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Involving elderly research participants in the co-design of a future multi-generational cohort study

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24 **Abstract**

25 **Objectives:**

26 To report as a case study the process of involving study participants and how this impacted the
27 co-design of a proposed multi-generational research study, using a novel standardised reporting
28 tool.

29 **Background:**

30 An advert was sent to 14,268 participants of the ASPREE-XT observational cohort study to
31 invite them to be involved in co-design and planning of a future multi-generational research
32 study. Participants were aged 74 years and older from Australia, located in both metropolitan and
33 rural locations.

34 **Interventions:**

35 We used participatory action research to involve elderly research participants in the co-design of
36 a proposed multi-generational cohort study between 2017-2019 using newsletters, telephone
37 interviews and an in-person workshop.

38 **Primary and secondary outcome measures:**

39 We used the ‘Standardised Data on Initiatives (STARDIT)’ Alpha Version to plan and report
40 how participant involvement activities positively impacted the study design.

41 **Results:**

Fifty-nine ASPREE participants were interviewed by telephone and 18 participants attended a face-to-face workshop event. Involving participants positively impacted the proposed study design by improving the research objectives, developing protocols, influencing funding decisions and improving ethics applications. Learning points included the importance of maintaining the ideals of the ASPREE study (respect, quality and transparency); research participants' preference for the option of receiving results (including genetic results); participants' need for involvement in decisions about recruitment, data access, governance and other ethical issues; and the preference for different communication methods, including both face-to-face and online methods. The process was highly valued by all stakeholders, including research participants, study staff and lead investigators. Involvement of participants was described by the lead study investigator as "enormously helpful".

Conclusions:

This case study provides evidence that including participants in the design of a research study positively impacted the study design, participants and researchers. Using a standardised reporting tool to describe the methods and impacts provides a way for learning from this case study to inform future research studies planning to involve people.

Plain English summary [required 250 words max]

Research which follows a population of people over time is a valuable way to learn about what contributes to both wellness and disease. Some studies of this kind look at multiple generations and are called multi-generational, longitudinal cohort studies. Such studies have had significant positive impacts on public health. However, such studies are challenging to establish and expensive to maintain.

65 It has been proposed that the existing ASPIrin in Reducing Events in the Elderly Extension study
66 (ASPREE-XT) would be a good basis for a future multigenerational research study (MGRS).
67 ASPREE-XT participants are all over the age of 74.

68 There is evidence that involving potential participants in co-designing research can improve the
69 quality of the study design, recruitment and acceptability of the research. A team of people
70 including current a ASPREE-XT participant were involved in co-designing how potential
71 participants would be involved in the co-design process. An advert was sent to 14,268
72 participants of the ASPREE-XT observational cohort study to invite them to be involved in the
73 process of planning a future multi-generational research study.

74

75 Fifty-nine ASPREE-XT participants were interviewed by telephone and 18 attended a face-to-
76 face workshop event. We used the ‘Standardised Data on Initiatives (STARDIT)’ Alpha Version
77 to plan and report how participant involvement activities positively impacted the study design.

78 Involving participants positively impacted the proposed study design by improving the research
79 objectives, developing protocols, influencing funding decisions and improving ethics
80 applications. This case study provides evidence that including participants in the design of a
81 research study positively impacted the study design, participants and researchers.

82

Introduction

The ASPIrin in Reducing Events in the Elderly (ASPREE) trial (2010-2018), a randomised controlled trial for aspirin in healthy older people, recruited 19,114 participants from Australia and the USA¹⁻⁴. The median age at recruitment was 74 years. The ASPREE Healthy Ageing Biobank is a sub-study which has collected biospecimens from over 15,000 ASPREE participants, alongside detailed medical records, lifestyle, cognitive function and physical testing data⁵. ASPREE researchers are conducting various types of genomics and biomarker research. ASPREE had a remarkably high retention rate, with only 1.2% of participants withdrawing from the study and 90% still attending annual visits after an average of 4.5 years follow-up^{6,7}.

ASPREE XT is a follow up observational study that was established in 2018 to continue to collect data from ASPREE participants for another 5 years. ASPREE-XT participants are over the age of 74 (defined as elderly⁸) and Australian participants are located in both metropolitan and rural locations. Multiple stakeholders, including a participant advisor, proposed a new multi-generational research study (MGRS) which could recruit direct descendants of ASPREE-XT participants, as part of a longitudinal observational study alongside their relatives for two generations or more. MGRS are challenging to establish and expensive to maintain, yet their value to medical and epidemiological research is significant. ASPREE-XT has been proposed as a platform for a multi-generational study. Recruiting ASPREE-XT participants and their descendants to a MGRS would provide a large starting population by international standards.

Previous multi-generational, longitudinal cohort studies have had significant positive impacts on public health. Examples include the Framingham Heart Study,⁹ Lothian Birth Cohort¹⁰ and Dubbo Osteoporosis Epidemiology Study,¹¹ all of which had substantially lower starting

populations than ASPREE-XT¹². ASPREE-XT provides a rare opportunity for such a study in Australia, already combining high-quality medical record data with genomic data on a large number of elderly Australians¹². Additionally, the cohort is already well engaged, with all surveyed participants supportive of a proposed MGRS.

The aim of this study was to report as a case study the processes of involving ASPREE-XT participants in the co-design of a proposed multi-generational research study, and how this impacted study design. The research objectives were to involve potential participants in the co-design of a new MGRS; plan the process in a standardised way; identify themes and preferences; and evaluate then report the process using a standardised reporting tool¹³.

ASPREE-XT's unique focus on healthy ageing adds substantial value to public health, medical, epidemiological and geriatric research. Previously collected genomic (including epigenetic) data, combined with detailed medical data and ongoing cognitive assessment, allow that a MGRS with the ASPREE-XT cohort would be of considerable value to science for decades to come, for example in dementia research¹⁴. The proposed study would examine health outcomes in a large, well-monitored cohort; and provide data to help inform our understanding about the genetic and environmental determinants of health and disease, across multiple generations. However, this is only possible if people choose to participate.

Many clinical research studies are underpowered due to poor recruitment and retention¹⁵.

Involving participants and the public in research design has been shown to improve the recruitment^{16,17}, quality and relevance of research^{18,19}. The concept of involvement is research being done 'with' people rather than 'on' them²⁰. Involving the public, patients, research

participants and other stakeholders in actively contributing to the research process can lead to a range of positive impacts and outcomes ²¹.

In human genomics research, the need to involve the public and other stakeholders is a crucial aspect of responsible research practice ^{19,22,23}. The term ‘stakeholder’ here means anyone who has a ‘stake’ in the research, in particular those with important knowledge, expertise or views that should be taken into account ^{22,24}. This includes ASPREE-XT participants, study staff and academic research investigators and the wider public. At the earliest stage of the research cycle (the conceptual stage), some current ASPREE-XT participants were invited to be involved in the co-design of a new MGRS. In this paper we aim to outline how people were involved in the co-design process in order to appraise the methodology and inform future best-practice.

Terminology

We have used consistent language in this paper to describe concepts such as ‘involvement’. To aid readers, Table 1 provides definitions of important terms used consistently throughout this paper.

Involvement – The words ‘**involvement**’ or ‘being **involved**’ describe the concept of people being ‘involved’ in research. This is when research is carried out ‘with’ people rather than ‘on’ them ²⁰. ‘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 ²¹. It is the ‘active involvement’ in shaping and guiding research, rather than only providing data ^{25–27}.

Participant – a person who participated in the process of sharing views and perspectives about the proposed MGRS, including sharing views about preferences for any future involvement. The term ‘ASPREE-XT participants’ will be used when specifically referring to participants from the existing study.

Participant advisor – before inviting people to become participants, it was necessary to involve a small number of participants to help advise and plan the process. These participants were chosen from an existing reference group of ASPREE-XT participants.

Stakeholder – 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 ^{22,24}. In this paper it refers to participants, participant advisors and ASPREE-XT study team members (including researchers, ASPREE-XT participant assessors and lead investigators).

Study team – this process was guided by the study team, who consisted of academic researchers, ASPREE-XT participant assessors, a participant advisor and a lead investigator.

The process – this term will be used to describe the process of involving ASPREE-XT participants by inviting them to share views and perspectives about a potential future MGRS. This process includes the co-creation of this case study with participant advisors.

Participatory action research (PAR) - this term describes a number of related approaches, including forms of action research which embrace a participatory philosophy and include ‘co-design’ and ‘co-production’ of research ^{28(p1)}.

148

149

Materials and Methods

Study design

A participatory action research (PAR) method was chosen to guide the process with co-design and reporting guided by a number of frameworks^{29–33}. PAR is an umbrella term which describes a number of related approaches, including forms of action research which embrace a participatory philosophy and include ‘co-design’ and ‘co-production’ of research^{28(p1)}. During the study design, we applied this co-design process where researchers and other relevant stakeholders (including research participants) “work together, sharing power and responsibility from the start to the end of the project”,³⁴ including knowledge generation and translation^{34,35}. Figure 1 summarises the process we used (Fig 1). The ASPREE participant advisor was an integral member of the study team, through each stage.

Guiding Frameworks

The process was guided by a number of international PAR methodology frameworks^{36–38}, including the International Collaboration for Participatory Health Research³⁹. The PAR method was also informed by an international review of involvement in genomic research carried out by some members of the study team¹⁹. Learning from this review informed the subsequent development and application of an Alpha version of ‘Standardised Data on Initiatives’ (STARDIT)¹³. STARDIT includes a tool to map people’s preferences for involvement in a standardised way, including mapping views on who should be involved and how. STARDIT was then used to guide co-design of the process, and to subsequently report how people were involved, using standardised data. An Alpha version of the STARDIT framework was also used

in parallel with the thematic analysis to organise data into pre-defined ‘super-categories’ which allow consistent comparison with other data using this reporting framework¹³.

We used a case study research methodology to record and describe the process of involving participants in the co-design of a proposed MGRS. Case study research methodology is a form of empirical inquiry and this study is presented as an instrumental case study, where the purpose is to understand the particular case and to provide data that could produce useful generalisations by using inferences from the data ^{40(p109)}.

The selection of the population for this case study was informed by a number of factors which were appraised by the study team, including ethical, pragmatic and population considerations^{41,42}. One of the study team members (PL) is an investigator with the ASPREE-XT study, which was used as a starting point by the study team to explore the appropriateness of this case study.

Data collection and reporting

The data collection and reporting was informed by a number of frameworks for reporting involvement in research^{13,22,30,31,38,43–45}. Qualitative and quantitative data was collected from multiple sources, including meeting notes, audio recordings, documents, emails and surveys. data sources were analysed using a number of methods including thematic analysis. Further information about how data sources were analysed is in ‘Table 2. Summary of data analysis’.

Stages of research

Fig 1. Process timeline of the participant involvement

[see file ‘Figure 1’ – ‘Process timeline of the participant involvement’]

Stage 1: Planning

The study team held four meetings to co-design the involvement activities. One participant advisor was involved in a number of tasks including reviewing and improving the written information, telephone interview questions, and the facilitation plan for the event.

Stage 2: Recruitment and telephone interviews

An advert was placed in a newsletter to 14,268 ASPREE participants. The process we used for recruiting and involving ASPREE-XT participants has been divided into four chronological stages, summarised above in Figure 1 (Fig 1).

After the recruitment and consent process, participants were interviewed by telephone. A summary of the questions and responses from the telephone interviews is shown in Table 3. For a list of all questions asked, see Additional file 2. Participants were asked about their willingness to provide feedback throughout the study, and to be involved in study design, as well as preferences for modes of communication. The definition of involvement below used in the script was co-designed with participants for subsequent interviewees, with versions 1 through 1.3 piloted with 15 participants.

Pre-question script: ‘Traditionally, research studies have been designed and conducted only by researchers, and people invited to participate. We are challenging this idea of researchers being the only experts.’

Question: Would you be willing to provide feedback and share your views and perspectives by commenting throughout the research process?’

216 Twenty relevant interviews were transcribed, coded and categorised (JN), with relevant
217 interviews identified by two investigators independently (JN, PL) ⁴⁶. To reduce any unconscious
218 selection bias, a sample of over 10% of the interviews was selected at random.

219 **Stage 3: Event**

220 Eighteen participants attended a four-hour workshop event in central Melbourne, led by JN, who
221 is an experienced facilitator. The event was co-designed by the study team, and was informed by
222 interview data and international best-practices for involvement events ^{37,47}.

223

224 The event included an introduction to the proposed MGRS by the lead ASPREE-XT genomics
225 researcher (PL); a plain-English introduction to genomics by an expert in genomics who is also
226 an ASPREE-XT participant (BH); a summary of the telephone interview results by the
227 interviewer (MS); and an interactive session which included open questions about the types of
228 information participants would like returned and recruitment of family members (JN).

229

230 The final session included a presentation and interactive discussion about involvement in
231 research, led by the event facilitator (JN). This session explored preferences about how people
232 would like to be involved, with open and closed questions. Questions included preferences about
233 tasks and modes of communication.

234

235 Throughout the event, participants shared their views on a range of issues through interactive
236 discussions, voting (by show of hands) and anonymous written feedback.

237

Stage 4: Evaluation and analysis

Table 2. Summary of data collection and analysis

Data source description	Analysis method
Meetings – including meeting notes, recordings and relevant documents	Qualitative (content analysis)
Telephone interviews – including recordings, notes made by interviewer, summary documents and related emails	Qualitative (thematic analysis, content analysis) Quantitative (number of responses)
Event – including audio and video recordings, interviews, written notes and feedback forms	Qualitative (content analysis) Quantitative (number of responses)
Study team surveys – responses to open ended questions by email	Qualitative (content analysis)
Other data – this included reflexive research diaries, relevant emails, financial and other relevant documents	Qualitative (thematic analysis) Quantitative (analysis of cost)

Evaluation and reflection

All members of the study team (except JN) were surveyed six months after the face-to-face event in order to integrate the valuable views and perspectives of those involved in co-designing and delivering the process. Design of surveys was informed by frameworks for planning and reporting public involvement^{30,31}. The study team were asked 11 questions (available in Additional file File 1) and the data from the four interviews was coded and categorised (JN) using STARDIT¹³.

Data analysis

The stages of qualitative data analysis included data mapping and familiarisation; transcription; coding; searching for themes; reviewing themes with study team members; labelling and summarising themes; and reporting the findings⁴⁶. In order to enhance validity of the analysis, two authors (JN, MS) independently analysed the data thematically, which was then ‘member checked’ by a third author (PL)^{46,48,49}. Standardised categories (STARDIT) were used during content analysis of the data in order to facilitate comparison with other research projects¹³. More information about the data sources and a STARDIT report available is in [Additional file 1](#).

Results

The results are presented in three sections. Section one provides results from the stages outlined in Figure 1 (Fig 1), section two shares the perspectives of ASPREE-XT participants and other stakeholders, and in section three we describe how participant involvement impacted study design in seven ways. We quote participants directly, assigning each a unique number. Figure 2 summarises the entire process, results and impacts (Fig 2). For quantitative data from the telephone interviews, see [Additional File 3](#).

Section 1: Results from stages 1-4

Results from Stage 1: Planning

The input of the participant advisor during the early co-design stage had clear benefits, in particular in identifying the best approaches for engaging the broader ASPREE population; improving the wording of participant information resources and improving question design for the interviews.

Results from Stage 2: Recruitment and telephone interviews

269 After reading the advert in the newsletter, 76 ASPREE-XT participants agreed to participate in
270 the process. We interviewed 59 people by telephone, with the remaining 17 not responding to a
271 number of follow-up calls.

272

273 Interview participants expressed a strong interest in receiving results from genetic research. The
274 strongest preference was for genetic results of medical significance (rather than ancestry or drug-
275 response).

276

277 All telephone interview participants expressed willingness to provide feedback throughout the
278 process of planning the proposed MGRS. While most participants understood the concept of
279 involvement, 9 of the 20 analysed expressed self-doubt about how they could be involved. Of 20
280 analysed telephone interviews, five participants seemed unclear about what tasks they could be
281 involved in, suggesting the concept of participant involvement was new to them. Six participants
282 sought clarity about what was expected of them when being involved, or discussed the skills and
283 knowledge required. Further, participants stated that the goals and processes of involvement
284 needed to be clear in order to avoid “spending too much time” and were willing to be involved as
285 long as the task had purpose and was not “just for the sake of chatting” (P1).

286

287 During the telephone interview stage, the interviewer initially “struggled to explain” the concept
288 of involvement (as distinct from participation in an active study) (MS). Co-designing and making
289 changes to the language used in the telephone interviews appeared to improve participant
290 understanding of the concept of involvement. Changes included providing a clear definition and
291 pre-question script and including actual examples of involvement (such as overseeing ethical
292 decisions about data access), thereby avoiding jargonistic abstract concepts such as
293 ‘involvement’.

294 **Table 3: Summary of data from telephone interviews**

Questions (closed)	Results	% of interview participants
1. Do you think participants like yourself should be involved in helping design research projects, or just researchers?	Just researchers	39
	Participants involved	46
	Unsure	11
2. Would you be willing to provide feedback and share your views and perspectives by commenting throughout the research process?	Yes	100
	No	0
3. Would you like to be more involved in study design rather than just participating?	Yes	65
	No	12
	Unsure	23
4. Would you be more or less likely to participate in research if participants were involved in design	More likely	25
	Less likely	0
	Unsure	13
	Wouldn't influence participation	13
5. What is your preferred mode of communication for being involved	Face to face event	15
	Post	31
	Online discussion	3
	Online questionnaire	15
6. If genetic research results were made available, which types of genetic testing would you be interested in? *	Medical	97
	Ancestry	97
	Drug response	94

7. If only one option for genetic testing was	Medical	65
available, which one would you prefer?	Ancestry	12
	Drug response	6
	No preference	18

295

296 *Note that percentages indicate the number of participants who responded with that response. On
 297 question six, participants could select multiple answers, so totals do not add up to 100%.

298 **Results from Stage 3: Event**

299 Eighteen of the invited 76 participants attended the face-to-face event. When specific tasks were
 300 discussed at the event, 10 participants expressed interest in being involved in recruitment and
 301 communication tasks, 7 in data access decisions, and 11 in ethical decisions. When surveyed
 302 after the event, the study team reported that involving participants positively impacted the
 303 proposed study design by improving the research objectives, developing protocols, influencing
 304 funding decisions and improving ethics applications. Videos of the event discussions and
 305 interviews with participants will be shared in the public domain.

306 **Results from Stage 4: Evaluation and reflection**

307 Participant feedback from the workshop event was analysed, with participants reporting it as a
 308 positive experience, one describing it as “brilliant” (P7). When surveyed, all members of the
 309 study team reported that the event achieved its intended aims and was a positive experience. No
 310 negative impacts were reported from any participants or study team members at any stage of the
 311 process.

312

One study team member described that through involving people, their perceptions “significantly changed” as they learned how participants could have “valid, interesting, and often novel ideas that researchers may not have considered”. The lead investigator (PL) noted:

“What I learnt is that sometimes the researchers’ assumptions about certain things can be off or even incorrect, and that researchers can miss critical points that are important to participants and the study”.

During the process, both participants and study team members reported changed views about the value of involvement in research, demonstrating ‘transformative learning’ and co-construction of knowledge^{39,50,51}

A number of significant learning points were identified by the study team when responding to the question “do you have any advice to other researchers planning involvement for their research”. Significant learning points are summarised in Table 4.

Table 4: Summary of learning points from study team

Summary of learning points from study team

1. Fund and prioritise involvement, make it a requirement
2. Ethics processes take time, but can improve plans
3. Know your audience – don’t make assumptions
4. Value diversity in experience and knowledge
5. A supportive team improves the experience for all

Cost and value

The entire process of involving people was estimated to cost \$10,000 AUD, including staff time, catering and event venue hire. The value of the process was summarised by the lead investigator who stated “I learnt a lot from the process and am very glad we made the effort”.

Section 2: Perspectives of ASPREE participants and other stakeholders

Recurring themes were identified by the study team from the data sources analysed, and seven specific impacts were reported as a result of involving participants (summarised below). The analysis is divided into two sections: (1) participant views about the proposed study, (2) stakeholder views about involvement in the proposed study.

1: Participant views about the proposed study

Participants were very positive about the proposed MGRS, with one stating ‘I think it sounds very good’[P20]. Altruism was a primary motivator for participation, with participants suggesting that outcomes of a MGRS could include benefits to themselves (personally and for their families); improving healthcare more generally; and the potential for saving lives, preventing diseases, improving quality of life, and improving future research.

Participant views on types of information they would like returned from genomic research and how the study should be funded (commercial funding versus public funding) were diverse and did not always align with the study team’s prior expectations. For example, two thirds of event participants wanted access to their own genomic data, which was described by the lead investigator as ‘very at odds with the current system’ (PL).

2: Stakeholder views about participant involvement in the proposed study

351 Participants were supportive about being involved, with all participants supportive of being
352 involved by providing feedback throughout the research process (100%, 32/32), with a typical
353 participant response being ‘I’d be happy to be involved’ [P3]. Views about enablers were shared
354 in three of the 20 interviews coded, by all 18 of the event participants and all study team
355 members surveyed. Views about barriers were shared in eight of the interviews coded and by
356 half of the study team surveys. Further data is categorised in Table 5. Additional mapping of
357 preferences for involvement using the STARDIT-PM tool can be found in Additional File 1 ¹³.

358 **Table 5: Enablers and barriers for involving participants**

359 [\[Insert 'Table 5: Enablers and barriers for involving participants'\]](#)

360

361 The lead investigator stated “the feedback has been so valuable” and that it will be “built into the
362 design” of future research. However, the study team also identified barriers to involving people
363 which exist for researchers. One study team member reported that at the start of the project they
364 were “unsure how people without a science or health background” could be involved. They
365 reported a personal shift during the involvement process from not understanding how
366 participants could be involved and being concerned about “asking too much” to believing that,
367 “with adequate resources (financial, training, time...), people can be involved in all aspects of
368 genomics research”.

369

370 Other barriers identified by the study team included delays in obtaining ethical approvals for
371 involving people and the cost of involvement in both time and money. One study team member
372 reflected in the follow-up survey that they had “worried too much” about the time-burden of
373 involving participants. The concern of “not putting further pressure” on participants was a theme

374 in email communication between the study team when making decisions to limit contact with
375 participants throughout the process of involving them.

376

377 Survey data from the study team also suggests that adequate funding, a supportive team,
378 involving participants in the very earliest stage of research planning and co-designing inclusive
379 methods of involvement all contributed to the impacts reported.

380 **Section 3: Impacts on study design from stakeholder involvement**

381 Involving stakeholders in the co-design process impacted the study in seven specific impacts
382 ways. By asking for participants' views on aspects of the proposed study design, the study team
383 gained insight into participant preferences and opinions. While there was diversity in views, the
384 process allowed the study team to improve aspects of the study design. These impacts are
385 summarised in Table 6 'Summary of impacts on study design'

386 **Table 6 ‘Summary of impacts on study design’**

Impact on planned research	Summary of impact
1: Recruitment and sample collection	Recruitment and consent for the MGRS will occur online wherever possible, and saliva samples (rather than blood) will sent by post to be used as biospecimens for DNA analysis.
2: Participant communication	A short video and ‘information pack’, which will explain the MGRS study, will be created to assist with recruiting family members.
3: Participant involvement in governance	Participants will be invited to be involved in overseeing governance, including funding decisions.
4: Data access	Study participants should be involved in controlling data access decisions and policies.
5: Communication and ways of involving participants	Participants will be included on study advisory groups, including for study recruitment and communication, data

access and ethical oversight using multiple communication modes.

6: Provide feedback to participants about the research

Participants will be informed about the impact of the research, and how their involvement has affected the design and management of the study.

7: Create learning and development opportunities

Learning and development opportunities will be created for potential participants, researchers and other stakeholders.

387

388

389 **Impact 1: Recruitment and sample collection approach**

390 A discussion about participants' adult children being 'time-poor' highlighted the importance of a
391 study design which minimised the time burden on younger generations. Only a third of
392 participants thought their children would be willing to do a blood test. This allowed the research
393 team to make a more informed decision about 'trade-offs' between the data that can be collected
394 via blood or saliva, versus the potential effect on recruitment.

395 **Impact 2: Participant communication**

396 Most participants reported willingness to be involved in recruiting family members to a new
397 study, if given appropriate information and supporting documents. Relatively inexpensive
398 information resources, such explanatory videos, could have a significant impact on the success of
399 the recruitment and the study as a whole. One participant also advised that information produced
400 for participants by researchers can be confusing, and that laypeople can help simplifying it.

401 **Impact 3: Participant involvement in governance**

402 Event participants unanimously agreed that they should be involved in all aspects of the research,
403 whereas 11 of participants from the 20 analysed interviews thought that they should be involved

404 in study design. One interview participant felt only researchers should be involved as they are
405 “the qualified people” (P8). Five of the interview participants expressed the view that non-
406 researchers are required in research as they provide an important alternative perspective.
407
408 Participants had mixed views about commercial organisations funding research. Four event
409 participants were against it, some were ambivalent, and the majority indicated no objections. One
410 participant was concerned about the risks posed by involvement of commercial organisations
411 with opaque “vested interests”, asking “what is in it for them?” (P2), referring to individuals and
412 organisations with real or perceived conflicting, competing or commercial interests. Participants
413 suggested that involving people in governance (including funding and ethical oversight) was a
414 way of mitigating this risk.
415
416 Study team members reflected that public funding would be preferable to commercial funding, as
417 the responses at the event suggested that a commercially-funded study might negatively impact
418 recruitment. As a result, the study team altered the proposed design to involve participants in
419 governance, oversight and funding decisions.

420 **Impact 4: Data access**

421 Participants shared the view that they would like different kinds of genetic information returned
422 from the research (see Table 3), including personal medical, ancestry and pharmacogenomic
423 results. Two-thirds of event participants wanted access to their own genomic data, and had mixed
424 views about who else should have access. All event participants stated they were comfortable
425 with their data being held by academics, and none were comfortable with data being held by a
426 commercial company. However, one participant suggested not “ruling private companies out
427 completely” from research (P9).

428

429 General practitioners (GPs) were generally trusted to access and interpret genomic data, but
430 participants felt GPs should not have access to data that they did not. All but two event
431 participants agreed they should exclusively control access to their own data, with those
432 disagreeing mentioning cognitive decline as a reason for a co-managed access model.

433

434 Some participants had concerns about themselves or their biological relatives (especially
435 offspring) finding out information they “might not want to know” (P6), or having it imposed on
436 them.

437 **Impact 5: Communication and ways of involving participants**

438 Preferences for communication mode differed between interview participants and event
439 participants. Interview participants preferred questionnaires via post (30%, 18/50), and face-to-
440 face events (25%, 15/59), over online questionnaires (17%, 10/59) and online discussions (8%,
441 5/59). Event participants suggested face-to-face meetings were helpful but only when there was
442 an “occasional need” (P3). Participant responses also suggested that limiting face-to-face events
443 where possible (in favour of telecommunication) may mean involvement is more inclusive.

444
445 Event participants felt certain tasks (such as reviewing information) could be done “more online”
446 (P3). Participants spontaneously suggested using online, moderated forums and suggested that
447 these should be hosted by trusted institutions (such as universities) rather than commercial
448 organisations, as some ‘don’t trust’ social media companies [P17].

449 **Impact 6: Provide feedback to participants about the research**

450 Event participants stated that keeping people informed about what has been learned from the
451 study is a good way of keeping people engaged in the study and improving retention. For
452 example, ASPREE-XT sends a regular newsletter to participants. Participants also stated that
453 they would like to be informed about when their involvement has made a difference.

454 **Impact 7: Create learning and development opportunities**

455 Participants often stated their willingness but also their uncertainty about how they could be
456 involved. Learning and development opportunities were identified as an e of involvement by
457 both a participant advisor.

458 **Fig 2. Co-designing multi-generational genomics research**

459 [See file – ‘Fig 2’ - Co-designing multi-generational genomics research]

460 **Discussion**

461 The participatory action research method gave insights into participants’ preferences that
462 measurably impacted on the proposed study design. The improvement of the interview design

463 using the co-design process illustrates the value of a flexible and iterative approach to
464 involvement in a study.

465
466 Participants' preference for being informed about both the study outcomes and the outcomes of
467 their involvement is supported by other studies which suggest that communicating about the
468 research regularly and sharing results may improve retention ⁵²⁻⁵⁴. This is particularly relevant
469 for those planning involvement in genomics research, which may span decades ¹⁹.

470
471 Such regular communication should also be combined with learning and development
472 opportunities, which could also help facilitate participant involvement by ensuring that people
473 understand the values which motivate the tasks they are being asked to be involved in ^{19,55}

474
475 Participant views about data storage and access aligns with a 2015 survey by Genetic Alliance
476 Australia, which indicated people mostly trusted universities and research institutes to use
477 personal genetic information for research, with commercial companies least trusted ⁵⁶. Even
478 participants' unanimous preference for having access to their own data and a general trust for
479 GPs to access and interpret genomic data also aligns with findings from other studies ⁵⁷⁻⁵⁹.

480

481 Participants' concerns about unintentional disclosure of data to biological relatives who 'might
482 not want to know' certain information highlights the challenges of asking participants about
483 information preferences, and recognises that this ethical decision extends beyond individuals
484 [P6]. This issue is common to almost all ongoing and proposed genomic research studies and
485 should be urgently addressed by contemporary individualist ethical frameworks ⁵⁸.

486 **Study strengths**

487 By asking participants their preferences, the study team gained useful insights to inform the
488 design of the proposed study. Participants preference for being involved in decision making
489 about funding sources, data management and ownership, and what information to share with
490 participants will help ensure any future study design aligns with participants' values, ensuring
491 the design is culturally safe and culturally competent ⁶⁰. Similarly, the participants preference for
492 being involved in reviewing participant information aligns with other research which suggests
493 that involving potential participants in reviewing information can help improve recruitment ⁶¹.

494

495 The effective involvement of ‘stakeholders’ also includes involving all relevant staff and health
496 professionals at all levels of an initiative, who may have important knowledge or perspectives
497 that senior research staff do not.
498
499 The ‘transformative learning’ during the process reported from both study participants and the
500 study team was an important impact captured by the participatory action research (PAR)
501 method^{39,50}. The process showed that it was valuable to create regular involvement opportunities
502 for each stakeholder. Reporting this process in a standardised way using ‘Standardised Data on
503 Initiatives’ (STARDIT) meant that impacts such as transformative learning could be reported and
504 that this case study can be compared to similar studies in the future [49].

505 **Study limitations**

506 Because the 76 participants who responded to the original advertisement were self-selected
507 volunteers, our findings may not necessarily be accurate for the whole ASPREE-XT cohort. Our
508 sample may reflect a sub-set of individuals who feel more strongly supportive of a proposed
509 MGRS than the cohort average. However, the data from this process is still useful and valid⁵⁷.

510

511 The number of interviews which were transcribed (20) and analysed was high for a case study ⁶².
512 As there is no agreement in case study literature on whether to code all or some of the data, our
513 methodology balanced exhaustive examination of the data with the capacity bias imposed by
514 time constraints ^{46,63}. Therefore, data transcribed and coded from the interviews does not include
515 all the data collected in the interviews.

516
517 While the mixture of written and verbal feedback at the event ensured a range of ways to give
518 feedback, the voting process (which involved people raising their hands in front of everyone)
519 may have given different results if it was an anonymous ballot⁵⁹. Future research should explore
520 and compare preferences for different methods of voting.

521
522 While the process of involving people described here did not exclude people based on
523 language,⁶⁴ the original ASPREE study required a certain level of English language skills in
524 order for people to participate in some of the cognitive assessments and public events. The
525 process described here may therefore have excluded people who cannot read English or do not
526 feel confident speaking in English, such as people who speak English as a second language.

527 **Conclusions**

528 This case study provides evidence that including participants in the design of a research study
529 positively impacted the study design. As many research studies are negatively impacted owing to
530 poor recruitment and retention, such evidence is increasingly important for informing
531 involvement in future studies. The process of involving ASPREE-XT participants in the design
532 of a new MGRS was highly valued by stakeholders, and was positively impactful for both
533 participants and the study team. The lead investigator stated “the feedback has been so valuable”
534 and that it will be “built into the design” of future research. Learning from the case study
535 suggests that adequate funding, a supportive team, involving participants in the very earliest
536 stage of research planning and co-designing inclusive methods of involvement all contributed to
537 the impacts reported.

538
539 The process of involving people can be viewed as a learning experience for both the participants
540 involved and study team members. The process changed participant and study team members’
541 views about the value of involvement. For example, one a study team member stated they
542 ‘learned that participants have valid, interesting, and often novel ideas that researchers may not

543 have considered' and their 'perceptions of involving participants in planning medical research
544 have significantly changed', which can be viewed as an impact of 'transformative learning' ^{39,50}.
545 Using a standardised reporting tool to describe the methods and impacts provides a way for
546 learning from this case study to inform future research studies planning to involve people,
547 including studies beyond the discipline of public health genomics.

548

549

550 **Declarations**

551 **Ethics approval and consent to participate**

552
553 The Ethics Committee of the Alfred Hospital (Melbourne, Australia) granted approval for
554 this project, which was considered Low Risk Review and approved on 16/04/2018.

555

556 **Project No:** 180/18

557 **Project Title:** ASPREE Genomics Engagement: Planning of Future Multi-Generational
558 and Family Studies

559 **Principal Researcher:** Dr Paul Lacaze

560

561 Participants gave informed consent to participate, with further consent being granted by
562 participants for sharing of recordings from the process.

563 **Consent for publication**

564 Not applicable

565 **Availability of data and materials**

566 All relevant data has been anonymised and shared in the additional files. Monash University is
567 storing all raw data according to the relevant ethics policies, and invites requests for more
568 detailed data. STARDIT reports will be added to databases in the future. Additional videos of the
569 events will be shared in the public domain.

570 **Competing interests**

571 The authors declare that they have no competing interests.

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573 Funding for the process described here was provided by Monash University.

574 **Authors' contributions**

575 JN wrote the manuscript and analysed the data, facilitated face to face events, designed and
576 carried out the evaluation of the process and the reporting (including STARDIT and GRIPP2
577 reports). MS edited the manuscript, conducted telephone interviews, ran sessions at the face to
578 face event analysed data and checked data analysis. DA advised on methodology and edited the
579 manuscript. BH was a participant advisor during the process, ran sessions at the face to face
580 event, advised on the process and edited the manuscript. JT advised on the process and edited the
581 manuscript. PL checked data analysis and edited the manuscript and ran sessions at the face to
582 face event. All authors helped co-design the process, read and approved the final manuscript.

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588

589

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777 **Additional files**

778 **Additional File 1 - Data and Analysis** - Detailed description of the data sources in this case
779 study, how they were analysed, including STARDIT reports.

780 **Additional File 2 - Telephone Questionnaire - Version Comparison** - A comparison of the
781 versions of the telephone questionnaire used.

782 **Additional File 3 - Quantitative results** - A summary of the quantitative results

783 **Additional File 4** - GRIPP2 report

784 **Additional File 5** - STARDIT report

785

786 Tables

787 **Table 5: Enablers and barriers for involving participants**

Enablers	Quotations	Barriers	Quotations
Financial remuneration for people's time; financial support for travel and accommodation	Running a business 'limits me and my time' [P15]	Living in rural areas and travel logistics a barrier to participation in face-to-face events	'I'd like to be involved online rather than face to face because of travel difficulties' [P6]
Learning and development opportunities for participants and researchers	'if you tell me what would be useful, I could do it' [P13]	Poor explanation of abstract concepts such as 'involvement', which can be jargonistic	'there seemed to be confusion, or a lack of understanding of what this 'involvement' would actually look like' (MS)
Small groups at events gave more people a chance to share perspectives	'Small group' discussions at the event 'worked well'	Self-doubt about their skills or knowledge mean they don't think they could be involved	'I'd probably ask a stupid question' [P2]
Early notice of events	'give me enough notice' [P10]	Some people not comfortable being part of a face-to-face group	'I get very uncomfortable in a group of people' [P12]

Clear information about timings and time commitments, frequency of involvement and available support	Ensure ‘people are advised what’s going to happen at the workshop’ [P8]	Lack of clarity about expected time commitments	Being involved ‘depends on what’s involved and time’ [P1]
Clear information about purpose and expectations of involvement, feeling their involvement has consequences	‘What’s the endpoint – what’s the goal?’ [P11]	Unclear about what tasks they could be involved in	‘I don’t know how I could but willing to help’ [P13]
Independent facilitator when working in groups (either face-to-face or online)	I’d feel more comfortable if I had someone who was facilitating [P14]	Face to face discussions ‘dominated’ by more confident or knowledgeable people	‘Participants from professional backgrounds to some extent dominated some of the discussions’ (Study team)
Short events ensure people do not get fatigued	‘any longer and I think fatigue would have dampened the enthusiasm’ (Study team)	People feel they have limited time, are busy with work or social commitments	‘I don’t have a lot of time left in life’ [P1]
Having access and literacy in using computers and online tools	‘if I could negotiate [online discussions] I’d be happy to do that’ [P15]	Lacking access, literacy or trust in using computers, smartphones or	‘I’m hopeless with computers’ [P16], ‘I don’t have internet’ [P12]

online tools (including social media)

A selection of flexible communication modes (such as face to face and facilitated online discussion forums)	'I'd be happy to be involved – more online but if there was an occasional need to come into the city I'd be happy to do that' [P3]	Only one mode of communication, such as expecting people to travel to events	'online is often easier', face to face only 'as long as it's not too far' [P22], 'travel distance is an issue' [P20]
Involving people in research ethics and governance	'ethics is the difference between right and wrong – you know what's right and you don't do what's wrong' [P18]	A 'researchers know best' attitude that doesn't value the process of involving people (MS)	Researchers 'don't see the forest for the trees' [P19]

Figures

Figure 1

Process timeline of the participant involvement

Co-designing a multi-generational genomics study

Figure 2

Co-designing multi-generational genomics research

Supplementary Files

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

- [AdditionalFile1DataandAnalysis.pdf](#)
- [AdditionalFile2TelephoneQuestionnaire.xlsx](#)
- [AdditionalFile3Quantitativerevents.xlsx](#)
- [AdditionalFile4GRIPP2.pdf](#)
- [AdditionalFile5STARDITAlphaVersionReport.pdf](#)