Dynamic Consent: An Evaluation and Reporting Framework

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Keywords: biobanking; digital; dynamic consent; evaluation; genomics; informed consent; reporting;
research participation; trials.

Abstract

Dynamic consent (DC) is an approach to consent that enables people, through an interactive digital interface, to make granular decisions about their ongoing participation. This approach has been explored within biomedical research, in fields such as biobanking and genomics, where ongoing contact is required with participants. It is posited that DC can enhance decisional autonomy and improve researcher-participant communication. Currently, there is a lack of evidence about the measurable effects of DC-based tools. This paper outlines a framework for DC evaluation and reporting.

The paper draws upon the evidence for enhanced modes of informed consent for research as the basis for a logic model. It outlines how future evaluations of DC should be designed to maximise their quality, replicability and relevance based on this framework. Finally, the paper considers best-practice for reporting studies that assess DC, to enable future research and implementation to build upon the emerging evidence base.

Introduction

Innovations in information technology, the increased ability to gather and reuse datasets in research, and changing ethical, legal and regulatory requirements, have resulted in new approaches to consent in a range of research disciplines. Dynamic Consent (DC) has drawn attention because of its potential to facilitate participant consent and engagement in research activities over time. DC refers to an approach that engages individuals about the use of their personal information or tissue samples, enabling both granular consent decisions and ongoing communication between participants and researchers. It utilises an interactive interface that supports a competent individual in making an autonomous decision to alter their consent choices in real time (Kaye et al., 2015). DC can accommodate different approaches to consent (for instance, both broad and atomistic), depending on research context (Budin-Ljøsne et al., 2017). Through the online platform, participants can, for example, agree to or decline new research opportunities, record preferences for sharing data with third parties, self-report health information, and reflect on their existing consent decisions. The DC approach has been described elsewhere in the literature (Budin-Ljøsne et al., 2017; Kaye et al., 2015; Williams et al., 2015). It may be especially useful in the context of biobanking, genomic research and large cohort studies, where the future uses of tissue samples and data may be unknown at the time participants are recruited. It could also have relevance for other research disciplines, such as computer science and social network research (for example, Norval & Henderson 2017).

It is posited that DC will empower individuals and improve their experience in research through providing better flexibility and control, enhance communication and engagement between researchers and participants, and improve both recruitment to and retention in studies (Javaid et al., 2016, p. 819; Melham et al., 2014; Teare et al., 2017; Teare, Morrison, Whitley, & Kaye, 2015, p. 8). However, concerns have also been expressed, including that by permitting granular decision-making DC will lead to 'consent fatigue' (Hutton & Henderson 2015; Steinsbekk, Kåre Myskja, & Solberg,

2013), and that the relative ease of consent withdrawal will actually reduce retention rates. There are a range of potential risks and benefits of DC in relation to equitable participation in health research, such as an improved capacity to provide information that is translated or tailored to different audiences, as against the challenges of providing access in remote locations or catering for group-based consent (Pictor, Teare, & Kaye, 2018). Since DC was first conceptualised, it has been subject to competing claims of risks and benefits (Steinsbekk, Kåre Myskja, & Solberg, 2013); meanwhile its measurable effects on participants, researchers and research organisations are yet to be defined through empirical research. More specifically, there is a growing need to compare DC's effectiveness in facilitating and engaging participation with traditional approaches to gaining consent. In this paper, we propose a framework for evaluating and reporting on the effectiveness of DC that complements existing evaluation and reporting approaches for research. Our proposed framework can be applied consistently across studies that use DC, to build the evidence base for this tool.

We take the position that DC, like other mechanisms of obtaining informed consent, is an activity with multiple components designed to affect individual experiences of recruitment and participation in research. Such activities can be organised, modified, and tested in comparative studies with the overarching goal of improving their effectiveness. They are also influenced by context including, in the case of DC, the larger research project for which consent is being sought. This paper situates DC within the literature examining the effectiveness of informed consent methods more generally. It maps the parameters that researchers should consider when designing formal evaluations of DC, and it sets a research agenda for future comparisons of DC and other types of consent for research participation.

Establishing an evaluation and reporting framework for DC is particularly important at a time when adoption is growing, and new applications of and contexts for DC are emerging, making the question of its effectiveness more pressing (Pictor, Teare, Bell, Taylor, & Kaye, 2019). There is a risk that ad

hoc studies of DC, without an overarching conceptual and evaluative framework and the necessary attention to methodological conduct and reporting standards, will result in low-quality, poorly-reported, one-off evidence that cannot easily be applied elsewhere. Ultimately, this could undermine normative justifications for using DC. This framework will also be important as a basis for identifying the essential components of DC and distinguishing between instances where DC has been implemented and those where the label is claimed but the underlying approach fails to include the relevant components. It is important to note, however, that we do not seek to stipulate a narrow definition of DC nor to set out a single model to which researchers must adhere. Rather, we aim to promote the detailed reporting and evaluation of specific iterations of DC. In this way, the field may progress towards a more precise definition over time. The framework we outline here focuses on quantitative evidence. This does not, however, negate the need for robust qualitative research exploring users' perceptions and experiences of DC. Further normative analysis around key concepts of DC, and normative reflection on empirical findings, is also required.

This paper establishes a research agenda in support of transparent, well-reported and replicable evaluations of DC. There are already numerous randomised trials and several systematic reviews assessing diverse ways of improving informed consent for healthcare research. Our evaluation framework will, ideally, enable discrete studies of DC eventually to be combined into systematic reviews and meta-analyses. Taking this approach is important because well-conducted systematic reviews of research studies can provide answers to questions about the effectiveness of specific tools in a way that minimises bias, offering a robust basis for decision making. We aim, through this approach, to inform future conceptual development and implementation of DC for optimal research governance and participant engagement.

This paper is in two parts. The first, and most substantial, focuses on the development of an evaluation framework for DC. It proceeds by first examining the existing evidence base for similar consent approaches and then presents a logic model for DC. Emerging from these are

recommendations about study design for DC evaluation, and the selection of outcome measures.

We also examine contextual factors that may influence the measurable effects of DC. The second part of the paper draws on best-practice methodological guidance to make recommendations about how such evaluations should be reported, to ensure that future instances of DC can build upon this emerging evidence base.

Evaluation Framework for Dynamic Consent

Existing Evidence

To our knowledge, DC has not been subject to formal quantitative evaluation. While the DC approach arose at least in part from concern about participants' exercise of their legal rights of consent and withdrawal (Kaye et al., 2015, pp. 142, 144), it also draws upon the decades-long push to improve the quality of consent processes for research (Flory & Emanuel, 2004; Nishimura et al., 2013). This includes work to expand and tailor information provision about the research project or clinical trial, and to promote the decisional capacity of participants (including their understanding of information, appreciation of its significance, the ability to use it in a reasoned way and to express a clear decision) (Dunn, Nowrangi, Palmer, Jeste, & Saks, 2006, p. 1323). Future evaluation of DC will benefit from drawing upon this substantial body of research, while recognising that DC also has distinctive features; for example, it has a long duration – over the whole life of a research project – compared with traditional, paper-based informed consent approaches which occur once, when a participant joins a research study.

Typically, quantitative research to improve aspects of research consent has adopted comparative methodologies such as controlled trials (with or without randomisation) and controlled observational studies (Ryan et al., 2013). In all of these, some people participate in standard informed consent procedures whilst others experience some form of 'enhanced' process. The latter includes a wide range of different methods, for instance: extra face-to-face discussion; the addition of a technological aid such as a DVD or computer-based multimedia presentation; a test/feedback

loop to check people's understanding and address any gaps; and the provision of supplementary or differently-presented written information. Individual studies might assess one, or a combination, of these different methods compared to standard consent processes.

In this research area, it is common for studies to be simulated or hypothetical (Nishimura et al., 2013; Synnot, Ryan, Prictor, Fetherstonhaugh, & Parker, 2014), in which people are asked to imagine they are considering participating in a research project. This may be in recognition of the complexity of real-life studies of informed consent which usually involve attaching an evaluation of an informed consent process to another trial of an intervention (which we will describe as the 'parent' trial, but is sometimes also known as the 'host' trial (see Treweek et al., 2018)). This practice may elicit concern about the effect on recruitment rates and timeliness of this parent trial. It will therefore be useful to draw on guidance emerging from the Studies Within a Trial (SWAT) initiative in designing evaluations of DC (Clarke, Savage, Maguire, & McAneney, 2015; Treweek et al., 2018).

As one alternative to traditional paper-based consent, (non-enhanced) digital consent (often referred to as e-consent) has also been the subject of research (Jackson & Larson, 2016). We do not consider, however, that this can assist the development of an evaluation framework for DC, as it is an approach that conceptually is no different to standard written consent. As Henry et al. (2009) note:

there is no reason to expect consent text presented on a computer screen to be superior to a printed document, unless the computer software ... is used in a way that compensates for known strengths of that technology and/or known deficits in standard presentation modes (p. 3).

Systematic Reviews of Informed Consent Processes

DC is a complex tool, assembled from many components, and the ways in which these components interact both with each other and with contextual factors will shape the effects of the tool (Clark,

Briffa, Thirsk, Neubeck, & Redfern, 2012). It is important to try to both describe and disentangle DC's elements and examine the ways in which these may maximise benefit and minimise harm for participants in different contexts. The future planning, implementation and assessment of DC should be based upon a strong foundation of analysis of the evidence base (Craig et al., 2008). There are no trials nor systematic reviews of DC to date, but the related field of enhanced informed consent methods for research has advanced through several systematic reviews in the past fifteen years, discussed below. Researchers utilise the systematic review methodology "to collate all empirical evidence that fits pre-specified eligibility criteria [...] thus providing more reliable findings from which conclusions can be drawn and decisions made" (Higgins & Green, 2011, sec. 1.2.2). The following summary of these reviews will inform the DC reporting framework presented later in this paper.

Flory and Emanuel's 2004 review on ways of improving research participants' understanding of information disclosed in the consent process identified 42 studies (including some randomised trials), dating back to the late 1960s. They grouped the consent processes into five categories: (1) multimedia, (2) enhanced consent form, (3) extended discussion, (4) test/feedback, and (5) other, comparing these with standard processes. The effects on people's understanding, satisfaction, and willingness to enrol in the parent study were mixed.

In 2013, Nishimura et al. conducted an updated systematic review and meta-analysis, including only randomised trials. They identified studies reporting on 54 enhanced consent methods which met the selection criteria. This review showed that improved consent forms and extended discussion tended to benefit understanding, and that overall the approaches either benefited, or at least did not worsen, participant satisfaction and accrual. A year later, Synnot et al. (2014) updated a previous Cochrane systematic review of randomised and quasi-randomised studies of trial consent processes that incorporated audio visual information (Ryan, Pictor, McLaughlin, & Hill, 2008). Even within the narrower scope of 'audio-visual' enhancements to consent processes, sixteen studies were

identified, providing low to very low quality evidence of small improvements in knowledge and understanding of the parent trial, little or no difference in participation rates or willingness to participate, and possibly greater satisfaction with the information provided – but not with other aspects of the recruitment process. Other reviews including those by Cohn and Larson (2007) and Palmer, Lanouette and Jeste (2012) show similar trends in findings to those reported above.

Lessons from systematic reviews

These reviews show that there is diversity in the formulation and implementation of enhanced research consent processes as well as in the choice of the ‘usual care’ controls (Nishimura et al., 2013) and in outcome measures across the field of research. Importantly, no single, standardised measure of ‘informed consent’ exists. There are also substantial gaps across the evidence base to be addressed, such as the failure to identify a conceptual basis upon which altered processes are expected to achieve improvements in informed consent. Synnot et al. (2014) and Palmer et al. (2012) both comment on the absence of a conceptual framework of human information processing to underpin the interventions. Synnot et al. point to research by Sheridan et al. indicating that successful informational interventions in health have a clear basis in theory (Sheridan et al., 2011, p. 50). This view is reinforced by the guidance from the UK Medical Research Council (MRC) on developing and evaluating complex interventions, which states that connecting with appropriate theory “is more likely to result in an effective intervention than is a purely empirical or pragmatic approach” (MRC, 2006, p. 9). Our logic model for DC, described below, linking purposes with activities and intended outcomes, offers this required conceptual grounding.

Lack of participant engagement in design is another problematic issue. The systematic reviews described above indicate that end user involvement in the development and pilot-testing of methods to improve informed consent does not occur uniformly. For instance, in the review from Synnot et al., less than half of the included studies reported that research participants were involved in developing the informed consent tool (2014). The role of users, for instance in advising on or pilot

testing such tools, is generally unclear. One older, broadly-framed systematic review provided some limited evidence that healthcare recipient input to the development of patient information materials might enhance information clarity and improve the knowledge of people who read the materials; there remains much scope for further empirical research into user involvement in enhanced informed consent tools (Nilsen, Myrhaug, Johansen, Oliver, & Oxman, 2006).

Finally, the studies included in the reviews of informed consent are located almost exclusively in high-income countries. They also often exclude potential participants who have low literacy skills (Synnot et al., 2014). These factors severely limit the generalisability of the results (Prictor et al., 2018).

In summary, the field of research on ways to improve informed consent for biomedical studies (including clinical trials) is heterogeneous both in types of informed consent processes and in results, while being only narrowly applicable in terms of setting and participants. Those lessons on the importance of a conceptual basis and user involvement have helped to guide the development of our evaluation framework.

A Logic Model for DC

As the field moves forward, a common organising structure for evaluation or ‘logic model’ will assist in the gathering of systematic information about how DC compares to traditional consent approaches. The use of logic models are proven tools to help in evaluation (Frechtling, 2007; Knowlton & Phillips, 2013). Although they are typically applied to program planning, the ‘logic’ of logic models as an evaluation framework can also apply here. Logic models depict the causal chain of events affecting an outcome, and the hypothesized links between causes in affecting an outcome. Logic models can be text or conceptual depictions, and in the case of how DC is intended to work, link the intended outcomes of DC with the processes thought to achieve these outcomes, as well as the theoretical assumptions of the DC approach. A logic model of DC should provide a framework for describing 1) the relationships between the resources needed for a DC approach, 2) the activities

required for this approach, and 3) 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. In this context, logic models provide a way to comprehensively integrate three important areas: the planning of DC studies as well as the implementation and evaluation of these studies. This is particularly important as DC studies will likely be embedded in larger observational studies or trials, so a logic model approach could assist in distinguishing what is required by the embedded DC study versus the parent project.

Figure 1 depicts a logic model for evaluating and reporting on DC. This type of logic model links ideas to explore the underlying assumptions of a DC approach. It starts by examining *assumptions* which relate to the necessary elements that would need to be in place to execute a DC approach. *Inputs* refer broadly to the resources needed to conduct an embedded DC study. *Activities* relate to the processes, tools, events and technology for the study as well as the infrastructure needed to accomplish the research. *Outputs* describe the results of program activities. They relate to process measures like the number of participants in the study or study retention. *Outcomes* relate to specific changes in attitudes, behaviours, or skills one might expect from the experience in a DC study. *Impacts* relate to the expected changes that may result from an embedded DC study. These could be either short-term or longer-term changes to improved research policy.

<<Insert Figure 1 here>>

Recommendations for DC Evaluation

Trial Methodology

One important finding from the systematic review literature is that randomised trial methodology can be applied to evaluate informed consent processes. Randomisation is endorsed in the MRC's guidance on developing and evaluating complex activities (or 'interventions'), which states that researchers "should always consider randomisation, because it is the most robust method of

preventing the selection bias that occurs whenever those who receive the intervention differ systematically from those who do not, in ways likely to affect outcomes” (MRC, 2006, p. 10). We can also draw upon guidance from the Studies Within A Trial (SWAT) initiative, which encourages the use of randomisation in studies whose purpose is to assess the effects of alternative ways of conducting trials (Treweek et al., 2018, p. 3). The Cochrane Risk of Bias Tool provides guidance on how to assess bias in randomised research design, in five domains: selection, performance, attrition, reporting biases as well as other bias arising from a particular study protocol (Higgins, Altman, & Sterne, 2011). Specifically to minimise the risk of bias in the enrolment process, true randomisation methods (such as coin toss, or computer-generated randomisation) should be adopted in preference to quasi-random methods such as alternation, day of the week, clinical record number or study site (“Randomisation and randomised trial,” n.d.). Concealment of allocation up to the point at which participants are assigned to groups should also be maintained (Higgins et al., 2011). Blinding of participants and personnel involved in such a study may not be practical but blinded outcome assessment should be maintained if possible.

Choosing Outcomes to Measure: The Challenge of Heterogeneity

The choice of outcome measures is an important one in evaluating informed consent processes, if the findings are to support decision-making in policy and practice and reflect what is of importance to stakeholders. No single measure of 'informed consent' for research exists (Gillies, Duthie, Cotton, & Campbell, 2018; Joffe, Cook, Cleary, Clark, & Weeks, 2001), and various proxy measures are commonly utilised. Some measures have been developed that apply only in certain settings, such as in clinical cancer research (Joffe et al., 2001). The associated problem of heterogeneity in outcomes – both in terms of the outcomes specified (e.g. understanding, satisfaction with decision) and the measures used to assess them – has been identified in at least two recent systematic reviews of enhanced informed consent tools (Nishimura et al., 2013; Synnot et al., 2014). This heterogeneity reflects that these tools are expected to have effects across a wide range of domains (Williamson et

al., 2012). It makes it difficult, however, to look across studies of comparable interventions to draw conclusions about their pooled effects. It is also connected with the problem of outcome reporting bias, defined by Williamson et al. as: “results-based selection for publication of a subset of the original measured outcome variables” (2012, p.1). Outcome reporting bias means that outcomes with more favourable results are more likely to be reported than those showing no difference or a harmful effect of an intervention.

In informed consent research, various aspects of understanding (Flory & Emanuel, 2004; Gillies et al., 2018; Nishimura et al., 2013; Synnot et al., 2014) and recruitment rates are quite commonly reported, with a long ‘tail’ of other outcome domains such as satisfaction (in the research or the researchers, or with the consent information or decision), trust, decisional conflict or regret, time taken to administer the informed consent process, locus of control (Lavelle-Jones, Byrne, Rice, & Cuschieri, 1993, p. 889) and cost measures. Even for the most commonly-assessed outcomes, however, there is enormous variation in the specific concepts being evaluated and the tools used to do this. As Gillies et. al noted in a recent systematic review,

There is no universally agreed measure of ‘good’ informed consent for clinical trials that might be used to objectively evaluate whether the potential participant has understood what trial participation means for them, or indeed ensured that all other considerations appropriate to ensure informed consent has been achieved have been met (2018, pp. 2–3).

The outcome of understanding, for example, as surveyed by Sand, Kaasa and Loge in a systematic review, is variously reported as comprehension, recall, knowledge, perception, awareness, view, therapeutic misconception, effectiveness, making sense of, and other constructs (2010, p. 5). The type of information being understood or recalled also varies, as do the actual tools used to assess this and the timing and frequency of assessment. The high degree of heterogeneity in this, one of the most frequently measured outcome domains, extends even further, across the whole range of other outcomes relevant to informed consent research. The ongoing debate over whether detailed

participant recall of what they have agreed to, as a means of ensuring respect for persons in research, is also relevant (Robinson, Slashinski, Wang, Hilsenbeck, & McGuire, 2013). DC may be a useful mechanism for engaging with this debate.

This summary demonstrates the difficulty of establishing a single, comprehensive standard for informed consent evaluation. Considering this, the specification of a core set of outcomes can provide a practical alternative.

Core Outcome Sets

Core outcome sets are increasingly recognised to promote quality and consistency in evaluation and reporting. These are “an agreed standardised collection of outcomes...which should be measured and reported in all trials for a specific clinical area” (Williamson et al., 2012; See also Clarke, 2007). They represent minimum standards which can be added to for individual projects. The use of core outcome sets may reduce bias, improve the relevance of research evidence to its users, and increase the statistical power of evidence syntheses (ultimately reducing wastage in research expenditure). It has been driven by initiatives such as Core Outcome Measures in Effectiveness Trials (COMET), and endorsed by health research funders, clinical trial registries and regulatory authorities (<http://www.comet-initiative.org/cosuptake>).

The ongoing ELICIT project by Gillies et al will identify a core set of outcomes for the future evaluation of informed consent methods applying to potential participants in randomised trials (Gillies et al., 2015). While DC is not limited (nor perhaps even best suited) to randomised trials, nonetheless this project will provide the strongest available foundation to guide the choice of outcomes when evaluating DC. Moreover, the use of a methodologically sound core outcome set in DC research will facilitate the development of evidence syntheses to guide future implementation of DC-based approaches. It is anticipated that the ELICIT research will be completed in 2019 and we recommend that the findings be used to guide outcome evaluation in DC research.

Recommending Outcomes for Reporting in DC Evaluation

In evaluating DC, the choice of outcomes should be informed by input from researchers and research participants about what is meaningful for them. The choice should reflect the purposes for which DC has been applied in the given project. For example, if DC was implemented primarily to improve research participant engagement over an extended period, then measures of that engagement over that period should be included. Likewise, if a DC tool was mainly intended to improve participants' understanding of the research, this should be a primary outcome for assessment. Further, the outcomes selected should be consistent with a core outcome set, if available. If none is available they should be consistent with outcomes that are commonly reported in systematic reviews of similar interventions, to minimise heterogeneity across the research field. Evaluations should specifically include cost-effectiveness measures and participation and withdrawal rates for the parent study, as well as usage measures for the DC tool itself (e.g. click paths, bounce rate, time spent using the tool). To minimise bias, outcomes should be selected a priori and fully reported against (*see also **Reporting Framework** section*) and utilise reliable and validated measures where possible. To check the biases in the study design, statistical adjustments can also reduce bias in the outcome (Emam, Jonker, Moher, & Arbuckle, 2013). Finally, they should be evaluated at time points that are meaningful and reflect the long-term nature of DC applications.

Contextual Factors Influencing Evaluation Outcomes

DC's impact depends heavily on context, including the nature of the parent study, the level of understanding of research held by potential participants, and the reliability and accessibility of the digital technology. Disentangling participants' perceptions and experiences of DC from perceptions and experiences of the parent study may be challenging. For example, a study participant who receives a genetic test result of concern may record reduced satisfaction with the DC tool because of the concerning result rather than the tool itself. Participants with some knowledge of DC may have pre-conceived ideas about its use (Thiel et al., 2015), for example, about DC taking longer or

requiring a higher level of ongoing interaction with the study. Performance bias may also be introduced if participants in the control group are aware that they are not in the DC group.

Another potential confounding factor is whether DC itself has an educative effect around informed consent that changes how participants evaluate the tool. It is well documented that participants usually do not recall key points about the consent process or the study itself (Fortun, West, Chalkley, Shonde, & Hawkey, 2008; Rebers, Vermeulen, Brandenburg, Aaronson, & Schmidt, 2018; Sherlock & Brownie, 2014). A clearer understanding (or critical appreciation) of consent processes through participation in DC could affect evaluation outcomes either positively or negatively.

In addition to these examples of confounding factors affecting evaluation *within* studies, confounding factors *between* studies may also be relevant. As an example, in genomic medicine research participants may be worried about the results they will receive as part of the research, and the waiting time to receive results could be long. This could conceivably lead to anxieties not experienced in other types of studies. Extra consent choices or complicated decisions relating to particular kinds of research could cause study participants additional stress, for example, making choices about permissions for secondary use of their genomic and health data. However, one study recently showed that study participants view their DNA and genomic information in the same way as any other medical information (Kelly, Spector, Cherkas, Prainsack, & Harris, 2015), so concerns about such confounding factors in genomic studies may be unwarranted.

Another factor in evaluating DC is accounting for the effects of participants' pre-existing level of comfort using web-based technology and in communicating personal information in that way. The 'digital divide' may be given as justification not to implement DC with certain participant groups; it is important that DC evaluations address rather than avoid this issue. Eliminating bias in the form of these potential confounding factors is not always possible, but researchers planning an evaluation should identify confounders and apply appropriate study design and analysis methodologies. Truly randomised studies with a sufficiently large sample size are recommended to control for

confounders, and analysis may include stratification and multivariate regression analysis (Pannucci & Wilkins, 2010). This approach will be key to reliably determine the kinds of research and participants groups for whom DC is best applied, which will become clearer as the evidence accumulates.

This paper has, thus far, considered the evidence base for informed consent approaches and developed a logic model for DC. On this basis we have outlined recommendations about the design of future studies to assess the effects of DC, including guidance on how outcome measures should be chosen. The second part of this paper outlines how such studies should be reported so that DC can progress in terms of future evaluation as well as in practice.

Reporting Framework for DC

In recent years, evidence-based guidelines for the reporting of research have been developed and promulgated. Such guidelines are designed to promote transparent trial conduct and to overcome the problems caused by inadequate research reporting, including avoidable waste in research. They have been widely endorsed by leading editorial organisations and journal editorial boards. We recommend that evaluations of DC should adhere to appropriate reporting frameworks.

Reporting on Randomised Controlled Trials of DC

If a randomised trial methodology is adopted to evaluate DC (as we recommend), the latest version of the Consolidated Standards of Reporting Trials (CONSORT) Statement (Schulz, Altman, & Moher, 2010) should be utilised to ensure comprehensive reporting of the DC trial. This includes information on the study design, participants, interventions and outcomes. It also requires comprehensive reporting on: methods including randomisation, allocation concealment, and blinding; participant flow; and results. Further, the CONSORT statement has an extension (updated in 2017) for non-pharmacologic treatment (NPT) interventions (Boutron, Altman, Moher, Schulz & Ravaud, 2017), and another for embedded recruitment method trials (Madurasinghe & Eldridge, 2016), which should be adopted in reporting trials of DC. Two especially useful elements of the CONSORT-NPT extension are

the requirement for a detailed description of the components of the intervention as it was planned and delivered and reporting on the adherence of participants to the intervention.

An additional resource for researchers evaluating DC is the Template for Intervention Description and Replication (TIDieR) guidance, developed to address concerns about poor quality descriptions of interventions which impede their replicability. The TIDieR checklist should be addressed in reporting on DC studies to make transparent the key elements of the intervention including its underlying rationale, informational components, processes, the involvement of personnel, and the location, timing and duration of people's engagement with the tool. Further, considering this guidance informed by the rich literature on the design, implementation and evaluation of complex interventions in health will ensure a firm foundation for this research (Craig et al., 2008; Lysdahl & Hofmann, 2016; Moore et al., 2015; Petticrew, 2011; Richards & Rahm Hallberg, 2015).

Reporting on Other Study Designs Evaluating DC

Researchers should refer to the EQUATOR Network (<http://www.equator-network.org/>) to identify reporting guidelines suitable for other study designs that might be used to investigate DC. These include qualitative research (O'Brien, Harris, Beckman, Reed, & Cook, 2014) and observational studies (von Elm et al., 2007).

Reporting on the Parent Study

Since DC evaluation is usually attached to a parent study, it is important when reporting on DC tools to also report on elements related to the parent study. This enables the evaluation of DC's effectiveness to be understood in context. The report of the parent study may include information about the DC evaluation. Otherwise, a standalone report of the DC study should include background information on the parent study and, in that context, the rationale for testing DC. It should also reference any published information about the parent study.

Publication

To advance the field of DC it is imperative that the findings of evaluations are made publicly available. Borrowing again from the SWAT guidance, this could occur within the reporting of the parent study, or as a standalone paper (Treweek et al., 2018). In time, such reports should be collated in a well-conducted systematic review, reported according to PRISMA guidelines (Moher, Liberati, Tetzlaff & Altman, 2009), so that the science around DC can be advanced, and its benefits optimised and harms minimised.

Research Agenda

This paper establishes a research agenda for the evaluation of DC in order to develop the field. To advance the evidence base, projects that are planning to implement DC in a research setting should, ideally, embed a high-quality randomised controlled trial of their DC implementation compared with the usual consent method in that setting. This has been the approach adopted by Australian Genomics in its 'CTRL' project involving genomic testing for participants with rare diseases (Pearce, 2018).

Best Practices

Based on this evaluation and reporting framework, future studies of DC should adopt trial procedures (such as true randomisation and blinded outcome assessment) that will minimise the trial's risk of bias. Studies should report on a consistent range of outcomes using validated measurement tools, with attention to the posited benefits and risks of DC from the literature (such as whether DC improves recruitment, retention, understanding and engagement, avoids causing consent fatigue, and is cost effective). The potential confounding factors outlined above should be considered. If the use of a randomised trial design is not possible, careful choice of study design to maximise the quality of the resulting evidence remains vital. Studies should be reported fully according to relevant guidelines. The findings of research into the effects of DC should, in time, be combined in systematic reviews with meta-analyses of outcomes data. If formal evaluation of DC is not possible, we encourage researchers adopting a DC approach to publish descriptions of their

implementation so that others considering whether and how to implement DC can draw upon this literature.

Educational Implications

This consideration of the elements for evaluating and reporting on DC offers an opportunity to improve the awareness of researchers and ethics committee members about the importance of thoroughly considering consent processes, content and desired outcomes, and making deliberate choices about these in overall study design.

Conclusion

Interest in DC is growing. In the past five years several papers have been published outlining its potential in biomedical and other forms of research (Budin-Ljøsne et al., 2017; Hutton & Henderson 2015; Kaye et al., 2015; Norval & Henderson, 2017; Teare et al., 2017; Williams et al., 2015), and reports are beginning to appear from patient groups and organisations calling for its widespread adoption (Hazelton & Petchey, 2015). At a time when more emphasis is being placed on digital health, and the opportunities for connecting health data online are growing (Australian Digital Health Agency, 2018), there is strong interest in tools that will allow individuals to have greater control over how their data are being used for multiple purposes. This could further drive support for approaches like DC. On this basis, it is crucial that we better understand how DC influences the research participant experience, and whether it brings additional value compared with other consent mechanisms.

Implementing a clear approach to evaluation, to allow groups to appropriately plan the evidence-collection phase will be hugely beneficial to ensure robust data are collected, that can be compared between studies. If such evaluation demonstrates clear patient benefit this could encourage widespread uptake of DC, and open conversations with ethics review bodies to better understand how to support rollout and implementation.

The evaluation approach outlined within this paper will enable DC to be considered as a distinct tool within a study and compared to other mechanisms of consent. This will promote collection of unbiased data about how individuals experience the consent process and whether the specific processes used can improve or influence participant experience and understanding. This may usefully demonstrate that DC can fulfil the expectations that have been described in other papers, for example as depicted by Budin-Ljøsne et al. (2017), or equally examine questions that have been raised in criticism of DC by documenting whether, for example, users tend to disengage with consent decisions, and whether information provision is overwhelming (Steinsbekk, Kåre Myskja, & Solberg, 2013). Either way, by gathering quantitative data relating to the primary outcomes of a project's implementation of DC, we will have a means by which to compare studies and refine processes to help improve consent mechanisms and support participants to make informed decisions.

These evaluation studies will be highly relevant for biomedical research and more broadly for digital health in clinical care, as patients are increasingly required to make decisions about the use of their health data online, in biomedical research, clinical care, as well as more broadly. It is our hope that teams developing DC tools will carefully consider how they evaluate their approaches to start contributing high-quality data to this field, including reporting instances where there is no clear advantage, to progress our collective understanding of the effects of DC.

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<https://doi.org/10.1186/1745-6215-13-132>

Acknowledgements

The idea for this article emerged from a workshop entitled ‘Requirements for implementing Dynamic Consent’ funded by the Daiwa Anglo-Japanese Foundation which was organised by Dr. Harriet Teare, Professor Jane Kaye and Professor Kazuto Kato. The workshop was coordinated in conjunction with the International Symposium on Genomics and Society (Genome ELSI Kyoto 2017) (part of the ELSI Leader Research Program funded by the Japanese Agency for Medical Research and Development [AMED], Japan). This event would not have been possible without the generous support of the DAIWA Anglo-Japanese Foundation Grant (Ref: 11037/12349) awarded to Professors Kato and Kaye. We gratefully acknowledge the other participants at the workshop: Tiffany Boughtwood, Alastair V. Campbell, Donald Chalmers, Jim Davies, Nao Hamakawa, Kazuto Kato, Su Yoon Kim, Atsushi Kogetsu, Eric Juengst, Fuji Nagami, Soichi Ogishima, Go Yoshizawa.

Author Contributions

MP, MAL, AJN, MH, HJAT made substantial contributions to the conception and design of the work and drafted the article. SB, HK, MK, JM, FM-G, BY and JK substantively revised the article. All authors read and approved the final manuscript.

Declaration of Conflicting Interests

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

Funding

The author(s) received no financial support for the research, authorship, and/or publication of this article.