

Co-Designing Genomics Research With A Large Group of Donor-Conceived Siblings

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Research article

Keywords: STARDIT, genomics, research

Posted Date: November 10th, 2020

DOI: <https://doi.org/10.21203/rs.3.rs-100595/v1>

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Version of Record: A version of this preprint was published at Research Involvement and Engagement on December 1st, 2021. See the published version at <https://doi.org/10.1186/s40900-021-00325-7>.

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2 large group of donor-conceived siblings
3

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14 3,912 words

15

16 Plain English Summary

17 Human genomics research is growing rapidly. There is evidence that involving potential participants
18 and the public in co-designing research can improve the quality, recruitment and acceptability of the
19 research. However, more evidence about effective methods for involving people is required,
20 especially those in vulnerable sub-populations of inherently high interest to medical research. In this
21 study, we worked with a large group of donor-conceived siblings who share the same sperm donor.
22 We sought their views regarding participation in possible future research. We co-designed a way of
23 involving them in discussion about their own “terms of engagement” with research. Online
24 discussions gave group members an opportunity to share their views, and take initial steps towards
25 developing their own research governance model. We used the ‘Standardised Data on Initiatives
26 (STARDIT)’ Alpha Version to report involvement, allowing findings to be compared with other
27 studies. Group members who participated reported enjoying the experience, remarking that online
28 discussions had advantages over other methods, including more time to reflect on answers and learn
29 collectively. Most participants’ preferences about who should be involved in research design
30 ‘widened’. Participants’ learning from the process also informed subsequent discussions in the
31 sibling group about participation in research, including about how to make informed decisions about
32 participating. Involving people in this way has the potential for a ‘transformative learning’ impact,
33 where people’s perspectives and attitudes change as a result of being involved. This is particularly
34 important for people in vulnerable or under-represented sub-populations which are of high interest
35 to genetic research.

36 Abstract

37

38 **Background:** Human genomics research is growing rapidly. More effective methods are required for
39 co-design and involving people, especially those from vulnerable sub-populations of inherently high
40 interest to medical research. This case study documents how we worked with a large group of
41 donor-conceived siblings who share the same sperm donor father, to explore how they might want
42 to engage with and influence any future genomic research.

43 **Method:** A participatory action research process was used to explore the views of a group of 18
44 known donor-conceived siblings; who are part of a larger group of up to 1000 people who share the
45 same biological father. The discussion explored views about how the group would like to be involved
46 in future research. Five members participated in co-design; 12 completed a pre-discussion online
47 survey; and six participated in an online discussion forum and evaluation survey. The online
48 discussion was led by one facilitator, supported by the study team.

49 **Results:** Co-design informed the research process. Participants reported enjoying the overall
50 experience of the surveys and discussion forum, which were perceived as inclusive and flexible. Most
51 participants' views regarding the value of involvement in research changed during the process, and
52 'widened' about who should be involved. Participants were supportive of future research being done
53 with the siblings group. All who completed the final survey requested to remain part of the co-
54 design process. Other themes in the online discussion included concerns about conflicting interests
55 and a desire for research participation to improve the situation for people affected by assisted
56 conception. The process informed later discussions in the sibling group about participating in a self-
57 managed biobank and informed decision making about participating in genomics research.

58 **Conclusion:** Findings from this study help inform how people from certain sub-populations should be
59 involved in planning and defining their participation in genomic research; particularly those from
60 more vulnerable or high-interest populations. This process provides a replicable and practical
61 method of involving potential participants in co-designing genomics research using online
62 discussions, with reported positive outcomes. Reporting this study using 'Standardised data on
63 initiatives (STARDIT)' to report the process allows comparison with other studies.

64 Introduction

65 Human genomics research involves defining sub-populations, measuring DNA changes within them
66 and linking them to traits or outcomes in order to understand how DNA variation can contribute to
67 human health and disease. The more genetically similar people are, the more likely it is that they will
68 share the same DNA variations that contribute to a given trait such as wellness or disease.

69 Therefore, historically human genomic research has focused on restricted populations who share
70 common biological ancestry, including large families or founder populations, where less genetic
71 variation has led to more clear links between DNA and disease¹.

72 Since the advent of affordable online ancestry DNA testing, increasing numbers of people are taking
73 DNA tests to understand genetic ancestry (genealogy). However this DNA testing can sometimes
74 result in returning unexpected information, including the revelation of being sperm donor-
75 conceived². These genetic results can also immediately link people to a broader group of biologically
76 related people who also share the same sperm donor². An increasing number of large groups of
77 donor-conceived siblings are now being discovered, some with over 100 half-siblings^{3,4}. In some
78 cases, people from such sibling groups were conceived at a time before regulation or clear legal
79 oversight of assisted conception services, with some countries still having no clear legal regulation or
80 oversight⁵.

81 Large groups of individuals who share a common sperm donor may provide new insights into
82 medical, genetic, sociological and psychological studies, and therefore are of inherently high interest
83 to research. However, they also represent a potentially vulnerable group. There are prior examples
84 of other groups with shared biological ancestry having research conducting “on them” rather than in
85 partnership “with them”⁶⁻⁸. People within these groups are of inherently high interest to medical
86 research are therefore often more vulnerable⁹⁻¹¹, with the best ways of involving them in shaping
87 future research yet to be established⁶.

88 Here, we involved members of a donor-conceived sibling group in co-designing research. This case
89 study documents a participatory action research process with members of one of the largest
90 biologically-related (donor-conceived) family groups ever documented (hereafter ‘the sibling
91 group’)^{3,12-15}. We report the outcomes using the ‘Standardised Data on Initiatives (STARDIT)’ Alpha
92 Version for planning and reporting involvement¹⁶. Members of the sibling group are all offspring of
93 one sperm donor, the scientist Bertold Wiesner, a consulting biologist at the Royal Northern Hospital
94 in the 1940s¹⁷. He may have fathered up to 1000 offspring between 1942 (or earlier) and 1967¹³⁻¹⁵
95 through the medical practice of his wife Dr Mary Barton, despite a 1945 British Medical Journal
96 paper where Barton and Wiesner stated they set an ‘arbitrary limit of 100 children for each donor’¹⁸.
97 More information about this sibling group and contextual information can be found in Additional File

98 1.

99 Methods

100 Study Design

101 A participatory action research (PAR) method was chosen for the study with co-design and reporting
102 guided by a number of frameworks¹⁹⁻²¹. PAR is an umbrella term which describes a number of
103 related approaches, including forms of action research which embrace a participatory philosophy
104 and include 'co-design' and 'co-production' of research²². It is a process where researchers, relevant
105 stakeholders and sometimes the public "work together, sharing power and responsibility from the
106 start to the end of the project"²³, including knowledge generation and translation²³. The term
107 'stakeholder' means anyone who has a 'stake' in the research, in particular those with important
108 knowledge, experiences, expertise or views that should be taken into account^{24,25}.

109

110 After one of the investigators (JN) discovered his biological grandfather was Bertold Wiesner, the
111 study team invited a researcher who had previously worked with members of the sibling group (MC)
112 to join the study team. The biological relationship of the investigator JN was then used as a starting
113 point by the study team who then worked closely with both potential participants and experts in
114 human research ethics from the La Trobe University to ensure the method was acceptable and no
115 one (including the study team) would be exposed to avoidable risk. Guidance on ethical involvement
116 of potential participants in research design remains unclear¹⁹, in particular when it involves
117 individuals conducting research with biological relatives. Accordingly it was decided by the study
118 team to commence co-design only after ethics approval was obtained.

119

120 In order to co-design the study from an early stage²⁶, volunteers from the sibling group were
121 recruited and undertook a number of tasks including reviewing the proposed research design and
122 giving feedback on participant information, online survey questions, and the online discussion
123 facilitation plan. Involving the public, patients, research participants and other stakeholders in
124 actively contributing to the research process has been shown elsewhere to improve the recruitment
125 ^{27,28}, quality and relevance of research^{6,29} and lead to other positive outcomes³⁰. As some elements
126 of this study were co-designed in parallel with another similar study, some aspects were inflexible
127 and thus 'co-refined' rather than 'co-designed', for example, the choice of the online discussion
128 software.

129

130 The study had four stages: 1. Co-design; 2. Recruitment and pre survey completion; 3. Online
131 discussion and post survey and 4. Analysis, with stakeholders involved in tasks at each stage (Figure
132 1).

133 **Figure 1: Stages of research**

134 [insert file 'Figure 1 – Stages of research']

135 The data categories for the two surveys were informed by other similar studies to allow
136 comparison³¹. Questions relating to 'Who should be involved in research' were included in both
137 surveys to allow comparison with other standardised data¹⁶. Participants could choose from the
138 categories outlined in Figure 2, with a change in direction towards more people being involved
139 labelled as 'widening', the inverse as 'narrowing'.

140 **Figure 2: Who should be involved in research?**

141 [Insert file 'Figure 2 - Who should be involved in research']

142 Online methods of discussion are appropriate where a group of people is geographically dispersed
143 and face to face discussion is impractical³². We selected an asynchronous online discussion format
144 for this study as it allowed flexibility, especially regarding participants contributing from different
145 time-zones. An open source software web application called 'Loomio'³³ was installed on virtual
146 machines hosted by an Australian Government initiative called 'Nectar Cloud'³⁴. Participants could
147 securely log-in from anywhere in world and participants' data was stored securely on servers
148 physically located inside Australia.

149

150 The online discussions were facilitated for 14 days and led by one facilitator (MC). Participants were
151 invited to co-create their own boundaries for the group discussion at the start by reviewing provided
152 statements and suggesting amendments. The facilitator judged when to introduce new topics
153 (depending on the engagement with each topic) with the recommend schedule below in Table 1
154 used as a template. The facilitated discussion period also included the provision of learning materials
155 (short videos, infographics and information about research terms).

156 **Table 1: Discussion overview**

Question	Suggested staging point
Agreeing boundaries	Day one
What do you understand by the word 'research'?	Day one
What do you understand by the phrase 'genomic research'?	Day one
Why do we do research?	Day two

Which aspects of any future research genomic research should be influenced by the which groups of people? Day four

What methods do you think could be used to involve those people in future genomics research? Day six

Do you have any ideas, thoughts or reflections that have not been shared yet? Day seven

Discussion closed Day 14

157

158 The learning resources provided during data collection drew on the outcomes of an analysis of
159 educational materials by the Australian Genomics Health Alliance³⁵. Resources were selected by the
160 investigator team in partnership with the Australian Genomics Health Alliance and refined following
161 feedback from the volunteers. Detailed information about which learning resources were shared at
162 which stage of the study can be found in Additional File 1.

163

164 We collected and analysed both qualitative and quantitative data during the involvement activities,
165 informed by a number of frameworks and standards^{36,37}. Coding and thematic analysis of
166 qualitative data was carried out by two authors independently (JN, MC) and checked by another
167 author (PL), which is best practice for enhancing validity in qualitative methods³⁸. For the online
168 discussion, we also used case study methodology to record and describe our experience of involving
169 participants. This process is presented as an ‘instrumental’ case study, where the purpose is to
170 understand the particular case, and attempt to provide data that could produce useful
171 generalisations by using inferences from the data³⁹. In order to aid analysis and comparison with
172 other case studies, we used ‘Standardised Data on Initiatives’ (STARDIT) Alpha Version to
173 consistently map preferences for involvement and report ways people were involved in the PAR
174 process¹⁶.

175

176 As well as analysing data from the pre- and post- surveys and online discussions we also analysed
177 meeting notes, emails, surveys of the study team, comments shared by two of the team (JN, MC)
178 during the online discussion and reflexive research diary entries of one member of study team (JN)
179 (Table 2). Design of the study team survey was informed by frameworks for reporting and assessing
180 public involvement^{20,21}.

181 Table 2: Summary of data analysis

Data source description	Analysis method
-------------------------	-----------------

Meetings – including meeting notes and relevant documents	Qualitative (thematic analysis)
Online survey – text data from open and closed questions	Qualitative (thematic analysis, STARDIT-PM) Quantitative (number of responses)
Online discussion – text data from a facilitated and moderated online discussion of both participants and a separate one for the study team	Qualitative (thematic analysis, STARDIT-PM) Quantitative (number of responses)
Study team surveys – responses by email	Qualitative (thematic analysis, STARDIT-PM)
Other data – reflexive research diaries,	Qualitative (thematic analysis)

182 **Participants and recruitment**

183 The selection of the particular shared ancestry population for this case study was informed by a
 184 number of factors which were appraised by the study team, including pragmatic considerations
 185 coupled with extensive consultation with professional ethics advisors and other experts⁴⁰.

186

187 In October 2018 one member of the sibling group forwarded an email invitation to the other 17
 188 known members to join the study. The invitation to participate contained a link to the participant
 189 information document, a plain English summary about the study, and some learning resources about
 190 genomics research and an informed consent form. We offered the choice of anonymity (through
 191 using pseudonyms or temporary email accounts) in survey completion and the online discussions to
 192 allow people to participate without disclosing sensitive or personal information.

193

194 Those who gave consent were invited to complete the online pre-discussion survey and were then
 195 contacted by email directly by a member of the study team (MC), who shared information about
 196 joining the online discussion alongside relevant learning resources. This included a short 60 second
 197 online video about the study, giving information about the context and purpose, and a one-page
 198 infographic summary of a scoping review about involvement in genomics research.

199

200 **Results**

201 **Figure 3: Summary of the research process**

202 [Insert file 'Figure 3 - Summary of the research process']

203 Stage 1: Co-design

204 Five members of the sibling group gave feedback during the co-design process (Stage 1), three of
205 whom went on to participate in the study. Their input during the planning and co-design stage had
206 clear positive impacts, particularly in improving educational resources and ensuring the pre-
207 discussion survey used appropriate and acceptable wording to describe the sibling group, the
208 members and the sperm-donor. During the co-design process it was agreed that the initial study
209 should only involve the siblings, excluding any of their offspring. Owing to the ethically novel
210 situation of one of the investigators (JN), it was agreed during the co-design process that direct
211 communication with the sibling group would be conducted by a biologically unrelated member of
212 the study team (MC) once the study commenced. Figure 4 summarises how many people
213 participated in the stages of the study. Of the 18 known living siblings in 2018, 14 participated in the
214 co-design stages or the study.

215 Figure 4: Sibling group involvement and participation

216 [Insert file 'Figure 4 - Sibling group involvement and participation']

217 Stage 2: Recruitment and surveys

218 Of the 18 members of the donor sibling group, 12 gave consent to participate and completed the
219 pre-discussion survey of whom six participated in the online discussion. All six completed the follow-
220 up survey in May 2019 and shared consistent identifying data at all stages, including the initial
221 survey, the online discussion and the follow-up survey, allowing a comparison of results throughout
222 the process.

223 Of the 12, seven were female and five were male. All stated that they were comfortable describing
224 other descendants of their biological father as 'half-siblings'. Self-reported educational attainment
225 was mostly 'degree (bachelors), diploma or post-graduate', with one participant having professional
226 experience in medicine and genomics. All were aged between 50-74, with most living in the UK.
227 Most agreed with the statement that they have a shared interest in discussing future research which
228 might affect them, including genomic research. More information about participant demographics
229 can be found in Additional File 1.

230 From the six participants who completed both the pre-discussion and follow-up surveys, a total of 54
231 responses were given. Of these, 35% showed a change towards 'widening' involvement (N=19/54),
232 8% 'narrowed' (N=8/54) and half stayed the same (N=27/54). More detailed data is provided in
233 Additional File 1.

234 **Stage 3: Online discussions**

235 Six people participated in the online discussion in October 2019. Only one chose to use a pseudonym
236 in the online discussion, so the study team could identify them. Some of those who gave consent to
237 participate did not make comments but logged-in multiple times and read comments. Table 3 shows
238 how many comments were made by each participant in the online discussion. The themes where
239 most participants shared views are summarised in Table 4. The most discussed themes included the
240 unique nature of the sibling group, that anyone should be involved in research, questioning who
241 decides ethical oversight in research, questioning research for profit and sharing concerns about
242 genomics research being used for political purposes.

243 Table 3: Number of comments in online discussion

Participant ID	Number of comments
P2	12
P4	14
P5	42
P6	15
P7	9
P9	13
Facilitator (MC)	65

244

245

246 Table 4: Quantitative summary of themes

Theme	Number of participants
Anyone should be involved in research	6
Research with sibling group is unique and complex but important	6
Those affected by research should be involved	6
Research for profit and 'bullying' by 'big pharma'	6
Who decides who decides what is ethical?	5
Concerns about genomics research being used for political purposes	5
Finding out they are part of sibling group has been a positive experience	4
View on topics for research	4
Participants reported changed views and perspectives as a result of participating	4
Desire to improve situation for people affected by assisted conception	3
Interested in learning what other siblings think and discuss issues	3
Concerns about control of knowledge and data	3
Questioning which groups should have 'equal influence'?	3
Questioning eugenic attitudes to genomic variations	3
Views on participation in genomics research	3
Participants learned about genomics	3
Motivation for participation to help researchers and sibling group	2
Uncertainty about what they can offer but happy to help	2
What is the purpose of research?	2
Experts should be involved (over seen by ethics boards)	2
Developments in genomics have significant implications for society	2

247

248 Stage 4: Analysis

249 Participant feedback in the post-discussion survey was positive, including that the experience was
250 'interesting', and that they 'enjoyed thinking about the questions posed and reading the responses
251 of others' and the 'perceptive comments' of the facilitator [P7] [P4]. Two reported experiencing
252 some usability issues with the online platform. All who completed the survey opted to stay involved
253 in the next stages of the PAR process.

254 Table 5: Summary of pre and post survey responses

255 [Insert: 'Table 5: Summary of pre and post survey responses']

256 Outcomes from the process

257 There were 8 outcomes reported from this process, which are summarised in Table 7, with
258 additional information available in the accompanying STARDIT report (Additional File 2).

259 Table 6: Summary of outcomes from the process

260 [Insert: 'Table 6: Summary of outcomes from the process']

261 Participant views about genomic research and involvement

262 Participants demonstrated improved understanding of the difference between participation in
263 research and involvement in research by the end of the discussion, although initially some were
264 confused by the distinction. One stated 'I am a strong supporter of patient involvement in medical
265 care' and that 'involving members of the public' in genomic research was important in order to 'have
266 their views, reactions, interpretations, questions, concerns sought, interacted with, and considered'
267 [P11].

268 All six participants of the online discussion thought that anyone should be involved in research, with
269 one saying 'everyone should have a voice not just scientists and researchers' [P5]. Another added
270 that it is a 'good idea to involve research subjects in formulating the research questions' [P10].

271 Methods of involving people were discussed in detail with a number of options explored.

272 Participants explored ideas around the purpose of research and one stated that research
273 participants should be involved in 'agreeing purpose, parameters and methods' [P7]. All expressed
274 concern about research for profit and those with financial interests influencing research without the
275 public being involved.

276 One participant noted that being 'highly educated' was an enabler for involvement as was having a
277 'bit of time on their hands' [P4]. Another noted they didn't feel 'qualified' to 'comment on aspects of
278 science itself' but felt 'strongly' that they should be involved in ethical decisions and sharing personal
279 experiences to help inform research [P5].

280 One participant asked 'there will be many interested groups so which ones will be listened to?' [P4].
281 Another noted they felt that certain pharmaceutical companies were responsible for 'bullying',
282 contributing to 'disinformation; ignorance and inflexibility of medical and scientific professions'
283 [P12].

284 **Participant views about proposed research with sibling group**

285 Participants were supportive of future genomics research with the sibling group. One participant
286 stated it would be 'worth the effort', and another stated they 'wholeheartedly support the
287 involvement of the next generation' in any future research with the sibling group and noted any
288 study design should ensure new siblings and their offspring should be involved to ensure they can
289 'become part of the research' [P9] [P5].

290 In reference to future research with the sibling group one participant stated that ideally 'we would
291 be able to exert control over the use' of data [P7]. Conversely, one participant stated in the follow-
292 up survey that they 'could not at all care whether my genomics are public or not. I do not see that
293 my genome is a matter for privacy concerns' and recognised that others may feel differently [P9].

294 Participants spontaneously shared their views about what possible areas of research topics could be
295 explored in studies they could participate in and how these could be conducted. These included
296 'mental health' [P6] and pharmacogenomics [P4], with non-health related topics including 'career
297 choices' and hobbies [P4]. One participant felt they should be involved in 'seeking answers to old, or
298 not yet thought of questions' and 'looking beyond the known into a murky unknown' [P6].

299 **Stakeholder views about the process**

300 Participants reported their motivation for participating in this research process was altruistic, to help
301 researchers and the wider public. Some also recognised that their participation might directly
302 benefit members of the sibling group itself. One participant also stated 'it is commendable' that
303 'participatory research' was being used, in reference to the research methods used by the study
304 team [P9]. While online discussions were perceived as having advantages and 'worked well'[P7], two
305 participants reported usability issues with the online platform.

306

307 Four participants identified specific things about the way this study was conducted that enabled
308 their involvement. One said the entire process was 'assiduous' and that the 'intent of this project'
309 was 'obviously thoughtful and interesting' [P9]. One participant said the 'system seemed to work
310 well' [P7]. Another added that being used to online platforms like Loomio, or having previous
311 experience of similar platforms and 'used to' that way of communicating might facilitate
312 involvement using such a communication mode [P5].

313

314 Four participants reported specific things about the way this study was conducted that were barriers
315 to their involvement. The pace of the discussions was mentioned as moving 'too quickly' with
316 another adding 'more time' was needed and that the study team should 'reconsider the pace of the
317 research' [P7] [P4] [P5]. A discussion about creating boundaries in the discussion revealed that some
318 participants felt that they should avoid 'topics which might trigger emotions which are stressful'
319 whereas others thought this could be viewed as 'restrictive, even censorious' [P7].

320 The study team noted a 'critical mass' effect in online discussions, with the pace of comments
321 seemingly affected by the number of posts. One study team member noted the difficulty in
322 achieving 'the balance of being prescriptive' (for consistency) and giving freedom to facilitators to
323 initiate discussions and follow emergent themes. Support for the facilitator by the study team was
324 identified as an important enabler of the research process by the facilitator. Further barriers and
325 enablers are summarised in the accompanying STARDIT report (Additional File 2).

326 Discussion

327 In this study we used a participatory action research process to explore the views of a group of 18
328 known donor-conceived siblings, with participants reporting enjoying the overall experience of the
329 surveys and discussion forum. Online discussion forums were reported as an inclusive way of
330 involving people which was more flexible for communities which exist across time zones.
331 Participants reported it also gave them more time to reflect on answers, learn collectively as a group
332 and provided the freedom to ask questions and share ideas throughout the process.

333

334 Co-designing the study ensured it was more likely to meet the needs of potential participants.
335 Involving participants in co-designing the research process resulted in a number of improvements to
336 the study design, including improving language used in recruitment and learning resources and co-
337 creating acceptable online discussion boundaries. The process improved participants' understanding
338 about genomics and research.

339

340 Participation in the process led to eight outcomes, including participants 'widening' their views
341 about who should be involved in research to include more people. Participants reported changed
342 views about the value of involvement and an improved understanding about how to be involved in
343 genomics research. Some participants reported via the co-authored STARDIT report that learning
344 from this process informed subsequent discussions in the sibling group about participation in

345 research, including a proposed self-managed biobank and helping them make informed decisions
346 about participating in genomics research.

347

348 During the online discussion, the facilitator made significantly more comments than any one
349 participant, reflecting the level of work and engagement required to facilitate a discussion. The
350 'critical mass' effect of a certain number of posts in a discussion encouraging others to post aligns
351 with findings from other studies which have explored participants' hesitancy in posting in online
352 discussion forums⁴¹. Only one participant used a pseudonym during online discussions, and this may
353 reflect that the other group members felt comfortable sharing views with others in the group and
354 trusted the security and privacy of the platform, however it is unclear why some participants logged
355 in but did not comment.

356

357 While the focus of the online discussion was involvement in genomics research, the discussion
358 entered additional areas which reflected the interconnected nature of the subject, including the
359 ethical, legal and social implications of all genomics research. The discussions thus served as an
360 exploratory focus group, mapping participants' views in these diverse areas.

361

362 The study team was faced with ambiguous policies for the ethical involvement of people in co-
363 designing participatory research, which can hamper the degree of control that potential participants
364 have in research. Members of the study team reported they felt that limitations in the ethics process
365 affected the extent to which the sibling group could be involved in the PAR process. This is reported
366 in more detail in the accompanying STARDIT report (Additional File 2). Additionally, an unplanned
367 delay of 9 months in collecting follow-up data (related to ethics processes) may have affected the
368 quality of data collected post-discussion, with one participant adding 'given the lapse in time, I
369 cannot answer' in response to a question about their experience of participating in the online
370 discussion.

371

372 Further clarity from ethics committees and relevant organisations about the ethics of PAR will
373 enhance power sharing at this crucial stage of research, with standardised reporting of data allowing
374 direct comparison of ethical methods of involvement. Transparent ways of sharing power and
375 control in genomics research is particularly important for vulnerable populations and other sub-
376 populations where people share recent ancestry such as some Indigenous populations⁹⁻¹¹, and
377 populations of people affected by rare diseases caused by similar genomic variations⁴².

378 **Strengths**

379 Measuring outcomes such as ‘transformative learning’ can be challenging. This process overcame
380 such challenges by collecting standardised baseline and follow-up data about views, which meant we
381 were able to show that involving people in online discussions about involvement in research appears
382 to change people’s views about who should be involved in research, including participants
383 ‘widening’ their views to include more people. Similarly, using STARDIT for standardised preference
384 mapping and reporting of involvement meant outcomes from the process could be mapped more
385 effectively^{16,43}, including outcomes beyond the date of the initial data collection.

386

387 The importance of using STARDIT-PM to map the potential participant’s preferences when co-
388 designing involvement was demonstrated by participants stating their preference for online
389 discussion methods over face to face discussion or interviews. Such preference mapping helped
390 ensure the research design met the needs of potential participants and the STARDIT report meant
391 that as an additional 24 people have discovered they were part of the sibling group after this study
392 had ended, their views and those of future members could potentially be integrated using future
393 STARDIT reports.

394 **Limitations**

395 While 12 siblings gave consent to join the study, only 6 participants made comments in the online
396 discussion. Some of those who did not make comments logged-in multiple times and read
397 comments. While the follow-up survey attempted to capture the views of all 12, this was
398 unsuccessful so it is hard to understand these patterns. The involvement of relatively small numbers
399 in this study mean that although there is useful learning from our findings, their significance can
400 only be enhanced by standardised data sharing that can then combine with results with other
401 studies¹⁶.

402

403 As the study was designed in parallel with other studies, some aspects of the study were inflexible
404 during the co-design process (such as the specific open-source discussion platform that was used).
405 Some aspects were therefore ‘co-refined’ rather than ‘co-designed’, thus limiting some areas which
406 could be influenced by the co-design process.

407 **Conclusions**

408 The process described here provides a replicable and practical method of involving potential
409 participants in co-designing genomics research using online discussions, with reported positive
410 outcomes. Co-designing the study ensured it was more likely to meet the needs of potential

411 participants and resulted in improvements to the study design. Reporting this study using
412 'Standardised Data on Initiatives (STARDIT)' to report preferences, plan and report involvement and
413 evaluate participatory research methods and outcomes allows comparison with other studies. Such
414 reporting facilitates learning from this case study and contributes to data to inform evidence-based
415 decision making when planning future research. Learning from this study contributes to the current
416 evidence-base used to inform future ways of involving people in genomics research. Such evidence
417 can be applied in the context of research such as self-managed biobanks for vulnerable populations
418 and other sub-populations where there is a perceived power-imbalance between researchers and
419 potential participants.

420

422 Table 5: Summary of pre and post survey responses

Question	Results
What made you decide to respond to our invitation to participate in this project? 12 participants responded (pre-discussion)	Participants wanted an ‘opportunity to be involved in research’ [P3], wanted to ‘learn more’ [P7] and regarded involvement as a ‘civic duty’ [P10]. Others stated this study may be useful to ‘future donor offspring’ [P11] and one participant stated ‘curiosity’ was a reason for participation. Four participants noted a familial connection to a member of the study team (JN).
What do you hope to get out of participating in this discussion? Do you have any specific expectations? 8 participants responded (pre-discussion)	Participants were ‘interested in hearing what their half siblings think’ [P4] and wanted an ‘opportunity to discuss’ and ‘think through the issues involved’ [P4] [P6]. One participant said they wanted ‘the satisfaction of knowing that I may have contributed’ to the study [P10]. Another wanted ‘to be useful to the researchers’ [P11]. One participant noted an expectation of anonymity while participating.
Do you have any ideas about how the different people could influence future research? 7 participants responded pre-discussion and 3 post-discussion	Five participants said ‘everyone should be involved in research’, with one adding ‘not just scientists and researchers’ [P5-pre]. One participant said ‘anyone with an opinion’ should be involved [P3-pre]. Another stated ‘researchers and those who are affected by what is being researched’ [P4-pre]. One participant stated ‘People who know their subject but do not have hidden motives or agendas’ should be involved [P2-pre]. One participant said the answer depended on ‘what kind of research it is’ [P7-pre] and one mentioned ‘ongoing discussions’ using online tools [P5-post]
Is there anything in particular you liked or thought was helpful about how the discussion was conducted? 5 participants responded (post-discussion)	One participant stated they ‘liked and appreciated the opportunity to participate’ [P6]. One participant stated the process ‘seemed to work well’ [P7]. One participant added ‘I think it is commendable that there is a concern about participatory research’ [P9]
Is there anything you didn’t like, thought was unhelpful or could have been improved about how the discussion was conducted? 5 participants responded (post-discussion)	Two participants reported finding the platform ‘complicated’ and problematic [P5]. Two participants stated they would have liked ‘more time’ for the process [P4].

Did you have any expectations from participating in this research that were met or not met? 6 participants responded (post-discussion)	Four participants stated their expectations were met. One responded that they ‘found some of the questions very complex and had difficulty answering them’ [P5].
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423 Table 6: Summary of outcomes from the process

Outcome	Summary and learning point
1. Improved understanding of genomics informed participation in future research	Participants reported their understanding about genomics research increased as a result of participating in the study. Learning from this process informed subsequent discussions in the sibling group about participation in research, including a proposed self-managed biobank.
2: Learning resources useful	Participants reported finding the information resources and videos useful. Learning point: Creating learning resources in multiple formats (hyper-text, infographic summaries, videos with subtitles animations) will ensure that information is more useful for people with neuro-diverse learning needs.
3: Changed views and perspectives as a result of participation	Participants reported their views and perspectives changed as a result of participating. Learning point: Online discussion facilitated collaborative learning and the changed views of participants can be viewed as an impact of ‘transformative learning’ ^{32,44,45} .
4: Participants asked to stay involved in the research	All participants who completed the follow-up survey opted to stay involved in the research process. Learning point: Participants demonstrated ‘critical reflexivity’, a stage of PAR which asks people involved to reflect on their views and the causes of problems and to be involved in exploring any solutions and the actions that people can take to bring about improvements ^{43,44,46} .
5: Participants enjoyed the online discussions	Participants stated the experience of participating was ‘interesting’ and they ‘enjoyed’ it [P7] [P4], despite some usability barriers. Learning point: Some participants stated they preferred online discussions over face to face discussion or interviews, which highlights the importance of mapping potential participants’ preferences when co-

designing involvement, to ensure research methods meet the needs of participants.

6: Improved understanding of how to get involved in research

Participants reported improved understanding of how to get involved in research; this helped inform decision making for individuals when invitations were sent to members of the group to participate in genomics research after this study and unconnected to this study. **Learning point:** Pre and post discussion surveys and standardised reporting are effective tools for mapping changes in understanding and views^{16,43}.

7: Co-design changed study design

Feedback from participants resulted in changes to the study design including improving language used in recruitment and learning resources. **Learning point:** Involving participants in helping co-create learning resources can improve them.

8: Method for future research co-design established

Participants stated that the methods used in this process could be helpful when co-designing future stages of proposed genomic research with the sibling group. **Learning point:** STARDIT can be used to map preferences and impacts from future co-design processes¹⁶. Learning from this process is relevant to sub-populations where people share recent ancestry such as some Indigenous populations⁹, and sub-populations of people affected by rare diseases⁴².

424

425

426

427 **Additional files**

428 Additional File 1 - Data and Analysis

429 Additional File 2 - STARDIT report

430 Additional File 3 – GRIPP2 report

431

432 List of Figures

- 433 1. Figure 1 – Stages of research
- 434 2. Figure 2 - Who should be involved in research
- 435 3. Figure 3 – Summary of the research process
- 436 4. Figure 4 – Sibling group involvement and participation

437 Declarations

438 Ethics approval and consent to participate

439 The La Trobe University Human Ethics Committee approved this study.

440 Project number: HEC18256

441 Project Title: Genomics Research and Involving People: Exploring the views and perspectives of
442 people with shared ancestry about being involved in genomics research

443 Consent for publication

444 Not applicable

445 Availability of data and materials

446 All relevant data has been anonymised and shared in the additional files. La Trobe University is
447 storing all raw data according to the relevant ethics policies, and invites requests for more detailed
448 data. STARDIT reports will be added to databases in the future.

449 Competing interests

450 The authors declare that they have no competing interests.

451 Funding

452 Funding for the process described here was provided by La Trobe University.

453 Authors' contributions

454 JN wrote the manuscript and analysed the data, designed and carried out the process (including the
455 facilitation plan), evaluation of the process and the reporting (including STARDIT and GRIPP2
456 reports). MC gave feedback on the process design, facilitated online discussions, advised on
457 methodology, edited the manuscript, analysed data and checked data analysis edited the
458 manuscript. PL gave feedback on the process design, checked data analysis and edited the
459 manuscript. All authors helped co-design the process, read and approved the final manuscript. All
460 participants who opted to stay involved were invited to give feedback on this manuscript.

461 **Acknowledgements**

462 All authors contributed to the creation of this article. Thank you to Becky Gardiner, David Gollancz
463 and Michael Bywater for giving feedback during the co-design stage. The authors wish to thank
464 everyone who participated in the co-design process, gave their time to participate in the research
465 process, helped complete the STARDIT report and shared their skills and knowledge, including
466 Shirley Brailey, Barbara Nunn, Adrienne Smith and Barry Stevens.

467

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Figures



Figure 1

Stages of research



Figure 2

Who should be involved in research?



Working in partnership with a group of **half-siblings** who share the same donor father, from an **international group** of 40+

We explored using **online discussions** as a way of **involving people** and sharing power by shaping future **genomic research being done with them**



Involving participants in the process resulted in multiple impacts

The online discussion was reported as a positive and helpful experience by participants.



Involving people changed views about the value of involvement and improved understanding of involvement in research.

Online discussions were also perceived as a more inclusive and flexible way of involving people, for example those in different time-zones.



People's views about who should be involved in research 'widened' to include more people.



Standardised reporting of involvement in research

STARDIT Reporting and analysis of involvement was standardised

•Who was involved in which tasks?
•Who is funding it and why?
•What is the outcome or output?
•How will data be shared?

STARDIT is being develop by the **Wikimedia Foundation's** open-access journals. The ongoing co-design process is hosted at ScienceForAll.World/STARDIT



Figure 3
Summary of the research process

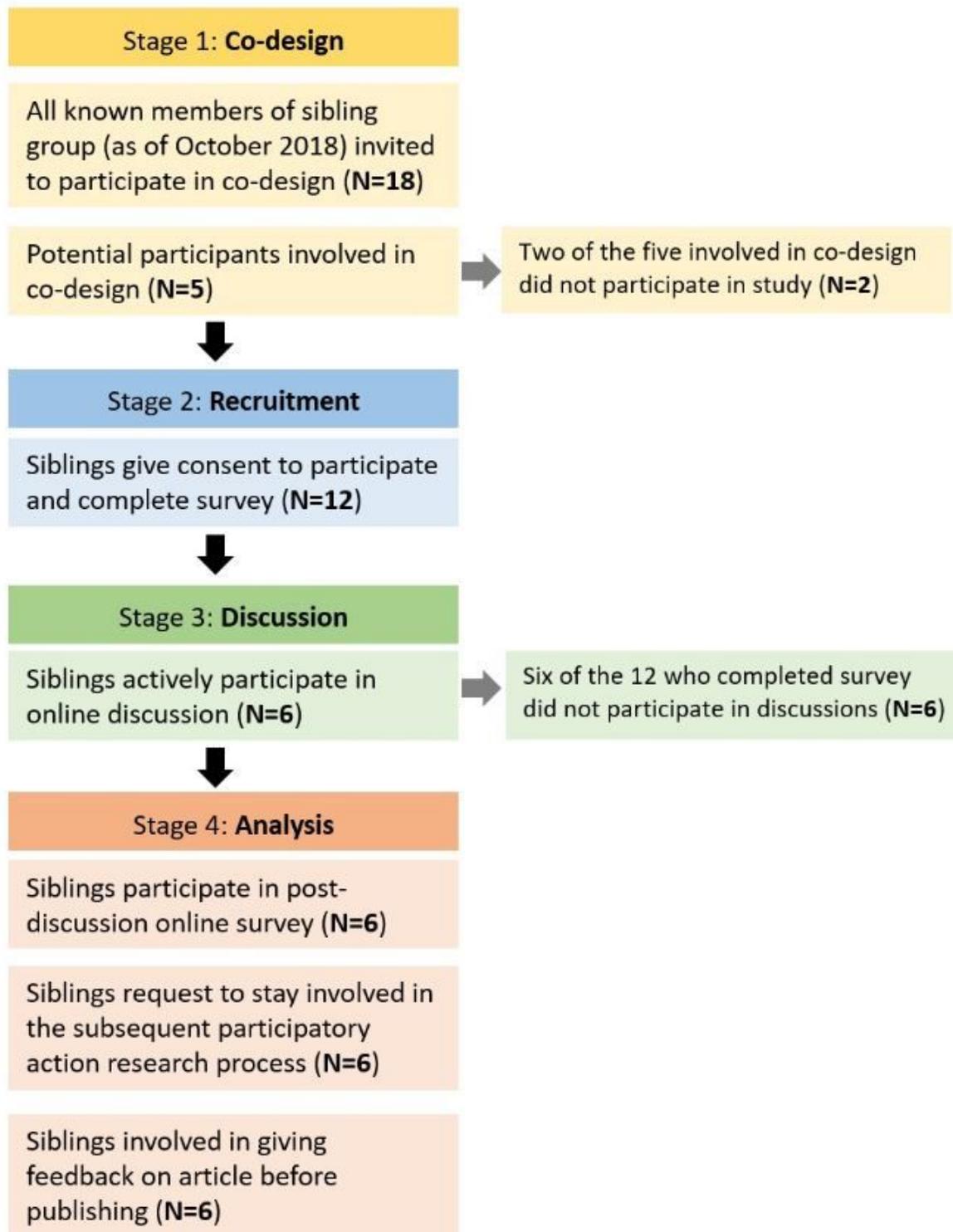


Figure 4

Sibling group involvement and participation

Supplementary Files

This is a list of supplementary files associated with this preprint. Click to download.

- [AdditionalFile1DataandanalysisSAV25.3.pdf](#)
- [AdditionalFile2STARDITreportSAV25.4.pdf](#)
- [AdditionalFile3GRIPP2reportSAV25.pdf](#)