would do before they clicked on it, and they were asked to describe their actions as they carried them out. This was to compare their expectation to their actual experience. I reported findings from these tests back to the RUDY team along with a prioritised list of small changes that needed to be made. As with the original requirements, RUDY's project lead approved these changes. Once these were made, the project lead signed off on the development work and the enhanced feedback intervention was made public.

User testing

Focus-group participants who had expressed an interest in taking part in further research activities were invited to participate via email. When a participant responded, they were sent a copy of the user testing worksheet (Figure 23) and a test was scheduled. The user testing document drew from the original intervention proposal and the requirements elicitation process with RUDY researchers. A list of tasks was created that users needed to describe their experiences of, and complete. For example, where the intervention proposal suggested "make a current projects section on the website", the corresponding user task was to identify current projects happening within RUDY.

Figure 23: Extract from user testing worksheet.

User testing worksheet

There are two parts to today's session. During the first you will be exploring changes to the website. You will then login to the test site. As you attempt the tasks below, bear in mind that all feedback is good feedback...

Which means that if you find anything difficult to do or understand, please point it out.

	Task description	Can you find it?
0	Go to https://research.ndorms.ox.ac.uk/rudytest	Yes / No / Not easily
1	What is the aim of RUDY?	Yes / No / Not easily
2	What does RUDY do?	Yes / No / Not easily
3	Who does RUDY work with?	Yes / No / Not easily
4	How many people are signed up to RUDY?	Yes / No / Not easily
5	What does RUDY do with participant information?	Yes / No / Not easily
6	What are some of the current projects going on?	Yes / No / Not easily
7	Is there any information missing from the top of the page?	N/A
8	Is the information you want to see easy to find?	N/A
9	Have you learned anything about RUDY you didn't know before?	N/A

This document was sent ahead of time so that participants could look through the task and prepare any questions they had. For the user testing document in its entirety, see Appendix 5. Two user tests were done online, two via the telephone and one via email. I took notes throughout the online and phone calls, consolidating these notes and email feedback in Nvivo. From this, I generated a report on key findings, which included a prioritised list of edits to be made before the changes were published. The RUDY team agreed to implement the list in its entirety, with the project and development leads commenting on the suggestions made.

5.2.3 Analysis and validation

Focus group transcriptions were loaded into Nvivo, with an initial reading providing an overview of the data. As with the previous analyses, extracts of interest were highlighted. The focus groups provided more information about RUDY's function and the features available to participants. Table 29 shows the categories used to group these extracts.

Table 29: Categories used to group data collected from online focus groups.

Categories for online focus-group data	
1. How participants currently use RUDY	Experiences regarding questionnaires
	Experiences regarding the website
2. Motivation to take part in RUDY	Related to research
	Unrelated to research
3. How participants think RUDY should communicate	
4. Features that participants would like to have	
5. Comments that surprised me	

Analysis of the qualitative data collected through the online focus groups and user test sessions was done as a form of thematic analysis. Three validation steps were taken, adding rigour to the findings gathered using this methodology. The intervention proposal was approved by participants, user tests were carried out after the initial development effort, and a second round of development effort ensured that the intervention study was ready for data collection to proceed.

5.3 Results

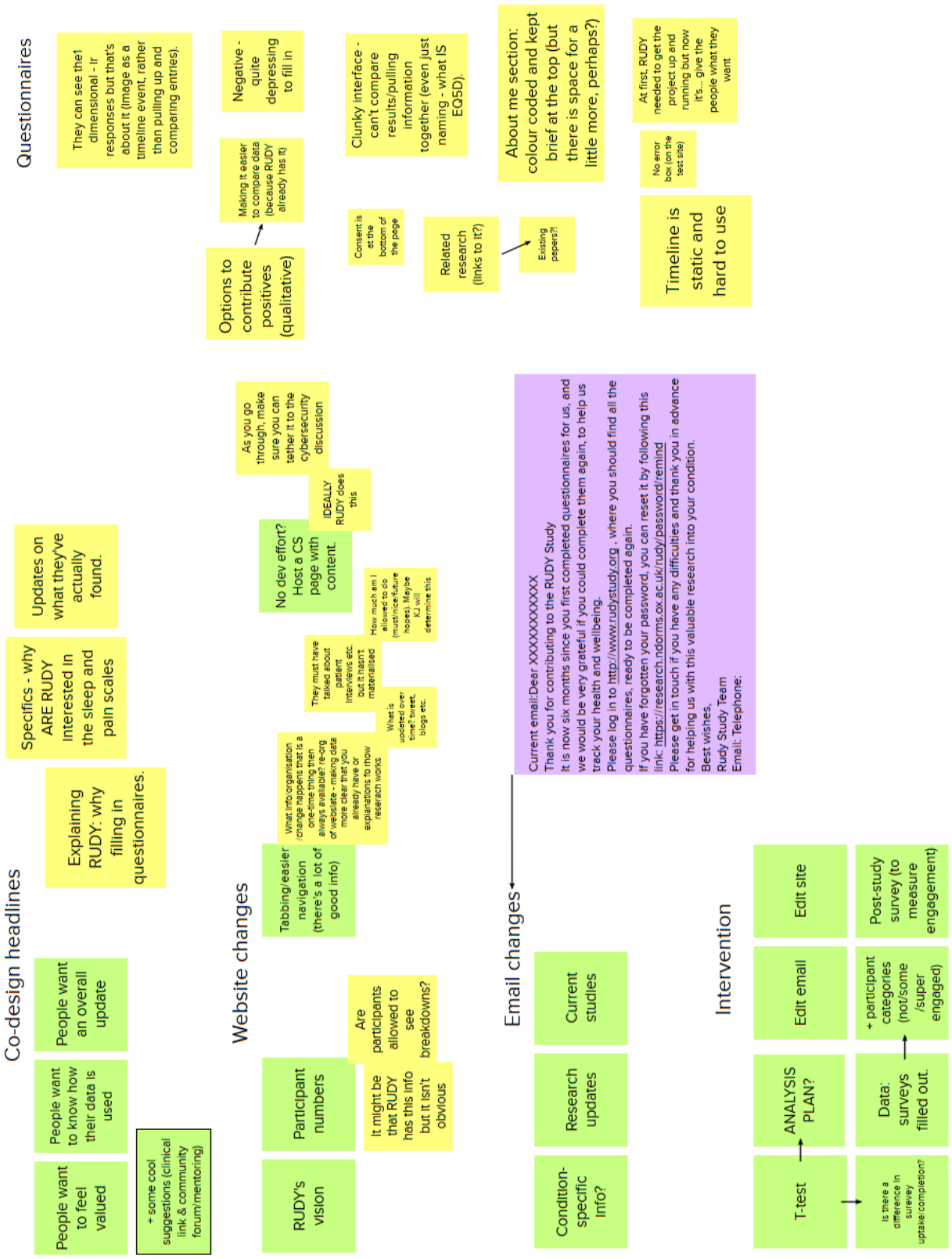
5.3.1 Online focus groups

One participant suggested that there were three types of participant enrolled in RUDY: active and enthusiastic, active and disengaged, and inactive and uninterested. These profiles, based on research activity and engagement levels, highlighted the group of participants that an enhanced feedback intervention would hope to influence. Active and enthusiastic participants were unlikely to be affected by increased communication from the study, and inactive and disengaged individuals were unlikely to notice. The people described as active and disengaged were those who were filling out questionnaires but did not know much about the project. In other words, the people who had been interviewed in Chapter 4.

There were seven areas that participants suggested were important and not currently communicated effectively. The highest priorities were communicating data-uses and research outcomes. The former was described by participants as a desire to understand their research contribution, and the latter related to making research articles available and more accessible. The other changes that participants identified as steers for the intervention included providing information about the project such as overall vision, funding information, upcoming work, how useful the research was, regular updates, and the number of participants. Participants also discussed peer support in the form of a forum or buddy system, and wanted information to be provided so that they could make decisions about medical care. The first of these would be an entirely new feature and outside the scope of this project, although RUDY Japan has implemented such a community (Hamakawa *et al*, 2021). Regarding the latter, I lack the medical expertise required. This feedback was passed to the RUDY team in case it could be useful to them in the future.

Figure 24 shows the output of a whiteboard session between HT and I after the focus groups data had been collected. The aim of this discussion was to create an intervention proposal to deliver to the RUDY team. Comments were grouped by “website”, “email” and “questionnaire” to identify which changes could be made to the project website, the reminder email sent out to participants, and the questionnaires available through the online portal.

Figure 24: Whiteboard discussion of key points raised in focus groups.



Appendix 3.

Table 30 shows how these ideas were developed into a proposal recommending changes in three areas. For the proposal in its entirety, see Appendix 3.

Table 30: An overview of the intervention proposal.

The RUDY website	Make a "current projects" section on the website and ask researchers for a video or quote.
	Put important information at the top of the website and make content easier to navigate.
	The number of participants in RUDY should be at the top of the website.
Email sent to remind participants to fill in questionnaires	The email should include the number of RUDY participants (or with a similar condition).
	The email should include RUDY's vision ("helping us improve research in rare diseases").
	The email should give an example of a collaborator who used data from the surveys.
Questionnaire descriptions (online portal)	Survey pages should give an example of how a RUDY collaborator has used that survey.
	Survey pages should describe how researchers use the data from surveys.

5.3.2 Software development

Table 31 shows how I worked with RUDY's research team to develop technical requirements. For this discussion in its entirety, see Appendix 4.

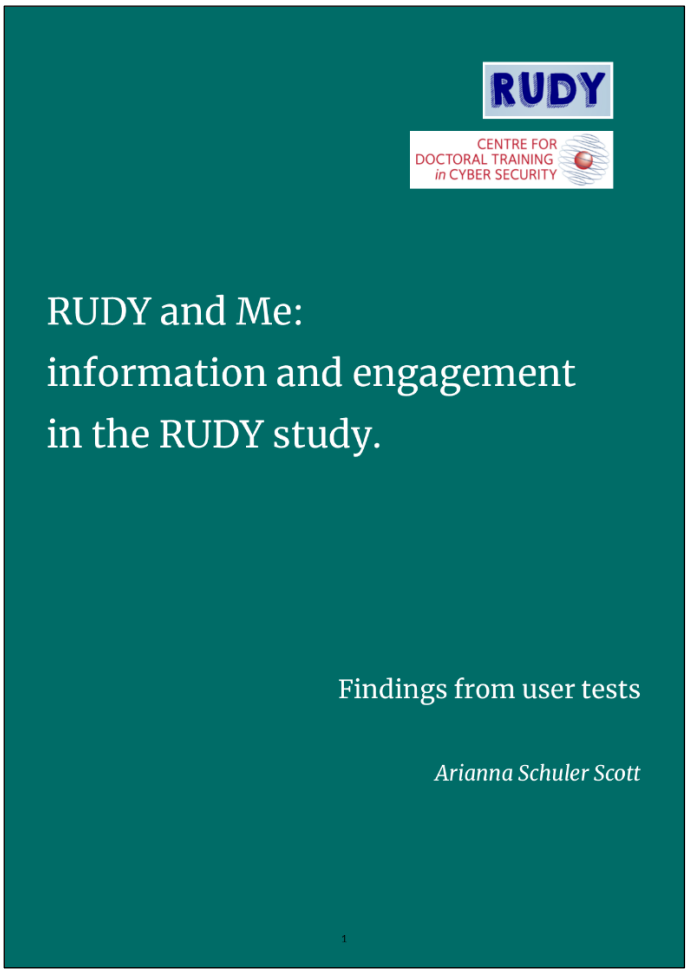
Table 31: Feasibility check of proposed changes, with RUDY team.

Proposed change	RUDY comments	My comments
Web site		
What we have done with the data so far (summary videos).	Can do – KJ Do you have more detailed information on what we have done with the data? (perhaps a 'blog' or news page via homepage would be a good addition) – sure I'll draft one.	Certainly a "news" page with text and video updates from collaborators and RUDY. A blog needs more oversight and resources (e.g., blog writers).
Email reminder		
Add by disease group number recruited.	We will prototype some work on this –possible, but we have concerns about current groupings needing to be manually curated.	Brilliant, thanks! Let's review this in our next meeting.
Questionnaire description		
Add a "find out more" box that describes why we use this questionnaire and what it will be used for.	In development for the new site – should be possible on existing site, but we will need to weigh up.	A brief overview, rather than in-depth description (e.g., "this questionnaire is used to get an idea of…").

User testing documents were sent ahead of time, and three participants prepared points they wanted to raise in advance. Their notes clarified the tasks at hand and highlighted issues that I had not noticed (e.g., some headings were long and complicated). These issues were important to identify and fix, as RUDY participants may only visit the website twice a year and if they cannot understand the website then they will not be able to navigate it. The highest priority changes in the user test report were those which prevented users from carrying out tasks described in the intervention proposal. The lowest priority issues were unrelated to the intervention's original aims. Figure 25 shows the user testing report in its entirety.

To summarise, the document is in three parts: "This document", "Suggested changes" and "Further details". Section one gives an overview of findings, with a summary at the top of the page, methods outlined below and a breakdown of user tasks. This table was used to show that participants found certain tasks more difficult than others, focusing the reader's attention. The report uses four tests because the user test carried out via email was done after the report had been written[29]. Section two is the prioritised list of edits to be made. There are two categories – "highest priority changes we should make before going live" and "minor edits that would significantly improve participant experience". High priority edits were those that were needed to ensure that we met the original brief provided by the intervention proposal. Minor edits emerged in the tests as issues that held participants up, confused or frustrated them but were not part of our brief. Section three simply shows all feedback, for the RUDY team's reference. I thought it important to show how much positive feedback there was.

Figure 25: Findings from user tests (4 pages).



[29] But corroborated findings from the other tests.

This document

In the last week I have worked with 4 RUDY participants to collect feedback on the changes implemented on the website. Positive feedback was received for these pages and the redesigned home page, but there are issues with navigating content.

This document is intended to help prioritise final edits to the RUDY website for the intervention study in collaboration with Arianna (Computer Science DPhil).

On a 30-minute call, I worked through a list of fact-finding tasks with the participants. I wanted to know if they could identify the changes made, and whether these changes were useful. This is because they were involved in designing these changes. After the exercise, I asked whether the users would "accept" these changes, as in whether or not they thought the changes met the original brief "to communicate useful information about RUDY back to participants".

Task description	Test 1	Test 2	Test 3	Test 4
1. What is the aim of RUDY?	Yes	Yes	Yes	Yes
2. What does RUDY do?	Yes	Not easily	Not easily	Yes
3. Who does RUDY work with?	Not easily	Not easily	Not easily	Yes
4. How many people are signed up to RUDY?	Yes	Not easily	Yes	Yes
5. What does RUDY do with participant information?	No	No	No	Not easily
6. What are some of the current projects going on?	Not easily	No	Not easily	No
7. Is there any information missing from the top of the landing page?	Yes	Yes	No	Yes
8. Is the information you want to see easy to find?	No	No	No	No
9. Have you learned anything about RUDY you didn't know before?	Yes	Yes	Not easily	Yes
10. Acceptance: do these changes achieve the original brief?	**Yes***	**Yes***	**Yes***	**Yes***

*conditional—"the content is there, but edits should be made".

Suggested changes

These are the highest priority changes that we should make before going live.

Issue	Suggested change
It is not clear that RUDY is a research project. *I couldn't find that RUDY is a research project, I assumed it was... there is nothing about the research process!*	FROM: "Help us improve research in rare diseases" TO: "Patients and researchers working together for better rare disease care"
The patient-centric nature of RUDY is not clear. *The wording is not inclusive... not obvious that they want me because I have a rare disease.*	FROM "Sign up today" FROM: "More information" TO: "Join us now" TO: "Find out more"
Key information is obscured by unexplained terms. *"Phenotype", what is that? Can you describe it?* *The page has everything we need but it could be more user-friendly or non-academic.*	On "What is Rudy": FROM: "To determine a detailed **phenotype** of patients with rare diseases." TO: "To determine how different symptoms present themselves in patients with rare conditions" FROM: "To identify unique patient subgroups within each disease cohort" TO: I have no idea what this means—can you simplify it? FROM: "personal", "understanding" and "research cohort" being in bold. TO: the above terms not being bold.
The landing page has to have all key information on it, or prompt people to read more. *It looks like, after FB/Twitter buttons, that's it. Can you add a down arrow? Encourage them down the page!* *I could only [find] answer [s to] what I could see.*	Decide whether this is something that you have time to implement. The feedback suggests that attention is not captured and most people will not scroll down the page. Signposting further down the page would be good to have.

Minor edits that would significantly improve the user experience:

FROM: "280 recruited so far"	TO: "280 joined so far", or "We have 280 people taking part"
FROM: "We have a lot of information about Rudy in the Library, that hopefully will answer most of the questions you may have. If you don't find an answer then please..."	TO: "Our Library will give you an idea of the kind of work we do as a research study. If there are any questions you can't find answers to then please..." **AND: change "Rudy information" to "Library" on landing page.**
FROM: section "Which diseases are we interested in" has no information about diseases.	TO: move the list of diagnoses from "How far have we come" to "Which diseases are we interested in".
FROM: section title "How far have we come".	TO: title "What do we have going on?" or "Project news".
FROM: section "What is Rudy" does not say the video is different.	TO: include a description like "find out more from Dr Javaid, who leads this project", or "watch this video to find out more about what we do, and the benefits of taking part".
FROM: section "What is Rudy" has erratic typography.	TO: make "Primary objectives", "Secondary objectives" and "Benefits of participation" the same heading level. AND: put the numbered points and bullet points at the same level.

☺ Positive feedback.
☹ Negative feedback.
? Question or comment.

Further details

Landing page (research.ndorms.ox.ac.uk/rudytest)

☺ Good use of animation.
☺ Clear boxes.
☺ I like it. My initial impression is "this is nice", it's user friendly and looks better but I can't say why!
☹ The "RUDY information" button is called the "Library" elsewhere and this is confusing. I like library but either works.
☹ No complete list/not a lot of info on rare diseases at first glance.

New pages (research.ndorms.ox.ac.uk/rudytest/more_information)

☺ I like the menu on the side, it's all in one place.
☺ There is a lot of great stuff, the video, the information.
? My first thought is that it looks fine but irrelevant to my condition.
? I don't know what the patient forum is. Am I on it?
? "How far have..." what do the numbers on the right mean?
? The "Library" mentions research without any of the details. People want information that's quite detailed. When I was on the [co-design] call there was a lady who had obviously put a lot of work into understanding her condition. Perhaps the information presented does not give credit to how engaged patients are with their health. You could have bits for non-experts, and bits that say "here's a research report, you are going to need a dictionary".
? "What is RUDY": on the first look I did not click the video because I thought it was the same one as the one on the landing page. Perhaps you could give it a heading like "find out more from the leader of the project", or "watch this video to find out more about [list Kassim's main points]".
? Could you provide a transcript of this video? On other website I have seen there is a button that takes you to a transcript of a pdf file of the transcript.
? "What will I have to do": the first thing someone will click on is the boring bit. Could "Registration instructions" not lead to some of your new stuff?
? It looks like, after the Facebook and Twitter buttons, that's it. Perhaps you could add a down arrow? Everything needs to be there. Encourage them down the page!
☹ "Help us improve research in rare disease" doesn't ask "are you living with a rare condition?" Why am I important?
☹ Rather than "help us improve", say "join to improve medical research... working together for better care".
☹ There are issues with the consistency of typography: Rare diseases, or rare diseases? RUDY or Rudy? The RUDY study, or RUDY study? Study, or study? These things need to be the same throughout.
☹ There are a couple of boxes where the writing is small, for no reason ("secondary objectives" and "benefits", this seems like important information).
☹ "Patient forum": bad grammar on the bullet points. Editing this should make it more concise and clear.
☹ "Which diseases..." doesn't list the diseases.
☹ What is a "phenotype"? What are "functional outcomes"? This is jargon and I don't know what it means.
☹ There are words highlighted in bold that are not clickable, explained, described, or defined. They seem important—are they?

Other:

? When you Google RUDY the search result description doesn't mention rare disease + the blurb is academic which is a problem for someone looking up RUDY who is not a researcher. As a patient I do not feel it is aimed towards me, so use the short description as a whole statement, e.g. Rudy, a study for rare disease, is patients and researchers working together for better care.

Table 32 shows extracts from the feedback provided by RUDY's project lead on the website changes. Table 33 shows extracts from the feedback provided by RUDY's project lead on the changes to the reminder email.

Table 32: Final edits agreed for changes to the website.

Issue	Suggested changes	PL's recommendation
It is not clear that RUDY is a research project.	FROM: "Help us improve research in rare diseases" TO: "Patients and researchers working together for better rare disease care"	Tweak to "Individuals and researchers working together for better rare disease care" – not all people with rare diseases see themselves as 'patients'
The patient-centric nature of RUDY is not clear.	FROM "Sign up today" TO "Join us now" FROM: "More info." TO "Find out more"	Agree

Table 33: Final edits agreed for changes to the reminder email.

Draft email	PL's recommendation
Dear Test User Thank you for contributing to the RUDY Study, you are one of [x] people taking part! [I would insert a space and separate these sections] It is now 12 months since you first completed questionnaires for us, and we would be very grateful if you could complete them again, to help us track your health and wellbeing. Please log in to http://www.RUDYstudy.org, where you should find all the questionnaires, ready to be completed again. By completing these questionnaires, you are helping improve understanding of rare diseases. ~~Thank you for your participation, completion of~~ The information you provide by filling in questionnaires such as	Agree make this two sentences. "Thank you for contributing to the RUDY Study. You are one of [x] people taking part from for the group of rare [disease] conditions" Agree

the Adl questionnaire is invaluable, so thank you for your time. The completion rate for Adl is currently at 61% for your disease group, which is [disease group].

If you have forgotten your password, you can reset it by following this link: https://research.ndorms.ox.ac.uk/RUDY/password/remind

Please get in touch if you have any difficulties and thank you in advance for helping us with this valuable research into your condition.

Best wishes,

RUDY Study Team

Email: XXXX@XXXX.ox.ac.uk

Telephone: 07XXX XXX XXX

Your RUDY ID: RD0062

RUDY: improving research in rare diseases since 2014.

The information you provide by filling in questionnaires such as the Activity of Daily Living questionnaire is invaluable, so thank you for your time. The completion rate for this questionnaire is currently at 61% for the disease group of rare [disease] conditions.

5.3.3 Enhanced feedback

Analysis of data collected throughout the co-design process with participants provided ideas for improving the feedback RUDY offered research participants. The co-design process involving the RUDY team led to the development of an enhanced feedback intervention. Enhanced feedback communicates project aims, the type of data being collected for research purposes, how that data is used, value of participant contributions, and research outcomes. Interview findings (Section 4.3) showed that RUDY participants did not know about research findings or project updates, indicating that there was information they would quite like to know. Focus-group participants reported that they wanted a more positive experience. One participant wanted to be able to report positive experiences, and several people reported their research involvement as a negative experience:

> "Life goes on but the forms emphasise the negative aspect", P11.

The task of filling out questionnaires was described by another participant as:

> "Ticking boxes is brain dead, it's all negative about pain and stuff... I have positives in my life", P1.

Table 34 shows that some participants described research participation as offering a sense of purpose:

"When you have a long-term health condition [it impacts work negatively]. [Taking part in research] Feeling of contribution to society. A positive role", P6.

Table 34: Focus group responses showing a preference for positive reinforcement.

Call prompt	Participant responses
When I think of RUDY I think of...	"My condition. A lot of the time you don't think of it. Numerous forms. A lot of forms to fill in. I would rather spend that hour and get it all done. Filling in forms can be a negative. Pain scale off the chart. Life goes on but the forms emphasise the negative aspect", P11.
On the website I use...	"Ticking boxes is brain dead, it's all negative about pain and stuff – I have positives in my life", P1. "Participation. Involvement. Thinking of how this will help in the future. Positive. Something… taking responsibility – questioning – empowering", P4. "Negative form filling but I can compare results over time. Also, I feel positive. When you have a long-term health condition [it impacts work negatively]. [Taking part in research] Feeling of contribution to society. A positive role. Often work for ourselves because we can't be reliable", P6.
I would like the RUDY website to have…	"Nice to have positive information on how we have helped. How many of us are on the study? Bit more personal. Log in, it doesn't say it's you. "Hi name" + state condition. Positive outcomes and how research is being used", P6. "You do fill out the form. Nice to see positive information. I have charts, but as a community it would be nice to know what they are taking out of the study. Even if the research is being done now, tell us that's being done", P11.
When filling out questionnaires…	"I get positives out of having my condition – the nurses at the hospital are lovely, I meet nice people in hospital. There are privileges people with my condition enjoy but I can't describe them anywhere. What would be hugely helpful is if RUDY could tell me why the research uses this data", [P1].
Next steps...	"If there was more info about research and what's happening with my data. A positive that something is happening. I don't mind a text once a month. A general email. If I am not up for looking at the website, I won't. Email with the questionnaire could say "also, while you're on there, check out…", [P11].

Another participant suggested that there was a more general desire for positive feedback across the community of people with rare conditions:

> "As a community it would be nice to know what they are taking out of the study. Even if the research is being done now, tell us that's being done", P11.

Participants expressed a desire for positivity within their research participation. They wanted to receive more good news stories and to be able to report positive experiences as well as the negative ones they were asked to report. RUDY collated and shared more data after the focus groups had finished. They shared graphs with me that compared responses among three health questionnaires: EQ5D (quality of life), PainDetect (measures pain) and HAD (anxiety and depression). At this point in time (2nd July 2020), RUDY had 2,727 participants and 30,963 completed questionnaires.

Figure 26 shows first-time questionnaire submissions, by year. Over time, completion rates for EQ5D and PainDetect seem to drop more than HADS, where numbers remain more consistent. Figure 27 shows completion of questionnaires after six months. The drop in completion rates for HADS are markedly lower than for EQ5D or PainDetect.

Figure 26: Graph showing first questionnaire completion by year of consent (%).

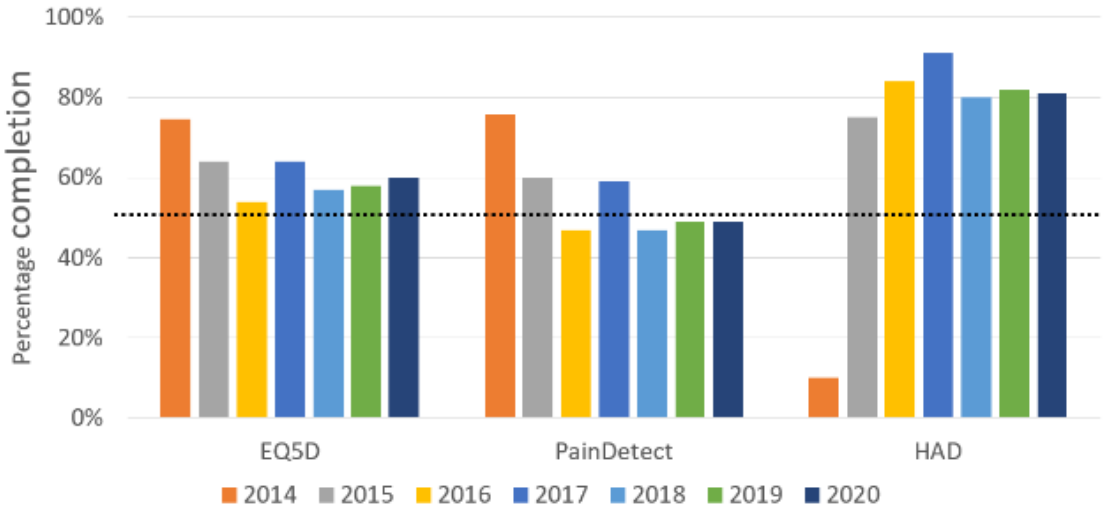
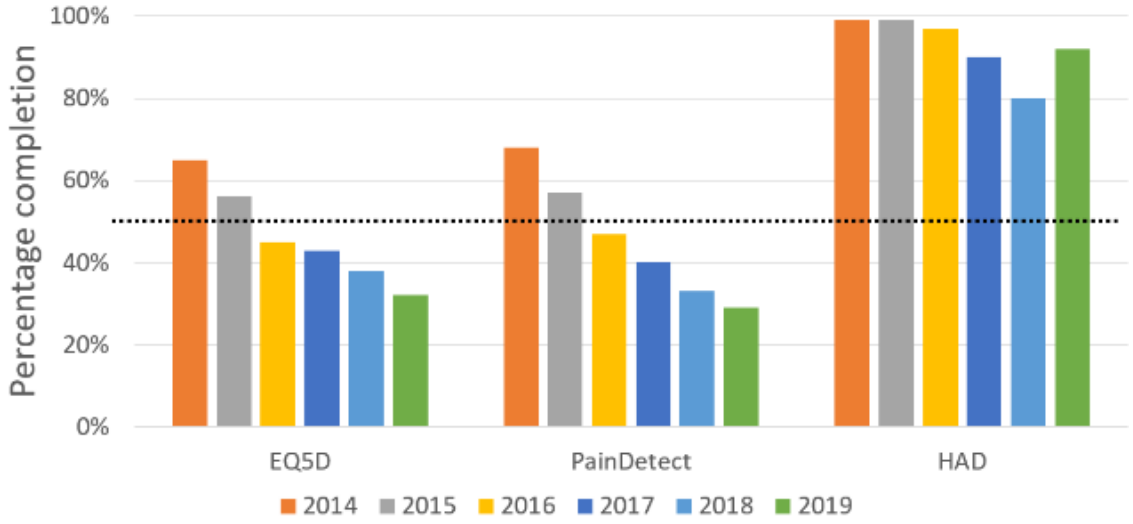


Figure 27: Graph showing subsequent questionnaire completion by year of consent (%).



The next three figures also suggest that completion rates over time drop more for EQ5D and PainDetect than they do for HADS. One reason for this, which is more of a heuristic measure informed by participant feedback during the focus groups than a rigorous hypothesis, is that the HADS questionnaire uses positive language, asking questions such as "when was the last time you laughed?". EQ5D and PainDetect focus exclusively on pain and the inability to complete daily tasks.

Evidence from the focus groups suggests that framing and communication might affect how people respond to the reminder email RUDY sends out. Participants described the questionnaires they had to fill out as boring and negative, expressing a desire to record positive aspects of their conditions. In the following graphs, where the title indicates "completion rates by issue", this means the number of times a questionnaire has been issued to a participant. E. g., in Figure 28 the data point at issue 4 is the completion rate for the fourth time someone has been asked to submit an EQ5D questionnaire. Figures 28 to 30 show completion over time.

Figure 28: Graph showing EQ5D completion rates, by issue.

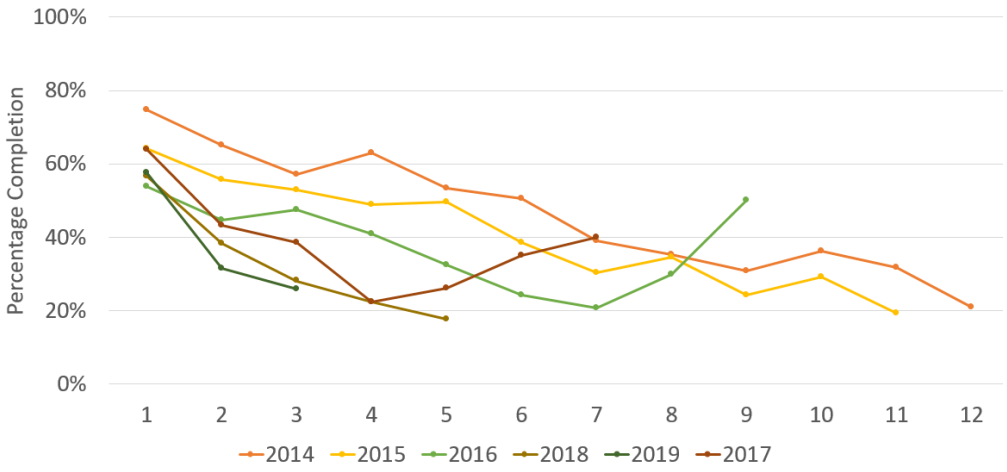


Figure 29: Graph showing PainDetect completion rates, by issue.

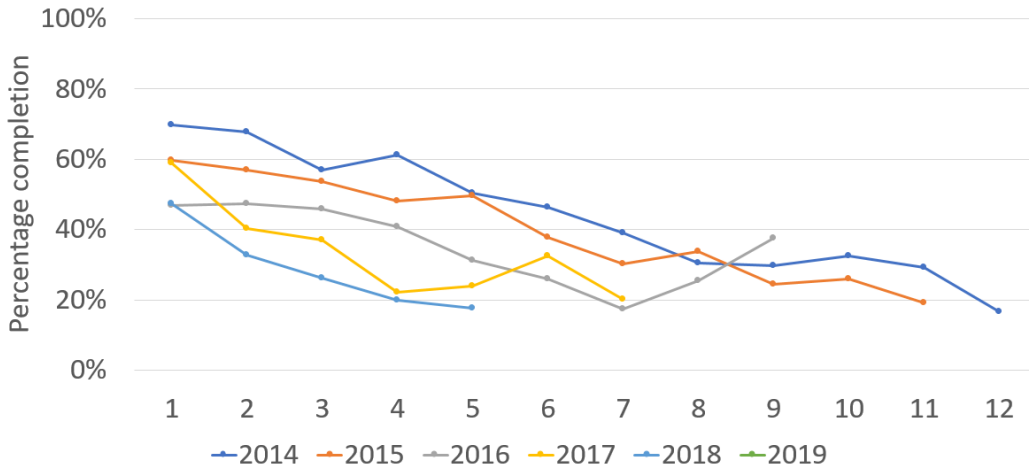
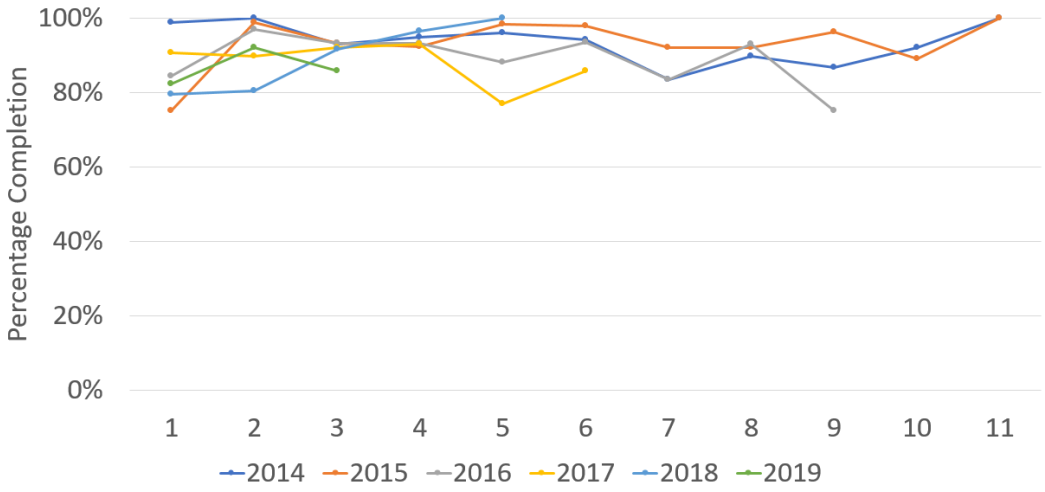


Figure 30: Graph showing HAD completion rates, by issue.



5.3.4 The value of co-design for PPI

Researchers actively resist sharing power with participants (Boaz *et al*, 2016). While patient and participant involvement (PPI) is 'good to have', it is not essential to research operation. However, participants can generate ideas and guide research (NHS, 2009). Requirements engineering literature offers one explanation as to why participants should be actively involved in developing research processes –their experiences provide information about the challenges and issues they face (Ambreen *et al*, 2018). The work described in this chapter justifies assertions from the research team in Section 4.3.2 that PPI is mission-critical for RUDY. One example that emerged from the focus groups was how participants had considered mental health as a factor worth investigating, which researchers had not thought of:

> "We as patients had come up with an idea that the researchers had not even thought of [a mental health study]. Patients thought this should be looked at… but we haven't heard anything since that meeting. I've forgotten when it was, we were told we had the opportunity to contribute towards writing a research paper. We submitted bits and pieces. You come away thinking "gosh we're making a difference" but we have heard nothing. We do as much as we can…", P4.

Also highlighted in this extract is an opportunity-cost. This participant never received feedback validating the importance of the information they had provided. The opportunity to engage with participants in this way is invaluable, leading to impactful research because it is relevant to those it describes, and serves. As a longitudinal study, RUDY must think about how participants are encouraged to feel involved[30], otherwise research participation become less of a priority. The account below is one of many from this chapter, demonstrating a need for feedback:

> "There is a willing band of volunteers around. They need to know what they can bring to the table that brings purpose or value. I have chosen not to prioritise RUDY before because I would be valued more elsewhere. When you have limited resources, RUDY needs to be a priority for you to spend those resources on it. I need to feel that I'm doing something useful here", P3.

The study described in this chapter made use of PPI, providing evidence to support the assertion that involving different stakeholders in software development leads to

[30] Or "drawn in". Felt involvement is a facet of engagement (O'Brien & Toms, 2010).

outcomes accommodating the interests of those stakeholders. Consulting participants led to insights that I, as a researcher, would not have been able to synthesise. Collaboration with the RUDY team meant that participant suggestions could be ranked by how desirable they were as a feature of RUDY and how feasible they would be to implement. Participants validated these changes further during user tests as they navigated the "improved" website. This work identified that researcher and participant needs needed to be negotiated, and highlighted trade-offs between the two. For example, participants wanted advice about care and forums for discussion and to meet others with rare conditions. Both of these requests lay outside RUDY's scope as a clinical research project. Requests for project-related information however, were easy to implement because it was generated as part of project design, and simply needed to be translated into more accessible language.

Co-design also demonstrates the value of Dynamic Consent as an interface through which participants can be linked to a "wealth of information that already exists for other purposes, such as lay descriptions" (Kaye *et al*, 2015). One participant suggested that in addition to research results, they wanted to see project-specific information:

> "Access to funding submissions… periodic reviews would be helpful", P1.

During a user test, another participant asked:

> "Could you provide a transcript of this video? On other websites I have seen there is a button that takes you to a transcript of a pdf file of the transcript", P4.

Kaye *et al* describe "hyperlinks to… alternative forms of presentation [which] may [overcome] consent form length and comprehension". P4 has spent time online, encountering tools that have helped them navigate digital spaces more effectively. This kind of feedback could be helpful to researchers; participants want to engage and may simply lack the necessary tools. Autonomy is especially important in communities with communication issues such as hearing loss, sight loss and reading difficulties, and participants know their value:

> "I have something to add", P3.

> "I feel like the team values us", P5.

This study demonstrates that RUDY values engagement enough to spend resources on it, and that participant input can be used to develop participant-relevant research processes. What is not clear, from this qualitative work, is whether such development effort is justified. This is why a quantitative intervention study is crucial. Both kinds of

empirical evidence are required to draw any conclusions about how useful Dynamic Consent is to research (Prictor *et al*, 2020b).

When asked directly, focus-group participants suggested that they would like to receive more information about how the data they provide is used for research purposes:

> "What would be hugely helpful is if RUDY could tell me why the research uses this data. What are the outcomes?", P1.

While researchers are cautioned that overwhelming participants with data acts as a barrier to engagement (Bester *et al*, 2016; O'Brien and Toms, 2008), there is room for communication that does not underestimate the intelligence of the public:

> "A little more information about using research - how information is used, what it's used to do and why those things are done – and how things are improved because of it. There's joy in finding out new things", P5.

There were some ideas as to how RUDY could individualise the participant experience, and the overarching theme of this discussion was that participants wanted, but did not receive, personalised feedback:

> "When you log in, it doesn't say 'Hi [name]' or state my condition", P6.
>
> "We know what we are doing, we just don't know why. I don't know what I'm bringing. I thought "maybe I should back out [of RUDY]" and would like some clarification about what my participation means and why", P3.
>
> "I would like to them to update us on what's going on. I want feedback on what they are doing with the data. Some feedback on how data is being used – even if vague. Just so you know it's worth your while. Numbers of people signed up or who have done surveys or have been helped in some way – so you can see your part of the bigger picture" P4.

Online focus groups examined issues with engagement within RUDY from a participant perspective. The website did not seem to be a useful resource, as two participants describe:

> "Dr Google is there. RUDY is not a resource for me", P1.
>
> "I am looking at it as a website getting information from me, I don't look at it as something I would get something back from", P5.

Participants described their experience of using the website as marginal:

> "I only use the website when I've been prompted that there are new questionnaires", P8.

> "[I] only go when prompted", P11.

> "I only use RUDY website for ticking the boxes", P2.

When participants went to the website, it was usually after they had received an email prompting them to fill in questionnaires. To access questionnaires, participants had to visit the website and log in to the online portal. One participant was aware of research outputs:

> "[I] look at results. To library to see if anything has been published", P13.

Participants knew what information they wanted RUDY to report:

> "I don't need information on the disease or health stuff, need much more information on why the data is important so it's more about how the research happens and why its adding value", P2.

The focus groups also provided insight into participant experiences of filling out questionnaires. Participants described the experience as repetitive, negative and time-intensive:

> "Tick boring boxes", P1.

> "Always more than happy to fill forms in, but it is repetitive", P8.

> "Filling in forms can be a negative. Pain scale off the chart. Life goes on but the forms emphasise the negative aspect", P11.

> "I bookmark the RUDY page. Don't do it all in one sitting", P3.

One participant also showed frustration that questionnaires did not allow self-expression:

> "In the questionnaires you are not always able to put a tick where you want to. A sliding scale might be more indicative. Is there an area where you could tick two boxes and add a qualifying comment? I just go with it. Online surveys are… not nuanced", P3.

This highlights an issue that the RUDY team faces in digitising validated questionnaires. The questionnaires they make available are taken from academic literature, which ensures that the data collected is rigorous and comparable to other data. Rigid research methods are in direct opposition to participant-friendly methods. To counterbalance the burden of filling in complex and largely negative questionnaires, RUDY has created tools

aimed solely at participants. This includes a timeline that compiles all completed questionnaires, entirely for the benefit of participants. The timeline is cited as a benefit:

> "If I was trying to recruit [people to RUDY], the only thing I could say to them at the moment would be that information is recorded in one place", P5.

RUDY participants are happy to delegate the protection of their data, and future decisions about data-use to the research team, who they consider to be experts (Nissen *et al*, 2019). However, informed consent is not informed if participants do not know how their data is being used, or the value of their contributions. Delegating choice fractures the relationship between a participant and their role in research:

> "[I am] Really interested to see what they make of all the answers. Just don't connect to it. Quantitative research has no nuance. Answers are not possible. Doubtful of their usefulness", P1.

> "I agree with P1; nuance is not in the questionnaire. No shades of meaning… Why do they want us to tick boxes? It's a black hole. I am happy to do it but I have absolutely no idea why. I trust they are doing it for a purpose but it would be good to know. Something like 'these questionnaires may seem the same, but the data we collect is… and we use it to…'", P4.

One participant chose to focus blocks of their time on RUDY questionnaires, which suggests that some participants compartmentalise their research activity:

> "Filling in the questionnaire, if I have a couple of hours spare I do it. I don't mind the time but I have to set time aside for it. Can't just do it for 10 minutes", P5.

This corroborates findings from Chapter 4, that some participants consider taking part in research as a form of community service. In putting time aside to fill in RUDY questionnaires, the participant fulfils their duty to the rare condition community, and society in general:

> "Use site when prompted. Not thought about it. Sit there – all in one go. An hour or whatever", P8.

> "Numerous forms. A lot of forms to fill in. I would rather spend that hour and get it all done", P11.

It follows then, that RUDY is not at the forefront of a participant's mind outside of this allotted time. Where someone might engage with a banking application regularly, a RUDY participant who fills in questionnaires when prompted will log into the online

portal twice a year. Analysis of data reiterated that basic information about RUDY is not retained over time – more effective information strategies are needed (Griffin *et al*, 2006). This chapter provides such a strategy, reminding participants of the value of their contribution, as well as RUDY's overall vision, goals and achievements.

One online focus group consisted of patient forum members. The patient forum is a group of participants that RUDY researchers consult for feedback about the project in general, and new features. These participants are a particularly engaged subset of the wider RUDY population but even they expressed a lack of clarity around the research process, as indicated by these participants:

> "I don't know what the data is for. *My history is on there. If someone (a doctor) might want to access it, it's there. Would a consultant have permission?", P5.
>
> "[RUDY] Collects data on rare conditions. Resource for patients as well. Whoever can access it", P6.

Participants talked about a meeting that they had all attended. There had been a patient forum meeting to provide a project update, and an opportunity for RUDY's project lead to consult participants:

> "We as patients had come up with an idea that the researchers had not even thought of – a mental health study. Patients thought this should be looked at… but we haven't heard anything since that meeting. I've forgotten when it was, we were told we had the opportunity to contribute towards writing a research paper. We submitted bits and pieces. You come away thinking "gosh we're making a difference" but we have heard nothing. We do as much as we can…", P4.
>
> "After a forum meeting I asked [project lead] whether the meeting was useless to them, and it was not as he had a book full of notes! Talking gives you information that quantitative work doesn't. Discussion groups as part of research – isn't that what RUDY is advocating as well? A personal experience of research", P1.
>
> "After this [patient forum] meeting, the plan was… but there was no follow up… The meetings seem to be a RUDY update but I am not sure of my use. Research is slow and without an incremental reward, it is hard to stay the course. Participants may never see a benefit or the end of a study", P3.

Analysis of this data shows that even the most engaged participant group within RUDY has communication issues. One participant expressed a desire for information in order to recommend the study at a support group they attended:

> "Meeting with researchers would be very good. Or getting information from researchers. Advocates would then be able to go and advocate at support groups. Instead of the "black hole" of filling in questionnaires. There is a forum we don't really know the focus of", P4.

This indicates that participants do not necessarily require researchers to act as translators. They want information that allows them to understand their own role in the project, and will help them introduce others to research participation.

There was evidence that RUDY participation had a positive effect on people's behaviour. One participant described increased note-keeping:

> "I try and make a note when I have micro-fractures. Previously, if I didn't go to hospital or it didn't take me out of action I wouldn't record it. Now, I make a note for RUDY. I tell professionals about RUDY!", P3.

RUDY was also described as offering positive opportunities for involvement and community building:

> "Another opportunity to help", P13.

> "I tell people about RUDY… There is a Facebook page for RUDY [which has] happy things", P6.

Participants also celebrated the project because it allowed them to consolidate information about their condition:

> "I asked members of my support group about filling in questionnaires. I only had one year of paperwork when I started. For others who have earlier diagnoses or who have been living with a condition for a long time, it's time-consuming to do so. I think RUDY has changed that now – making it more user-friendly" [P4].

> "I feel like the team values us… If I was trying to recruit [people to RUDY], the only thing I could say to them at the moment would be that information is recorded in one place" [P5].

> "handy as a resource to see timeline – you don't always have access to that yourself. As a patient, don't have access to that information" [P8].

RUDY offers a novel, and entirely functional, research approach that values participant satisfaction and engagement. Participants seem to be satisfied that their participation is

of some (undisclosed) value, which reflects RUDY's ethos that key to retaining participants is that they value being part of RUDY (Javaid *et al*, 2016). However, Chapter 4 indicated that while participants wanted information, this desire was tempered by a resignation that research participation holds no personal benefit. This chapter describe a similarly jaded outlook, illustrated by one participant's compartmentalisation:

> "I want to do it all at once/one time and move on with the rest of my life", P4.

Low expectations of research involvement meant that participants did not value their data-contributions. Focus-group participants did not expect to receive any feedback from RUDY, but when asked, they wanted researchers to be clear about research aims, how participants can take part, and what participant contributions entail. This is information that most ethical applications require so, rather than placing the burden on participants to seek out information, this data should be easy to reference.

5.4 Reflection

This chapter corroborates findings from a review of data security and consent within the NHS that emphasises the importance of leadership and a motivated team (NDG, 2016). Without acceptance from RUDY's project lead, and without the rapid, agile development efforts from RUDY's development team, this intervention could not have been created. Also corroborated is the review's conclusion that:

> "The public's knowledge about how… data is collected, protected, and used… is limited. It is therefore clear that future communications cannot make any assumptions about existing knowledge of data processes and uses", (NDG, 2016).

The self-selecting focus group members were convinced of RUDY's importance and trusted the research team to use data however they saw fit, for research purposes. Analysis of the focus group data suggested that privacy was not an issue for participants – there was no evidence that participants made trade-offs between taking part, and their own privacy. In fact, a commonly held view was that RUDY offered an opportunity to learn more about themselves and the condition they live with. Patient forum members knew more about RUDY in general; they knew who the project lead was, or remembered meeting him in person. They also described attending meetings where their opinion was sought, but despite this level of involvement participants were still unaware of the value of their contributions (contributions without which, RUDY would not function).

In this chapter, we have moved from data-contributions that consist solely of filling out questionnaires, to a more substantive level of research involvement where patient group participants are providing ideas to support research direction. Pertinent to note, is that as research moves online, the type of implicit trust demonstrated in this chapter will be an ineffective way to mitigate the risk of data-harms. This means that the role of experts in designing responsible data practices will become even more important, and this will need to be driven by research principles – the RUDY team's enthusiasm in being directed by participant input, for example, was indicative of their wider emphasis on PPI.

The social contract (Section 2.3.1) must evolve alongside research practices that are becoming more "open-ended and ongoing" (Kaye *et al*, 2015). The technologies supporting these practices, such as Dynamic Consent, must be designed to accommodate participant involvement over time. Researchers must be open to feedback and criticism from participants and in return, their participant pool is committed and productive (Budin-Ljøsne *et al*, 2017). This study provides such an example; researchers certainly welcome feedback and participants are both committed and productive, but a lack of communication seems to be causing the latter to feel quite underappreciated. When asked to expand on this point, three demands were made of researchers: clarity, precision and conviction. This work shows that participants are not used to being asked what they want, but when prompted, they can provide a litany of information requirements. What may be the case is that participants do not know what to ask for because they do not know what is possible.

Dynamic Consent literature describes how active participation, rather than the more "inactive" participation reported in focus groups can only be facilitated by researchers. Participants cannot, and should not be expected to design research methodologies. A RUDY participant might only access the portal twice a year, so the research team must make this space easy to navigate as decide how information about the project will be communicated. Analysis of data in this chapter once again emphasises the two-way nature of research. Research participation was peripheral for participants, i.e., a task that was not done regularly or prioritised in daily life. When participants sat down to fill out questionnaires, they relied on information that was immediately available, such as the reminder email they had received, prompts from the portal when they logged in, and their own assumptions about research as a whole. Responsibility lies with those who ask for consent to provide project updates and signpost how data is being used. Analysis of focus group data suggested that this information was not being communicated.

In a departure from theories like the 'privacy paradox' which claim users are uninterested in their personal data, this chapter posits that the results of data-use (within RUDY) and sharing (with authorised collaborators) should be communicated to data-subjects

because they want to know how data is being used. A RUDY participant might not want to understand how research is implemented or receive feedback as work progresses, but they may want a lay summary describing what their data-contribution has helped achieve.

This chapter has described the co-design process for an intervention study investigating the research value of participant engagement. This process involved online focus groups with RUDY participants, resulting in a proposal for enhancing feedback within the study. The communication requirements gathered from these focus groups were used to drive a software development cycle with the RUDY development team, enhancing the study's feedback mechanisms. User tests were carried out with focus group attendees and people who had not attended the calls, to ensure the changes matched the proposal and met the original requirements. I worked with the RUDY team to make changes to the reminder email, website and online portal, directed by participant suggestions. These changes were validated through user tests with focus group members. This increased confidence that the changes made were relevant and useful to both participants and researchers.

There are limitations to this work. First, its small sample size (15); while indicative, these findings can be strengthened by further work. Second, I was conscious of over-representation of white and middle-aged participants. While these categorisations seem to reflect the RUDY population, it is not necessarily representative of the wider population of people with rare conditions. Rare genetic conditions affect people, regardless of their age, ethnicity or socio-economic status. Finally, online focus groups and user tests meant that interactions were not as rich as they could have been in person. Online methods can also serve as a barrier to participation, excluding people with additional accessibility and communication requirements, or simply the confidence to use digital technologies.

The key finding from this chapter was criteria for enhanced feedback: researchers must describe their project aims and the data that they hope to collect, they must say how this data is to be used, state the value of participant contributions, and make sure to report research outcomes in a way that participants can understand. The impact of this enhanced feedback intervention is described in the next chapter.

Chapter 6

Study 3: enhanced feedback intervention.

"Unlike carrier pigeons, information does not automatically seek out relevant audiences", O'Neill, 2014.

6.1 Introduction

Members of the public can engage, disengage and re-engage with research projects. I argue that the burden of communication lies with researchers to communicate their work clearly, concisely and regularly, rather than on participants to understand dense academic terms. The conversation between a research participant and the study they are taking part in starts during the informed consent process, so informed consent needs to be structured in a way that keeps participants updated over time. Regulatory oversight demands that participants should be informed, and evidence from Chapters 4 and 5 indicate that participants want to be informed. This chapter describes an intervention study designed to measure the effect of enhancing information feedback on research participation. Evidence from this chapter will suggest that providing information about RUDY in an accessible way increases overall participation and reduces drop-off in

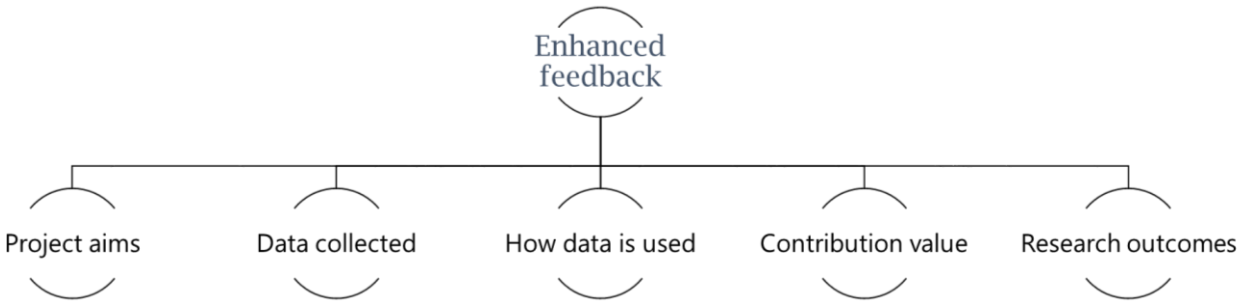
participation over time. This is a valuable finding that positions this work as a signpost towards good practice for researchers working with human participants.

Participant input can contribute to the research process, but it is only as useful as researchers allow it to be. Previous chapters identified that RUDY has a process in place to seek participant input (the patient forum), but project-level feedback needed improving. Participant accounts indicated that the research conversation went only one way: from them to the study. Chapter 5 described how participant experiences were gathered to engineer communication requirements for a communication intervention. This chapter describes the impact of that intervention. This work provides an empirical justification for the value of data practices that provide feedback about how data is used; responsive data-practice. This is part of a wider piece on engagement within online longitudinal studies, where data-practices should be inclusive (seek input) and responsive. There are many descriptions of participant engagement; for the purposes of this work a research study is engaging if participants are interested in the study, there is knowledge-transfer, and there are opportunities for active or further participation. RUDY participants are interested in the study and highly motivated to take part, and there are opportunities to participate. What is missing is the transfer of knowledge. This work demonstrates that researchers can use responsive data-practices to transfer knowledge, and to inform participants over time.

6.2 Methods

Changes were made to RUDY's website, reminder email and online portal, enhancing the feedback available to participants (Figure 31). The hypothesis for this study was that enhanced feedback would positively affect research participation, and this hypothesis was accepted. Keeping participants informed is important because people's recollections fade over time, of what they have agreed to do (Joan M. Griffin *et al*, 2006). Informing participants over time is especially important in a project like RUDY where six months will pass between participant submissions. This intervention was designed to make simple messages available on the website and to include key takeaways in the email reminder.

Figure 31: Modelling enhanced feedback.



This study measured the number of questionnaires submitted to RUDY over time. Data was collected by the RUDY study, gathered from RUDY participants whose record showed consent to share data for research use. A non-randomised, interval control group, pre-test, post-test experiment design was employed. The experiment was designed as an interrupted time-series (Figure 32) of data-samples collected over time.

Figure 32: Time-series interval data collection (adapted from Field & Hole, 2013)

The dependent variable was research participation, the metric for which was "completion rate". Completion rate was measured as the number of questionnaires submitted as a percentage of the total number of questionnaires assigned to participants. The independent variable was the enhanced feedback intervention that improved the quality of information relayed to RUDY participants. As described above and elaborated upon in Chapter 5, this enhancement meant making changes to the project's website, email reminders and online portal. Making these changes involved re-organisation of data rather than creating new content.

The participants under study were the people with rare genetic conditions, filling out and returning questionnaires via RUDY's online portal. The sample data provided by RUDY did not include demographic information, but a breakdown of participant age, ethnicity and recruitment method is provided in Section 6.3. The intervention was implemented, or 'pushed to production', on 24 September 2020. Figure 33 shows the data collection intervals. Pre-intervention intervals were from 24 September to 31 December 2019[31], and from 1 July to 23 September 2020[32]. Post-intervention data was collected from 24 September to 31 December 2020. Another interval was added to explore changes in submission due to the impact of a national lockdown caused by the COVID-19 pandemic: 1 July to 23 September 2019.

[31] Comparing post-intervention patterns to the year before.

[32] Comparing post-intervention patterns to earlier that year.

Figure 33: Illustration of pre- and post-intervention control groups.

Pre-intervention	Pre-intervention	Pre-intervention	INTERVENTION	Post-intervention
01 July – 23 Sept 2019	24 Sept – 17 Dec 2019	01 July – 23 Sept 2020		24 Sept – 17 Dec 2020

6.2.1.1 Sample data

RUDY provided sample data indicating low submission rates, which helped to justify a need for this study to RUDY's data-access panel. This panel entertains applications from external parties seeking access to RUDY data, usually research collaborators. I was provided with an initial dataset of questionnaire submissions between 1st June and 31st December 2019, for all adult participants consenting to information use (n=2352):

> "I agree that the anonymised data that is collected about me during the study may be looked at by both national and international academic researchers approved by the RUDY Data Access Committee that contribute to the aims and objectives of RUDY. I permit these individuals access to my research records"[33].

For each participant record in the sample their date of consent was provided, as well as dates that they had been assigned, had started, or had submitted questionnaires. Figure 34 shows the breakdown of participants in this sample by the year they consented to take part in RUDY. The two most recent cohorts, 2018 and 2019 represent over two thirds of this sample.

RUDY's project lead suggested that I explore "old" (consenting before June 2019) and "new" (consenting after June 2019) completion rates. Table 38 compares old and new participant completion rates for 12 questionnaires for the ADL questionnaire, 1603 were assigned and 636 submitted, giving a total completion rate of 39.7%. 63.2% of new participants asked to submit an ADL submitted one, and 29.4% of old participants asked to fill out an ADL submitted one. I could not reliably conclude that the earlier a participant had consented to take part in RUDY, the lower their completion rate was likely to be.

[33] Taken from the consent options available on RUDY's test website, where changes are published before being made widely available. It was inappropriate for me to create an account on the live RUDY platform to access this information.

Figure 34: sample data participant consent cohorts (n=2,352).

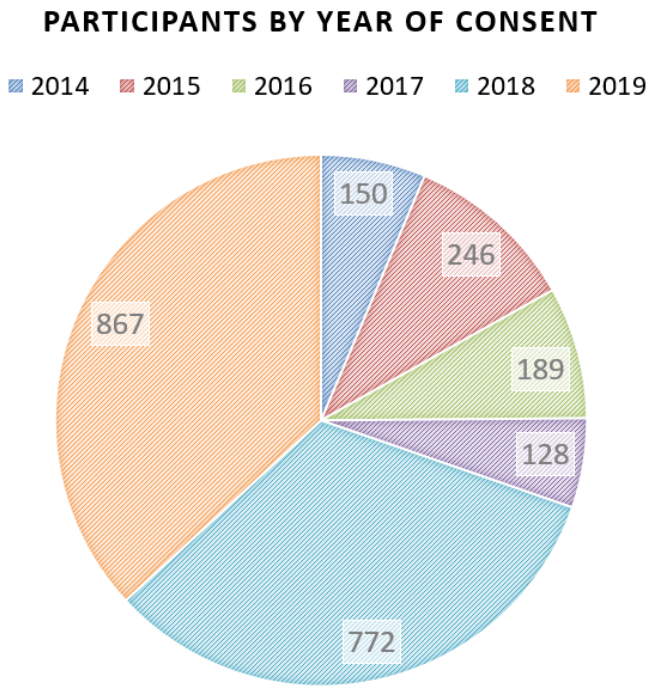


Table 35: Completion rates compared by pre- and post-June 2019 cohorts.

	ADL	EQ5D	PSQI	MCGILL	SF36	PAINDETECT
All	39.7%	38.2%	33.6%	34.5%	36.7%	33.7%
New	63.2%	60.6%	50.3%	52.4%	57.5%	50.3%
Old	29.4%	28.4%	26.3%	26.7%	27.6%	26.5%

	FACIT	HADS	EPWORTH	FATIGUE	WALK 12	OTHER
All	34.9%	33.4%	34.1%	35.4%	41.9%	24.7%
New	52.6%	50.5%	50.9%	54.4%	68.0%	39.1%
Old	27.2%	25.9%	26.8%	27.2%	30.5%	18.4%

Table 39 stratifies completion rates by consent cohort, showing that cohort 2016 had the lowest completion rates, rather than cohort 2014. Cohort 2019, the most recent, had the highest submission rates. This may be due to overrepresentation in the sample data, however. Light shading indicates lower cohort completion rates and darker shading indicates higher lower cohort completion rates. For the ADL questionnaire, cohort 2019 demonstrated the highest response rate and cohort 2016 the lowest. Analysis of this data suggests that response rates drop over time, but does not show that length of participation is any guarantee of performance.

Table 36: Completion rates by consent cohort; response rates tend to drop over time.

	ADL			EQ5D			PSQI		
	Assigned	Submitted		Assigned	Submitted		Assigned	Submitted	
ALL	1603	636	39.7%	1603	612	38.2%	1603	539	33.6%
2014	126	36	28.6%	126	36	28.6%	126	34	27.0%
2015	154	39	25.3%	154	37	24.0%	154	35	22.7%
2016	93	19	20.4%	93	18	19.4%	93	16	17.2%
2017	70	19	27.1%	70	17	24.3%	70	16	22.9%
2018	499	161	32.3%	499	155	31.1%	499	143	28.7%
2019	661	362	54.8%	661	349	52.8%	661	295	44.6%

	MCGILL			SF36			PAINDETECT		
	Assigned	Submitted		Assigned	Submitted		Assigned	Submitted	
ALL	1603	553	34.5%	1603	588	36.7%	1603	541	33.7%
2014	126	34	27.0%	126	35	27.8%	126	33	26.2%
2015	154	34	22.1%	154	36	23.4%	154	34	22.1%
2016	93	16	17.2%	93	16	17.2%	93	16	17.2%
2017	70	17	24.3%	70	17	24.3%	70	17	24.3%
2018	499	146	29.3%	499	151	30.3%	499	145	29.1%
2019	661	306	46.3%	661	333	50.4%	661	296	44.8%

	FACIT			HADS			EPWORTH		
	Assigned	Submitted		Assigned	Submitted		Assigned	Submitted	
ALL	1603	559	34.9%	1603	535	33.4%	1603	547	34.1%
2014	126	35	27.8%	126	32	25.4%	126	33	26.2%
2015	154	35	22.7%	154	34	22.1%	154	35	22.7%
2016	93	17	18.3%	93	16	17.2%	93	17	18.3%
2017	70	17	24.3%	70	15	21.4%	70	18	25.7%
2018	499	147	29.5%	499	143	28.7%	499	146	29.3%
2019	661	308	46.6%	661	295	44.6%	661	298	45.1%

	FATIGUE SCALE			WALK 12			OTHER		
	Assigned	Submitted		Assigned	Submitted		Assigned	Submitted	
ALL	1603	568	35.4%	1603	671	41.9%	1689	417	24.7%
2014	126	35	27.8%	126	39	31.0%	130	17	13.1%
2015	154	35	22.7%	154	40	26.0%	159	20	12.6%
2016	93	17	18.3%	93	20	21.5%	105	14	13.3%
2017	70	17	24.3%	70	20	28.6%	84	17	20.2%
2018	499	148	29.7%	499	165	33.1%	517	106	20.5%
2019	661	316	47.8%	661	387	58.5%	694	243	35.0%

6.2.1.2 Enhanced feedback intervention

This section describes the changes that were made to RUDY as part of the enhanced feedback intervention. These changes were published online on September 23rd 2020. Figure 35 shows RUDY's project website before and after the changes were made, which included bringing the number of participants into a more prominent position and re-phrasing the project's aim so it was easier to understand. Figure 36 shows the information pages created to communicate important information, e.g., project aims, participation opportunities and current work.

Project aims: researchers should communicate what they are trying to do with their work.

A video, delivered by a RUDY's project lead, outlining the project aims was put on the website. The project aims were re-formatted to make them clearer and easier to read. Header levels were edited for consistent formatting, and technical terms like "phenotype" were translated into more accessible terms ("different phenotypes" was changed to "differences between individuals with the same rare disease"). A section was added to describe the different genetic conditions RUDY is interested in, and RUDY's aims were added to the reminder email.

Data collected: researchers should say what kind of data they are collecting from participants.

RUDY had general descriptions of the kind of information they wanted (e.g., questionnaires). These were added to the patient-accessible questionnaires and edited to clarify what kind of data the questionnaire was designed to collect. Participants saw these descriptions as they opened each questionnaire.

How data is used: researchers should describe how they use the data they are collecting, who it is shared with, and the access-request process.

A video was added, describing how questionnaire data was used. A section labelled "Who we are" was added to give human points of contact for the study. Participants could see the people responsible for safeguarding their data. The questionnaire descriptions also included an example of how the data that was being collected could be used by researchers.

Value of contribution: researchers should emphasise how valuable it is to the project that people take part.

The most prominent change made to the website's home page was a re-phrasing of their aim from "Welcome to RUDY, help us improve research in rare diseases" to "Individuals

and researchers working together for better rare disease care". This emphasised the role of participants in the study. The number of participants was made bigger and put at the top of the page. A page was added that described the patient forum and what participation might entail, emphasising its value ("patient forum members are making a real difference to RUDY and we value their contribution").

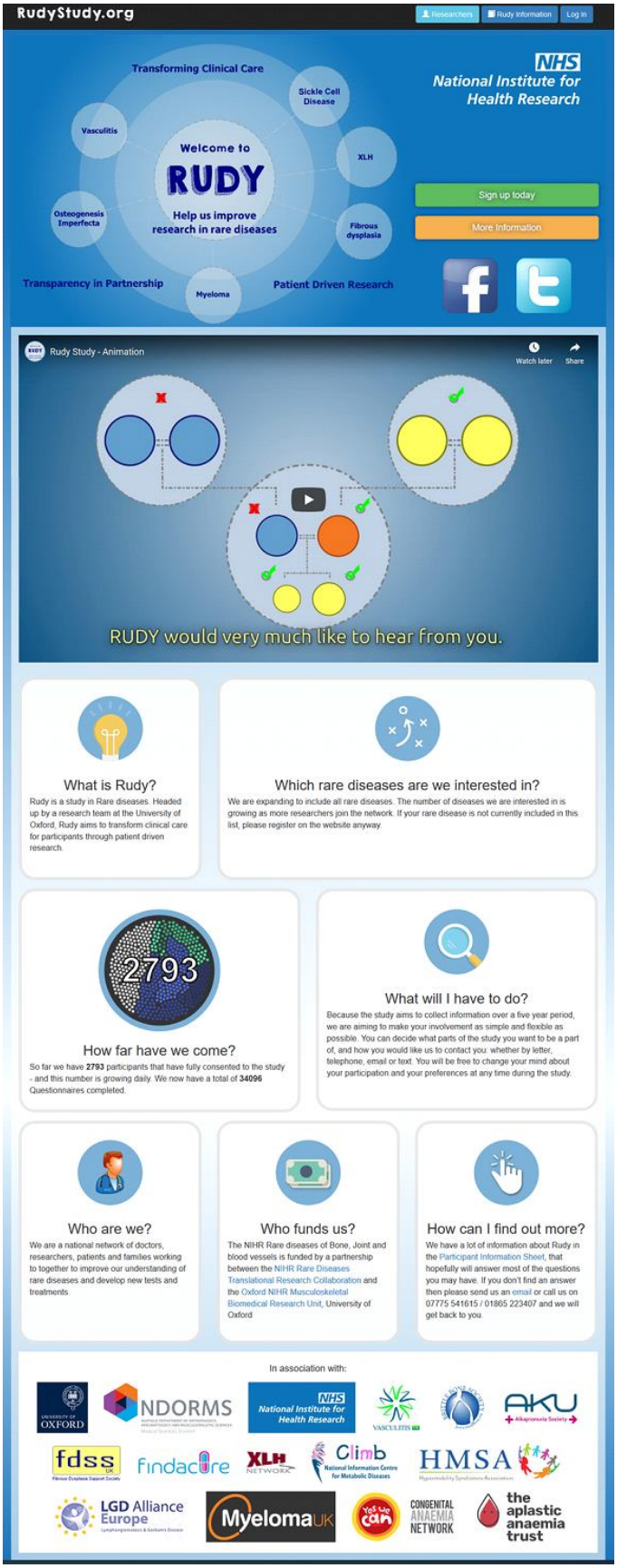
The remainder email was also re-phrased to thank participants for their time and demonstrate the value of their contributions. Information was provided designed to situate individuals within the wider context of the study: "you are one of [x] people taking part for disease [y]". The participant's role within the study was also reiterated: "by taking part, you are helping improve understanding of rare disease".

Research outcomes: researchers should communicate what they have found and what that means.

A "Project news" page was added, that included the number of participants and completed questionnaires. As of November 2020, there were two videos on this page: a collaborator working on Osteogenesis Imperfecta, and a RUDY collaboration related to Fibrous Dysplasia. This page has titles, lay descriptions and citations for five reseach papers that RUDY data has contributed to. Information was also included in the reminder email: "Completion rate for this questionnaire is currently [x]% for disease group [y]".

Figure 35 illustrate the changes made to RUDY's website. On the left is a screen capture of the website before any changes were made, and on the right is a capture of the post-intervention curation. Figure 36 showcases the different information pages that were created to communicate different aspects of the study. Some of the information had already been available, but feedback from participants shaped the curation of pages and available wording.

Figure 35: RUDY website pre- (left) and post-intervention (right).



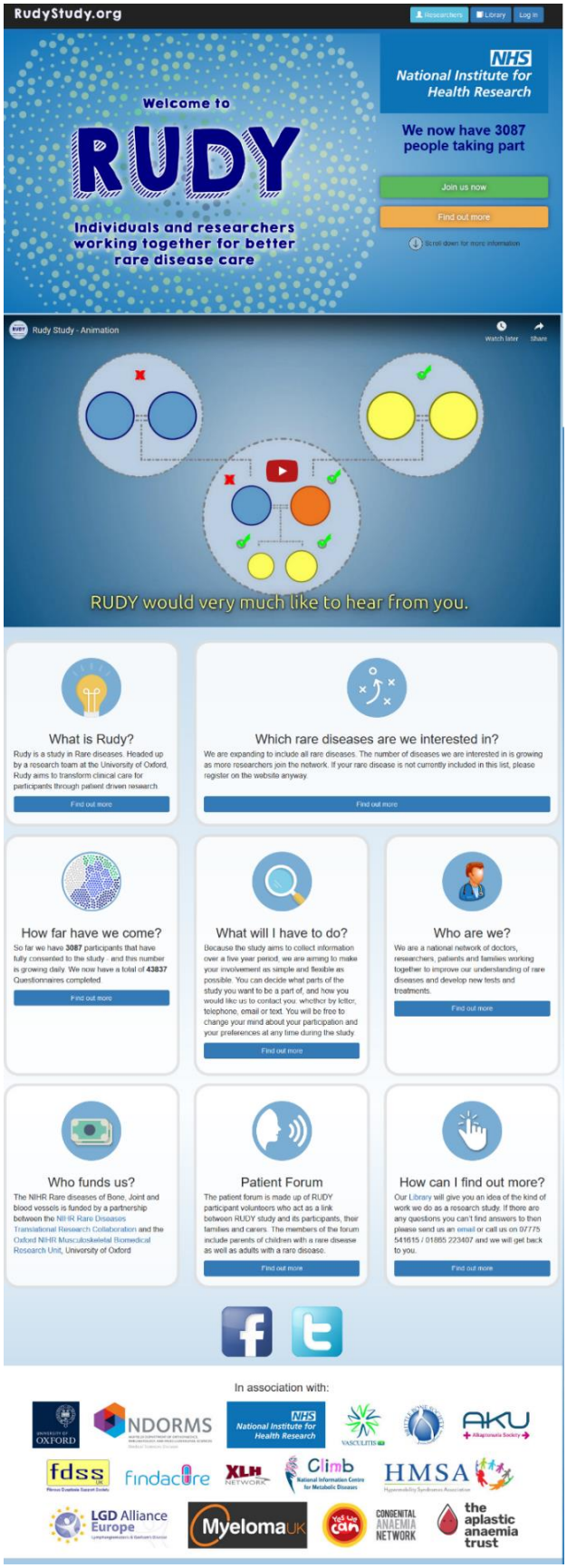
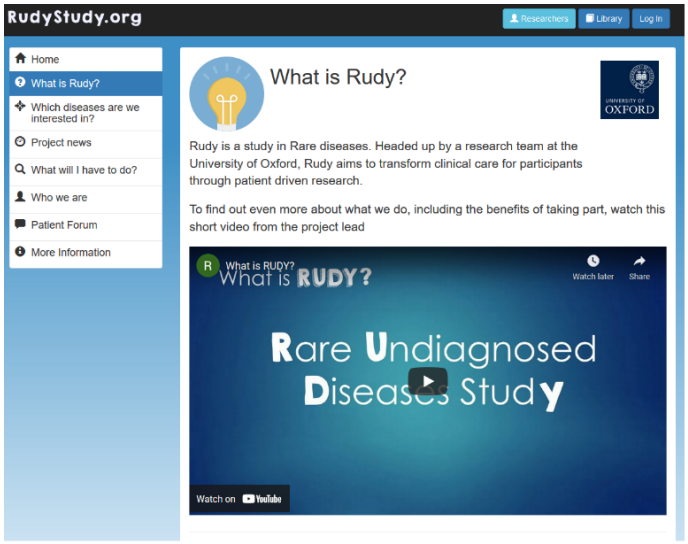
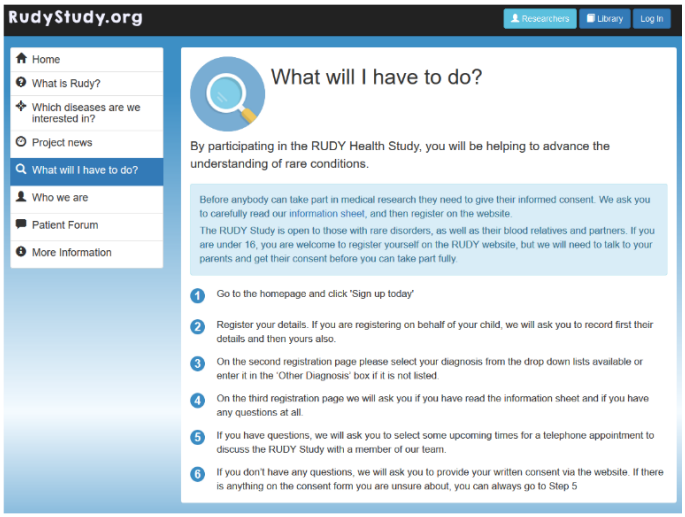
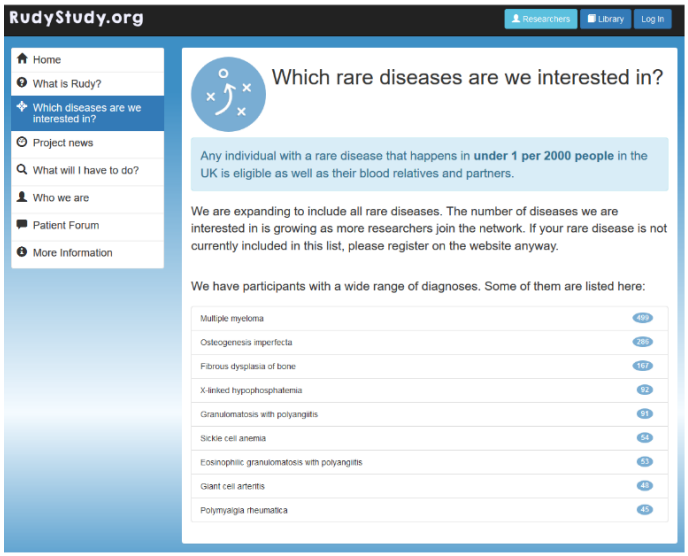
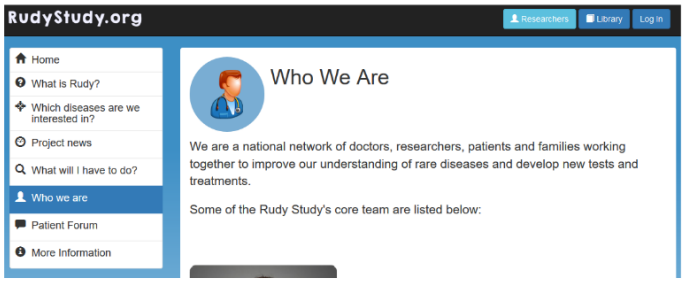


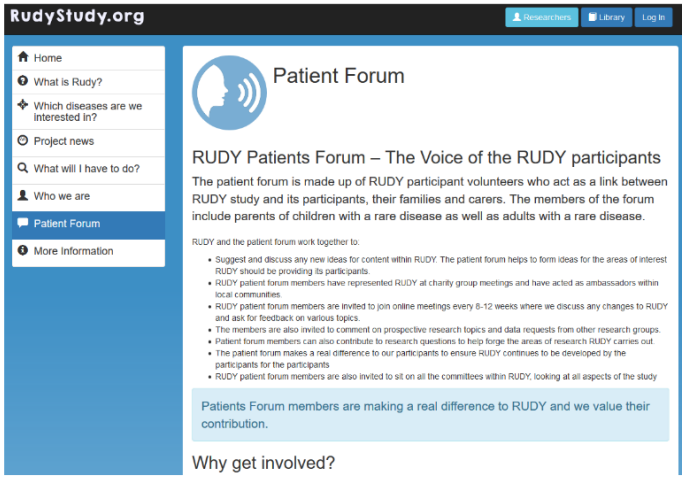
Figure 36: Information pages created (RUDY website > user clicks "Find out more").

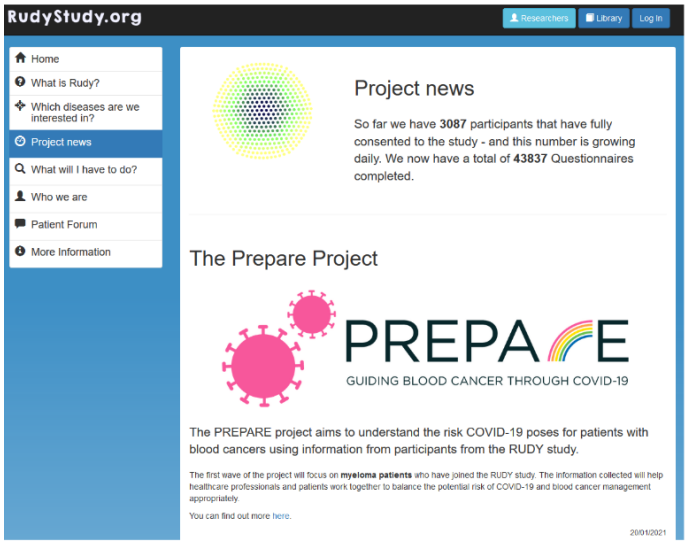


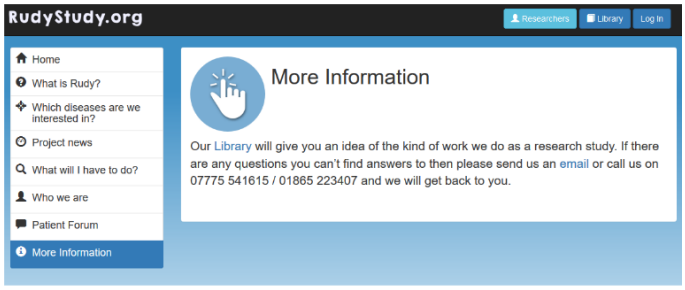












6.2.1.3 Research quality

This work implemented change within the RUDY project and the outcomes of these changes needed to be measured. This was for two reasons, to put findings into perspective (Glasgow *et al*, 1999) and to provide a way for other researchers to compare metrics (Proctor *et al*, 2011). Table 37 describes the implementation outcomes for this study. Note that under "measurement", "Consultation" has been added as a category. This does not appear in Proctor *et al*'s original description and has been included here as consulting participants is key to my work; particularly as I employ participatory methods such as co-design. This seems a relevant measure for this study.

Table 37: Evaluating success of this intervention's implementation (Proctor *et al*., 2011).

Outcome	Measurement	
Acceptability Is this intervention agreeable?	Qualitative data	The concept of this intervention was taken from interviews with RUDY participants and a focus group with RUDY researchers so that it would be acceptable to key stakeholder groups. Neither party disagreed with the intervention proposal or said that it was unacceptable.
Adoption Is evidence-based practice used?	Observation	I will see, from RUDY participant completion rates, whether this intervention has made any difference.
Appropriateness Is the intervention relevant (to RUDY)?	Focus groups	This intervention's implementation was co-designed with participants to ensure the changes were relevant. The implementation was done in collaboration with RUDY researchers to ensure the changes were appropriate.
Feasibility How can this intervention be used (by RUDY)?	Consultation	Consulting RUDY researchers was a key part of the development process for this intervention study, ensuring the changes were feasible (they are currently in use).
Fidelity Was the intervention implemented as intended?	Observation	Rather than design, implement and review, this study was validated at each design stage: research aim, evidence gathering, implementation and testing. We met regularly to review progress, which ensured implementation as intended.

Implementation cost What did this implementation cost?	Administrative data	A full-time DPhil student managed this project, and significant organisational and development effort was invested from the RUDY team. Designing and implementing the study took six months (April – September 2020).
Penetration How will this intervention be built into RUDY practices?	Consultation	The intervention has changed RUDY's website, online portal and email reminder. During the development process, the RUDY team reflected that they would benefit from a change management process!
Sustainability Can this intervention be institutionalised ?	Consultation	The success and rapid turnaround of this study is in no small part due to academic buy-in from RUDY's project lead. Without ongoing management effort, this work would not have been possible. This work can be institutionalised if there is leadership support.

One limitation to the use of implementation outcomes worth considering, is that using taxonomies or frameworks to measure quality does not guarantee any standard measure. Simply put, these measures may not be equally applied in other work. Their use may, in time, validate the reason for using these methods due to the level of rigour they offer and structure they provide for researchers to discuss different projects. Being able to assess research quality and apply similar methods across different studies provides a benefit that I think outweighs this limitation. Definitive proof concerning the usefulness of these frameworks may simply take time. I chose to use implementation outcomes as a way to describe the successes of this study because being able to define successful outcomes in this case is especially important. This is due to the way in which the empirical findings are complemented by the (context-specific) outcomes described in Chapters 4 and 5.

The intervention under study focuses on communicating information, so I have also provided a measure of educational quality. In this instance, the education RUDY participants are receiving as a result of the curation and creation of information on the website, portal and reminder emails. Individualisation, feedback and reinforcement are strong predictors of educational quality (Mullen *et al*, 1985) and their use in this study is described below:

Individualisation

During the design of this study, participants could ask questions or request accommodations by contacting I via email. Focus groups were designed to include room

for discussion to allow individuals to formulate and develop ideas. During the intervention study, participants could contact the RUDY team with any questions or to request accommodations directly, but this is standard practice for RUDY. Individual feedback was designed into the reminder emails as part of this intervention.

Feedback

During the design of this study, information was provided to participants about their role and the importance of their contribution via participant information sheets. Methods were validated through user tests where participants could see the consequences of the focus groups they had contributed to. Regular development meetings were held with the RUDY team to ensure they received feedback on progress. This study will summarise findings and thank people for taking part. This will be disseminated via RUDY.

Reinforcement

Previous findings showed that participants sought validation for the time they spent taking part in the RUDY project. This study has been designed to "reward" participation after it has occurred by making information available about the consequences of research participation. Positive reinforcement was implemented in three ways: describing the personal data collected and used in questionnaire descriptions, making academic language more accessible across the website and online portal, and reiterating research aims in the reminder emails.

Evaluating the success of an implementation means identifying the aspects that have worked, and identifying areas for improvement (Proctor *et al*, 2011). This allows other researchers to observe success and identify further research questions. Evaluating the success of this intervention is crucial as the Dynamic Consent literature is formative at this time, and results from this work may be replicated or investigated further.

6.2.1.4 Analysis plan

What follows is the procedure for analysing the data collected during this intervention study. Key areas of interest are outlined below, with the expectation that unplanned findings might emerge over the course of data-analysis.

Post-intervention data only

This information (Table 38) offered an overview of the data collected after the intervention was put in place. Age, ethnicity and "RUDY discovery method" provide insights into the participant sample. RUDY discovery method simply shows how a participant found their way to the study.

Table 38: Template for post-intervention initial data.

Characteristic	Assigned [n]	(% of total assigned)	Submitted [n]	(% of total submitted)
Age group				
<18		%		%
18 – 24		%		%
25 – 34		%		%
35 – 44		%		%
45 – 54		%		%
55 – 64		%		%
65 – 74		%		%
75 – 84		%		%
85 – 96		%		%
Method of discovering RUDY				
Charity		%		%
Doctor, Nurse or GP		%		%
Web search		%		%
Other		%		%
Ethnicity				
English / Welsh / Scottish / N. Irish		%		%
Irish		%		%
Any Other White Background		%		%
White and Black Caribbean		%		%
White and Asian		%		%
Any Other Mixed background		%		%
Chinese		%		%
Any Other Asian Background		%		%
African		%		%
Caribbean		%		%
Any Other Ethnic Group		%		%
N/A		%		%

Comparing 2019 and 2020 data

This information offered a way to compare completion rates across different time points. Table 39 shows how completion rates were compared between different consent cohorts.

Table 40 shows how completion rates were stratified by first-time submissions, versus subsequent ones. Table 41 shows how completion rates were compared between different questionnaires. Note that the entries for VOLP and SF36 v2 questionnaires are absent from 2019 data because they were added in 2020.

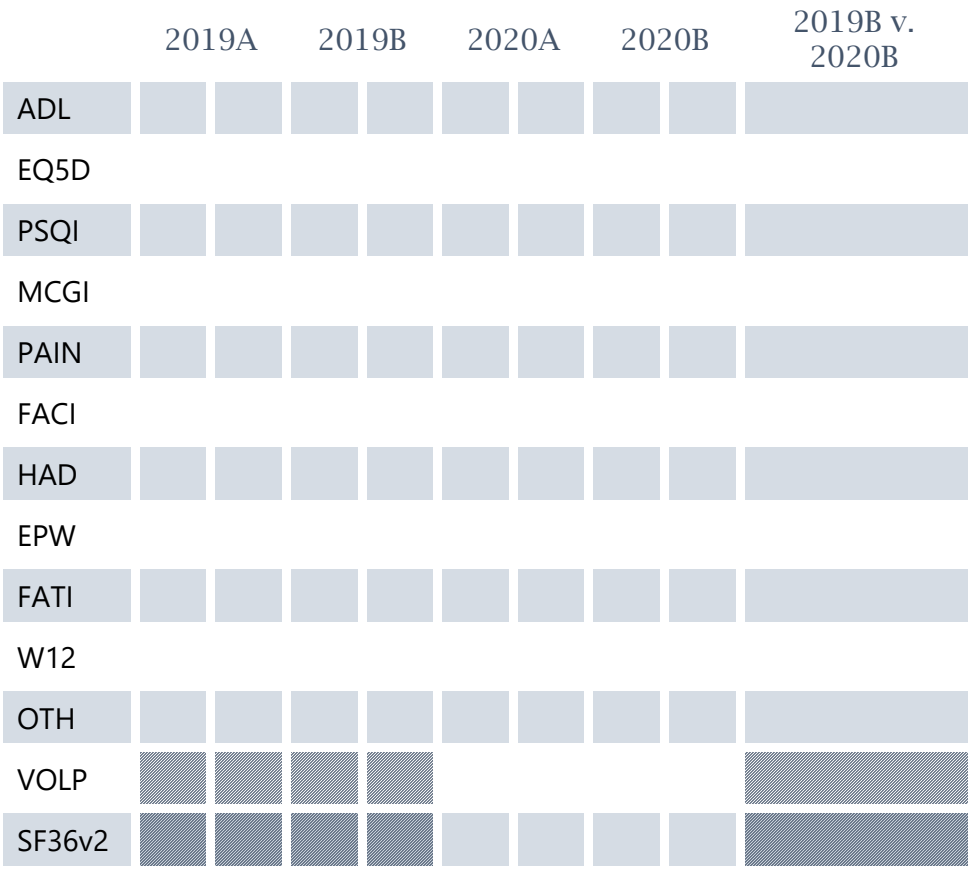
Table 39: Template for pre- and post-intervention data, stratified by year of recruitment.

		2019A	2019B		2020A	2020B		2019B→2020B
N(assigned)								
N(submitted)								
Completion rate								
Consent cohort response rate (number of participants)	*2014*							
	2015							
	2016							
	2017							
	2018							
	2019							
	2020							

Table 40: Template for pre- and post-intervention data, stratified by submission type.

		2019A	2019B	2020A	2020B	2019B→2020B
First-time	*Submitted (n)*					
	Completion (%)					
Subsequent	*Submitted (n)*					
	Completion (%)					

Table 41: Template for pre- and post-intervention data, stratified by questionnaire type.



6.2.1.5 Validation

When I finished analysing this data, I collated findings into a report for the RUDY team. During this meeting, it was suggested that I compare first time questionnaire submissions to subsequent ones. 'First-time' questionnaires are the initial set of questionnaires that participants are asked to complete (Javaid *et al*, 2016). This was an incredibly helpful suggestion that resulted in the initial/follow-up breakdown above. The team's general feedback from their experience of working with me was that this work had been useful. They indicated that there were not many opportunities to receive feedback from an external party on the study. This work had, the team suggested, highlighted two potential areas for development: a more sophisticated notification system, and a change management process for future development work.

6.3 Results

In 2020, 8663 questionnaires were assigned to participants pre-intervention, of which 2655 were submitted. This indicated a completion rate of 30.6%. 17352 were assigned post-intervention, of which 4797 were submitted, a completion rate of 27.6%. There seems to have been a 3% fall in overall completion rate. This may be due to the covid-19 pandemic, but cannot be proven (anecdotal evidence from the RUDY team suggested that this may be the case). There is somewhat of a relative decline in participants not completing surveys, despite an increased number of submissions in general.

In 2019, over the same time points: 8829 questionnaires were assigned earlier in the year, with 3215 returned (36.4% completion). 8780 were assigned later in the year, with 1974 returned (22.5% completion). There seems to have been a 13.9% fall in overall completion rate. The fall in completion rate seems to be lower over the intervention period.

Additionally, a comparison of the post-intervention completion rate (2020B, 27.6%) against the same time the year before (2019B, 22.5%) shows that the general completion rate was 5.1% higher. The potential impact of two national lockdowns, due to the COVID-19 pandemic, on research participation is uncertain.

6.3.1 Post-intervention data only

The only way to explore differences between participants was to use age data. Information about other characteristics was incomplete (ethnicity) or not provided (e.g., gender). People aged between 55 and 84 were over-represented in the data submitted, as they accounted for 69.6% of submissions despite only accounting for 54.4% of people to whom questionnaires were assigned (see Table 42).

From the data available, the method through which a participant discovered RUDY (through a charity, via their clinician, by searching online, or some other route) seemed to have had no demonstrable impact on completion. A number (61.1%) of participants to whom questionnaires were assigned had no ethnicity (marked N/A), but 67% of those recorded were English, Welsh, Scottish, Northern Irish, British, Irish, or any other white background. This group is also over-represented in the data as it only accounts for 37.4% of people assigned questionnaires.

Table 42: Characteristics for the post-intervention sample (24-Sept - 31-Dec 2020).

Characteristic	Assigned [n=1360]		Submitted [n=431]	
Age group				
<18	68	4.9%	9	2.0%
18 – 24	38	2.7%	8	1.8%
25 – 34	137	9.8%	29	6.4%
35 – 44	150	10.8%	29	6.4%
45 – 54	225	16.2%	61	13.5%
55 – 64	297	21.3%	120	26.6%
65 – 74	350	25.1%	148	32.8%
75 – 84	111	8.0%	46	10.2%
85 – 96	16	1.1%	1	0.2%
Method of discovering RUDY				
Charity	261	18.8%	91	20.2%
Doctor, Nurse or GP	579	41.6%	182	40.4%
Web search	100	7.2%	33	7.3%
Other	452	32.5%	145	32.2%
Ethnicity				
English / Welsh / Scottish / N. Irish	505	36.3%	294	65.2%
Irish	1	0.1%	1	0.2%
Any Other White Background	14	1.0%	7	1.6%
White and Black Caribbean	1	0.1%	0	0%
White and Asian	3	0.2%	2	0.4%
Any Other Mixed ethnic background	7	0.5%	3	0.7%
Chinese	1	0.1%	1	0.2%
Any Other Asian Background	2	0.1%	1	0.2%
African	3	0.2%	0	0%
Caribbean	3	0.2%	1	0.2%
Any Other Ethnic Group	1	0.1%	0	0%
N/A (information not provided)	851	61.1%	141	31.3%

Table 43 shows the post-intervention completion rate for each questionnaire. This suggests healthy completion rates when compared to the original data-access request justifying this work that cited completion rates averaging 19% (see Appendix 11). The average completion rate for the post-intervention period was 27.6%.

Table 43: Post-intervention questionnaire breakdown.

	Assigned [n=17352]	Submitted [n=4797]	Completion rate [average=27.6%]
ADL	1330	403	30.3%
EQ5D	1330	373	28.0%
PSQI	1330	356	26.8%
MCGILL	1330	357	26.8%
PAINDETECT	1330	351	26.4%
FACIT	1330	372	28.0%
HADS	1330	358	26.9%
EPWORTH	1330	356	26.8%
FATIGUE	1330	364	27.4%
WALK12	1330	407	30.6%
OTHER	1392	307	22.1%
VOLP	1330	412	31.0%
SF36 (v2)	1330	381	28.6%

6.3.2 Comparing 2019 and 2020 data

Data collection from July 1st to September 23rd 2019 is referred to as 2019A. Data collection from September 24th to December 31st 2019 is referred to as 2019B. 2020A and 2020B refer to data collection intervals from July 1st – September 23rd 2020 and September 24th – December 31st 2020 respectively. Table 44 shows how many RUDY participants were represented in the data shared by the study.

Table 44: Breakdown of unique participants who were assigned questionnaires (n=1936).

	A only	B only	A&B
2019 (n=1549)	709	710	130
2020 (n=1776)	416	957	403

6.3.2.1 Initial characteristics

Table 45 shows that people with similar characteristics were submitting questionnaires pre- and post-intervention. Age, route-to-RUDY and ethnic breakdown all show similar patterns of participation. The table uses number of submissions, rather than unique individuals, to illustrate who was submitting questionnaires. E.g., 65-74 year olds submitted the most questionnaires across all samples.

Where the table shows a number and a percentage, the value indicates the number of submissions from that group and the percentage value indicates that number as a proportion of the number of questionnaires submitted. For example, in 2019A 3215 questionnaires were submitted. 67 submissions were made by people aged 18 – 24. This means the 18 – 24 age group accounted for 2.08% of submissions between July and September 2019 (cell outlined in red).

Table 45: Breakdown for all samples.

	2019A [n=3215]		2019B [n=2979]		2020A [n=2655]		2020B [n=4579]	
Age groups								
<18	5	0.16%	2	0.07%	1	0.04%	3	0.07%
18 – 24	67	2.08%	46	1.60%	14	0.54%	39	0.88%
25 – 34	195	6.07%	322	11.17%	182	7.04%	289	6.53%
35 – 44	232	7.22%	283	9.82%	185	7.15%	282	6.37%
45 – 54	373	11.60%	540	18.73%	467	18.05%	506	11.43%
55 – 64	760	23.64%	621	21.54%	613	23.70%	1281	28.93%
65 – 74	1153	35.86%	865	30.00%	886	34.25%	1578	35.64%
75 – 84	419	13.03%	193	6.69%	239	9.24%	450	10.16%
85 – 96	11	0.34%	11	0.38%	0	0.00%	0	0.00%
Found RUDY								
Charity	684	21.28%	592	19.87%	540	20.34%	907	19.81%
Doctor, Nurse or GP	1118	34.77%	1353	45.42%	1048	39.47%	1860	40.62%
Web search	213	6.63%	194	6.51%	149	5.61%	306	6.68%
Other	1200	37.33%	840	28.20%	918	34.58%	1506	32.89%
Invited to RUDY	1128	35.09%	1543	51.80	1195	45.01%	2132	46.56%
Not invited to RUDY	2087	64.91%	1436	48.20%	1460	54.99%	2447	53.44%
Ethnicity								
Eng/Wel/Scot/ N. Irish	1925	59.88%	1788	60.02%	1642	61.85%	3131	68.38%
Irish	0	0%	0	0%	0	0%	13	0.28%
Other White	75	2.33%	33	1.11%	45	1.69%	74	1.62%
White & Black Caribbean	8	0.25%	0	0%	0	0%	0	0%
White and Asian	0	0%	0	0%	0	0%	2	0.04%
Other Mixed background	33	1.03%	1	0.03%	21	0.79%	29	0.63%
Chinese	11	0.34%	0	0%	0	0%	13	0.28%
Other Asian Background	5	0.16%	0	0%	0	0%	13	0.28%
African	11	0.34%	21	0.70%	17	0.64%	0	0%
Caribbean	21	0.65%	11	0.37%	25	0.94%	13	0.28%
Other Black/African/Carib	0	0%	1	0.03%	0	0%	0	0%
#N/A	1126	35.02%	1124	37.73%	905	34.09%	1291	28.19%

6.3.2.2 Completion rate data

This section compares pre- and post-intervention submissions (a) by consent cohort, (b) by first-time and subsequent submissions, and (c) by questionnaire.

Consent cohorts

"Consent cohort" refers to the year a participant was recruited and gave consent to take part in RUDY (e.g., cohort 2014 is the group of participants who signed up to RUDY in 2014). Table 48 compares completion rates across 2019 (2019A v. 2019B), 2020 (2020A v. 2020B), and 2019/2020 (2019B v. 2020B). Overall completion rates dropped by 13.9% in 2019, and only 3% in 2020. There was a 5.2% increase in general completion from 2019 to 2020.

Table 46: Pre- and post-intervention data, grouped by consent cohort[34].

		2019A	2019B		2020A	2020B		2019B→ 2020B
N(assigned)		8829	8780		8663	17352	+100.3%	+97.6%
N(submitted)		3215	1974		2655	4797	+80.7%	+143%
Average completion rate		36.4%	22.5%	-13.9%	30.6%	27.6%	-3.0%	+5.2%
Consent cohort response rate (%) [submitted/assigned]	*2014*	24.0% 177/739	30.4% 198/651	+6.5%	31.6% 255/806	22.8% 202/886	-8.8%	-7.6%
	2015	28.0% 219/783	21.1% 193/916	-6.9%	27.1% 251/927	24.3% 341/1406	-2.8%	+3.2%
	2016	26.1% 128/491	12.9% 69/533	-13.1%	13.9% 62/445	19.2% 168/877	+5.2%	+6.2%
	2017	33.4% 128/383	15.5% 62/401	-18.0%	22.5% 85/378	17.4% 126/724	-5.1%	+1.9%
	2018	32.3% 1047/3243	27.0% 612/2264	-5.3%	29.1% 659/2262	26.1% 974/3735	-3.1%	-1.0%
	2019	47.5% 1516/3190	20.9% 840/4015	-26.6%	29.5% 884/2995	28.3% 1670/5909	-1.3%	+7.3%
	2020	0%	0%	0%	54.0% 459/850	34.5% 1316/3815	-19.5%	0%

[34] Percentage changes are provided as a heuristic to illustrate change over time. The metric of interest is completion rate.

Analysis of this data shows participant retention, and reduced drop-off in participation over time. The breakdown of completion rate by consent cohort showed an increase (or a reduced drop) from 2019 to 2020 for every cohort except cohort 2014. Cohort 2014's completion rates would be worth investigating further, perhaps a focus group with participants from this cohort. Such work is unfortunately out of scope for this thesis. Figure 37 (n) and Figure 38 (%) illustrate cohort submission data. More questionnaires were submitted during the intervention period in 2020, but completion rates were lower than before the intervention. Cohort 2016 was the only group to show an increase in completion rate from 2020A to 2020B, which differs from the sample data (Section 6.2.1.1) in that they were the least engaged cohort.

Figure 37: Questionnaire submissions (n), by consent cohort.

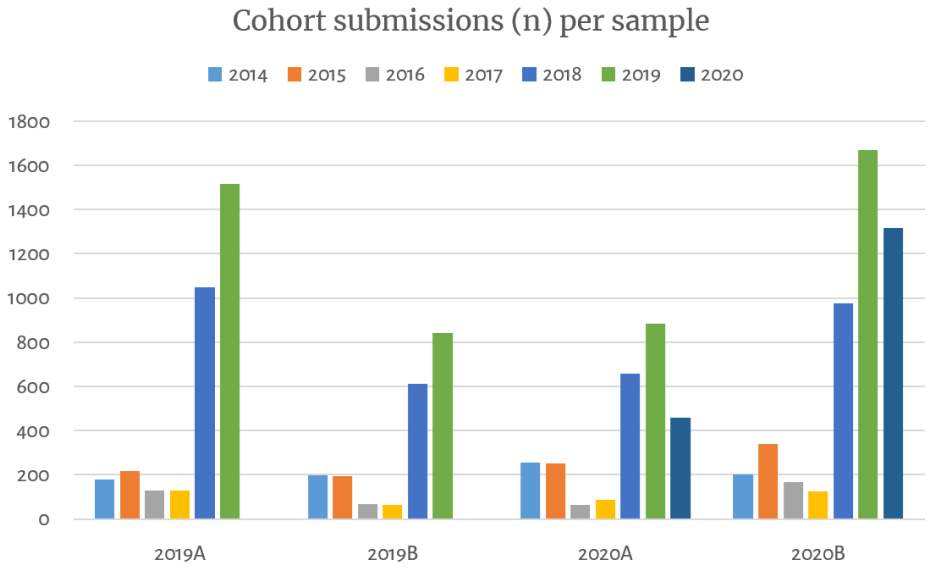
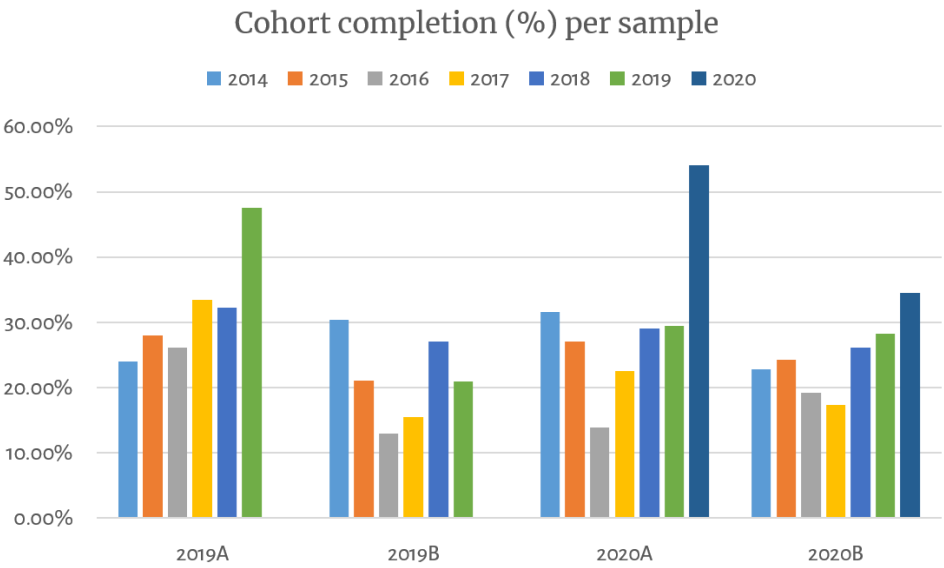


Figure 38: Questionnaire submissions (%) by consent cohort.



First–time and subsequent submissions

Table 47 shows that completion rates for first-time and subsequent questionnaire submissions increased from 2019 to 2020. This data also supports the assertion that the intervention reduced drop-off in participation over time: initial completion dropped by 32% in 2019 and 8.9% in 2020. Follow-up completions dropped by 15% in 2019 and increased by 4.5% in 2020. Figure 39 compares the number of initial and follow-up submissions (n). Figure 40 compares initial and follow-up completion rates (%).

Table 47: A comparison of first-time and subsequent submissions.

		2019 A	2019 B		2020 A	2020 B		2019B→ 2020B
First-time	*Submitted (n)*	1105	621	-43.8%	426	459	+7.7%	-26.1%
	Completion (%)	52.7%	20.5%	-32.2%	59.3%	50.4%	-8.9%	+29.9%
Subsequent	*Submitted (n)*	411	219	-46.7%	33	857	+2497%	+291.3%
	Completion (%)	37.5%	22.3%	-15.2%	25%	29.5%	+4.5%	+7.2%

Figure 39: Questionnaire submissions (n), by first-time and subsequent submissions.

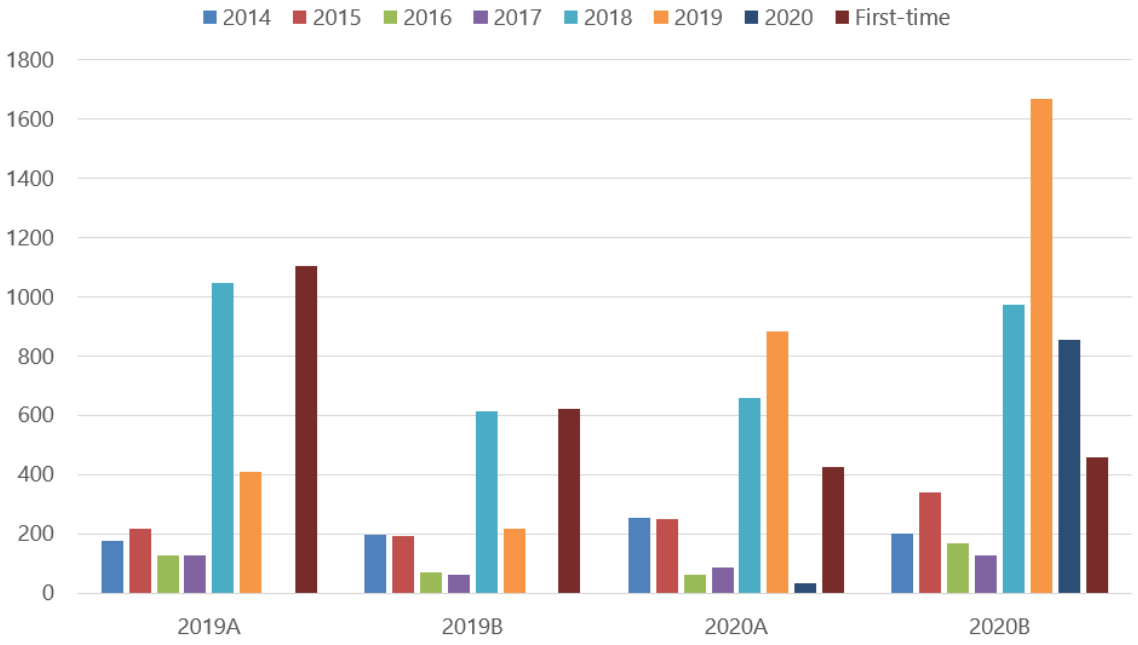
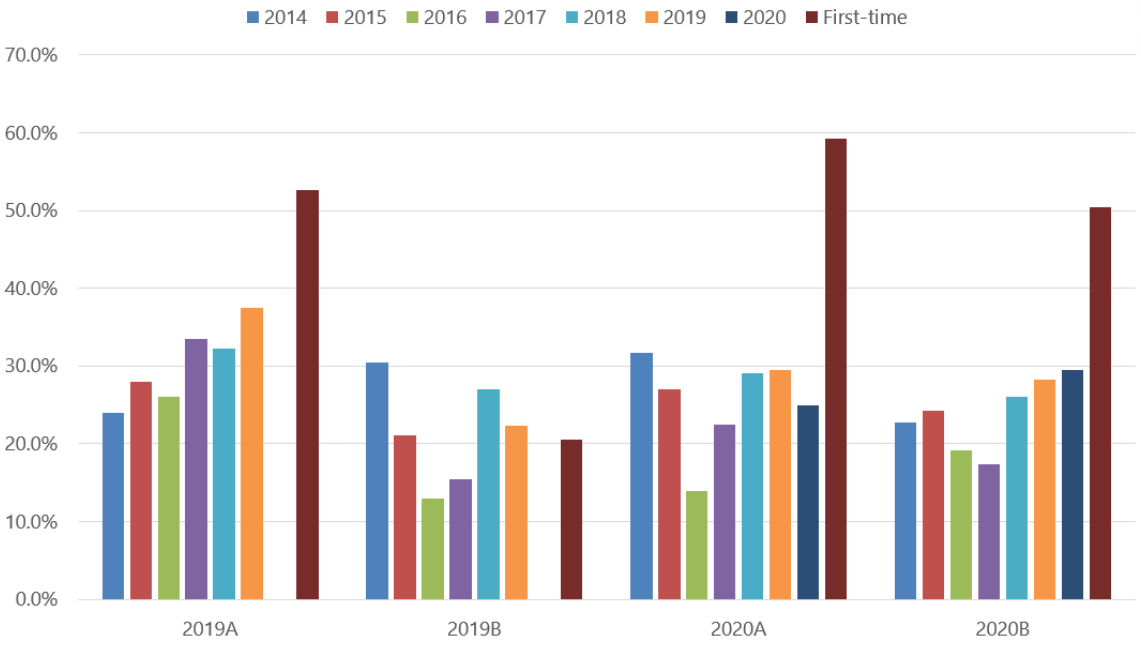


Figure 40: Questionnaire submissions (%), by first-time and subsequent submissions.



Breakdowns per questionnaire

Table 48 shows that grouping completion rates by questionnaire was not helpful for this analysis. While the number of questionnaires submitted increased pre- and post-intervention, completion rates did not. Completion rates dropped for all questionnaires over time.

Table 48: Submissions per questionnaire.

	2019A		2019B			2020A		2020B			2019B v. 2020B
ADL	328	41.1%	304	39.5%	-1.6%	260	33.5%	385	29.7%	-3.8%	-9.8%
EQ5D	317	39.7%	297	38.6%	-1.1%	250	32.3%	356	27.4%	-4.9%	-11.2%
PSQI	281	35.2%	258	33.5%	-1.7%	229	29.5%	339	26.1%	-3.4%	-7.4%
MCGI	290	36.3%	267	34.7%	-1.6%	231	29.8%	340	26.2%	-3.6%	-8.5%
PAIN	283	35.4%	259	33.6%	-1.8%	227	29.3%	334	25.8%	-3.5%	-7.8%
FACI	295	36.9%	268	34.8%	-2.1%	241	31.1%	355	27.4%	-3.7%	-7.4%
HAD	275	34.4%	264	34.3%	-0.1%	233	30.1%	341	26.3%	-3.8%	-8.0%
EPW	289	36.2%	260	33.8%	-2.4%	230	29.7%	339	26.1%	-3.6%	-7.7%
FATI	300	37.5%	271	35.2%	-2.3%	236	30.5%	347	26.8%	-3.7%	-8.4%
W12	345	43.2%	322	41.8%	-1.4%	269	34.7%	390	30.1%	-4.6%	-11.7%
OTH	212	25.3%	209	25.6%	0.30%	197	24.1%	295	21.7%	-2.4%	-3.9%
VOLP						36	76.6%	394	30.4%	-46.2%	
SF36v2						16	34.0%	364	28.1%	-5.9%	

6.4 Reflection

There is evidence in this chapter to support the study's hypothesis that enhanced feedback positively affects research participation. Analysis of the data suggested that enhanced feedback can be used to retain participants over time, and gather more data overall. Enhanced feedback seems to have positively affected research participation: people returned to take part, and more data was collected.

RQ2a: Is research participation affected by how information is communicated to research participants?

This is likely. More data was collected, so this increase in the number of questionnaires collected means that more data is available for research purposes. This is a valuable finding that speaks to the importance of developing feedback mechanisms into research practice. If this is not possible at the start of a project then, as this study demonstrated, an existing project can introduce feedback mechanisms during its lifetime. Sample data provided by RUDY indicated a general drop in completion rate, which is common in longitudinal data collection. One of the most exciting findings from this study was that analysis of data showed enhancing feedback reduced participation drop-off over time.

RQ2b: What is the relation between participant engagement and the richness of longitudinal research data?

The working definition of engagement used in this thesis is that engagement consists of interest, knowledge and active participation. The enhanced feedback intervention aimed to reward interest and improve the transfer of knowledge from the research team to participants by rephrasing information in a way that was easier to understand. Information was provided about current studies and collaborations, descriptions of the kinds of data collected by questionnaires were added and the website was re-designed to make important information immediately obvious. A series of information pages were created under a "find out more" button that included a description of the patient forum, and reminder emails were re-designed to thank participants and highlight the value of their data-contributions.

These changes seem to have resulted in more questionnaires being submitted, which suggests that enhanced feedback leads to richer data collection. There may have been confounding variables that I have not been able to identify, such as the influence a pandemic may have had on participants. In longitudinal studies like RUDY, data richness comes from participant retention as well as more data being collected in general, because information is collected from the same individual and insights are gathered over time. Analysis of the data collected during this study indicated that the drop in submissions

seen over time was reduced – the general drop in completion rate over 2020 (pre- and post-intervention) was less prominent than the year before.

There are several limitations to this study, and a number of areas to investigate further. First and foremost, the study was designed to operate on a simple metric: questionnaire response rate. This, along with incomplete measures of demographic data meant that a granular breakdown cannot be provided. Second, the findings described are not directly applicable to the wider research community because rare-disease research is a specialised sub-field that may not easily translate to other disciplines. The third limitation relates to how the study was designed. This intervention did not distinguish between the different types of changes made (e.g., website changes, changes to the portal, or to the reminder email), so this data is unable to support any conclusions about the independent effects of each change. This offers a rich avenue for exploration, supported by an existing literature that compares different communication methods (Dresselhaus *et al*, 2000). Table 49 suggests how the single hypothesis from this study could be broken down into sub-hypotheses for a more comprehensive investigation:

Table 49: Hypotheses for a more comprehensive feedback intervention.

Communicating project aims positively affects research participation.
Communicating what data is used positively affects research participation.
Communicating how data is used positively affects research participation.
Communicating the value of data-contributions positively affects research participation.
Communicating research outcomes positively affects research participation.

This work, and any similar projects, would be greatly improved and further validated by an analysis of data collected over a longer period of time. This is because compliance is best measured over time rather than at one or two individual points (Brieger *et al*, 2007).

The enhanced feedback intervention was designed as a direct response to areas of tension reported by participants, resulting in changes in questionnaire completion. Findings are useful to RUDY because they show that investing in engagement leads to richer data collection, which further justifies their current research approach. They adopted engagement as a design principle and have designed research activities that collect participant feedback, such as Skype calls with members of the patient forum. These findings are useful to the wider research community because they provide an example of where even engaging projects falter, highlighting areas for attention.

Dynamic Consent practices emphasise the importance of an individual's knowledge (Teare *et al*, 2015); the data that RUDY requires concerns how symptoms manifest and develop over time. People with rare conditions are the only individuals in a position to provide the information RUDY needs – analysis of the intervention data suggests that communicating this importance improved data collection. Some researchers find public communication difficult because they do not know at what level (of abstraction) to communicate (Schuler Scott *et al*, 2019a). This study suggests that researchers should seek feedback from members of the public, and demonstrates that inclusivity is valuable. Researchers should be open to constructive criticism and choose to consult participants.

The social contract between society and science must be demonstrable. When researchers demonstrate trustworthiness, the public's response to this is to trust. If members of the public are to take part in research and allow researchers to access personal information and generate further insights, then those research efforts must be of value. Findings from this study indicate that by enhancing their feedback mechanisms, researchers can retain participants over time, reduce drop-off in participation over time and collect more data overall. Participant engagement provides research value.

If the theory of Dynamic Consent is going to evolve, the fundamental principles it operates on must be clear. As more researchers adopt a Dynamic Consent approach, they seek to draw from existing theory and practice. Dynamic Consent is not a panacea and the onus for responsible research must lie with researchers and not their participants. Participant engagement involves capturing the interest of members of the public, transferring knowledge, and providing opportunities for active participation. People with rare conditions are already a highly motivated participant community, researchers must demonstrate that this participation is valuable. Tailoring communication to be accessible shows consideration and respect for this participation.

This chapter has presented key findings and described practical and procedural limitations of this work, such as the relatively short post-intervention period and the potential impact of the COVID-19 pandemic. Also identified is a need for materials that support the development of engaging research practices that invite interest, provide knowledge and enable active participation in research. The chapter ahead provides such materials.

Chapter 7

A tool-kit for engagement

"Over the course of their work on patient compliance, they came to define the major problem in medical practice not as the behaviour of patients, but as the behaviour of physicians" (Abedin & Warner, 2016).

7.1 Introduction

Taking a dynamic approach to consent is more representative of human choice; people change their minds over time, and any information they receive influences this. A significant issue with current Dynamic Consent is that its practicalities are under-reported, as we will discuss. This chapter describes how researchers can measure engagement within their own work. Quite simply, they must identify their own metric and decide how they will measure this throughout their study. In the previous chapter, engagement could have been measured as a complex variable drawing together website use, questionnaire responses and perhaps response times for individual participants. Instead, I chose a simple measure that provided a more general overview of how participants engaged with the study: questionnaire completion rates.

The conclusion that this chapter will draw is that engaging research data-practices are important and to be effectively reported, they must be planned in advance. Reporting

research findings after they have been discovered without describing the initial design work required is a barrier to good practice. Such reporting is limited (Curran-Everett & Milgrom, 2013) and makes findings harder to replicate. The GRIPP2 checklists for reporting patient and public involvement in research are an excellent example of a clear, concise and usable reporting tool (Staniszewska *et al*, 2017). The tools in this chapter encourage "ante-hoc" descriptions, created before work has begun. Such descriptions can be used as development aids, and are important because researchers can use them to create a working specification for engagement that can be compared with implementation steps (Michie *et al*, 2009).

Reflecting on the working definition of engagement for this thesis[35], researchers must pique the interest of their participants, provide new knowledge and facilitate active participation. This chapter provides three tools to aid such efforts. The first is a model that can be used as a design prompt to draw out strategic aims, and consider how data-flows operate from researcher and participant perspectives. The second is a framework that draws out specific assumptions made about Dynamic Consent, prompting the researcher to associate a metric in order to measure whether or not the assumption is met. Third, a checklist for implementing Dynamic Consent provides practical support. This tool-kit is demonstrated with a use-case that is similar in design and scope to the intervention study in this thesis.

7.2 Reviewing engagement

There are many reasons to prioritise engagement as a researcher working with human participants, other than its importance to Dynamic Consent. Before this discussion however, let us address the meaning of the phrase "working with" participants. This statement colours underlying research assumptions with My personal position: that research is a reciprocal and inclusive arrangement between researchers and participants. Such an arrangement is used in biomedical research (Gottweis *et al*, 2011; Hobbs *et al*, 2012; Meslin and Cho, 2010), and advocated within Dynamic Consent literature (Kaye *et al*, 2012). Such a position is, at present, a goal to strive towards rather than common practice. Let us revisit this opening statement: there are many reasons to prioritise engagement as a researcher collecting data from human participants. Such researchers might prioritise engagement for one of three reasons. First, engagement demonstrates trustworthiness (Aitken *et al*, 2016) and establishes a working partnership between participant and researcher. Second, engaging with participants serves as an opportunity

[35] Engagement = Interest + Knowledge + Active participation.

to celebrate public contributions instead of relegating them to "tinkering at the edges" (Boaz *et al*, 2016). Public celebrations also serve to redress the asymmetry of knowledge and control. Finally, as the previous chapter indicated, enhancing the feedback available to research participants enriches data collection. Designing engaging data-practices that include participant input and respond to their needs also demonstrates the social contract in action – Chapters 4 and 5 demonstrated that participants thought researchers had data-protection responsibilities: to use information appropriately, and to show consideration for personal data.

Demonstrations of trustworthiness are becoming, and will continue to become increasingly important in digital and online spaces where engagement is developed and operationalised without any discussion of RI[36]. Users want to know how their information is put to use (Coathup *et al*, 2016), and researchers know how to build engaging interfaces communicating these uses (O'Brien *et al*, 2018; O'Brien and Toms, 2010). Despite this demand and capacity for engaging research practices, their value remains tantalisingly out of reach. This draws parallels with security (and other non-functional) requirements in software development, often under-represented because they are difficult to describe (Mylopoulos *et al*, 1992). This work has shown that engaging research practices improve research quality, and developed criteria for implementation in the form of enhanced feedback. The enhanced feedback intervention was built in response to participants seeking information that they should already been able to access.

7.3 Modelling Dynamic Consent

A brief review of the relevant literature follows, before the tool-kit is described. There are multiple accounts of Dynamic Consent's use (Budin-Ljøsne *et al*, 2017; Dixon *et al*, 2014; Kaye *et al*, 2015; Prictor *et al*, 2020b, 2018; Sanders *et al*, 2014; Teare *et al*, 2015; Williams *et al*, 2015) but only a few practical examples (Haas *et al*, 2021; Pattaro *et al*, 2015; Teare *et al*, 2017; Teodoro *et al*, 2017; Thiel *et al*, 2015). Encore's first description of Dynamic Consent comes from a technical architecture. Data subjects could provide (or amend) informed consent in a fine-grained way, governing access to, and use, of confidential and personal data (Casassa Mont *et al*, 2012). A focus group gathering responses to a Dynamic Consent approach emphasised this granularity (Sanders *et al*., 2014):

> "Patients are not restricted to giving a universal consent or universal 'opt out' for use of their data"

[36] For more information, see Baldus *et al*'s discussion of "brand communities" (Baldus *et al*, 2015).

The authors also claimed that Dynamic Consent would allow patients to access project feedback, research results and an overview of who had used, and could access their data. Kaye *et al*'s definitive outline of dynamic consent benefits, limitations and challenges described two key components: a specific "Dynamic Consent" project, and a wider concept offering a new, personalised, digital approach to consent that was designed to meet the needs of the twenty-first century research landscape (Kaye *et al*, 2015). The fundamental aim of Dynamic Consent was to shift the balance of power in favour of research participants, with an interface facilitating two-way communication and a technical architecture protecting and ensuring responsible use of data. Engagement was a key tenet of this early thinking, demonstrated by Teare *et al*'s move towards "Engagement 2.0" (Teare *et al*, 2015). Revocation was another – patients could electronically control their consent over time (Williams *et al*, 2015). Two years later, the positive research impact of the type of ongoing communication that Dynamic Consent provided was described as:

> "Relatively easy to set up and maintain, its implementation will require that researchers re-consider their relationship with research participants and adopt new procedures", (Budin-Ljøsne *et al*, 2017).

These authors created a process flowchart (see Section 2.3.4) to help researchers develop their own Dynamic Consent approaches, spanning recruitment, informed consent and follow-up. A further methodological benefit of Dynamic Consent was reported as reduced barriers to research participation (Prictor *et al*, 2018). The most recent theoretical work has provided an evaluation framework to maximize the quality, replicability, and relevance of projects using Dynamic Consent (Prictor *et al*, 2020b).

Reports from projects implementing Dynamic Consent are rare. An initial pilot study received positive feedback and indicated that participants preferred to actively consent to data-use (Thiel *et al*, 2015). The Cooperative Health Research In South Tyrol (CHRIS) study is a longitudinal study. CHRIS was designed to investigate the genetic and molecular basis of age-related common chronic conditions and their interaction with life style and environment. In an initial report, the authors examined the legal and ethical attributes and challenges of Dynamic Consent but could not describe its effects as the study had not yet started (Pattaro *et al*, 2015). Later on, the authors briefly describe one use of Dynamic Consent:

> "The dynamic consent options for re-contact allowed us to re-invite the participants for the CHRIS-NAFLD study, collecting additional information and re-consent. Prior to participation, participants were informed about the objectives and extra procedures of this additional

> study, for which they provided written informed consent", (Motta *et al*, 2019).

The CHRIS study was also cited as an example of good practice. The clarity of governance and prioritisation of participant choice means that the study is equipped to adapt to new scientific discoveries and comply with new regulations (Piciocchi *et al*, 2018). RUDY started before CHRIS, but was reported slightly later (Javaid *et al*, 2016; Teare *et al*, 2017). The My Health My Data project used Dynamic Consent to drive data exchanges in a probative, secure, open and decentralized manner (Teodoro *et al*, 2017). Most recently, Dynamic Consent has been used to keep personal and contact details up to date; make consent choices (including indicate preferences for return of results and future research use of biological samples, genomic and health data); follow their progress through the study; complete surveys, contact the researchers and access study news and information (Haas *et al*, 2021).

There is, at present, no way to measure the quality or benefit of Dynamic Consent or standardise its reporting, despite efforts to develop an evaluation framework (Prictor *et al*, 2020b). Dynamic Consent is predicated on revocation and engagement, but current work citing Dynamic Consent sometimes ignores engagement, or in some cases divorces engagement from Dynamic Consent entirely (e.g., Prictor *et al*, 2020a). This is a considerable oversight because engagement is valuable. In this chapter a model, framework and specification are provided to aid researchers who want to develop engaging data-practices, which include Dynamic Consent. This tool-kit uses the model of enhanced feedback developed earlier in this thesis.

The following tools are a result of my own analysis[37]. These materials were created after the intervention study had been carried out and I had spent four years working closely with RUDY researchers and participants on the importance of providing feedback to research participants. With this design priority in mind, I returned to the literature. They had initially found the Dynamic Consent process model and evaluation framework difficult to use: in the design of their research process, and later, to evaluate and report their use of dynamic consent. The work supporting this project required tools that aligned with development goals, were more flexible, created less initial overhead and could be easily revisited over time. This tool-kit was developed to fulfil those requirements, highlighting a key design principle. The tools were created to centre user needs rather than emphasise technical design.

[37] For blank, template versions of these resources please refer to Appendix 12.

The tools are valuable because they have been adapted from existing academic solutions to methodological problems, and they directly address issues discovered in the course of this work. Development effort was spent on presentation: adding images and colour and reviewing layout and format to make the materials more user-friendly. Significant effort was spent on clarifying how these materials would help researchers conceptualise revocation and engagement, and build them into their data-practices. The tool-kit was developed to encourage high-level thinking and stimulate an overall view of the project, rather than direct effort towards immediate implementation. Chapter 4 highlighted that participants wanted feedback, Chapter 5 indicated forms in which information could be provided, and Chapter 6 demonstrated that such feedback provides research benefit. These ideas were woven into the following materials to support other researchers in implementing their own feedback mechanisms.

The first tool is a model designed to conceptualise Dynamic Consent as part of developing research data-practices. The second is a framework to draw out the reasons for using Dynamic Consent and assigning metrics to those assumptions. Logic[38] models and frameworks can be useful in reporting and comparing projects, and have been references in Dynamic Consent literature (Prictor *et al*, 2020b). The example used to illustrate these tools is based on a fictional research team developing a project that collects questionnaire data over time and uses digital recruitment and data collection. The digital methods being used are: a project website, an online portal for participants, and twice-yearly participation reminders via email. These methods are supplemented by a research team that can be contacted directly over the telephone or via email, to accommodate barriers to participation. The team wishes to implement a Dynamic Consent process where participants to change preferences at any time, and receive (enhanced) feedback. The researchers also want to set up a participant forum to build rapport and collect participant experiences of the study. This is a worked example that draws from previous chapters in this thesis.

The strength of a model lies in its usefulness as a development aid: building shared understandings and common vocabularies, anchoring development and identifying the "next steps" of a project (Knowlton & Phillips, 2012). Figure 41 shows a model that has been designed for such conceptual use, with the expectation that the ideas developed are revisited, reflected upon and redesigned throughout a project's lifetime. The work required in the course of scoping, refining and creating the intervention study provided insight into practical barriers to the application of existing Dynamic Consent literature

[38] For this discussion, "logic" means the systematic ordering of ideas as opposed to mathematical organisation.

(Budin-Ljøsne *et al*, 2017). Figure 41 draws from the dual-stakeholder approach used in Budin- Ljøsne *et al*'s process model. The authors' model was created to indicate where Dynamic Consent could provide value within various research processes, and to support researchers in building their own data-practices drawing from Dynamic Consent. The model was overly complicated and the basic principles of revocation and consent did not seem obvious design priorities. I drew out key components from this model: researcher and participant perspectives and a focus on recruitment, informed consent and follow-up activities. To emphasise revocation and engagement, the new model prompts researchers to think about ongoing communication and consent management as long-term processes. Design was simplified, and illustrations added. To emphasise the focus on data-use, I also included questions to prompt reflection on the type of data being collected and how that information would be used, shared and controlled.

Figure 41 demonstrates how the research team might model Dynamic Consent while their project is in its formative stage. This team must consider the recruitment, informed consent and follow-up processes from their own point of view as well as that of the participant. This presents an opportunity to think about data-flows[39] and consider how digital tools will be used across the study. In this example, the team have identified that an online recruitment process poses risk. This risk does not necessarily need to be dealt with at this time, it is enough at this stage to simply identify the issue.

This model encourages the research team to consider the categories of data they will ask participants for, and what the management process for that information looks like. They also use this model to conceptualise ongoing communication and consent management throughout the project's lifetime – a point of reference and subject to further review as the project matures. Communication has been modelled in terms of inputs and outputs to emphasise the requirement to feed information back to participants. Consent has been modelled as having start, middle and end points rather than simply being a one-time contact point at recruitment. Under "data-use", the team has outlined the type of information they want to use, their reasons for that use, what data-use entails and a description of data-sharing procedures. All of this information can be adapted into lay terms for use on the participant-facing website[40].

[39] Recalling Nissenbaum's work mitigating privacy violations by defining "appropriate" data-flows.

[40] Meeting the requirements for "enhanced feedback" as demonstrated in the intervention study.

Figure 41: Modelling Dynamic Consent as part of research practice.

MODEL	Recruitment	Informed consent	Follow-up
 Researcher	**1.** Online recruitment via the project website. Link can be shared with patient groups, collaborators and other researchers. **2.** Offline recruitment via posters, clinical recommendations, etc. **3.** Online access widens the pool of potential recruits, but introduces security concerns.	**1.** Online: possible, if a potential participant feels comfortable giving consent online. **2.** Online/offline: potential participants can request a call back to discuss the study. **3.** Information sheets are available to read and download, and videos are available to watch on the project website.	**1.** There is a participant forum that participants can choose to enrol in (a form of ongoing communication). **2.** Individual feedback is not within current scope (subject to change). **3.** The project website provides collaborator summaries, and we will collate an annual review of achievements.
 Participant	**1.** Potential participants can be recruited by their clinicians, through patient groups, or by visiting the project website. **2.** Geographic location will not prevent someone from taking part in this study. **3.** People need to know that participation leads to real impact.	**1.** The consent process is under constant review, participant input is sought for this purpose. **2.** Consent preferences can be reviewed and edited via the online portal, for further details, see "consent management" below.	**1.** Questionnaire reminder emails will thank participants, reiterate the value of their contribution, and highlight a fun fact. **2.** Participants can see projects they have taken part in (that have used the data they have contributed) via the online portal.

Ongoing communication →

Input sought: there is an open invitation for participants to join a forum, who the research team will consult for their experience of the study and their opinions on current work and future research direction.

Output to feed back: project aims, what we collect from them, how this data is used, the value of their data-contribution, and project research outcomes.

Consent management →

Initial consent: potential participants are asked to provide consent and if they have any questions they can discuss the study with a team member.

Consent review: "edit consent" is an option on a participant's home page. They can review and change data collection, communication and participation preferences.

Revocation: participants can withdraw at any time, online or by contacting the team.

Data-use

→ Personal data and questionnaire responses will be requested from participants twice a year.

→ We will use this data to understand how rare conditions affect daily life.

→ The entire dataset will not be readily available—collaborators must submit a request for data-samples.

→ Data-access requests (scheduling check-ins) must be submitted to our data-access committee.

Frameworks offer structure for project reporting. They support systematic comparisons between projects with the same goals but different focuses and implementation (Frechtling, 2007). Figure 42 shows a framework that has been designed to prompt a researcher to define their own criteria for Dynamic Consent and to identify how each criterion can be measured. The work required in the course of scoping, refining and creating the intervention study provided insight into practical barriers to the application of existing Dynamic Consent literature (Prictor *et al*, 2020b). Figure 42 draws from the inter-linked ideas approach used in Prictor *et al*'s logic model framework.

The authors' framework was created to support researchers evaluate and report implementations of dynamic consent by linking assumptions of Dynamic Consent to project inputs, activities, outputs, outcomes and impact. When trying to apply the framework, I invested significant time and effort in translating the framework provided into the context of my work. I had assumed that the framework was ready to use in its current form. The Dynamic Consent principles of revocation and consent did not seem obvious design priorities, however. I kept the original headings and refined the framework so that it prompted researchers to fill these headings in for themselves, rather than providing examples that would be irrelevant to some studies. To emphasise revocation and engagement, the new model prompts researchers to think about ongoing communication and consent management as long-term processes. Design was simplified, and illustrations added. To emphasise the focus on data-use, I also included questions to prompt reflection on the type of data being collected and how that information would be used, shared and controlled. Revocation and engagement were not designed into this tool, as it helps researchers draw out and provide evidence for their own assumptions about Dynamic Consent.

Figure 42 demonstrates how the research team might map out their underlying assumptions about Dynamic Consent ensuring that they do not use it as a panacea. At the start of the project, the team would be able to complete the "Inputs", "Activities" and "Outputs" columns. In this example the research team have decided to explore whether their use of Dynamic Consent has led to increased participation in the study. As their research relies on participants submitting questionnaires, their "input" measure is the number of questionnaires being submitted before the activity and their "output" measure is the number of submissions afterwards. The "activity" itself is a website update. The team have created a video clarifying that the type of data being collected is questionnaire submissions and personal details provided at the point of consent, such as age and ethnicity. The video also describes that this data is used by the research team and limited access is granted to collaborators who have been approved by an ethical oversight board. The research team have added a new section to their website called "Project outcomes"

with lay summaries of research papers based on data from the study. The team have asked collaborators to create short videos describing the work they have done, even if it is a work-in-progress report. As their project matures, the team will collect and analyse data. As they do this and start to report their findings, they will be able to complete the "Outcomes" and "Impact" fields. One "outcome" the team might observe is an increase in the number of questionnaires being submitted. This example indicates that the activity described improves participation, but attributing this research benefit to Dynamic Consent may not be possible within the context of one study. As is often the case, more research is needed. That is what this tool has been developed to support: somewhat standardised accounts of Dynamic Consent.

Figure 42: Framing underlying assumptions of Dynamic Consent in research.

FRAMEWORK					
Assumption	**Inputs**	**Activities**	**Outputs**	**Outcomes**	**Impact**
Using Dynamic Consent increases retention	Questionnaire completion rates (categorised by start date)	Emphasise contribution value on website and reminder email	Questionnaire completion rates (categorised by start date	Participants who started earlier are represented in the data	To retain participants, emphasise data's value
Using Dynamic Consent increases recruitment	Pre-intervention questionnaire completion rates categorised by start date	Clarify project aims on website, reduce jargon and make it easy to sign-up	Post-intervention questionnaire completion rates categorised by start date	People who signed up recently are submitting questionnaires	To increase recruitment, communicate goals and make self-signup easy
Using Dynamic Consent increases participation	Pre-intervention questionnaire completion rates	Clarify data used, how it is used, and research outcomes	Post-intervention questionnaire completion rates	More participants are submitting questionnaires	To increase participation, communicate how data is used
Using Dynamic Consent increases feedback	Pre-intervention amount of feedback via the forum	Clarify how the forum contributes to the study and make sign-up easy	Post-intervention amount of feedback via the forum	More participants are providing feedback via the forum	To increase feedback, describe its value, make it easy to do and show its impact
Using Dynamic Consent increases value to participants (understanding)	Interviews with participants indicating level of understanding	Website, email and questionnaire descriptions re-written to be accessible	Interviews with participants show increase in level of understanding	Intervention has led to a better understanding of the study	Engaging research practices (such as DC) are valuable
Using Dynamic Consent increases value to researchers (richer data)	Interviews with researchers indicating level of data-use	Intervention measuring effect of feedback on data collection	Interviews with researchers show increase in level of data-use	Intervention has led to richer data collection	Engaging research practices (such as DC) are valuable

7.4 Implementing Dynamic Consent

In theory, Dynamic Consent relies on revocation and engagement (Casassa Mont *et al*, 2012; Kaye *et al*, 2015; Teare *et al*, 2015). In practice, examples of engagement are sparse (Javaid, 2017), but starting to emerge in greater detail (Haas *et al*, 2021). We will come to Table 54 presently, that illustrates a specification for Dynamic Consent. Before we do, however, we will briefly explore my analysis steps and earlier validation work (Schuler Scott *et al*, 2019b). The specification was developed to guide design and provide practical examples of how to implement Dynamic Consent[41]. The same fictional use-case is used as a demonstration.

This specification was designed to help researchers acknowledge and address the limitations of one-time consent, namely that participants agree to vague future uses of data which they do not expect to be told about (Teare *et al*, 2015; Whitley *et al*, 2012). A design feature of this specification is that researchers are encouraged to show trustworthiness (Manson & O'Neill, 2007) by demonstrating accountability and transparency (Aitken *et al*, 2016). This may help them build public trust (Williams *et al*, 2015), perhaps retaining participants over time (Sullivan *et al*, 1996). The specification prompts reflection on inclusive and responsive practice. Researchers should seek participant input because this demonstrates reciprocity (Gottweis *et al*, 2011; Hobbs *et al*, 2012), improves general awareness (Ludman *et al*, 2010; Riordan *et al*, 2015) and builds trust (Lipworth *et al*, 2009). Research participants want to know how their data has been used (Sanders *et al*, 2014), so the specification also prompts researchers to think about feedback mechanisms.

The specification provides eight prompts, each of which is linked to several suggestions for implementation. These are provided to stimulate discussion, or reflection, where researchers are working alone. The prototype version of this form was a design aid, consolidating Dynamic Consent literature, created to build trustworthy research processes (Schuler Scott *et al*, 2019b, see Table 50). Developed before the intervention study, this list had no grounding in practical work. Since carrying out the intervention study, the list was updated to reflect a more pragmatic approach. I also re-wrote the tool to assume that the researcher has no knowledge of Dynamic Consent, which is why examples are provided. Table 53 illustrates how researchers are encouraged to think in the longer-term rather than apply a vague notion of Dynamic Consent, and the examples are grounded in the enhanced feedback structure developed over the course of this work.

[41] As with the model and framework, a blank template can be found in Appendix 12.

Table 50: Initial v. current framework iterations.

Previous work (Schuler Scott *et al*, 2019b)		Current work	
Design prompt	Implementation	Prompt	Examples
What will you feed back to participants?	☐ Information about the research process ☐ Where their data is used ☐ Who their data has been used by ☐ Parties data is shared with ☐ Other	What feedback will participants receive throughout the project?	☐ Project aims ☐ Information that has been collected ☐ How data is used for research purposes ☐ Participant contribution ☐ Research outcomes

The first prompt, "Why use Dynamic Consent?", asks the researcher to think about why they are using Dynamic Consent and what they expect it will help them to achieve. In this example, the team is not concerned with saving resources, they are expecting their dynamic form of consent to enrich data collection. Prompt two, "Who are the project stakeholders?", suggests that the researcher identifies parties with a vested interest in the success of their project. In this example, researchers, clinicians and participants commit to building and taking part in the study, to varying degrees. The general public can be described a stakeholder in the long-term, if the researchers go on to use their findings to create impact of some kind, but this is not a design priority at this point. Prompts number three and four focus on recruitment and informed consent processes.

The examples provided here are a list of features that the team may choose to implement, or reflect on before developing their own, as demonstrated. In this example, online recruitment means that participants are not limited by their location, and the process is standardised. If there are changes that need to be made, the team will need to think about how they manage this. For example, if the team adds a post-mortem option to the consent form ("if I die, withdraw my details from the study") they may choose to email all participants who were not asked that question when they joined. Change management is out of scope for this work, but these tools are meant to be revisited and updated throughout a project's lifetime. The team has not chosen to provide participant information in different formats, because it will be available on the website. This team

will be working with people who require video transcripts and large print options, so this is part of their standard practice.

Another team might consider these methods to be "various information formats". "Consent management" provides another list of options, and prompts six and seven have been designed to draw out exactly what researchers want from their participants. Research findings will be based on questionnaire data. The team want to identify co-design opportunities and ask participants about their experiences of the study at monthly participant forum meetings online. They have also decided on the type of feedback they plan to provide about the project – this information will be communicated on their website in lay terms.

Table 51: Specification for building Dynamic Consent into research practice.

Prompt	Examples
Why use Dynamic Consent?	☑ To inform over time ☑ To improve recruitment ☑ To improve participation ☑ To improve retention ☐ To save time ☐ To save money
Who are the (committed) project stakeholders?	☑ Researchers ☑ Clinicians ☑ Participants ☐ Public services
Recruitment process (drawing from a Dynamic Consent approach)	☑ I want to standardise recruitment ☑ I do not want geographical limitation ☐ Recruitment will be online ☐ Recruitment will be partly online ☑ People can express interest online and fully consent. If they have questions we will call them to discuss. ☑ Online recruitment will be one option, people can be recruited in-clinic and over the telephone.

Informed consent process (drawing from a Dynamic Consent approach)	☐ There will be various information formats (But participant information sheets will be available online, and a project overview video + transcript) ☑ Consent records are viewable online ☑ Informed consent will be online ☑ Informed consent will be partly online
Consent management (drawing from a Dynamic Consent approach)	☑ Electronic authorisation ☑ Standardised access to preferences ☑ Secure record storage ☑ Secure record access (researchers) ☑ Secure record access (participants) ☑ There is a revocation process
How do participants take part in the study?	☑ Providing questionnaire data online ☐ Providing feedback online (as part of forum)
How do participants take part in the research process?	☑ Co-design with participants ☑ Participants provide research process input ☐ Participants are on an oversight committee
What feedback will participants receive throughout the project?	☑ Project aims ☑ Information that has been collected ☑ How data is used for research purposes ☑ Their contribution ☑ Research outcomes

7.5 Validation

To evaluate these tools and validate the methods used, informal interviews were carried out with researchers asking whether the tool-kit was relevant or useful. For the document sent to experts ahead of time, see Appendix 13. The first interview was carried out with a qualitative researcher (V1) whose previous research examined collaborative practices in multidisciplinary healthcare teams. Her current work focuses on understanding and improving communication practices in healthcare settings, aiming to develop an evidence base of helpful and effective ways to deliver lifestyle advice. The second interview was carried out with a senior qualitative researcher who was also a programme director for a research funding stream. Her current work demonstrates an interest in how

people use the internet in relation to their health, and she uses social science informed qualitative methods.

V1 reported that this work had a valuable premise and was especially relevant in the context of healthcare. She described research challenges that she had faced, that the tool-kit could help others to address. Namely, a lack of ongoing conversation between researchers and participants, boring and dull consent methods and a tendency for researchers to overthink ethical requirements and over-complicate messaging. The expert also cited a lack of participant involvement:

> "In healthcare… [you are] rushing around and you're trying to chat/informed consent them. You give them information and sheets but are they paying attention?", V1.

V1 suggested that there is a need for research following on from this thesis. Longer-term work that observed the number of participants returning to RUDY's website once they had been consented onto the study, perhaps. While agreeing that "it's good to keep in touch", V1 said that more information was needed on whether participants were making use of the changes made to RUDY. V1 drew parallels between this and their experience on a 3-year study of pregnancy where she had needed to follow-up with participants over the telephone. V1 echoed a concern of this thesis regarding impact: applied research must be realistic. Her concerns related to participant burden, the trade-off between data protection and collection, and administrative research overhead. Regarding the first of these, V1 expressed concern that a long list of consent options overburdens participants. This concern was shared by the RUDY team in Chapter 4. To address this, the tool-kit created offers design prompts rather than an exhaustive list of "magic sentences" (Whitley *et al*, 2012), prompting researchers to think about what they are asking of participants, and why.

V1 was concerned that freedom to withdraw from a study would result in participants withdrawing, which could devastate research findings. She had recorded interviews in the past and quickly anonymised them, meaning that if a participant withdrew from her study she would not have been able to remove their interview after a certain period of time. My response to this was to suggest that researchers who design research processes must communicate in clear terms what participants are agreeing to. If the consent form states that participants have three weeks after the interview to withdraw entirely, then that provides a reference point for any further discussions. Such communication is not standard research practice, which tends to over-abstract data-sharing choices in a needlessly dictatorial fashion (Agrafiotis *et al*, 2010a; Ram, 2008). V1's final concern related to practical elements; how much time and resources would use of this tool-kit incur and could this work be outsourced? The design prompts in this tool-kit have been

designed to provide structure for those who have no experience in participant involvement, or those have some experience and want guidance. V1's final point was that research priorities must be supported by any tools that are used, because there is a risk of distraction and over-complication. My response to this was that the tool-kit has been designed to provide structure and support where necessary, and points of reference for further discussion.

> "Front-loaded consent doesn't seem to work for anyone. But it's a requirement", V2.

V2's research work involved collecting interview data for wider dissemination. V2 described how this process contained multiple contact points: researchers contacted participants before arranging an interview to summarise what would happen and answer any questions they might have, interviews were scheduled and carried out, and participants reviewed the interview output. V2 worked with a charity that published extracts of these interviews, and summaries of findings. As interviews were open-ended, consent was given under the expectation that the participant would review the recording or transcript. Between interviews and publication, participants could contact the research team and ask for material to be withdrawn, and at the end of the project, a celebration event brought participants and researchers together. Her description of this work process suggests high engagement:

> "There have been about 5000 interviews collected in this study and fewer than 5 people have ever withdrawn. This relationship [between engagement and participation] must be important", V2.

V2 reported that the work described in this thesis would have the highest impact during research design. In her capacity as a program director she did not think that there was a tradition of applicants needing to explain processes for retaining participants through engagement over time. Reflecting on recent funding applications:

> "I was looking at some final reports this morning and about a third of participants were lost to follow-up. Loss to follow-up is expensive… inefficient and may end up really biasing study conclusions", V2.

V2 described that, as a funder, she thought there were good reasons that researchers should consider using Dynamic Consent processes. She then reflected on research trends, specifically how PPI is now a standard part of practice, with members of the public on panels to provide their thoughts on taxpayer spending. It would, V2 argue, be inconceivable not to include PPI. The tool-kit would be useful in helping researchers build their cases for support when trying to persuade funding bodies to invest in their work. Such a tool could be used to demonstrate that engaging with their study would not

overload participants, would be appropriate to ethical requirements, and a continuous process throughout the study, Researchers could show that they wanted to lower the cost of participant loss by building methods aimed to keep people in the study. Using the tools to provide outcome measures would show that the applicant had carefully considered their research design for clarity of reporting and replicability. V2 described how research culture had recently shifted around three issues in particular: towards following patient needs, ethnic inclusion, and translational work. She suggested that if researchers were furnished with a research problem such as participant attrition, and tools to fix that issue such as the ones offered in the tool-kit, then those tools would be used. This evaluation provided an informal review of the tool-kit. Feedback from practitioners indicates that these tools are needed and would be valuable.

7.6 Reflection

Three tools were presented, developed for the purposes of implementing Dynamic Consent; a model, a framework and a specification. In choosing to use Dynamic Consent, a researcher commits to underlying principles of revocation and engagement but historically, engagement has been underserved in Dynamic Consent literature. The tool-kit was created to support the development and reporting of engagement in research practice, and the use of Dynamic Consent. This work directly relates to the third research aim of this thesis:

RQ3a: How can Dynamic Consent be described, implemented, measured and reported in research?

The description, implementation, measurement and reporting of Dynamic Consent rely on a united, rather than a modular approach. To describe why Dynamic Consent is being used, a researcher subscribes to its basic principles (Kaye *et al*, 2015) and must decide how this method contributes to their work (Budin-Ljøsne *et al*, 2017; Prictor *et al*, 2020b). This provides metrics, whether it is questionnaire completion rate or the number of recommendations made by participants that have been actioned by the research team. Knowing the expected impact of Dynamic Consent practices means that a researcher can implement these methods, observe the result, measure success and report findings. Such reporting provides a point of reference for the researchers responsible, and benefits the wider research community through publications, conferences and shared practice.

RQ3b: What kind of tools can support these processes and accommodate knowledge-sharing among researchers implementing, or trying to implement, Dynamic Consent?

A model was created to stimulate discussion at an early stage of project design. A framework was developed to encourage researchers to draw out their assumptions of

Dynamic Consent and develop metrics for measuring its impact. A specification was designed that provided examples of how these design choices could be implemented. These tools provide a standard template that could be put to use in different contexts – the use of these tools in the sharing of knowledge provides a rich area for further development but on further reflection lies outside the scope of this project.

The tools provided in this chapter draw from academic literature (Budin-Ljøsne *et al*, 2017; Casassa Mont *et al*, 2012; Javaid *et al*, 2016; Kaye *et al*, 2015; Prictor *et al*, 2020b; Teare *et al*, 2015), and the motivation for this work is taken from the demonstrable value of engaging research practice discussed in earlier chapters. Another motivation for researchers to seek tools to help them develop Dynamic Consent is to provide a more accurate and representative record of consent (Schuler Scott *et al*, 2019b), which is sorely needed in research where researchers must demonstrate their respect for participant needs (Manson & O'Neill, 2007). These tools have been designed to aid in so-called ante-hoc experiment design (Michie *et al*, 2009) and the measurement of implementation outcomes (Proctor *et al*, 2011). In the field of Dynamic Consent, the importance of revocation and engagement at design stages has been overlooked in favour of basic information controls (Elsum *et al*, 2019; Mamo *et al*, 2020; Norval and Henderson, 2019). This is especially important as, even for information communication specialists, there is little guidance available regarding the development of technologies that engage with users over time (EPSRC, 2018; ORBIT, 2018).

The tools presented in this chapter have been designed as a starting point from which to develop research conversations around consent practices, and how to engage with participants in a practical sense. As with most technologies, these tools do not offer a solution in isolation because their value lies in how they are applied.

Chapter 8

Thesis conclusions

"A key factor that may hinder patient engagement is the patient's perception of their role and status as subordinate" (Valderas *et al*, 2016).

8.1 Introduction

The purpose of this thesis was to explore the use of Dynamic Consent as a mechanism for privacy control. The work described in Chapters 4 to 6 showed that providing feedback as part of research data-practice enriched data collection. This finding had data-protection implications for researchers as it addressed a key failing in current practice: informed consent is unfit for purpose because participants are asked to make a one-off decision about data-use over time. Participants are asked to agree to unknown future uses of data, with no assurances from researchers as to what those uses might be. Through the development of an enhanced feedback intervention, this work has shown that making information available and accessible increases research participation and reduces drop-off in participation over time.

This chapter draws out the implications of findings from previous chapters to present the overall contributions of this thesis. The planned contributions were practical and theoretical. In fulfilling the research aims described below, three further contributions

were realised: an empirical justification of the research value of engagement, evidence to support the methodological value of co-design, and a value proposition: enhancing feedback is valuable to both researchers and participants. This final Chapter provides a description of each contribution, a critical review, and directions for further work.

8.2 Summary of research aims and findings

This work was driven by three research aims (Section 1.4). Firstly, to observe and evaluate an existing implementation of Dynamic Consent, identifying conflicts and similarities with academic literature. Secondly, to extend the theory of Dynamic Consent by providing empirical evidence strengthening the research requirement for engagement. Thirdly, to develop practical guidelines for researchers seeking to implement their own form of Dynamic Consent.

The first research aim was addressed in the description of similarities and differences between RUDY's implementation of Dynamic Consent and Dynamic Consent as described in academic literature. The project had an online portal through which participants could edit their information-sharing preferences and consent choices. Participants could withdraw consent by emailing the study, but there was no evidence that this was common behaviour. The project recruited motivated participants to take part in a patient forum, regular online meetings with RUDY researchers to discuss upcoming work, and to provide feedback on the study. Input from the patient forum had been invaluable, resulting in changes to the project, such as the addition of post-mortem data-sharing options, and research direction.

By all accounts, RUDY was a successful project that collected data from people with rare conditions over time, and demonstrated a participant-centric research approach. The project was an example of good practice for longitudinal studies collecting biomedical data, and this success was hard-won by a dedicated team who had worked together over a number of years. Additionally, 50 out of 52 participants interviewed reported that they enrolled for positive reasons (to help others, to help family members or to fulfil a civic duty etc.) and trusted researchers to act in their interests. Despite this, participants could not describe how their data contributed to the study. This was important, because participant data-contributions (questionnaire submissions and ideas concerning research direction) are fundamental to RUDY's academic impact. The dataset that RUDY used and allowed collaborators to access consisted of data that participants had chosen to provide. This simple message had been lost.

The second research aim focused on the importance of engagement, a core component of Dynamic Consent. An intervention study co-designed with participants and

researchers demonstrated that engaging data-practices enriched data collection. This finding justified the need for engagement, demonstrated in Dynamic Consent literature. Initially, analysis of data from participant interviews, and the researcher focus group, showed no evidence that information communication would have any impact on participation. Participants had expressed curiosity about research progress, but reported that they had completed and submitted questionnaires and demonstrated no requirement for feedback of any kind.

Requirements for enhanced feedback were elicited through focus groups, where participants suggested that there was information they wanted to know, but did not feel comfortable asking for. This highlighted a requirement for researchers to make information accessible to participants. RUDY did not need to create new information to produce the website updates, simply curate what they already held. The intervention explored the assumption that, to meaningfully engage with participants, research processes must make information freely available and easy to understand. This connected to the Dynamic Consent literature: for a dynamic form of consent to be realised, participants needed to be informed over time. Researchers did not have to disseminate information themselves, simply make it available and accessible. A communication strategy enhancing the information made available to RUDY participants was developed in collaboration with RUDY's development team. This communication intervention resulted in an observable change in participation.

The third research aim of this thesis was addressed by the tool-kit developed in Chapter 7. Findings from the intervention study indicated that more people submitted questionnaires and more participants returned in order to complete questionnaires after feedback was enhanced for clarity, accessibility and relevance. This demonstrated that making data-practices engaging enriched longitudinal research data. The tool-kit was created to support researchers in building engagement into their own research practices, and consisted of a model, a framework, and a specification for Dynamic Consent. These tools drew from academic literature, as well as from findings in previous chapters in this thesis. The model was created as a design prompt to draw out overall project aims and data-flows. The framework was created to encourage researchers to specify their reasons for using Dynamic Consent and identify metrics that would allow them to measure and justify these assumptions. The specification was designed to offer practical support in making research practice more engaging.

8.3 Contributions

Exploration of research aims generated theoretical, empirical, methodological and practical contributions, and a value proposition.

Theoretical

A critical analysis of contemporary literature citing Dynamic Consent showed a lack of reporting, and discussion, of engagement (Dankar *et al*, 2020; Mamo *et al*, 2020; Norval and Henderson, 2019). This was an important issue because developing a relationship with research participants is a key challenge area (Kaye *et al*, 2015). In wider research literature, current guidance on carrying out responsible research innovation showed a similar lack of reporting and discussion. Let us recall the 4P AREA framework that encourages researchers to think about process, product, purpose and people when they anticipate, reflect, engage and act (ORBIT, 2018). The guidance available for "Engage" contains significantly less supporting material than any of the other three: a key issue is that the section of this framework that describes the *purpose* of engagement is empty. This is indicative of a wider problem within research: engagement is often overlooked as a requirement.

Public engagement needs to happen as early as possible (Willis & Wilsdon, 2004), and should be designed into research processes as it is too complex to be an afterthought (O'Brien & Toms, 2008). The work described in Chapter 4 offered a definition of engagement, refined in Chapter 5, that combined participant interest, knowledge-sharing, and opportunities for active participation. A key finding was that active engagement required both inclusive and responsive practices (Stilgoe *et al*, 2013). Participant input must be sought, and researchers must demonstrate that they have responded to that input. RUDY researchers highlighted a need for participant engagement, and had developed mature, inclusive practices that sought participant insights. Responsive practices were lacking, which could have been used to demonstrate the depth, breadth and value of participant data-contributions.

RUDY's inclusive practices led to positive participant interactions, relatively high recruitment, and research processes tailored to participants. These methods were not responsive – they did not demonstrate that participant input had led to project changes. The majority of participants interviewed reported that they did not receive any feedback at all. Participants were unaware of key research goals and the importance of their data-contributions, volunteering their time because they felt a moral imperative to do so (Dixon-Woods *et al*, 2007).

Participants trust researchers. A further theoretical contribution of this work was a description of participant assumptions about researchers. RUDY participants expected to retain overall control of their data, delegating implementation of these preferences to researchers. Participants reported that they felt they had a duty to contribute towards research, recruit others and refrain from falsifying data. They saw participation as a contractual agreement that would be broken by a 'moral breach' on the part of researchers, or if they no longer thought the study was valuable. This work also provided a description of consent fatigue, identifying this as a research concern. RUDY researchers described consent fatigue as presenting participants with an overwhelming number of data-sharing choices, resulting in choice paralysis and disengagement with the decision-making process.

Empirical

One of the key findings of this thesis is that enhanced feedback mitigates drop-off in submission over time. The empirical contribution of this work is the demonstration that participant engagement provides research value. Chapter 6 demonstrated, through quantitative analysis, that participant engagement is an important part of practice for longitudinal studies collecting data online. Findings from this study indicated that feeding information back to participants about project aims, the importance of their contribution, and how data is used, reduced drop-off in participation over time and increased participation overall. The average drop in questionnaire submission rates over 2019 was 13.9%. These rates dropped over 2020 by only 3% and a comparison of post-intervention data to the same period in 2019 indicated a 5.2% increase in questionnaires submitted. The empirical contribution of this thesis is compounded by the longitudinal nature of the data collection. Findings were not based solely on pre- and post-intervention data collected during a national lockdown. This data was also compared to questionnaire submissions from the year before to explore behavioural disruption caused by the pandemic. This study was created as a service evaluation, illustrating a proof-of-concept: taking time to communicate information back to participants is a valuable research practice. This empirical work provides many avenues for further investigation, as indicated by the list of suggested hypotheses for future work.

An issue that the RUDY team pointed out early on was a loss of participation between initial and subsequent submissions. Over 2019, initial completions dropped by 32.2% and follow-up submissions fell by 15.2%. Over 2020, initial completions dropped by 8.9% and follow-up submissions increased by 4.5%. The most significant effect, however, was seen when comparing post-intervention data (2020) to the same period in 2019; initial completions increased by 29.9% from one year to the next and follow-up completions increased by 7.2%. This indicated that enhanced feedback affected participant retention,

which is an exciting finding. These findings speak for themselves and, more importantly, to the value of Dynamic Consent. This work empirically supported the importance and value of participant engagement in research (often overlooked, despite its research value (Teare *et al*, 2015). In justifying a need for engagement as part of research practice, these findings directly supported the theory of Dynamic Consent. Empirical support for engagement, however, does not unilaterally endorse Dynamic Consent. These findings are specific to an in-depth case study but further research is required to discover the degree to which they are more broadly applicable. The ideas developed here are likely to have some application in longitudinal studies; cohort studies, pain studies and other rare disease research. There are other types of studies that these findings will relate to less directly – cancer research, for example.

Methodological

The work described in this thesis made use of a novel, mixed-methods approach. Collaboration with RUDY provided an untried use-case for Dynamic Consent: an online research platform collecting questionnaire data. Dynamic Consent is usually cited in the context of biobanking where physical samples are collected[42]. RUDY is an unconventional example of an epidemiological study because data is collected directly from patients rather than about patients via a clinician (Section 4.3.2). The ontological grounding of this work led me to seek knowledge from different stakeholders to build a more accurate depiction of RUDY: from a participant perspective the study was not engaging, but the researchers who built the platform reported that their work was participant-centric. The disconnect between researcher expectation and participant experience drove the empirical investigation of the intervention study. Novelty is also demonstrated in the methodology underpinning this work, because responsible innovation and participatory research principles are embedded into its design (Section 3.2). They are then implemented and reported.

Reflecting on four of the six key RI principles: engagement was the subject under study; gender equality was considered but not part of the intervention; science education was provided as project updates to RUDY which were distributed to participants via social media[43]; and ethics approval was sought institutionally and directly from RUDY's data-access committee. The remaining principles of open access and impact on governance are aspirational at present, for this work. The relevance and acceptability of this work was ensured by seeking participant and researcher feedback regularly within the research

[42] Biobanking was one of three original use-cases for Dynamic Consent, as described by EnCoRe.

[43] Lessons learned from the software-development co-design work led to a public-engagement workshop for secondary school students, independent of this thesis.

process. Regarding participatory research, participants created the research agenda for this work, which I simply needed to refine and formalise by opening up areas for discussion. Input was sought from RUDY participants, RUDY researchers and other experts, as necessary throughout development of the research to improve relevance and validity of research outputs.

A key methodological contribution is that this work offers an example of co-design in practice. The online focus groups and software-development work, demonstrates that co-design resulted in research that was achievable within the allotted time, relevant to key stakeholders, and timely (Co.Create.Training, 2019; Dimopoulos-Bick *et al*, 2019). The improved relevance and validity mentioned above were due to my making a design choice early on in the development to seek input from RUDY's end-users. Participant focus groups resulted in previously unexplored insights, and seeking feedback on the findings of those calls confirmed that the proposed intervention directly addressed participant needs. These needs were tempered by recommendations from RUDY researchers, who knew what could be accomplished within the available time. The software-development process resulted in the final intervention and was validated during user tests. This also provided the research team with an opportunity to review their own working practice – identifying the need for a change-management process, for example.

The work described in Chapter 5 showed that co-design must be structured, flexible and invite user feedback (Goodyear-Smith *et al*, 2015). I sent information to participants ahead of time, describing tasks in full, clarifying the type of data that I would be collecting, and setting out ground rules of the interaction. I also communicated what participants could expect of me – I told them how often I would communicate with them, what the next point of contact was and how they could contact me directly.

Practical

This work offers an example of Dynamic Consent's implementation demands in terms of time, money and expertise (Kaye *et al*, 2015). Practical guidance, in the form of a tool-kit, was offered to guide other researchers in developing engagement into their own data-practices. The materials described in Chapter 7 were created to fill research gaps identified by the literature review, and their eventual design was based on findings described in earlier chapters of this thesis as well as existing Dynamic Consent theory (Budin-Ljøsne *et al*, 2017; Casassa Mont *et al*, 2012; Javaid *et al*, 2016; Kaye *et al*, 2015; Prictor *et al*, 2020b; Teare *et al*, 2015). The key research problem that this tool-kit addresses, as drawn from academic literature, is a lack of informed consent that reflects genuine preference and informs over time (Boulton and Parker, 2007; Joan M. Griffin *et al*, 2006; Ingelfinger, 1972; O'Neill, 2003; Schuck, 1994; Schuler Scott *et al*, 2019b). Expert

testimony highlighted the stage of research where this tool-kit may be most relevant and useful: funding applications. There is no requirement at present for applicants to explain how they will use engagement to prevent drop-off in participation over time. Chapter 7 offered tools to help researchers demonstrate efficient use of resources, long-term thinking, appropriate respect for participant needs, and reproducibility (Manson and O'Neill, 2007; Michie *et al*, 2009; Proctor *et al*, 2011). Such tools are especially important given current inadequacies in guidance for building engagement into research practice (EPSRC, 2018; ORBIT, 2018).

Research engagement can be difficult to define and implement. An important practical contribution of this work is the provision of a tool-kit to help researchers elicit their own requirements for engagement. Chapters 4, 5 and 6 offer an example of this process: evidence was gathered showing that participants felt disengaged and desired feedback, ideas for communicating feedback were explored with participants and then the benefits of providing feedback were demonstrated. The work described in this thesis offers methods that can be copied or adapted, such as the multi-stakeholder interview process and co-design. The tools support initial project design as well as change-management processes during an ongoing study, as demonstrated by this work, during an ongoing study. Such changes may be cheaper to implement the earlier they are considered, e.g., during ideation or ethical review. The guidance offered in this work has been designed to be flexible and easily aligned with development goals. Several desirable research-project attributes have been identified: projects must provide feedback, their requirements may change over time, and they should have low administrative overhead where possible. Also proposed are several design principles for inclusive and responsive data-practices. Such practices must be open to improvement, draw from responsible innovation and co-design, stay human-centred and focus on data-flows. Regarding Dynamic Consent, this work has focused on helping researchers conceptualise key principles of revocation and engagement to build them into data-practice.

Value proposition

This work has shown that providing feedback is important to participants and beneficial to researchers. In other words, the engagement process is valuable to both parties. This justifies engagement as part of the wider research process and reiterates the need for a social contract between the scientific community and society (Gibbons, 1999b; Meslin & Cho, 2010). The intervention study described in Chapter 6 provided researchers with richer data collection and demonstrated operational and informational transparency (Aitken *et al*, 2016) by describing study-specific information in clear terms. The study was designed to provide information validating participants' commitment to taking part and encouraging further involvement. This was a direct response to participant reports;

participants did not know how they contributed to the study and felt ignored, despite the research platform relying on online questionnaire submissions as a primary source of data. The enhanced feedback intervention articulated research goals and vision, demonstrated transparency and invited the public to get involved with the scientific process. This work offers an example of how researchers can demonstrate trustworthiness to engender the public's trust (Aitken *et al*, 2016; O'Neill, 2013).

This value proposition is especially important, as the Internet and digital tools are increasingly used to run large-scale research trials. Where these trials are run over long periods of time, participation may be inconsistent and the engagement process may need to account for disengagement and re-engagement. The value that engagement provides can be used to inform development of data-practices within research, like recruitment, informed consent, and feedback. These practices will only be valuable if researchers choose to implement them.

8.4 Critical review

This research was novel, and provided valuable insights, despite its limitations. Opportunities have been identified for empirical rigour and wider applicability:

Empirical rigour

Analysis of intervention data simply provided a comparison of completion rates. A deeper analysis of this information, using statistical tests to identify the power and strength of these findings, would provide further insights. As this work was largely a proof-of-concept, this depth of analysis was not prioritised. A further area for development would have been the use of surveys, asking RUDY participants about their experiences of the study before and after the intervention. This could have drawn from the interview scripts in Chapter 4, and provided standardised responses for analysis pre- and post-intervention. No interviews were carried out with participants after the study to gauge whether their experience of the study was different post-intervention. Unfortunately, the scope of this project was limited by resource constraints. I initially made provisions for developing a survey, but this would have required further recruitment and additional ethical oversight which would have needed clinical approval due to RUDY participants' patient status. As this thesis focused on data-use and communication, rather than RUDY's clinical context, survey development was abandoned in favour of data collection from participants whose records showed valid consent for data to be shared with approved RUDY collaborators. Finally, analysis of intervention data did not stratify by communication type (i.e., website changes, questionnaire descriptions and changes to the reminder email). I initially designed the study to do so, but the eventual study would

have exceeded the time requirements. Development capacity was also a concern; a complex intervention would have required more support from the RUDY development team, which was not available.

Wider application

This work addressed a specific use-case: online longitudinal research into rare genetic conditions. Findings generated in the course of this work, while indicative, are not widely generalisable. Based on the evidence provided, this work offers guidance to those outside this research area, but cannot provide any guarantees. Further work is needed to measure the impact of enhanced feedback on participation in other research areas. A further limitation is that, despite the title of this thesis, the research that has been carried out cannot support any of the broader claims around Dynamic Consent: that its use promotes scientific literacy, lets researchers target and recruit participants and can be tailored for specific situations (Kaye *et al*, 2015). One further area to highlight is the absence of input from patient advocacy groups, which might have provided insight on broader participant requirements. An invitation to take part in the online focus groups was sent, via RUDY, to patient groups but none attended.

8.5 Directions for future work

More work is needed to standardise the evaluation of projects that use Dynamic Consent (Prictor *et al*, 2020b), and further validation is required for the tools that have been created over the course of this research. The findings presented provide initial claims of the importance of engagement in research, and these claims must be substantiated if they are to retain their strength and purpose. There is an entire research area, related to this work, that also lies entirely out of scope and has remained untouched so far – revocation. There is evidence that informing participants about their revocation rights leads to more nuanced decision-making (Agrafiotis *et al*, 2010a), and my work has outlined conditions under which participants might revoke their consent (Section 4.3). Further empirical work is needed to explore consent decision-making, and measure observed behaviours. Further avenues for investigation include other Dynamic Consent challenges: culture change, partnership development and new technologies for operational control (Kaye *et al*, 2015).

Participants initially reported intrinsic motivations for taking part in RUDY, such as a sense of duty. The enhanced feedback offered an external motivator, emphasising the importance of participation and justifying the trust placed in the research team. This work could be construed as a form of positive reinforcement for 'good behaviour', meaning there is plenty of scope for further research into choice architecture in research, and in

general. Future work on making data-practices in longitudinal research engaging should expand on outcome measures. More generally, future work should focus on reporting the impact of engagement on research practices, especially over time. This thesis has provided somewhat of a theoretical evaluation of engagement, and indicated that engagement is related to retention. This could be expanded to experimental comparisons. The implications of this work are serious, because they assert a need for trustworthy research. I also claim that researchers can demonstrate trustworthiness by re-thinking how they engage with participants over time.

8.6 Reflection

I have taken two key learning points from this work. The first addresses the practicalities of research, and the second is more of a philosophical development in my approach. First, engagement requires investment from both parties, but this relationship requires further development effort from the party with more power. The entire premise of this research was predicated on total honesty from researchers willing to accept criticism, and from participants willing to renew their engagement with work they felt disengaged from. Power to change the situation lay with the RUDY research team, not with participants. In seeking truly informed consent, that genuinely reflects people's preferences, researchers need to prioritise information that is relevant to participants. This information should be provided in a way that participants can understand, and consent choices must be developed to avoid coercion. Researchers can ask participants what information is relevant to them, find out how they would like to receive information, and check their understanding of the project. To reduce the participant burden here, there is, of course, a wealth of relevant literature describing participant views on these matters.

As demonstrated in this work, even researchers seeking engagement may find their participant population disengaged. While Dynamic Consent provides no guarantees, its longer-term approach to relationship-building equips researchers with tools that they can adapt to suit their needs for richer data collection, and more impactful work. Second, there are shockingly few requirements for researchers to describe how they will engage with either their participants or the general public. The tool-kit was designed to directly address this omission in practice. This work showed that meeting participant communication needs improves data collection over time. The use-case of an online, longitudinal, publicly funded, biomedical study demonstrates that these findings apply in an area where low-retention, high-attribution, and a general lack of understanding are serious concerns. Engagement mechanisms would be easier to critique during ethical approval, and easier to implement if they were part of initial design. Moving forward, I

will take these lessons into every venture, identifying and negotiating the needs of multiple stakeholders in my approach.

The co-design work, described in Chapter 5, was carried out during a national lockdown due to the Covid-19 pandemic. The online focus groups took place during April and May 2020, a few weeks after travel restrictions and a stay-at-home order had been announced. Understandably, recruitment was low as RUDY participants had other concerns. However, as research methods had been designed to operate online, the pandemic did not prevent focus groups, or intervention development, from taking place. The software development took more time than it might have done in other circumstances, as the entire RUDY team needed to establish a work-from-home protocol. During this process, I was informally advised that participation had fallen over the lockdown period. This underscores the importance of this thesis' findings – that enhanced feedback encourages participation. Further work is needed to explore the full impact of the pandemic, but this work offers evidence that clear descriptions of participation opportunities, and the provision of feedback, encourages engagement despite difficult circumstances. The response to Covid-19 shaped this work in its latter stages due to the mass-adoption of remote working. This emphasised a need for asynchronous communication and making information available and accessible to those who need it. This was due to differences in work-patterns and availability.

8.7 Concluding remarks

Consent and privacy options establish what participants can expect of researchers, but such processes are designed to inform, rather than educate (Dixon-Woods *et al*, 2007; Ingelfinger, 1972; Ormond *et al*, 2009). This is problematic, given the nature of the social contract science holds with society (Gibbons, 1999b). This work explored how researchers engage with participants about data: demonstrating that communicating relevant information in a way that people understand holds their attention over time. This thesis holds recommendations for research practice, lessons for digital health, and suggestions for digital interactions across all sectors. Privacy policies are unfit for purpose (Graber *et al*, 2002; Mcdonald and Cranor, 2008), and informed consent is in great need of repair (Corrigan, 2003; Ingelfinger, 1972; Katz, 1977; O'Neill, 2003; Schuck, 1994; Skolbekken *et al*, 2005).

A great deal of the academic discussions surrounding informed consent focus on autonomy, but, in Chapter 4, participants did not want autonomy. Rather, they emphasised the importance of research purpose and trust (Hansson, 2005; O'Neill, 2002). Whatever someone's personal motivation to take part, the prevailing opinion was that researchers were best placed to decide on research implementation (Nissen *et al*, 2019).

This includes how data is used and shared. The design of informed-consent practices should focus on message content and communication, rather than participant autonomy (Manson & O'Neill, 2007). This focus introduces data-security as a requirement early on in the research development process. This work is very much applicable to other studies in two key ways and may help researchers think about security as they design their methodologies. First, studies seeking to use Responsible Innovation within their work can use this tool-kit to conceptualise how they will collect and use data from participants, and how they will inform the over time. Second, the tool-kit has been designed for anyone running or designing online longitudinal studies (e.g., UK Biobank, Medical Research Council cohort studies…) to pick up and use as part of the ethical approval process within their institution.

The public needs demonstrable evidence that research is in the public interest. This work offers a step-by-step guide to developing research data-practices that engage with people over time, showing that a project is trustworthy. Academic researchers enjoy a privileged position in society, where requests for personal data are fulfilled, usually with little to no expectation of feedback. Such feedback is valuable however, as it emphasises the value of participation, and serves as positive reinforcement. The central finding from this thesis is that researchers can enrich data collection by communicating five pieces of information to their participants: project aims, the type of data being collected, how that data is used and shared, the value of this data-contribution, and research outcomes.

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Appendices

Appendix 1: The roots of informed consent

Informed consent was created to protect people from harm, in response to atrocities committed in the name of medical research. A person's requisite freedom to voice agreement, un-coerced and based on the provision of useful information, was enshrined in the Nuremberg Code after World War II:

> "1. The voluntary consent of the human subject is absolutely essential.
>
> This means that the person involved should have legal capacity to give consent; should be so situated as to be able to exercise free power of choice, without the intervention of any element of force, fraud, deceit, duress, over-reaching, or other ulterior form of constraint or coercion; and should have sufficient knowledge and comprehension of the elements of the subject matter involved, as to enable him to make an understanding and enlightened decision. This latter element requires that, before the acceptance of an affirmative decision by the experimental subject, there should be made known to him the nature, duration, and purpose of the experiment; the method and means by which it is to be conducted; all inconveniences and hazards reasonably to be expected; and the effects upon his health or person, which may possibly come from his participation in the experiment.
>
> The duty and responsibility for ascertaining the quality of the consent rests upon each individual who initiates, directs or engages in the experiment. It is a personal duty and responsibility which may not be delegated to another with impunity", (Taylor, 1949).

Medical research provides countless examples of harm being caused by a lack of consent. Thirty years after the Nuremberg Trials, the United States issued the Belmont report. The report illustrated informed consent as a tool for direct application, rather than an ethical principle. The criteria for informed consent in the United States was re-written to focus on beneficence, justice and respect, with a practical emphasis on information, comprehension and voluntariness in demonstrating freedom of choice. This positioning underscored the importance of informed consent in medical research (DHEW, 1979) and was a result of studies that caused, rather than prevented harm: the involuntary sterilisation of poor women seeking birth control, black men being infected with syphilis and left untreated, and children being infected with hepatitis (Winslow, 2017).

Appendix 2: The privacy paradox

In 2001 researchers interviewed users about their internet use. Participants were asked about their general use of the Internet, their online shopping habits and community activities, and privacy emerged as a common theme (Brown, 2001):

> "… there was also a concern about their privacy. It is difficult to judge the severity of this concern however, since the participants were also happy to use store loyalty cards for only minimal gain. This is something of a 'privacy paradox'"

This 'privacy paradox' has spawned a considerable amount of empirical work (Norberg *et al*, 2007) and theoretical development (Kehr *et al*, 2015; Trepte *et al*, 2014), and worth addressing directly for two reasons.

1. The claim operates on fundamentally flawed assumptions – the privacy paradox may not be a paradox at all (Burkhardt, 2018), and was based on reported rather than observed behaviour (Kokolakis, 2017).
2. Second, the theory places responsibility for privacy protection on users rather than product developers.

Norberg *et al* emphasised the role of individuals as a front-line privacy control, declining to place any responsibility on the parties collecting personal data (Norberg *et al*., 2007):

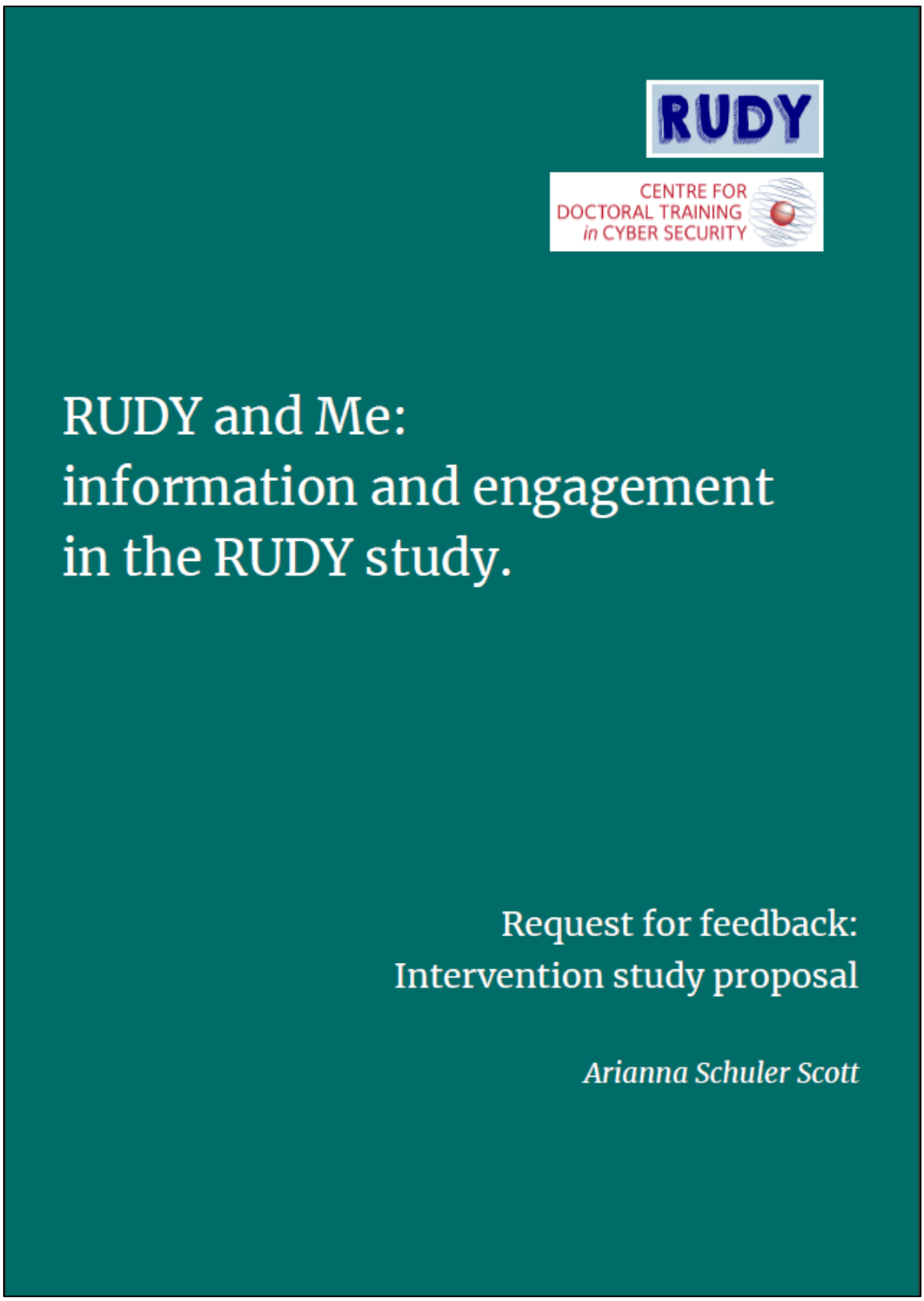
> "Unless consumers make the effort to truly understand what they are granting permission to, and to whom they are giving their personal information, their sense of personal privacy will continue to deteriorate… Enlisting consumers as the first line of defense to protect their own privacy may well emerge as the most efficient means to ease everyone's concerns with… data collection"

This position rests on the assumption that users can find information that will help them make privacy trade-offs. This is simply untrue, as these decisions are directly influenced by the interfaces users are presented with (Kehr *et al*., 2015):

> "… privacy assessment is influenced by momentary affective states… consumers underestimate the risks of information disclosure when confronted with a user interface that elicits positive affect"

Trepte *et al* criticised the simplicity of the privacy paradox's authors assuming that privacy could be measured as a single dimension, offering three: informational, social and psychological (Trepte *et al*, 2014). We must reverse-engineer the assumptions that the privacy paradox introduces, to re-think how users engage with data-sharing.

Appendix 3: Intervention study proposal



This document

1. **I think that participant engagement adds value to research, and technology can improve it**
2. **I would like to know what you think of my ideas for how the RUDY study can improve participant engagement.**

In 2018 I interviewed RUDY participants and the RUDY research team. I found out that the research team put a lot of development time and effort into designing the RUDY platform with opportunities for participants to feed into the research process, for example:

A. A group of patients (a"patient forum") who act as consultants for the research team.
B. A web portal that allows individuals to complete surveys.
C. Boxes for comments and error reporting on the website.

While RUDY participants are highly motivated to take part in the study, they do not know how important their contributions are.

Over the past few weeks I have run online focus groups to ask participants how this importance could be demonstrated or emphasised. My findings so far suggest that:

A. Important information could be put at the top of the website.
B. The email asking participants to fill out surveys could outline recent RUDY progress.
C. The online portal could describe how surveys are used by researchers.

You have this document because I would like your feedback on my ideas.

A. What are your thoughts?
B. What do you like?
C. What do you not like?
D. Have I missed anything?

Many thanks for your time, please let me know if you have any questions.

Arianna

Research findings (the headlines!)

Participants want to feel that their contribution is valued.

- Participants do not understand how their questionnaire responses are useful.
- Participants want to know about ongoing research and the overall status of RUDY.
- Participants want to know about research findings and condition-specific results.

Patient involvement is central to the RUDY study. My PhD work investigates the value provided by dynamic consent: granular control over data and participant engagement. RUDY's patient involvement model is an example of participant engagement and I want to run a study that measures whether or not engaging with research participants is valuable for research.

In online focus groups with RUDY participants I asked:

- What information do you want from the RUDY study?
- What information do you think RUDY wants from you?

My initial call was with members of the patient forum, the second was with non-forum participants. The first group wanted to **understand the research process** (how questionnaire data is used, and what it is used for, etc.), while the second group wanted to know **how useful their data was** (quote from the call: "What if a researcher comes up with something? We should be informed").

These focus groups confirm what I found in a series of interviews I conducted in 2018: RUDY participants do not feel engaged with the study, despite considerable research and development effort to provide ways to engage (i.e. a public-facing website and the patient forum.

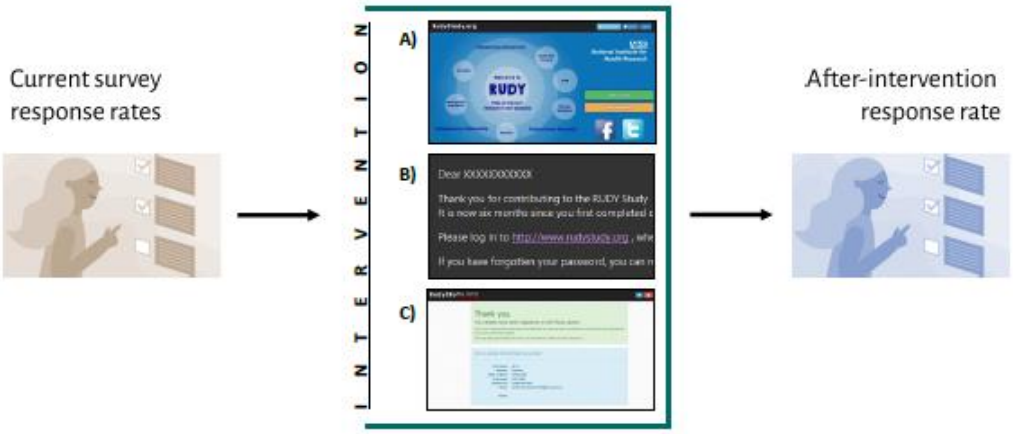
I would like to note that responses to RUDY are overwhelmingly positive. I have needed to draw out criticism and learning points ("I tell professionals about RUDY!", and "I feel like the team values us").

From the conversations I have had, these are the issues that prevent engagement:

- Participants do not know what RUDY's vision is.
- Participants are not sure how they are contributing.
- Participants only visit the website when they are prompted to fill out surveys.
- Participants find the surveys boring and repetitive.
- Participants are unable to identify with RUDY.

Intervention study proposal

I want to test whether enhanced feedback affects the number of RUDY surveys completed.



By "feedback" I mean feeding information back to RUDY participants. By "enhanced feedback" I mean the new ways to feed information back that I want to test:

A) Changes to the RUDY website

- Make a "current projects" section on the website and ask researchers for a video or quote.
- Put important information at the top of the website and make content easier to navigate.
- The number of participants in RUDY should be at the top of the website.

B) Email reminder signposting

- The email should include the number of RUDY participants (or with a similar condition).
- The email should include RUDY's vision ("helping us improve research in rare diseases").
- The email should give an example of a collaborator who used data from the surveys.

C) Survey description edits

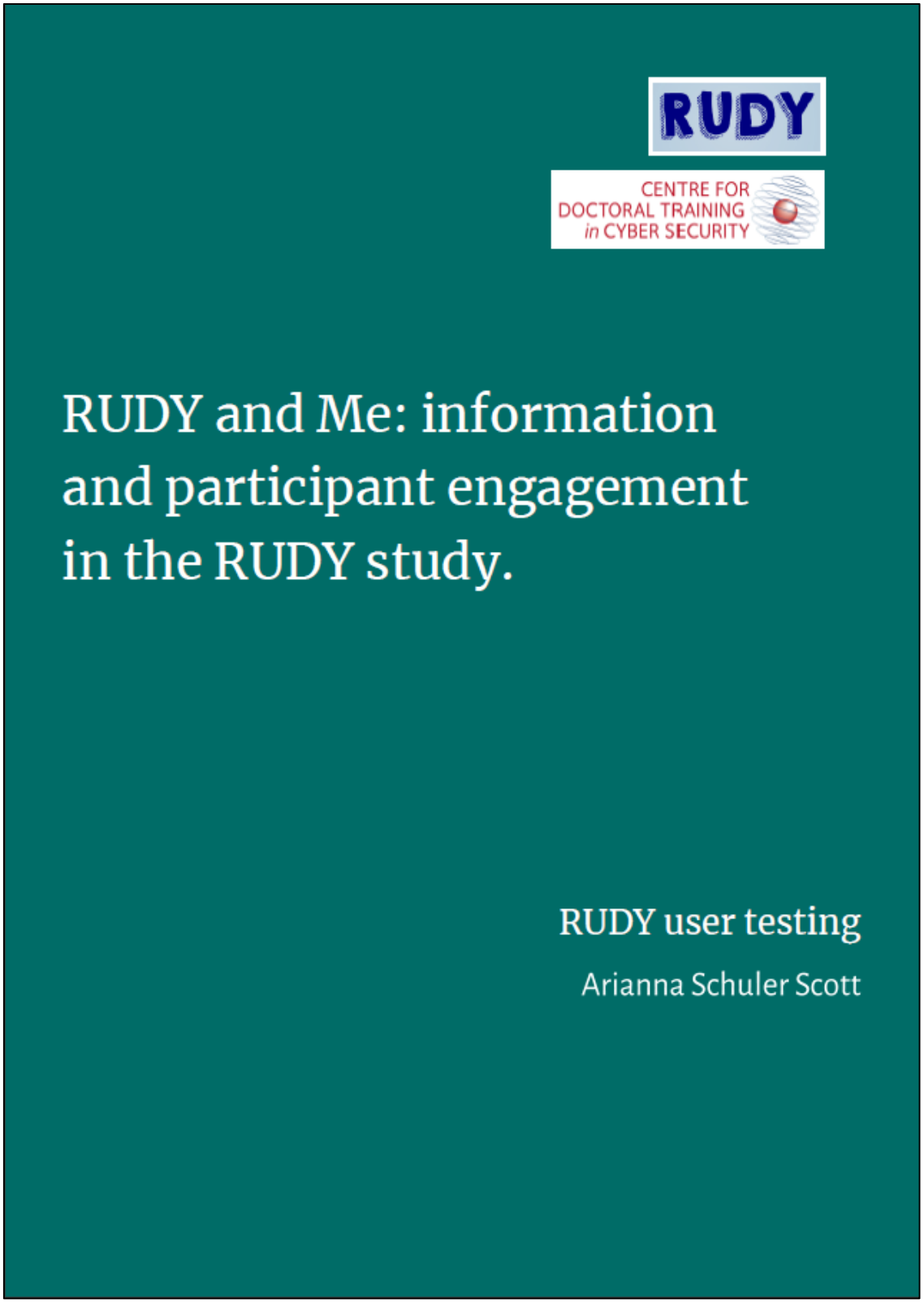
- Survey pages should give an example of how a RUDY collaborator has used that survey.
- Survey pages should describe how researchers use the data from surveys.

Appendix 4: Commentary on intervention proposal

Once I had submitted a list of proposed changes to RUDY's website, online portal and email reminder, these changes were passed to the software developers for comment. They would have a better grasp of how realistic these suggestions would be to implement, and whether they were feasible within the timeframe we were operating on.

Proposed change	RUDY development team	My comments
Arianna web site level changes		
Signpost the homepage with a subheading list in addition to sign up today and more information that allows fast scrolling – 1. animation that describes the study, 2. video that takes people through the registration and into the data collection and video that shows "how we use your data" 3. who we are / RUDY team 4. what our primary objectives are 5. what we have done with the data so far (summary videos), 6. active research collaborations (summary videos), 7. patient forum who, what and how to join / buddy system 8. links to patient groups by disease groups 9. update published paper library (Javaid *et al* 2016, Teare at al 2017, Javaid *et al* 2018 FD pathway),	1. Already present on RUDY 2. Fairly simple (and other people could create) 3. Easy 4. Easy – KJ can you provide some text? – yes I can do 5. Can do – KJ Do you have more detailed information on what we have done with the data? (perhaps a 'blog' or news page via homepage would be a good addition) – sure I'll draft one. 6. (+7) These are both more reliant on content from others. If we can be provided with text we can easily add this info to the site- sure, we will set up a reporting template, e.g., key points and format for the video 8. Possible – we just need providing with the details – will provide a table for patient groups. 9. Possible – we already have the relevant code – we just need the paper summaries and list of patient-id's that were used in the research	1. Brilliant, thanks! 2. I understand this to be a "how we use your data" overview. OK so need 2 videos 3. Brilliant, thanks! 4. Brilliant, thanks! 5. I love the idea of a "news" page with text and video updates from collaborators and RUDY. I think a blog would need more oversight (e.g., people to write blog posts) and might pull more resources. 6. I made a short video that could be used as an example – rather than prescribe what to do, I suggest giving prompt questions and a time limit (example below). 7. Not my area of expertise. 8. Not my area of expertise. 9. Not my area of expertise.
Add the number of recruited patients to the top of the home page – total and by disease group (allow duplicate counts)		

Appendix 5: User testing document



This document

Thank you for sharing your experience of the RUDY study. The RUDY team and I have been making changes and we would like to ask you what you think of them.

Your feedback is invaluable because you have been involved right from the start, and we want to know whether you think these changes are acceptable.

This session should take 30 minutes.

Getting input from people who are the target **user** of any kind of technology is important. This input is important because it gives technology makers an idea of whether what they have made works as intended.

The journey so far:

- I interviewed RUDY participants individually and found that they wanted to know that their contributions are valued.
- During online calls with participants, I found that participants are interested in understanding more about the research process and how RUDY uses their data.
- Changes have been made to do a better job of communicating information about RUDY to participants.

During this session we will:

1. Re-visit the **user testing info page**.
2. Work through the **user testing worksheet**.

Many thanks again for your time and attention,

Arianna

User testing page

This page describes what "user testing" is and what we would like you to do. Page 4 has the activity itself.

"User testing" gives an unfinished product to a small group of people, to check that it fits their basic requirements, before it is released into the wider world. A small group of shoppers may be given early access to an online shopping application (or **app**) so they can explore it, point out what works for them, and identify difficulties. Finding problems early saves time and effort.

You are being asked to test changes made to RUDY, because you helped develop these changes and have an idea of why they have been made. Changes have been made to the website and to the online portal. We would like to know three things:

- Can you can find these changes?
- Are these changes useful?
- Would you accept that these changes meet our original brief: "**to communicate useful information about RUDY back to participants**"?

Your feedback will be used to prioritise edits before these changes are made public. The kind of edits we can make at this point are small, such as:

- Phrasing tweaks.
- Rearranging text.
- Changing button links.

We will not be able to make all of the edits received, but if different people raise the same suggestion then this indicates that we need to address it!

When you are ready, please proceed to the **user testing worksheet** on page 4.

User testing worksheet

There are two parts to today's session. During the first you will be exploring changes to the website. You will then login to the test site. As you attempt the tasks below, bear in mind that all feedback is good feedback...

Which means that if you find anything difficult to do or understand, please point it out.

	Task description	Can you find it?
0	Go to https://research.ndorms.ox.ac.uk/rudytest	Yes / No / Not easily
1	What is the aim of RUDY?	Yes / No / Not easily
2	What does RUDY do?	Yes / No / Not easily
3	Who does RUDY work with?	Yes / No / Not easily
4	How many people are signed up to RUDY?	Yes / No / Not easily
5	What does RUDY do with participant information?	Yes / No / Not easily
6	What are some of the current projects going on?	Yes / No / Not easily
7	Is there any information missing from the top of the page?	N/A
8	Is the information you want to see easy to find?	N/A
9	Have you learned anything about RUDY you didn't know before?	N/A
10	**To be continued: we will go through the login process together as part of the call.**	

The point of these changes is to feed useful information about RUDY back to participants about the research process and how their data is used.

Do you think these changes achieve this goal?

This concludes the session,
thank you once again for your time and attention.

Appendix 6: Edits agreed with RUDY after user tests

Issue	Suggested changes	Project lead's recommendation
It is not clear that RUDY is a research project.	FROM: "Help us improve research in rare diseases" TO: "Patients and researchers working together for better rare disease care"	Tweak to "Individuals and researchers working together for better rare disease care" ~ not all people with rare diseases see themselves as 'patients'
The patient-centric nature of RUDY is not clear.	FROM "Sign up today" → TO "Join us now" FROM: "More info." → TO: "Find out more"	Agree
Key information is obscured by unexplained terms.	On "What is RUDY": FROM: "To determine a detailed **phenotype** of patients with rare diseases." TO: "To determine how different symptoms present themselves in patients with rare conditions" FROM: "To identify unique patient subgroups within each disease cohort" TO: I have no idea what this means—can you simplify it? FROM: "personal", "understanding" and "research cohort" being in bold. TO: the above terms not being bold.	use "To describe in detail how a rare disease affects individuals". use "To describe the differences between individuals with the same rare disease diagnosis" Agree
The landing page has to have all key information on it, or prompt people to read more.	Decide whether this is something that you have time to implement. The feedback suggests that attention is not captured and most people will not scroll down the page. Signposting further down the page would be good to have.	Agree add a scroll down icon under the "welcome to" section and move the twitter and facebook icons to below the How can I find out more? section.
FROM: "280 recruited so far"	TO: "280 joined so far", or "We have 280 people taking part"	Agree

FROM: "We have a lot of information about RUDY in the Library, that hopefully will answer most of the questions you may have. If you don't find an answer then please…"	TO: "Our Library will give you an idea of the kind of work we do as a research study. If there are any questions you can't find answers to then please... " AND: change "RUDY information" to "Library" on landing page.	Agree Agree
FROM: section "Which diseases are we interested in" has no information about diseases.	TO: move the list of diagnoses from "How far have we come" to "Which diseases are we interested in".	Agree
FROM: section title "How far have we come".	TO: title "What do we have going on?" or "Project news".	Agree
FROM: section "What is RUDY" does not say the video is different.	TO: include a description like "find out more from Dr Javaid, who leads this project", or "watch this video to find out more about what we do, and the benefits of taking part".	Use "To find out even more about what we do, including the benefits of taking part, watch this short video from the project lead"
FROM: section "What is RUDY" has erratic typography.	TO: make "Primary objectives", "Secondary objectives" and "Benefits of participation" the same heading level. AND: put the numbered points and bullet points at the same level.	Agree

Appendix 7: Interview structure for study 1

Section 1 of 5: introductory questions

A. Why did you join the RUDY study?

Please write here.

B. How did you hear about the RUDY study?

Please write here.

C. How long have you been taking part in RUDY?

Please write here.

D. What has your experience of the RUDY study been?

Please write here.

E. Do you use the information you enter in the RUDY forms for anything?

Please write here.

F. Have you ever withdrawn consent for information to be accessed or shared?

Please write here.

G. Is there anything that might make you withdraw consent?

Please write here.

Section 2 of 5: scenario 1

Manisha has a rare condition and has enrolled in a study at a local hospital where she can provide contact details, log symptoms and information about diagnoses.

She is largely housebound so a researcher enrols her over the phone. Once they have hung up, Manisha realises that she did not take the researchers name.

A. How could she get back in touch?

Please write here.

6 months after joining the study, Manisha wonders whether the researcher she had been talking to had collected enough people's information for them to do their work.

B. What might she have to do to find out?

Please write here.

C. What kind of information might she discover (what might you want to know of a study you took part in)?

Please write here.

Manisha is sharing her mobile number, email and symptom log and is happy to be contacted about other research trials. On the news, a 'data breach' at the hospital is announced.

D. What does ‘data breach’ mean to you?

Please write here.

E. Is a ‘data breach’ a problem?

Please write here.

F. What do you think the patient has a right to know?

Please write here.

G. What responsibilities do researchers have in this area, if any?

Please write here.

H. What is personal information and who does it belong to?

Please write here.

Manisha decides to withdraw consent for the study to access to her mobile phone number and share her information with other studies. She continues using the symptom log and after a couple of months allows the researchers access to her mobile number again.

I. Is control of your information and decisions made about you important to the delivery of your healthcare?

Please write here.

Section 3 of 5: scenario 2

Anna has signed up for a store credit card. Every time she shops at that store she collects points that add up to vouchers that she can spend in store. One day Anna notices that her credit limit has increased without her permission.

A. How could this have happened?

Please write here.

B. What should she do?

Please write here.

She decides to call the shop the next day but does not get round to it. Three days later she receives a call saying that a purchase had been made in another county that reached her (new) credit limit. The shop refunds her the amount.

C. Is this a problem?

Please write here.

Anna worries that her card details were taken via transactions she had made online or that her computer is infected with a virus that collects her information.

D. Do you think she is right?

Please write here.

E. Do you worry about providing data or interacting with people online?

Please write here.

F. What kind of information is important to be kept private or secure?

Please write here.

Section 4 of 5: finishing questions

A. When you think of RUDY who do you think of, if anyone?

Please write here.

B. Thinking about your involvement so far, how satisfied are you with RUDY and why do you think that?

Very poor								Very good
0	1	2	3	4	5	6	7	8
	9	10						

Please write here.

C. Do you trust the RUDY study?

Please write here.

D. If so, what do you trust RUDY to do?

Please write here.

E. Do you trust medical research?

Please write here.

F. If so, what do you trust medical research to do?

Please write here.

G. Has participating in RUDY been the right decision for you?

Please write here.

H. Would you make any changes?

Please write here.

Section 5 of 5: demographic information

A. What is your age?
- 18 - 24
- 25 - 34
- 35 - 45
- 45 - 54
- 55 - 64
- 65 - 74
- 75 - 84
- 85+

B. What is your gender?

- Male
- Female
- Non-binary
- Other: _____

C. How would you describe your background?
   - African
   - Caribbean
   - East Asian
   - Latino/Hispanic
   - Middle Eastern
   - Mixed
   - South Asian
   - White
   - Other: _____

D. What is the highest level of education you have completed?
   - Incomplete
   - Secondary school
   - College
   - Some university
   - Doctorate
   - Vocational qualification
   - Bachelor's degree
   - Master's degree
   - Professional degree
   - Other: _____

E. Are you happy for Arianna to follow up with a summary of this interview?
   - Yes
   - No

This is the end of the interview, your time is very much appreciated.

Appendix 8: Participant info sheets for study 1

CENTRE FOR DOCTORAL TRAINING *in* CYBER SECURITY

UNIVERSITY OF OXFORD

Arianna Schuler Scott
Doctoral candidate in Cyber Security
arianna.schuler.scott@cs.ox.ac.uk

An exploration of attitudes to dynamic consent in research

PARTICIPANT INFORMATION SHEET

PURPOSE

This study hopes to explore the relationship between those who collect, store and use information and the people who provide this information.

As someone who allows their data to be collected for medical or research purposes, we hope to ask you what you think about having control over information about you.

Dynamic consent is a term for a system like RuDY's (which you may be quite familiar with) because of the following:

- They ask for patient feedback about their experience as part of the project.
- They allow patients to withdraw consent for information to be shared.

WHY ME?

You are seeing this information sheet because it is likely that you are one of two kinds of people:

You have used a system that implemented dynamic consent.

You have been involved in designing or delivering a system for dynamic consent.

BACKGROUND

Those who collect, store, share and use people's personal data (referred to as 'data controllers') often have more control over how that information is used than the people providing it ('data subjects').

This study is the first step in exploring this relationship, asking about people's experiences and understanding of how their data is used.

AIMS

This study wants to find out what RuDY's version of dynamic consent does well by looking at two things:

1. Asking people who have used or developed the system about their experience.
2. Asking people about their expectations as to how information should be (or **is** currently) used.

DO I **HAVE** TO?

Put simply, no. But you can **ask questions** before deciding to take part, and you may withdraw yourself or your data **at any time** without a reason.

The only inclusion criteria for this study is that you have used a computer system that made dynamic consent happen in some way (This could mean being able to revoke access to your data, or that there was good communication around what was done with your information etc.).

If you withdraw at any point before the interview finishes your data will be deleted. If you withdraw once the interview is over, this will still be anonymised (only a number is associated with your interview transcript, not your name) and used in the study.

SO, WHAT NOW?

What happens to me if I take part? If you are happy to take part in this study, you will be asked to attend a single interview of about 45 minutes, and asked if you would be happy to follow that with a short 'check-in' email a few days afterwards:

Arrange an interview

Ask questions

Interview recorded

Keep in touch

A note on privacy: not everybody feels comfortable being recorded. You will be asked permission as to whether sound is recorded or not and quotes can be used.

RISKS

Consent can be a sensitive subject, especially when drawing from personal stories. You are free to take a break (or leave) at any time.

BENEFITS

This study is an opportunity to engage with research. We want to know about your experience. Questions are welcome at any time!

What happens to the data provided?

Research data is the information gathered from the interviews. Interviews will be anonymised (your name will not be associated with the gathered data). Recordings will be written up within 3 days and then destroyed. The transcripts and data that comes from them will be password encrypted on a password protected machine.

Personal/sensitive data is the information about you. This study will not put your name on a quote in public, but in order to look at the diversity of interviewees' demographic information will be retained. Only the researcher will have access to this. It will be password encrypted on a password protected machine.

Key points

- This study would like permission to use direct quotes.
- This study would like to use the anonymised data in future studies.
- All research data and records will be stored for 3 years after publication or public release of the work of the research.

LET'S DO THIS!

- Arrange an interview via this link: www.meetingbird.com/m/B1Zljltfm
- **OR** email: arianna.schuler.scott@cs.ox.ac.uk
- **OR** email RuDY to say you are interested!

LET'S TALK ABOUT RESEARCH

Will this research be published?

Initial results are expected by August 2018, with this work directing further research over the next two years. To support a commitment to the dissemination of research for the benefit of society and the economy, the University of Oxford has established an online archive of research materials which includes digital copies of successfully submitted postgraduate student theses. This work will be written up as a thesis and on successful submission it will be openly accessible both in print and online in University archives.

Who is organising and funding this research?

This research in funded by the Engineering and Physical Sciences Research Council (EPSRC). The results of the study may be published in academic conferences or journals, and will be published in the researcher's thesis.

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

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 email Arianna, who will do her best to answer your query. If your concern has not been acknowledged, or you have not been given any idea of how your concern will be handled, please contact the chair of the Social Sciences & Humanities Inter-Divisional Research Ethics Committee at the University of Oxford who will seek to resolve the matter in a reasonably expeditious manner:

- ethics@socsci.ox.ac.uk
- Research Services, University of Oxford, Wellington Square, Oxford, OX1 2JD

Further information and contact details:

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

Arianna Schuler Scott
Department of Computer Science
Robert Hooke Building, OX
arianna.schuler.scott@cs.ox.ac.uk

CENTRE FOR DOCTORAL TRAINING *in* CYBER SECURITY

UNIVERSITY OF OXFORD

Arianna Schuler Scott
Doctoral candidate in Cyber Security
arianna.schuler.scott@cs.ox.ac.uk

An exploration of attitudes to dynamic consent in research

CONSENT FORM

This is a form that we will ask you to sign or give verbal consent to. It gives you a summary of the Patient Information Sheet and gives us a record of your decision to take part.

You get a copy and the study keeps one on record. Please initial the statements you agree with.

1. I agree to take part in this study.	
2. I am participating voluntarily.	
3. I have read the information sheet for this study and had time to think about it. I am happy that any questions I have wanted to ask have been answered.	
4. I understand that I am free to withdraw at any time without giving any reason, without my medical care or legal rights being affected.	
5. I agree to audio recording and the use of anonymised quotes in research reports and publications.	
6. I would like to receive feedback about my interview and this study.	
7. I agree for my anonymised interview to be used in future research, which may involve commercial organisations.	

Name of Participant	Date	Signature
Name of Person taking Consent	Date	Signature

Appendix 9: Post study peer review session

Friday 29th March 2019

HCC Data-session

An Exploration of Dynamic Consent

Arianna, Cyber Security CDT

Good morning, thank you for taking the time to attend this session. Your feedback is greatly appreciated and will be pored over in detail! Please leave this pack with me at the end of the session.

You will find the following information in this pack:

- A brief introduction to Dynamic Consent.
- An outline of the "Exploration of Dynamic Consent" study.
- A sample interview in its entirety.
- Selected extracts from other interviews.

Session schedule

Time	**Activity**
10:30	Overview, sample interview given (for prospective coding).
10:45	Check-in, discussion of existing coding structure.
11:00	Further discussion.
11:15	End.

Study outline: "An Exploration of Dynamic Consent"

This study was meant to look at whether Dynamic Consent, an electronic consent tool for allowing participants to have greater control over their samples and data, influences participants' involvement in research. Participants were recruited from the RUDY study, a project implementing Dynamic Consent to learn about their experiences and whether this has influenced their attitudes towards data-sharing. Two participant groups were identified: the research team (as to their expectations of what dc would achieve) and participants (to see whether those expectations had been achieved).

The aim of this study was quite specific - as part of the initial analysis, I would like us to look at emergent themes and what came out of the interview data gathered from participants.

Dynamic Consent: engagement and revocation over time

- A mechanism used by one party to store another's personal data for later use, to keep that individual informed as to how their data is being used and allow them to revoke permission for that use as long as that personal data is on record.

- Dynamic Consent emerged from biobanking, the use of research frameworks that collect biological samples and derive data from them.
- Dynamic Consent is an important concept because it focuses on the relationship (and partnership, however temporary it may be) between those collecting data and those from whom that data is collected, for more engaging and impactful research.
- The "dynamic" part is important – it allows re-use with knowledge and consent, enables choices to be made in response to changing circumstances and allows people to be approached for different kind of consent or permissions.

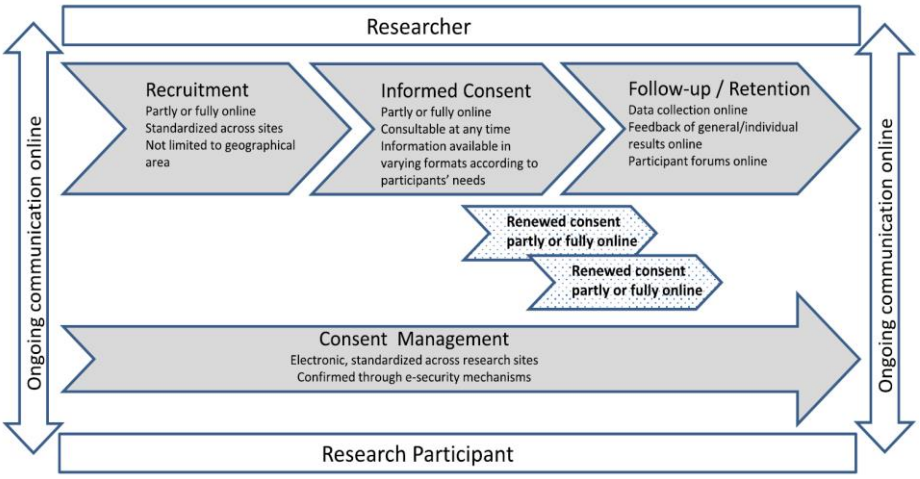


Figure 1: Dynamic Consent's contribution to phases of research (Budin-

5. Challenges of Implementation

The implementation of dynamic consent requires:

- A change in culture for both health professionals and individuals.
- The development of new kinds of partnerships between researchers and patients.
- New policies, standards and ways of working that recognise the valuable role of patients.
- New technologies and processes to provide operational control.
- Time, money, expertise and, most importantly, commitment from clinicians, researchers, health-care services, research and governments.

Conclusions drawn

Theme 1: Participation

- ➔ Desire for consultative process.
- ➔ Positive responses.
- ➔ Ownership of data.
- ➔ As a moral obligation/duty ("anything you can do").
- ➔ As a favour.
- ➔ Bad thing? Withdraw.
- ➔ No personal benefit but still doing it.

Theme 2: Communication

- ➔ Desire to be recognised.
- ➔ Desire for feedback.
- ➔ Face to face is better → communicates more information.
- ➔ Output isn't patient friendly.

Theme 3: Research expectation

- ➔ Moves things forward.
- ➔ Data kept in (contextual) "sphere".
- ➔ Researchers enable patient control.
- ➔ Researchers as professionals.
- ➔ Providing services for real people.

Theme 4: Trust

- ➔ Absence of mistrust.
- ➔ "No reason not to trust".
- ➔ Depend on system as there's no other option.

Theme 5: Consent motivation

- ➔ Context?
- ➔ Help future people (kids/non-personal).
- ➔ Give something back.
- ➔ Greater good.
- ➔ Data security ((implicit) assumption of security/explicit concerns).
- ➔ Pain.

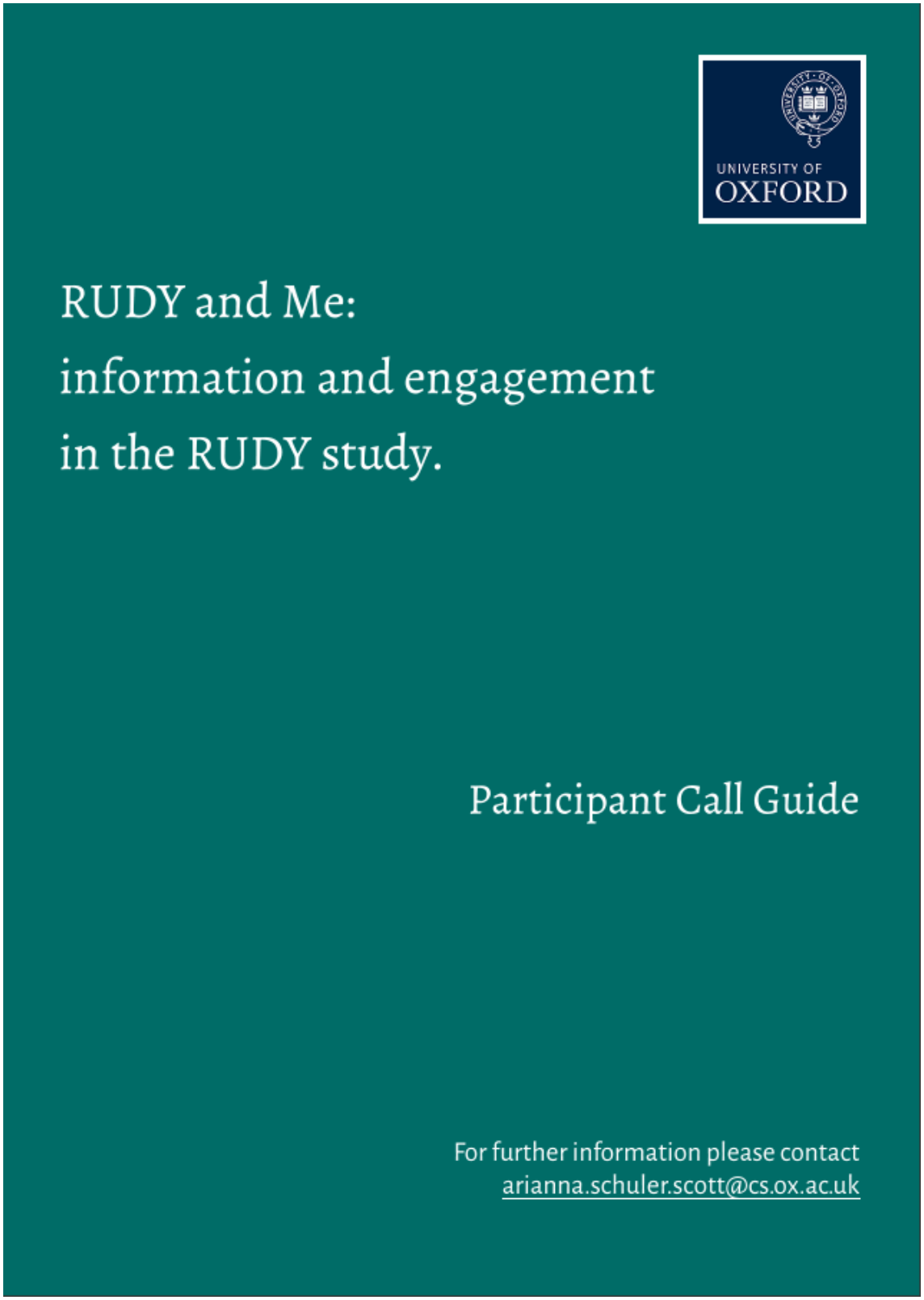
Theme 6: Data leakage

- ➔ General sentiment.
- ➔ Practical effects.

I think that what I will see emerge are themes of trust, data-use and communication across individual motivations, experiences and expectations of research:

	(PERSONAL) CONSENT MOTIVATION FOR RESEARCH	(PERSONAL) PARTICIPATION EXPERIENCE OF RESEARCH	(GENERAL) RESEARCH EXPECTATIONS
TRUST	→	→	→
DATA-USE	→	→	→
COMMS	→	→	→

Appendix 10: Co-design call guide



Your data

This is just to let you know about the information I am interested in and will be using.

I have your email address and if you have sent me other information, I have that also (phone number, etc.). This is not being shared with anyone else.

Your answers are in confidence. During our call, I will be making notes so I can remember the key points raised during our discussion. I will write a report that aggregates and draws from these notes—your answers cannot be linked to you in any way.

When this study is finished, I will email you a brief update, with your permission, and that is when I will delete your email addresses from my contact list.

I will also delete any emails with any other personal data you have sent me, such as your phone number.

GDPR notice

The University of Oxford is the data controller with respect to your personal data, and as such will determine how your personal data is used in the study. The University will process your personal data for the purpose of the research outlined above. Research is a task that we perform in the public interest.

Further information about your rights with respect to your personal data is available from https://compliance.admin.ox.ac.uk/data-protection-policy

Hello and welcome! Thank you for taking part in my research. Participation is entirely voluntary and you can leave at any time – just let me know, if you can!

In this call guide you will find a great deal of (hopefully helpful) information about our call on:

Wednesday 13th May, 14:00 – 16:00

This symbol invites you to provide input ahead of our call, e.g. a suggestion for Skype etiquette, or a note to let me know if anything is unclear.

Call guide contents

If you have any questions or I can make this experience more comfortable for you, please let me know.

Email: arianna.schuler.scott@cs.ox.ac.uk

Skype: https://join.skype.com/invite/jMndmzodFVUB

Introduction

Many thanks for your help with some work I am doing to try and improve the RUDY patient experience. Given your role as a RUDY participant I am so glad that you are interested in taking part in a Skype discussion to think over aspects of the information and engagement approach that RUDY takes.

By "the information and engagement approach that RUDY takes" I mean how the study provides information of any kind, but particularly information about the research they are doing or facilitating.

My name is Arianna, I am working towards a PhD in Computer Science and my research is all about data protection and consent. Last year I was very fortunate to be able to interview a number of RUDY participants about their experience on the study.

The findings from these interviews suggest that more could be done to provide information about what is going on in the RUDY study. This is something I would like to address, preferably with some ideas from you all about how best to do it.

Agenda

Many thanks for your help with some work I am doing to try and improve the RUDY patient experience. Given your role in the patient forum, I am so glad that you are interested in taking part in a Skype discussion to think over aspects of the information and engagement approach that RUDY takes.

This is an overview of what we will be doing in the call.

You can suggest changes to the agenda by having a read, and letting me know!

- [05 mins] Introduction
- [10 mins] Exercise 1: warm-up ("I am a part of RUDY because...")
- [30 mins] Exercise 2: fill-in-the-gaps ("the RUDY experience")
- [10 mins] Break
- [30 mins] Exercise 3: discussion ("engaging with the RUDY study")
- [10 mins] Summary and "What's next?"

Call etiquette

This "etiquette" is a short set of agreed rules about how to behave on a call. They are more an indication of what you can expect from me, and general expectation of politeness and order...

You can suggest changes to the call etiquette by having a read, and letting me know!

- Before a call starts, Arianna will message attendees.
- Arianna will welcome everyone—brief introductions at this point!
- No one should feel that they must show video, it is perfectly acceptable to take part using audio.
- If someone else is speaking, please mute your microphone in case there is noise in the background which might be distracting.
- If you are speaking, please unmute yourself so others can hear you!
- In general, we will just practice common courtesy and if there is any confusion, Arianna will facilitate.

Call prompts

These prompts are for letting me know if you need anything during the call and do not feel comfortable speaking. Just type, or copy and paste into the chat box during our call.

Here are some example prompts:

- "You have been speaking for a long time".
- "Please slow down".
- "I cannot hear you".
- "I am feeling tired".
- "I need a break".

Exercises

There are two exercises planned for this call, you will find an outline and the templates for these activities here. Our call has time built in to complete these activities but do feel free to prepare answers ahead of time. The most valuable part will very likely be the discussion we have around everyone's ideas.

You can suggest changes to the exercises by having a read, and letting me know!

1. Warm-up: I am a part of RUDY because...

- This exercise is optional.
- This exercise is planned to be 5 minutes long.
- The prompt is: "I am a part of RUDY because...".
- Everyone has a minute to finish the sentence.
- We go around the group, reading our sentences out.

Continued >>>

Exercises (continued)

2. Fill-in-the-gaps: the RUDY experience.

- This exercise is planned to be 30 minutes long.
- There are 5 statements:
  1. When I think of RUDY I think of _____.
  2. On the RUDY website I _____ .

     E.g. "On the RUDY website I look at the number of participants".
  3. I would like the RUDY website to have _____ because _____.
  4. When filling out RUDY surveys I use _____.

     E.g. "When filling out RUDY surveys I use my mobile phone".
  5. I would like RUDY to make _____ available.
- Everyone has a minute to fill in the blanks or think about what they have already written.
- We go around the group, question by question and discuss our answers.

Continued >>>

Exercises (continued)

3. Discussion: engaging with the RUDY study.

- This exercise is planned to be 30 minutes long.
- As a group we will discuss:
    1. What do you want to know from RUDY?
    2. What do you think RUDY wants to know from you?
- I am looking for a steer on what my next steps should be for this project—in the last ten minutes I will ask for one thing that you think would improve your experience of taking part in RUDY.

Call script

The following is a script of the call. I will be reading from this script and while I cannot write a script for the discussion parts, if you have an idea of how this call is planned then hopefully you will be more able to focus on the content and exercises themselves.

Introduction [5 minutes]

Good morning, my name is Arianna and I will be facilitating our call today. I would first like to thank you all for joining me and before we start I would like to take us through what the plan is for the next couple of hours and some points about general admin on this call as well.

A little bit about me: I am a Computer Scientist, I am interested in information security – things like privacy and consent. I am particularly interested in the RUDY study because of the way they gather consent in a sort of checklist.

If I want to do anything that affects the RUDY patient experience, I really should ask patients for input. That is the idea behind this "co-design" session. The point is to get different types of people involved during the planning phase of a research project. This means you have different perspectives about what the problem actually is, and whatever comes out of the project is hopefully more relevant and useful to the people who will eventually use it.

Continued >>>

Call script (continued)

In other words, rather than you (a research participant) simply doing what I (a researcher) tell you to do, I am asking you whether what I think is important is... actually important. Your experience is something I cannot make up. So, I am looking forward to working with you, if at any point you feel unsure about whether to say something... please say it!

I just want to make it clear that there is no "right" or "wrong" way to do anything today, I am hoping we will get creative!

If you find that you have any technology issues, please email me or give me a ring during the breaks on

Speaking of technology, we are going to test microphone muting now, and whether you can see and hear my timer. If everyone could mute their microphones now please, and try saying "hello" [allow 2 minutes to test muting microphones]. Excellent, thank you. Please try and mute your microphone when you are not talking as this makes it easier to hear whoever is talking.

Does anyone have a poor internet connection?

Alright, I am going to make sure we can all see and hear the timer we will be using. I have set the timer for 5 seconds and will play it now. Can everyone see or hear it?

Continued >>>

Call script (continued)

Right, I will talk about what we are doing today. In your call guide you should already have the agenda – I am going to pause so that you can open that up if you want to. Let's go through that now.

[**Go through the agenda**]

Most of our session today will be hands-on, I do not want to spend much time talking at you because I would much rather listen to what you have to say!

Our agenda is flexible because we may want to take slightly more break time, or find that we get into a particularly meaty discussion!

I would like to practice one more thing before we get going. In your call guide you have some call prompts. These are for you to let me know if you need anything (I am watching the "chat" feed. I would like everyone to try using one (or sending me a "SIMON SAYS" and an instruction). Please do so now.

Thank you for staying with me thus far, and without any further ado let's kick-off!

Continued >>>

Call script (continued)

Exercise 1: warm-up [10 minutes]

Okay, our warm-up exercise is to get us talking!

This is optional, so if you would rather sit this out to see how this all works and join us in a minute or two, please feel free to do so.

The prompt for this exercise is "I am a part of RUDY because... "

Now, you may have a pre-prepared answer but if not, I am going to set the timer to one minute and when that goes off we will go around the circle.

Just so you know, we will go around in order of whoever happened to log in, so that means I will go first and then... [establish reading order].

Right, brains at the ready! We have one minute starting now.

Okay, I will kick this off: I am a part of RUDY because...

[**We go around the group**]

Continued >>>

Call script (continued)

Exercise 2: engaging with the RUDY study [30 minutes]

Anyone who has been waiting in the wings, now is the time to take part—do we have everyone?

Right, this "fill-in-the-blanks" exercise is about your RUDY experience. You may have pre-prepared your answers to this or you may not. We have 5 statements which I will read through:

1. When I think of RUDY I think of _____.
2. On the RUDY website I _____ .

   E.g. "On the RUDY website I look at the number of participants".
3. I would like the RUDY website to have _____ because _____.
4. When filling out RUDY surveys I use _____.

   E.g. "When filling out RUDY surveys I use my mobile phone".
5. I would like RUDY to make _____ available.

Whether you have prepped beforehand or not, let's take a minute to think about (or complete) the exercise. Ready? Let's go.

Okay, we are going to take this question by question. We'll go round everyone's answer and discuss. I started last time, so is there anyone who would like to kick this exercise off?

[**We go around the group and discuss answers**]

We are going to take a break everyone. The plan is to come back in 10 minutes. Does that work for everyone? I will leave the timer running and will be here if you need anything—please mute your microphone and camera!

Continued >>>

Call script (continued)

Exercise 3: the RUDY experience [30 minutes]

Welcome back! I am hoping you had the chance to stick the kettle on or perhaps gaze out of a nearby window...

Our next exercise is a discussion. We have two prompts:

> "What do you want to know from RUDY?"
> "What do you think RUDY wants to know from you?"

As with our last exercise you may have already thought about this ahead of time, but if you haven't—don't worry!

Right, this call is really for me to get a steer, or strong impression, as to what my next steps should be for this project. In the last ten minutes I would like to go round and ask for your one thought on what could be done to improve communication in the RUDY study. Something I can take away and put into action.

Let's get stuck in.

[**Discussion**]

Thank you everyone, our time is almost up! What is your one thought on what could be done to improve communication in the RUDY study?

Continued >>>

Call script (continued)

How is everyone feeling at the moment, do we need a five minute break?

We have been sitting at our computers for a while now!

[Either] Let's come back in five minutes, mics and cameras off please.

[Or] Continue.

Action plan and thanks [10 minutes]

A quick summary of what we have been up to today: as someone interested in how RUDY communicates with patients and vice versa, I asked you about your experiences and whether there was anything you thought might improve things.

[**Our top ideas**]

The action plan is now for me to…[**give action plan**].

Would anyone like me to keep in touch about what comes out of this project (it's absolutely fine not to)?

[**If so, ask what would be best—e.g. an update at the end, or progress updates?**]

The last thing, I think, is for me to thank you all very much for your time and energy today. Please take care and I hope you get some rest after all this hard work today.

Many thanks again - goodbye!

Funded and supported by

Appendix 11: RUDY data-access committee request

STUDY 1

NIHR RD TRC Musculoskeletal - RUDY

Analysis Plan

The aim of RUDY is to increase our understanding of patients with rare diseases of the bone, joints or blood vessels

Title: An exploration of attitudes to Dynamic Consent in research.

1. Version no: 2.0
2. Date submitted: 9th May 2018
3. Primary Author (with e-mail): Arianna Schuler Scott (arianna.schuler.scott@cs.ox.ac.uk)
4. Sponsoring senior investigator: Professor Michael Goldsmith (Comp.Science), Dr. Harriet Teare

(HeLEX)

5. Potential co-authors: This is DPhil work at the moment. I would be more than happy to discuss work focusing on PPI engagement at a later time.

6. Optional : Institutions involved and roles (eg writing/analysis):

Ethics approval number (Department of Computer Science): Ref No: SSD/CUREC1A CS_C1A_18_011

The University of Oxford is the only institution involved at this point but if anything changes I will raise this with the RUDY Team submit a request for change to the Access Committee. I am a DPhil student and hope to carry out interviews. I will be the person talking to RUDY participants and carrying out the analysis/write up. My supervisor is Michael Goldsmith, based in the Department of Computer Science, and my cosupervisor is Dr. Harriet Teare who is based at the Centre for Health, Law and Emerging Technology. Dr. Teare has someworked with the RUDY project before.

7. Research Questions (Following PICO format if possible):

P (Problem or Patient or Population) : What do elements of Dynamic Consent look like in practice ?

I (intervention/indicator) : Interview participants who use/design/build the RUDY consent system.

C (comparison) : I will compare this to existing literature on data privacy controls and consent.

O (outcome of interest) : I want to know what public engagement looks like in practice, how it can be successful and whether that has any bearing on patient attitude to data protection or consent.

8. Patient significance:

I do not want to interview the RUDY patients because of their conditions, I would like to interview them about the infrastructure that RUDY has in place. I know that there is an engaged cadre of about 15 patients, a focus group that provides invaluable feedback.

9. Study design:

Description: this study will look at how Dynamic Consent, an electronic consent tool for allowing participants to have greater control over their samples and data, influences participants' involvement in research. We will recruit participants that have already been involved in projects using Dynamic Consent, to learn about their experiences and whether this has influenced their attitudes towards data-sharing. This project is the first step in exploring the power dynamics between those who collect, store, share and use (or lose) people's personal information (data controllers), and the people that information comes from (data subjects). No conflicts of interest identified.

Professional guidelines: while there are no specific guidelines listed for Computer Science, I will be taking the following as minimum (Willig, C. (2013). Introducing qualitative research in psychology. McGraw-Hill Education (UK).): work from the notion of informed consent, do not deceive, the participant has the right to withdraw, they are debriefed, and complete confidentiality is kept as far as possible.

Use of data/results: the results of this study will be used to direct further DPhil research. The work is being done across very different research groups so the result of this will be a 'check-in point' for the academics involved. The academics involved are all from the University of Oxford: the Cyber Security Analytics Group based in Computer Science, and the Centre for Law, Health, and Emerging Technologies which is part of the Nuffield Department of Population Health. The data will be held securely, with anonymised results being shared with these supervisors. Identifiers will be removed and no names or identifying information shared.

10. Subjects:

Inclusion criteria: Those who have taken part in a study using a system for Dynamic Consent (a research subject for example) or those involved in designing, implementing or delivering a system for Dynamic Consent (academics, project managers, and software developers).

Exclusion criteria: not being a member of the above.

11. Limitations to analysis/ design

If no participants sign up then I will not be able to carry out interviews and collect data.

12. If analysis involves using new measurements that are not included in original core protocol:

Description of new data to be collected: interviews as described, either over the phone or voice over IP (VoIP, such as Skype), in person.

Methods/ procedures to be employed:

Methodology: this initial foray will look at projects that are already using Dynamic Consent – where subjects can see what information of there is being shared, with the

option to make changes or revoke access to that. It will be important to look at both those using this kind of system, and those involved in creating it. Working with research teams that have conducted the studies using Dynamic Consent we will approach participants who have given permission to be contacted for future research, asking them to take part in a survey. This survey will include vignettes (a combination of scenario and survey questions) to investigate what consent means to interviewees and what they think of how their data is handled.

Methods:

- Snowball sampling (recruiting through contacts of existing participants).
- Participant completes questionnaire in hard copy.
- Participant completes online questionnaire or other online task.
- Using social media.
- Interview - as described, either over the phone or voice over IP (VoIP, such as Skype), in person.
- Audio recording of participant (you will generally need specific consent from participants for this).

Interviews: some participants will not respond well to being recorded, towards the end of the interview there will be a 'Chatham House rule' section where the audio recording is turned off. This is to try and capture from the participant any thoughts that they may have withheld, subconsciously or not. This data will not be used differently to that collected previously.

Administration/ data/ bio-specimen storage

Interview organisation: this project directs participants to email Arianna to arrange an interview or use a service called MeetingBird (https://www.meetingbird.com) to centralize interview or call organisation. There are many RUDY participants that will not be able to meet for a face to face interview and this plan hopes to facilitate this by having an option for calls either online or over the phone.

Sensitive data capture: All personal or one to one correspondence (such as scheduling interviews) will be conducted via university email to mitigate the risk of a personal email account holding participant emails, names, or other sensitive and personally identifiable information. The timetable itself and contact list will be stored as a password protected file.

The parts of interviews recorded on an audio device will be transcribed and checked against the recording before then destroying the recording. Transcriptions will be written using word processing software and kept as a password encrypted file on a password protected machine. These files will be backed up to an encrypted hard drive every time a new entry is completed.

The parts of the interview not recorded on an audio device will be annotated, scanned and transcribed. Both the scans and transcriptions will be checked against the paper notes (which will be shredded) and stored as above.

In terms of access and reuse of information, the only person with access to this data is the research student, Arianna. Data will be anonymised, no names are needed but

demographics will be taken. Instead of asking for addresses, more general terms will be used (e.g., 'Oxford' given as opposed to '123 Great Road, OX1 2AB').

Data will be retained for a minimum of three years after publication of the results, according to university policy.

Discussion of ethics/ consent/ patient information

The key areas of ethical concern are currently confidentiality and data protection. There is very little risk of participants meeting or discussing their interviews as this is one on one. The list of people taking part will be saved as a password encrypted file, on a machine that is itself password protected. Direct quotes will only be used with permission (given, or not, when informed consent is asked for). Participants' names are not required so participants will be anonymised using a numbering rather than a naming convention. Demographic data will also be collected: age, gender, nationality, ethnic information, profession and location. To mitigate potential identification broad ranges will be used where possible – age would use brackets, for example.

Research ethics: The research student has not attended a formal training session, but in developing the rationale for this work by exploring the philosophical theoretical perspective (that people's perspective is coloured by their own experience and existing structures, rather than there being an absolute truth to consent) and epistemological positioning (that good research is a product of collaboration and engagement rather than centering on the researcher entirely) it has been possible to think about and prepare the research ethics surrounding this research.

Online integrity and ethics materials provided by the university have been incredibly helpful as references as well, in this instance. In addition to this, the student has talked at length with others that have experience in qualitative research, and has sense-checked plans and intended methodology.

This study does not want to risk an increase in levels of participant paranoia (in who is keeping their data, and what is known about them in general), and neither are changes in behaviour or attitude towards consent in scope for this work. To avoid questioning bias and promoting specific behaviours, questions will be kept neutral. The interview will last approximately 45 minutes.

Recruitment: I am making contact with RUDY as a 'gatekeeper' allow access to their participants (and team) to take part in this study because as part of these projects, participants will have stated whether or not they would be happy to be involved in future research. Only those who have agreed to such contact will be contacted. The participant pipeline would be as follows:

- Recruitment: they receive a participant information sheep from the 'gatekeeper' (RUDY's research study and PPI coordinator).
- Contact is established via direct email or online scheduling service MeetingBird.
- An interview or call is scheduled.
- On the day, the participant is briefed, they give verbal consent to take part in the study if it is a phone or online call and a signed form if it is in person.

- Participant is interviewed and debriefed – they are given an overview of the project, they will be asked if they are happy to receive follow up a follow up email and whether their contact details can be kept for future use by the researcher, and encouraged to follow social media for research conclusions.

Consent decisions: Taking informed consent important for many reasons, especially as the General Data Protection Regulation dictates that without it, data cannot be stored at all. To complement the given criteria for informed consent, the idea of informed consent for this study rests on the following principles [1]:

- Process disclosure – the participant is told not only what they are doing, but about the research process as a whole.
- Benefits/risks and what refusal to take part in the study means.
- Voluntariness – the idea that unless the participant freely volunteers, informed consent is not given.
- Competence – the participant is able to make a decision about their own consent.
- Alternatives – to try and ensure the participant's choice is not engineered it will be made clear what happens if they say no (there is no consequence).

13. Target journal/ conference/ deadlines:

As a validation step I would like to take findings from these interviews to a workshop: the 13th International Federation of Information Processing (IFIP) Summer School 2018 on Privacy and Identity Management. The theme is 'Fairness, Accountability and Transparency in the Age of Big Data'. It is to be help on 20–24 August 2018 in Vienna, Austria.

14. References (Vancouver format):

[1] Faden, R. R., & Beauchamp, T. L. (1986). A history and theory of informed consent. Oxford University Press.

STUDY 2

NIHR RD TRC Musculoskeletal - RUDY

Analysis Plan

The aim of RUDY is to increase our understanding of patients with rare diseases of the bone, joints or blood vessels

Title: A co-designed study measuring the effect of information feedback on research participation.

Note: this is a request for data to shape my methodology. I would like to know what data RUDY collects about how participants use the online platform (what do the RUDY timelines look like, is meta-data collected about attempted log-ins, time spent on the platform and pages that are visited? Are different versions of consent tracked, and does it change?) alongside actual survey completion rates. The reason for this is that this study is an information intervention - what participants do not see may be just as important as what they do see. I am interested in self-reported and actual behavior. I realise that there may be information that I have not asked for that may be helpful so I am open to recommendations from the committee.

1. Version no: 5.2
2. Date submitted: 10th March 2020
3. Primary Author (with e-mail): Arianna Schuler Scott (arianna.schuler.scott@cs.ox.ac.uk)
4. Sponsoring senior investigator: Supervisor:
    a. Professor Michael Goldsmith (Department of Computer Science).
    b. Co-supervisors: Dr Harriet Teare (Centre for Health, Law and Emerging Technologies, Faculty of Law) and Dr Helena Webb (Department of Computer Science).
5. Potential co-authors:
    a. Dr Kassim Javaid, Alison Turner and members of the RUDY team.
    b. I am also looking to involve participants.
6. Optional : Institutions involved and roles (eg writing/analysis):
    a. I submitted a CUREC 1A application to the Department of Computer Science and received feedback : further discussion was necessary.
    b. I submitted this application along with further supporting evidence to K. Shepherd (clinical trials & research governance, CTRG), as advised by Oxford University Hospitals Governance (S. Hussain).
    c. K. Melham (Senior Clinical Research Support Manager, CTRG) is now my liaison for this project and has worked with Dr Javaid to advise on the ethical aspects of this project.
7. Research Questions (Following PICO format if possible):
    a. P (Problem or Patient or Population) : RUDY participation rates show a drop-off after an initial period of engagement (see Table). Interviews with adults consented within the RUDY study have reported that participants feel research is a one-way conversation.

i. Table 2 below gives a tentative indication as to current RUDY survey completion rates. An anonymized dataset of all RUDY participants with active consent was provided – completion rates (the number of forms submitted as a percentage of the total number of forms assigned) seem low but this preliminary data likely includes people who have consented to take part in RUDY but have not yet filled in any surveys at all.
ii. Take the HADS survey, for example. The completion rate in 2020 for everyone due to complete surveys in May/June is 21%. If we want to try and double this rate (42%) over this intervention study, we would need to run it over 2 months (May and June), and recruit 111 people for the control and 111 people for the intervention arm.

Table showing core survey completion rates.

	ADL	EQ5D	PSQI	HADS	WALK12
Completion rate (joined May/June 2019) n = 84	13%	13%	13%	13%	13%
Completion rate (joined May/June 2018) n = 77	24%	20%	16%	16%	24%
Completion rates (joined in May/June 2014-19) n = 273	26%	23%	21%	21%	27%

b. I (intervention/indicator) : a before/after intervention study measuring whether providing feedback about RUDY to participants (via email or by putting descriptions on the RUDY portal they can see when they log in) affects their level of participation in the study (the number of surveys completed).
c. C (comparison) : I want to compare empirical measures of participant engagement (survey completion rate) before and after enhanced feedback is given. "Enhanced feedback" is feedback about the RUDY study that participants suggested they would like to know. E.g., a summary of general information about RUDY and studies they have taken part in, how they can get involved, and specific information about their own condition.
d. O (outcome of interest) : whether communicating information about a research study (such as current progress and lay summaries) improves completion rates (of core questionnaires such as EQ5D and SF36) of RUDY participants.

8. Patient significance:
    a. The focus of this study is to investigate how users of technological systems can be encouraged to engage in how their data is used. Improving the participant experience within RUDY by showing the

benefits of participation provides participants with relevant and useful information. A better participant experience also provides researchers with more complete data.

9. Study design:
    a. In the last study of this collaboration, RUDY participants were individually interviewed and members of the RUDY team took part in a focus group. This study found that the majority of participants do not feel engaged with the study and suggested research feedback they "wouldn't mind" receiving. "Feedback" included overall progress, how participation has helped, and upcoming opportunities. This intervention will measure whether survey completion increases when RUDY provides information about the study's progress in layperson's terms (summaries, social media posts, etc.).
    b. Pre-study co-design:
        i. RUDY sends out an email inviting members of the user group to participate in co-design session 1. Co-design sessions are likely to be a "webinar"[44]. Materials will be send out for review ahead of time in line with a flipped learning[45] approach. Participants will be able to dial into the call, discuss, ask questions and propose ideas. The session will be recorded for those who cannot attend, to watch in their own time and provide feedback.
        ii. Co-design session 1(see appendix 3 for a guide to co-design sessions): members of the group help develop the study (development of objectives and sense-checking) and give feedback on proposed content.
        iii. RUDY sends out an email inviting participants from the previous study to participate in co-design session 2.
        iv. Co-design session 2: returning participants give feedback on the proposed intervention content (based on the qualitative interviews).
        v. RUDY sends out an email inviting patient group partners to take part in co-design session 3.
        vi. Co-design session 3: patient groups give feedback on how the intervention content is being communicated.
        vii. Applicant refines study design. Applicant + RUDY team prepare to run study (please note: I am aware that this may require software development effort and content creation, and may be able to request funding from my own Centre for Doctoral Training in Cyber Security for essential research costs).
    c. Intervention study

This study is a before/after service evaluation of RUDY.

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[44] Webinars are…

[45] Flipped learning is…

i. Recruitment:
    1. RUDY participants who have consented to their data being shared for research use will be automatically enrolled onto the study.
    2. RUDY participants who have not consented to their data being shared for research use will receive a request to participate.
ii. There will be 2 control groups ("before" and "after") and an intervention arm:

Data collection period	Study arm
May/June 2019	Before control
July/August 2019	Before control
March/April 2020	Before control
May/June 2020	Intervention
Jul/August 2020	After control

iii. Intervention start (late April, to accommodate a May start).
iv. Intervention ends (early July, to accommodate a June finish).
v. Post data collection, intervention participants will be asked to complete a survey reflecting on their experience of research so far.
vi. Analysis.

10. Subjects:
    a. Control group inclusion criteria: any adult RUDY participant who has consented to share their data for research purposes and was due to fill out core surveys within May/June 2019 and July/August 2019.
    b. Intervention inclusion criteria: any adult RUDY participant who has consented to share their data for research purposes and is due to fill out core surveys within March/April 2020, May/June 2020 and July/August 2020.
    c. Exclusion criteria: RUDY participants who have not consented to taking part.
11. Variables
    a. Predictor(s): use of enhanced feedback.
    b. Outcome(s): completion of core survey/s.
    c. Adjusting and stratifying co-variates: unknown at this time.
12. Statistical issues: unknown at this time.
13. Limitations to analysis/ design:
    a. Co-design takes time. This may overrun and is being mitigated by having a research plan with regular check-ins (DPhil student/advisor, DPhil student/RUDY, DPhil student/advisor/RUDY).
    b. Interdisciplinary work takes time. The risk of "scope creep" (adding new ideas or features and extending project length) is being mitigated by

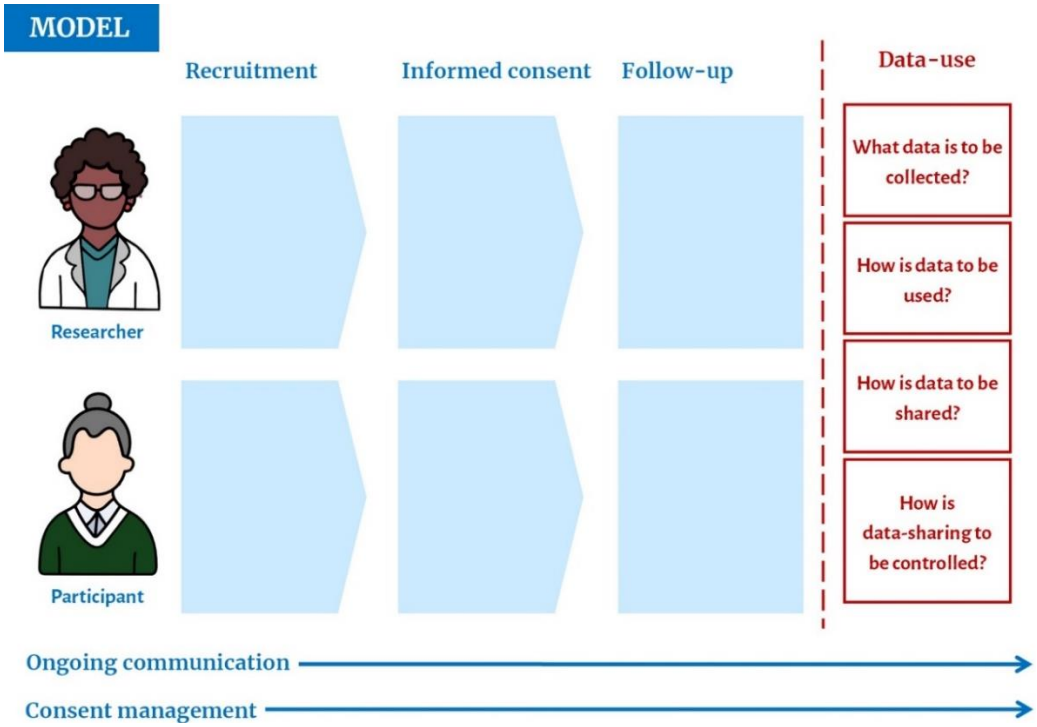
having a clear study plan before beginning work (this application) and a liaison within CTRG to advise on governance/research issues.

14. Mockup of key table/figure: Unknown at this time.
15. If analysis involves using new measurements that are not included in original core protocol:
    a. Description of new data to be collected: This application is requesting access to information that the RUDY project already has access to (completion rates and demographic information).
    b. Methods/procedures to be employed: this is an intervention study that will use co-design to involve members of the user group and RUDY participants who took part in a previous study.
    c. Administration/data/bio-specimen storage + discussion of ethics/ consent/ patient information: patient information is absolutely confidential. Intervention participants will be assigned an identification number and patient information will be stored in a password-protected file on a password protected university computer.
16. Target journal/ conference/ deadlines:
    a. ACM Health
    b. Aging Clinical and Experimental Research
    c. Suggestions welcome!
17. References (Vancouver format):

Please email completed analysis plans to RUDY@ndorms.ox.ac.uk

Appendix 12: Tool-kit template

A conceptual model to aid development of Dynamic Consent in practice, adapted from (Budin-Ljøsne *et al*, 2017).



A framework to aid further development of Dynamic Consent in practice, adapted from (Prictor *et al*, 2020b).

FRAMEWORK

Assumptions	Inputs	Activities	Outputs	Outcomes	Impact
Assumption 1	What data is being gathered to test this assumption?	What will be done with this information?	What are the metrics you will put to use?	Record outcome. Does assumption 1 hold?	What does your outcome mean, who is affected?
Assumption 2	↓	↓	↓	↓	↓
Assumption 3	↓	↓	↓	↓	↓

Appendix 13: Tool validation prompt

Below are first and last pages of a document sent to researchers ahead of time to prompt reflection on the tool-kit described in Chapter seven. For brevity, tool-kit illustrations are not included.

CENTRE FOR DOCTORAL TRAINING *in* CYBER SECURITY

EPSRC Engineering and Physical Sciences Research Council

HeLEX

RUDY

Validating tools to aid development of engaging research practice*.

*by offering enhanced feedback: information about project aims, the data being collected, how that data will be used and shared, the value of these contributions and research outcomes.

Thank you for your time!

I am looking for initial thoughts on some tools I have developed to help researchers think about participant engagement in practical terms (feel free to use p7 to structure feedback).

I carried out an intervention study from July to December 2020 testing the effect of "enhanced feedback*" on research participation.

I found that feeding information back to people improves participation and retention.

The following model, framework and specification have been created to help develop research that feeds information back to participants as well as collecting data from them.

Each template is followed by an illustrative example.

Please direct any questions to arianna.schuler.scott@cs.ox.ac.uk

Your thoughts on these tools

I am looking for first impressions and reflections.

1. What is your area of expertise (feel free to use a collection of key words)?
2. a) What do you think the model is for? Do you think is it useful?

   b) What are your first impressions of the model?

   c) Do you have any reflections that differ from your first thoughts?
3. a) What do you think the framework is for? Do you think it is useful?

   b) What are your first impressions of the framework?

   c) Do you have any reflections that differ from your initial thoughts?
4. a) What do you think the specification is for? Do you think it is useful?

   b) What are your first impressions of the specification?

   b) Do you have any reflections that differ from your initial thoughts?

Many thanks for your time.