

# Data and Analysis: Co-designing genomics research with a large group of donor-conceived siblings

## About this document

This document contains additional data relevant to the case study 'Co-designing genomics research with donor-conceived siblings: 'Into a murky unknown''. In addition it contains a more detailed description of the data sources in this case study. This document includes the preferences mapping data (STARDIT-PM), and other data about this initiative<sup>1</sup>. The corresponding Standardised Data on Initiatives Alpha Version (STARDIT) of the report can be found in 'Additional File 2 - STARDIT report'.

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## Definitions of terms

We have used consistent language to describe concepts throughout this case study. The list below defines important terms used throughout.

<p><b>Involvement</b> – The words ‘<b>involvement</b>’ or ‘<b>being involved</b>’ describe the concept of people being ‘involved’ in research. This is when research is carried out ‘with’ people rather than ‘on’ them.<sup>2</sup> ‘Involvement’ can also be defined as when other people aside from the research team, such as the public, patients, research participants and other stakeholders, actively contribute to the research process.<sup>3</sup> It is the ‘active involvement’ in shaping and guiding research, rather than only providing data.<sup>4-6</sup></p>
<p><b>Engaged</b> – participants in the online discussions are described as ‘engaged’ if they are reading and commenting in discussions, and ‘disengaged’ if they are not.</p>
<p><b>Enablers</b> – enablers are things which ‘facilitate’ certain things happening. For example, something which enables someone to participate in research.</p>
<p><b>Facilitator</b> – in this article it refers to a person facilitating online discussions. For example, “<b>Facilitators</b> shared views about <b>enablers</b> of involvement”</p>
<p><b>Online community</b> – as the half-siblings discovered one another through various direct-to-consumer ancestry services, they self-created an online community which used a mailing list to an email group as the mode of communication. Participants were recruited from this online community.</p>
<p><b>Online discussion</b> – the study team recruited participants to an online discussion, specifically created for this study and hosted on the secure platform Loomio.</p>
<p><b>Participant</b> – a person who participated in the process of sharing views and perspectives about the genomics research, including sharing views about preferences for any future involvement.</p>
<p><b>Potential participant</b> – before inviting people to become participants, it was necessary to involve a number of potential participants to help advise and plan the process.</p>
<p><b>Shared Ancestry groups</b> – refers to people who have shared ancestors and have been grouped into these sub-populations by genomic researchers.</p>
<p><b>Sibling group</b> - refers specifically to the community of shared interest defined by people who shared the same sperm-donor father, Bertold Wiesner.</p>
<p><b>Stakeholder</b> – this term includes anyone who has a ‘stake’ in the research, in particular those who have important knowledge, views or perspectives that should be taken into account.<sup>7,8</sup> In this paper it refers to participants, representatives, patients, parents and carers of patients, potential patients and the study team (including researchers and representatives) and the wider public.</p>
<p><b>The study</b> – the study refers to the formal research described in this case study, which was overseen by the ‘Ethics, Integrity and Biosafety team’ team at La Trobe University and the La Trobe University Human Research Ethics Committee.</p>
<p><b>Study team</b> – this process was guided by the study team, who consisted of academic researchers and members of the sibling group. The work of the study team was also advised and overseen by the ‘Ethics, Integrity and Biosafety team’ team at La Trobe University and the La Trobe University Human Research Ethics Committee.</p>

**Sub-populations** - This term refers to any grouping of people below the population level. Groupings can include communities of shared interest defined by shared genetic variation. These can include groupings of people who are half-siblings.

**The process** – this term will be used to describe both the study and the co-design process which involved members of the sibling group by inviting them to share views and perspectives about genomics research, including sharing views about preferences for any future involvement.

## Case study background and context

The practice of artificial insemination existed in a legal and ethical grey area for many years<sup>9</sup>. For example, in the UK in the 1950s, the legitimacy of children conceived from a donor father was unclear as husbands were registered as the fathers, which was legally an offence<sup>10</sup>. Despite the recognised ‘immense social, moral and medical questions’ raised by this process<sup>9</sup>, there was no legislative oversight and the practice was self-regulated by the individuals managing clinics carrying out the procedure. Members of the UK’s House of Lords suggested that artificial insemination of married women with the husband’s consent be classified as adultery as late as 1954<sup>9</sup>. Such contemporary attitudes created potential ethical, legal and social issues for families conceiving in this way<sup>9</sup>. Subsequently, there was a requirement for discretion for all involved, including the identities of donors which were surrounded by ‘complete secrecy’<sup>9</sup>. Additionally, it was the view of some doctors that parents should not know the identity of the donors as it was ‘incompatible with secrecy’<sup>10</sup>. Accordingly, many parents were encouraged to never disclose the paternity to the offspring.

The total number of donor conceived people in the UK by 1958 was estimated to be 7500, and 100,000 in the United States<sup>9</sup>. One pioneering clinic mentioned in debates of the UK Parliament in the 1950s was the Dr Mary Barton’s medical practice, which operated in London from the 1940s to the 1960s and was responsible for at least 433 children with Dr Mary Barton stating that she had seen 600 prospective parents between 1944 and 1954<sup>11–13</sup>. The Barton practice used donors from ‘intelligent stock’ and ruled out donors where there was ‘inheritable disease on that side’ or ‘criminality’, introducing concepts of ‘the eugenic quality of the donor’s stock’ into the very earliest years of the practice<sup>10</sup>. While attempts were made to find suitable donors that were a ‘match’, (including parents choosing whether or not they wanted a ‘Jewish’ donor<sup>10</sup>)<sup>13</sup>, some early practitioners of artificial insemination used donors from ‘a very small panel of donors’<sup>10</sup>, often from their own immediate social circles<sup>13,14</sup>.

One prolific donor was Dr Barton’s husband, the scientist Bertold Wiesner, a consulting biologist at the Royal Northern Hospital in the 1940s<sup>15</sup>. According to some estimates Wiesner may have fathered up to 1000 offspring during the time the clinic was operational<sup>16</sup>, despite a 1945 British Medical Journal paper where Barton and Wiesner stated they set an ‘arbitrary limit of 100 children for each donor’<sup>10</sup>. While a Government register of donors was proposed in the UK as early as 1949<sup>9</sup>, it was never created. Subsequently, a number of people who have discovered they are the offspring of Wiesner have discovered each other by various means in subsequent years (including using direct-to-consumer genetic testing services) and formed an online community. Some members of the group have disclosed their biological relation to Wiesner (and thus other siblings) in the public domain through media, including documentaries<sup>14,17,18</sup>, and advocated for the rights of people who are donor conceived<sup>19</sup>.

# Survey questions

## Pre-discussion survey questions

After participants had read the Participant Information and given consent to participate, they were asked to complete the following information. The questions below are worded exactly as participants read them.

1. Full Name (if you would prefer to not use your real name you may use a pseudonym)
2. Email address (Please note this needs to be a working email address. If you do not have one leave this blank and we will contact you by your preferred method of communication.)
3. Phone number (optional)
4. Any other preferred method of communication? (Please share any other preferred method of communication if email or phone are not preferred)
5. Age
6. Gender (Choose from 'Male', 'Female', 'Transgender', 'Intersex', 'Other', 'Prefer not to say')
7. Educational background (tick all that apply)
  - a. Middle school qualifications (up to age 16) ('lower')
  - b. High school qualifications (ages 16-19) ('middle')
  - c. Degree (bachelors), diploma or post-graduate ('higher')
  - d. I have qualifications or professional experience in genomics (professional)
  - e. Prefer not to say
8. Please tick which of the following statements that you agree with:
  - a. I feel comfortable describing other descendants of my biological father as 'half-siblings'
  - b. I would describe our email group as an 'online community'
  - c. Members of this email group potentially have a shared interest in discussing future research which might affect them, including genomic research
  - d. If you do not feel comfortable describing other descendants of your biological father as 'half-siblings' please share any term (or terms) you prefer.
9. In which country do you live (or spend most time)?
10. What made you decide to respond to our invitation to participate in this project?
11. What do you hope to get out of participating in this discussion? Do you have any specific expectations?
12. There are many benefits of involving people other than researchers in the co-design of research studies at every stage of the research cycle. Research suggests that involving people improves the quality and the relevance of the research. Involving people can also improve participant experience and increase participation. **Who do you think should influence what kind of genomic research should be done in the future?**
13. What makes you say that? (why did you give that answer?)
14. Do you have any ideas about how the people from your previous answer could influence future research? (For example, what tasks could people affected by EGID be involved in?)
15. **Which aspects of any future research genomic research should be influenced by the following** (participants were presented with a grid of tick boxes, the horizontal axis being who should be involved, the vertical a list of tasks. The horizontal was as follows)

Everyone (any member of the public who is interested)	Anyone who might be indirectly affected by the research	Only people who are directly affected by the research	Only people who are participating in the research	Only people with a professional role in research
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- a. All aspects mentioned below (leave others blank if ticking this)
  - b. Finding questions to ask (identifying research topics)
  - c. Deciding which questions to prioritize and fund
  - d. Deciding how to try and answer the question (the research method)
  - e. Attempting to answer the question (carrying out the research, including collecting information)
  - f. Trying to understand if it is possible to answer the question (analyzing the information)
  - g. Sharing the information that has been found, and any answers that may have emerged (dissemination and publication)
  - h. Ensuring that any information or answers are able to be used to help people in practice, policy or future research (sometimes called research translation)
  - i. Deciding if the way of asking the question and all the other stages of the research were appropriate (evaluating the research method and any impacts)
  - j. Designing how people are involved in the research
16. Have you ever participated in research in the past? (by participation, we mean as a research subject – for example part of a trial)
- a. Yes
  - b. No
  - c. Prefer not to say
  - d. Unsure
17. Have you ever participated in research in the past? (by participation, we mean as a research subject – for example part of a trial)
- a. Yes
  - b. No
  - c. Prefer not to say
  - d. Unsure

## Post-discussion survey questions

1. How would you rate the following? (chose from 'Excellent', 'Somewhat good', 'Neither good nor bad', 'Somewhat poor', 'Extremely poor')
  - a. Your overall experience of participating in the online discussion
  - b. Your assessment of how we conducted the survey and discussion format
  - c. The support you received to be involved (for example, practical support such as instructions for using the online tools)?
  - d. Information and learning materials you were given before the event
2. Did you feel you meaningfully contributed to the discussion?
  - a. Yes
  - b. No
  - c. Unsure
3. Is there anything in particular you liked or thought was helpful about how the discussion was conducted?
4. Is there anything you didn't like, thought was unhelpful. or could have been improved about how the discussion was conducted?
5. Do you have any other thoughts, ideas or comments?
6. Would you like to be updated about the progress of the research and offered chances to be involved where possible? (Choose 'yes' or 'no')

7. Did you have any expectations from participating in this research that were met or not met?
8. Have any of your views and perspectives about involving people in genomic research changed since participating in this research? If so, please describe.
9. There are many benefits of involving people other than researchers in the co-design of research studies at every stage of the research cycle. Research suggests that involving people improves the quality and the relevance of the research. Involving people can also improve participant experience and increase participation. **Who do you think should influence what kind of genomic research should be done in the future?**
10. What makes you say that? (why did you give that answer?)
11. Do you have any ideas about how the people from your previous answer could influence future research?
12. For example, what tasks could people be involved in?
13. **Which aspects of any future research genomic research should be influenced by the following** (participants were presented with a grid of tick boxes, the horizontal axis being who should be involved, the vertical a list of tasks. The horizontal was as follows)

Everyone (any member of the public who is interested)	Anyone who might be indirectly affected by the research	Only people who are directly affected by the research	Only people who are participating in the research	Only people with a professional role in research
---	---	---	---	--

- a. All aspects mentioned below (leave others blank if ticking this)
  - b. Finding questions to ask (identifying research topics)
  - c. Deciding which questions to prioritize and fund
  - d. Deciding how to try and answer the question (the research method)
  - e. Attempting to answer the question (carrying out the research, including collecting information)
  - f. Trying to understand if it is possible to answer the question (analyzing the information)
  - g. Sharing the information that has been found, and any answers that may have emerged (dissemination and publication)
  - h. Ensuring that any information or answers are able to be used to help people in practice, policy or future research (sometimes called research translation)
  - i. Deciding if the way of asking the question and all the other stages of the research were appropriate (evaluating the research method and any impacts)
  - j. Designing how people are involved in the research
14. Full Name (Optional- if you would prefer to not use your real name you may use a pseudonym)
  15. Email address (optional)
  16. Phone number (optional)
  17. Age
  18. Gender (Choose from 'Male', 'Female', 'Transgender', 'Intersex', 'Other', 'Prefer not to say')
  19. Educational background (tick all that apply)
    - a. Middle school qualifications (up to age 16) ('lower')
    - b. High school qualifications (ages 16-19) ('middle')
    - c. Degree (bachelors), diploma or post-graduate ('higher')
    - d. I have qualifications or professional experience in genomics (professional)
    - e. Prefer not to say
  20. Please tick which of the following statements that you agree with:

- a. I feel comfortable describing other descendants of my biological father as 'half-siblings'
- b. I would describe our email group as an 'online community'
- c. Members of this email group potentially have a shared interest in discussing future research which might affect them, including genomic research
- d. If you do not feel comfortable describing other descendants of your biological father as 'half-siblings' please share any term (or terms) you prefer.

21. In which country do you live (or spend most time)?

## Facilitator survey questions

The Facilitator (MC) was surveyed 6 months after the online discussion in order to integrate the valuable views and perspectives of those involved in planning and delivering the process. Design of surveys was informed by best practice frameworks for public involvement<sup>20,21</sup>. This method was informed by the Public Involvement Impact Assessment Framework Guidance (PiiAF)<sup>20</sup> and the questions were informed by sections 7 and 8 of the GRIPP2 reporting checklist<sup>21</sup>. The Facilitator was asked 11 questions and the data was coded and categorised, including using the STARDIT framework<sup>1</sup>. The data was then compared and integrated with the other data from the interviews and checked by other study team members (JN and PL).

1. Please describe your tasks in the process of involving people in planning of the study
2. What did you learn from the process of involving participants in the research planning phase?
3. Please describe specifically what worked well or was useful about the way the study was conducted (including how people were involved)
4. Please describe specifically what did not work well or was not useful about the way the study was conducted (including how people were involved)
5. Were there any barriers or enablers to conducting the study or involvement activities? (institutional or otherwise)
6. Do you think the involvement activity achieved its intended aim(s)?
7. Do you think the study achieved its intended aim(s)?
8. Do you have any advice to other researchers planning involvement for their research?
9. Do you have any advice to other researchers planning to involve people using online discussions?
10. Describe the impact you think involving people had (positive/negative - on the research, staff or participants)
11. Who do you think should influence the kind of human genomic research done in the future, and why? (e.g. the public, participants of research studies, doctors, school children, politicians etc)
12. Which stages of future genomic research should be influenced by people other than researchers (if any)? (e.g. concept planning of new studies, study design, conducting the research, presenting the results etc)
13. Other comments

## Data

### Data sources

This table summarises all the data sources used for the case study.

Data Category	Data point description
<b>Diary</b>	Research diary of lead investigator (JN) – including reflections during the process
<b>Emails and meeting notes</b>	Email, meeting notes and notes from planning and discussion. This included involvement of potential participants in co-designing and co-refining the study.
<b>Online pre-discussion survey</b>	Informed consent and pre-discussion survey data
<b>Learning resources</b>	Learning resources for participants and the Facilitator giving information about genomics and using Loomio (see section ‘Learning resources’)
<b>Online discussion with participants</b>	Text data from online Loomio discussion with participants
<b>Online discussion with facilitators</b>	Text data from online Loomio discussion between Facilitators of two parallel studies
<b>Online post-discussion survey</b>	Post-discussion survey data from participants
<b>Follow up survey for facilitators</b>	Post-discussion survey data from Facilitators and additional emails with further reflections

## STARDIT Preference Mapping (STARDIT- PM)

This table uses the Alpha version of the Standardised Data on Initiatives Preference Mapping (STARDIT-PM) to categorise the data into certain areas<sup>1</sup>. Preferences were recorded from all data sources, including the initial survey, online discussion with participants, online facilitator discussions, follow-up surveys with participants and with facilitators. Facilitator comments in the online discussion were not included. If the same participant made the same point at different stages, this was counted as one view. The standardised categorisation is intended to facilitate comparison with other studies. Accordingly, the content may be similar to other sections of the qualitative thematic analysis.

STARDIT-PM area and quantitative data	Qualitative summary
<p>Views on who should be involved:</p> <p><b>10 participants</b> shared views about who should be involved</p>	<p>Participants stated that anyone should be involved in research, with a experts, people affected by the research directly and the public all sharing perspectives in the context of research carried out with ethical oversight. Six participants stated that anyone should be involved in research, with one participant stating ‘everyone should have a voice not just scientists and researchers’ [P5]. Another participant stated ‘it needs to be a wide-ranging discussion so that the benefits and possible problems can be fully explored’ [P4]. One participant said it can depend ‘what kind of research it is’ and what the purpose is [P7], with another adding ‘we all need to have a voice’ as ‘we may not be “experts” in genomics but our opinions must be respected and have validity’ [P5].</p> <p>Six participants stated those affected by research should be involved, however others challenged this saying this could provide ‘a rather one-eyed perspective’ [P4]. One participant stated ‘some research will benefit certain people - those people should probably influence it if they are an identifiable group’. Another participant noted that ‘we’re all biased; whoever is affected by a condition is likely to want it prioritised’ [P12]. Another participant concluded ‘we’re all biased; whoever is affected by a condition is likely to want it prioritised’ [P12].</p> <p>One participant stated that people (including the public and research participants) will have a ‘variety of professional and technical and creative skills’ which will be useful, with the most useful one being ‘knowing ourselves’ [P7].</p> <p>One participant stated ‘I am a strong supporter of patient involvement in medical care’ and that ‘involving members of the public’ in genomic research was important in order to ‘have their views, reactions, interpretations, questions, concerns sought, interacted with, and considered’ [P11].</p> <p>Two participants stated that experts (including ‘scientists’ [P9]) who ‘know what they are doing’ should be involved [P2], with ‘research reviewed by ethics boards’ [P9].</p> <p>There was a recognition that different groups in society might have different interests and influence. One participant articulated groups including ‘medical scientists’, ‘social scientists’, ‘psychologists’ and the</p>

general public as being groups which should influence research, but noted that not all 'groups should have equal influence'[P10]. One participant asked 'there will be many interested groups so which ones will be listened to?' [P4] One participant stated that 'people who are not looking for personal gain, but who have a desire to improve quality of life and help us understand ourselves' should influence research [P6].

Views on specific tasks people involved could do:

**8 participants** shared views about specific tasks people involved could do

One participant said that research participants should be involved in 'agreeing purpose, parameters and methods' [P7]. Another asked 'whatever format is decided upon who would decide on the points for discussion?', implying that participants' tasks should include deciding this [P4]. One participant added that it is a 'good idea to involve research subjects in formulating the research questions' [P10]. Another participant stated they should be involved in 'having a say in what research is supported by public money' and 'making sure that the uses and purposes to which the research is put are responsible and allied with the laws and mores of our society' – which includes 'ethical oversight' [P7]. One participant also added that the public should have a voice in how 'science and research can better involve' people [P5]. In reference to future research with the sibling group one participant stated that ideally 'we would be able to exert control over the use' of data [P7]. One participant felt they should be involved in 'seeking answers to old, or not yet thought of questions' and 'looking beyond the known into a murky unknown' [P6]. The discussion also explored who should be involved and in which tasks. One participant noted they didn't feel 'qualified' to 'comment on aspects of science itself' but felt 'strongly' that they should be involved in ethical decisions and sharing personal experiences to help inform research [P5]. They also stated experts 'need to drive research' but they 'cannot do it in vacuum' as the public need them and they need the public [P5].

Views on modes of communication:

**4 participants** shared views about preferred communication modes

One participant stated 'moderated face to face discussions (through video if need be) remain the best method in my opinion for focussed outcomes and decisions with groups of people' [P7]. Another participant stated that 'most of our group would be able' to use video-conferencing platforms [P5]. Face-to-face synchronous discussion was ruled out by another participant as there are 'too many voices' which are across multiple time-zones [P4]. Communicating via a verbal interview was perceived as taking less time than anything which 'requires a lot of writing' [P4]. One participant said they were 'happy to contribute in any way in which is practical online' [P5], with another adding 'email is still the best way' [P4]. One participant suggested there would be 'differing preferences for how such a project should be organised' and suggested agreeing on some ideas (not using email) and then sending these ideas to the group once decided [P4]. One participant concluded that 'a forum for considered comments' online can be useful as long as enough time is allowed [P7].

Views on what methods should be used to involve people:

**5 participants** shared views about what methods

The participatory research method was described as 'commendable' [P9]. Participants suggested the idea of using one to one interviews as way of involving people (including using telecommunications) [P5], however, one participant noted that one-to-one interviews can restrict discussion and 'be quite straight jacketed with circumscribed questions', compared to online discussions [P4]. A 'group of "special interest" people involved in a group discussion' was suggested as a 'simple but effective method of encouraging debate', if participants can

should be used to involve people

'can dip in and out' [P4]. They also stated group discussion would not work as 'there are too many voices and some would be drowned out' [P4]. This participant also stated 'we will have differing preferences for how such a project should be organised' and asked 'would it be possible to agree on some ideas and then post them to the group' in order to involve people in co-creating how they will be involved and participate [P4].

One participant suggested that discussion, interviews, surveys and 'documents and videos shared for feedback' would all be viable methods of involving people [P5].

Another participant suggested a professional 'market researcher' who was 'tasked with finding a cross-section of people' might be a helpful method to involve people [P4].

One participant shared highly-specific views about the method that should be used, stating 'It should not be a plebiscite' nor 'self-electing moral Praetorian guard', adding that 'the more diverse the debate, the more dilute the effect of irrational preconception and ethical incompetence should become. The model of representative democracy seems to me the best available' [P8].

Another participant stated that 'ongoing discussions using social media and specific pages' such as Facebook pages, could be a good method to involve people [P5].

Another participant concluded that 'Moderated face-to-face discussions (through video if need be) remain the best method in my opinion for focussed outcomes and decisions with groups of people', adding that 'a forum for considered comments which are neither binding nor meant to be conclusive such as this can certainly be online' as long as enough time is allowed' [P7].

Views on enablers (facilitators) of involvement:

**7 participants and 3 Facilitators** shared views about facilitators of involvement

One participant noted that being 'highly educated' was an enabler for involvement and that having a 'bit of time on their hands' was also an enabler [P4]. Being 'respectful' when involving 'those affected by genomic research' will facilitate research as the 'more brains applied to research, the more likely answer to puzzles will be found' [P11]. Similarly, another participant stated 'the more diverse the debate, the more dilute the effect of irrational preconception and ethical incompetence should become' [P8]. One participant stated that whatever model was chosen, it should be 'as flexible as possible' [P5].

Four participants reported specific things about the way this study was conducted that facilitated their involvement. One participant said the entire process was 'assiduous' and that the 'intent of this project' was 'obviously thoughtful and interesting'[P9]. One participant said the 'system seemed to work well' [P7]. Another added that being used to online platforms like Loomio, or having previous experience of similar platforms and 'used to' that way of communicating might facilitate involvement using that communication mode. One participant suggested an alternative discussion format where the participants

discussed a thread for 2 days and then had a 3 day break before coming to another thread [P7].

The Facilitator (MC) noted that regular contact with the study team and timely support was essential and they 'could not have done it without this'.

Views on barriers of involvement:

**6 participants and 3 Facilitators** shared views about barriers of involvement

Barriers to involvement in research identified by participants included public fear and 'hysteria' caused by a lack of understanding, which may 'hamper' research, involvement and general public support for research [P5]. Synchronous discussion was highlighted as another barrier if participants 'are across time zones' [P4]. One participant mentioned that they felt that their emotional response to some issues made it difficult to get involved in some ways [P6]. Being required to watch lengthy videos was identified as a barrier by one participant. One-to-one interviews were mentioned as being 'quite straight-jacketed with circumscribed questions' compared to more open online discussions [P4]. They also stated 'I don't think that a group discussion would work as there are too many voices and some would be drowned out' [P4]

Four participants reported specific things about the way this study was conducted that were barriers to their involvement. A discussion about boundaries revealed that some participants felt 'avoiding topics which might trigger emotions which are stressful or unpleasant' could be viewed as 'restrictive, even censorious' [P7]. The pace of the discussions was mentioned as moving 'too quickly' with another adding 'more time' was needed and study team should 'reconsider the pace of the research' [P7] [P4] [P5]. Updates from the discussion were sent to participants according to their preferences, and one stated they 'lost track of emails' and were sometimes unsure if they were 'responding to the right part' [P6]. Two participants stated the 'platform presented technical difficulties' [P4] and that it was 'complicated' [P5]. One participant stated the 'premise and the purpose of the study could be clearer' and that the various discussion threads were 'difficult to untangle sometimes' [P7]. They also mentioned it was 'hard to be able to guarantee to do this every day for a period' and that not doing so meant they 'got lost' [P7]. Another participant added that 'it's a difficult subject to discuss in a vacuum, without real life examples' [P4]. One participant expressed 'trepidation' at sharing views about research and compared the feeling to getting an answer wrong in an 'exam' [P6].

The Facilitator (MC) stated that they felt more time was required in the co-design process. In addition, the administrative processes surrounding the unplanned change of Chief Investigator and related administrative processes in relation to the ethics process (outside of the control of the study team) meant they felt support was 'non-existent' and was 'wholly inadequate' for the participatory research process being used.

Views on what the outcome or output of the involvement could be

One participant stated they wanted to know that their involvement had been 'useful to the researchers' [P11], with another stating an outcome of being involved would be the 'satisfaction of knowing that I may have contributed' [P10]

## 2 participants

shared views on what the outcome or output of the involvement could be

Views on which stage of the research people should be involved:

**1 participant** shared views about which stage of the research people should be involved

One participant noted that changing study design in the later stages of a study not always possible, posing the question 'can we be both subjects and supervisors - at the beginning yes, later, maybe not' [P7]. The participant also stated that compared to 'highly-qualified scientists who know what's possible' some 'relatively ignorant lay people' will be limited in what they can offer at some stages [P7]. The participant suggested that getting the 'purposes, the parameters and the methods agreed with all participants' at the design stage might be most appropriate [P7].

Views on who should the data from this project shared with?:

**3 participants** shared views on who data from this project should be shared with

Participants shared views about who data should be shared with, with One participant stated explicit concerns about sharing data for political or financial use [P5], with other participants agreeing. One participant stated 'Research is for humankind. Its benefits should be available to all. Information should be for the most part easily available. If it's publicly funded, it must be publicly available [P9]'. Cultural conventions around ownership of knowledge such as 'patenting' were challenged as forms of knowledge control which are 'unethical' in some contexts.

Views on how think learning from this research could be used

**8 participants** shared views about how learning from this research could be used

Eight participants shared multiple views on how learning from a proposed study could be used, including predicting human traits (for example, risk of diseases or mental health problems). One participant indicated the study could help individuals in the group understand 'what unwelcome genes we might have inherited' [P4]. Participants also indicated the research could be used to improve understanding of how things like personal experience, socioeconomic circumstances and culture interact with genomics. One participant added 'longitudinal studies might well be set up to study a cohort of babies with certain genomic sequences which predispose them to certain diseases' [P4]. Another participant stated research with the group 'could have implications for all' people [P3].

## Demographic information

Category	Pre-discussion survey	Post discussion Survey
<b>Gender</b>		
Female	7	3
Male	5	3
<b>Age</b>		
50-54 years	1	0
55-59 years	2	0
60-64 years	0	0
65-69 years	6	4
70-74-years	3	2
<b>Educational background ('highest' only counted)</b>		
Degree (bachelors), diploma or post-graduate ('higher')	11	5
High school qualifications (ages 16-19)	1	1
<b>Number who agreed with following statements</b>		
I feel comfortable describing other descendants of my biological father as 'half-siblings'	12	N/A
I would describe our email group as an 'online community'	9	N/A
Members of this email group potentially have a shared interest in discussing future research which might affect them, including genomic research	11	N/A
<b>In which country do you live (or spend most time)?</b>		
Canada	3	2
Greece	1	0
Spain	1	1
United Kingdom	7	3

## Views about who should be involved in research

Participants were asked the same questions before and after the online discussion. When asked who should be involved in various tasks in research, participants could choose from the categories outlined in Figure 2 in the main article. A change in direction is described as ‘widening’, the inverse as ‘narrowing’. Widening was calculated as being a move towards an attitude that more people should be involved in research, whereas narrowing was calculated as a move towards an attitude that fewer people should be involved.

A total of 54 responses were given by 6 participants where participants completed answers to questions of both the baseline and follow-up survey. 35% of responses showed a change towards ‘widening’ involvement (N=19/54) while 8% ‘narrowed’ (N=8/54). 50% of responses stayed the same (N=27/54).

### Widening and narrowing for each question

Who should influence which aspects of research?	Change to wider	No change	Change to narrower
Finding questions to ask	2	4	0
Deciding which questions to prioritize and fund	2	3	1
Deciding how to try and answer the question (the research method)	2	2	2
Attempting to answer the question (carrying out the research, including collecting information)	2	3	1
Trying to understand if it is possible to the answer the question (analysing the information)	3	3	0
Sharing the information that has been found, and any answers that may have emerged (dissemination and publication)	2	3	1
Ensuring that any information or answers are able to be used to help people in practice, policy or future research (sometimes called research translation)	2	3	1
Deciding if the way of asking the question and all the other stages of the research were appropriate (evaluating the research method and any impacts)	2	4	0
Designing how people are involved in the research	2	2	2
<b>Change totals</b>	<b>19</b>	<b>27</b>	<b>8</b>

## Participant experience

<b>How would you rate your overall experience of participating in the online discussion</b>	
Somewhat good	4
Neither good nor bad	1
<b>How would you rate how we conducted the survey, and discussion format</b>	
Excellent	1
Somewhat good	2
Neither good nor bad	1
Somewhat poor	1
<b>How would you rate the support you received to be involved (for example, practical support such as instructions for using the online tools)?</b>	
Excellent	2
Somewhat good	2
Neither good nor bad	1
<b>How would you rate the information and learning materials you were given before the event</b>	
Excellent	3
Neither good nor bad	2
<b>Did you feel you meaningfully contributed to the discussion?</b>	
Yes	3
Unsure	3

## Investigator shared learning group

During the online facilitation of the two online discussions, a shared learning group was established for facilitators and the study teams of two similar projects being run in parallel. The study teams shared reflections and learning about the process of facilitation online, as well as offering and receiving support regarding technical and practical issues. The data was coded and categorised, including using the STARDIT framework<sup>1</sup>.

## Learning resources

A number of different learning resources were shared with participants at different stages of the process. This included a short 60-second online video about the study, giving information about the context and purpose<sup>22</sup>, a one page infographic summary of a scoping review about genomics research<sup>23</sup>.

Learning resources were both co-created and selected by the study team, working in partnership with the Australian Genomics Health Alliance and co-refining the selection with potential participants. In addition, in order to support the facilitators in providing good quality information, a number of were curated into a list to be available for facilitators to share during the online discussion, if they became relevant to aspects of the discussion in order to help inform people. The next section summarises the resources used, with references using Internet Archive links to future-proof the content of the learning resources as well as the URL.

## Summary of Learning Resources

Stage	Title	Media	Summary	Authorship
<b>Stage 2: Before consent</b>	What is genomic testing?	Portable Document Format (PDF)	Simple infographic explaining the basics of genomics research in plain English	Australian Genomics Health Alliance <sup>24</sup>
	What is genomics?	Online video animation with audio narration and subtitles	A 6-minute video outlining the principles of genomics research	Genome BC <sup>25</sup>
<b>Stage 3: Before online discussion</b>	Definitions and explanations	PDF	These definitions and explanations were used as a glossary to explain the main concepts of this research project.	Study team (see 'Learning resource example 1' below)
	Genomics and involvement	Online video with hard-coded text	A 2-minute video exploring why people should be involved in genomics	Jack Nunn <sup>22</sup>
	Infographic summary of scoping review	PDF (infographic – images and text)	A one page summary of the main findings from a recent scoping review about involving people in genomics <sup>26</sup>	Jack Nunn et al <sup>23</sup>
	Guide to using Loomio	PDF (text with hyperlinks)	A co-created learning resource giving practical advice for using the online discussion platform Loomio	Study team
<b>Additional resources available to facilitator</b>	Inheriting genetic conditions (chapter)	Webpage	An additional resource if participants wanted more information about inherited conditions	U.S National Library of Medicine <sup>27</sup>
	Data in the 100,000 Genomes Project	Online video animation with voice over and subtitles	An example of the bioinformatic pathway (specific to Genomics England) but generalisable (talks about access review committee)	Genomics England <sup>28</sup>
	Ethical issues in human genetics and genomics	PDF (text with hyperlinks)	Additional resource for a relevant ethics discussion from a medical perspective	Centre for Genetics Education <sup>29</sup>
	Genes, DNA and cancer	Webpage (text with hyperlinks)	Good plain English information about genes in relation to cancer.	Cancer Research UK <sup>30</sup>
	How to Share Genetic Test Results With Family	Webpage (text with hyperlinks)	Good information about sharing genetic test results with family.	American Society of Clinical Oncology <sup>31</sup>
	How do you sequence a human genome?	Image file (infographic – images and text)	Infographic about the stages of genome sequencing	Genomics England <sup>32</sup>

## Learning resource example: Definitions and explanations

The following definitions and explanations were used to explain the following concepts throughout this research project. This document was formatted as a PDF with references.

### Genomics

The study of all the DNA in the genome together with the technologies that allow it to be sequenced, analysed and interpreted is collectively called genomics, or genomic medicine if applied to patients<sup>33</sup>. The study of genomics can include other types of “omics”\*, such as ‘proteomics’ and ‘metabolomics’ – which for simplicity will be referred to under the term ‘genomics’. When the term ‘genomics’ is used in this project, it refers exclusively to human genomics.

*\*Other types of “Omics” include transcriptomics (all the RNA molecules in cell or organism), proteomics (all the proteins in a cell or organism) and metabolomics (all the metabolites in a cell or organism).*

### Genomic research

Genomic research refers to any kind of activity which is intended to increase our current understanding of genomics. It is distinct from genomic medicine or other routine services which use existing knowledge, rather than add to it. However, this distinction is not always clear.

### Involvement in genomic research

Research should be conducted ethically and to benefit people. Involving people as equal partners in genomic research has been identified as the most crucial aspect, as the benefits include improved public trust<sup>8</sup>. The concept of ‘public involvement’ in research is defined as research that is carried out ‘with’ people rather than ‘on’ them<sup>2</sup>. This allows people to have ‘active’ rather than ‘passive’ roles in research, which can lead to better research outcomes. Involving people in research in this way is now promoted by many governments as a right and is predicted to increasingly be an obligation in biomedical research, with research funding initiatives already recognising evidence of involvement as a key criterion<sup>3,34,35</sup>. Involving people can give greater public influence over research directions and conduct<sup>36</sup>, ensuring research is both acceptable, accessible and meets people’s needs by reflecting and balancing the diversity of priorities<sup>35,37,38</sup>. Tasks that people can be involved in include identifying areas of benefit, helping design and plan studies, helping in raising funds and analysing results.

### This project

This project is part of Jack Nunn’s PhD, ‘Genomics research and involving people’ which is focused on exploring how we can better involve people in all stages of genomics research, including the best methods to do so.

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