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Towards an Inclusive Genomics

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Towards an Inclusive Genomics

Executive Summary

This document reviews the literature relevant to designing an inclusive approach to genomics research. The Centre for Population Genomics (CPG) aims to build a genomic reference database that represents the diversity of the Australian population. As existing large reference databases, such as gnomAD, contain mainly genomic data from people of European and American descent, many Australians from non-European and non-American ancestry groups are less able to benefit from genomic medicine. The mandate of the Centre's Inclusive Genomics team is to use transparent communications and respectful engagement processes to support significant numbers of Australians from diverse backgrounds in deciding to include their genomic data in CPG's planned reference database so that all Australians will be able to benefit from genomic medicine. This literature review will inform the design of the reference database project.

A key challenge for this work is that ethnic minority groups in high-income countries are known to be less likely to participate in research. This underrepresentation has less to do with a reluctance amongst minority ancestry groups to take part in research, and more to do with how research, as it is usually carried out, is set up to cater to the majority population. Research-as-usual presents barriers to the participation of minority ancestry groups. These include:

- Not being reached through mainstream recruitment efforts
- Researchers not accommodating different language and literacy needs
- Failure to communicate in ways that resonate with minority ancestry groups
- Research processes that do not align with communities' values
- Not addressing logistical barriers to participation (e.g. time, convenience, cost)

Mistrust of researchers can also be a barrier to participation, though this will vary between communities depending on historical experiences. Addressing mistrust and building trusting relationships is central to greater inclusion of minority ancestry groups in medical research. CPG will need to understand the nature of mistrust in the different groups we work with, and address concerns and fears in a transparent and upfront way. Community-based participatory approaches are a well established way of building trust and of developing an inclusive approach that facilitates the participation of minority ancestry groups.

Developing culturally and linguistically relevant project materials and platforms will be a central part of the Inclusive Genomics team's work. Catering to different language needs and designing materials that will suit different literacy levels will be essential. Tools such as health questionnaires and digital participant portals may need to be tailored to different groups to ensure their validity and accessibility. CPG will also need to take into account preferred styles of communication, terminology, and sources of information in the development of communication strategies and education materials. Further, materials should be developed in a way that is culturally relevant by incorporating and building on community understandings of health, illness, and heredity.

The suitability of standard consenting frameworks, materials, and processes will need to be considered in working with diverse populations. While broad consent may be encouraged as an ideal because of its scientific benefit, providing this as an option amongst others may be reassuring to participants, particularly in communities where trust for research is low. Broad consent can be encouraged through conversation and dialogue with communities about its value. Innovative digital approaches that replace the traditional text-heavy participant information sheet can be more effective in supporting informed and ongoing consent. Digital education and consent options can be useful in supporting ongoing contact with communities in between in-person encounters, though they should not replace face-to-face engagement with communities.

Participation in CPG's reference database project will ideally require the donation of a venous blood sample. Compared to other biospecimen collection methods such as saliva, requiring a blood draw is likely to have an impact on recruitment. As with the majority population, fears around the process of blood collection may be barriers, and in some communities, the meanings associated with blood may make donation of blood a greater burden. Researchers can support participants to donate blood by addressing concerns and acknowledging the significance of the donation for community members. Additional resources can be put into lowering other barriers and facilitating participation to compensate for the increased challenge of recruitment in requiring venous blood samples.

While the future benefit of participation for communities can be a motivating factor in the decision to take part, these are abstract benefits that are less tangible to prospective participants. Altruism may be a strong motivator in some communities where there is the concept of a selfless gift, while in others, framing a donation as a 'gift' may raise expectations of reciprocity. Incentives in the form of reimbursement or compensation can be useful in helping community members to participate where there may be financial barriers to taking part. Being able to receive information about one's health (both specific and general) can also serve as an incentive to participate. Additional incentives (e.g. benefit sharing) may need to be considered to encourage broad consent as community members may be wary of the prospect of research conducted for profit.

CPG will address the underrepresentation of minority ancestry groups through targeted work inviting specific communities to be part of our reference database project. In our approach to community engagement and research, we aim to privilege the perspectives and meanings of the communities we engage with. Through co-design, we will work with communities to develop suitable approaches to tackling the challenge of recruiting large numbers of participants from Australia minority ancestry groups.

CPG's community engagement and research work will seek to lower barriers and facilitate participation by first gaining an in-depth understanding of the needs of the community in taking part in the project. This will occur through a recognition of, and respect for, differences in community norms, knowledge, beliefs, language, literacy, motivations, fears, and concerns. Through co-design in partnership with communities, we will build on these understandings, drawing on the expertise of key community members to develop pathways to participation that meet these needs. This work will lay the groundwork for an inclusive genomics that secures a future where all Australians will be able to enjoy the benefit of developments in genomic medicine.

Introduction

CPG's Inclusive Genomics (IG) team's mandate is to use transparent communications and respectful engagement processes to support significant numbers of Australians from diverse backgrounds in deciding to include their genomic data in CPG's planned reference database. This is a core part of CPG's work. It is important because existing large reference databases, such as gnomAD, contain mainly genomic data from people of European and American descent. This means that many Australians from non-European and non-American ancestry groups are less able to benefit from genomic medicine.

Data from the last census show that Australians report having a very wide range of ancestries:

- Oceanian peoples (including Aboriginal and Torres Strait Islander, Maori, Fijian, Samoan, Papua New Guinean, and other peoples),
- North African and Middle Eastern peoples (including Egyptian, Iranian, Iraqi, Lebanese, Palestinian, Sudanese, Turkish and other peoples),
- North Asian peoples (including Chinese, Korean and other peoples),
- South-East Asian peoples (including Filipino, Indonesian, Cambodian, Vietnamese and other peoples),
- South Asian peoples (including Indian, Sri Lankan and other peoples),
- Sub-Saharan African peoples (including Ethiopian, Ghanaian, Kenyan, Mauritian and other peoples),
- as well as European and American peoples

There were 4.9 million reports of ancestries other than European or American by 23.4 million Australians in the last census (with more than one ancestry possible). By building a new database of genetic variation that more fully captures the diversity of the communities living in Australia, CPG will lay the foundation for a future in which all the peoples of Australia can benefit from genomic medicine.

A key challenge for this work is that ethnic minority groups in high-income countries are known to be less likely to participate in research. This is true in medical research generally (George et al., 2014), in biobanking research (Kim and Milliken, 2019), and in genomics research (Hindorff et al., 2017; Popejoy and Fullerton, 2016). The underrepresentation of groups referred to in Australia as culturally and linguistically diverse (CALD) - people not of Anglo-Celtic or Indigenous Australian descent - is well established (Low et al., 2019; Minas et al., 2013; Smith et al., 2018). Yet this underrepresentation has less to do with a reluctance amongst minority ancestry groups to take part in research, and more to do with how research, as it is usually carried out, is set up to cater to the majority population.

Studies conducted in the US suggest that there is little difference between the willingness of ethnic minority groups to take part in research when compared to the majority population (Wendler et al., 2005). Rather, participation is limited by opportunity and access (Hagiwara et al., 2014). Research-as-usual presents barriers to the participation of minority ancestry

groups. These barriers can range from not having been reached through mainstream recruitment channels in the first place (Davis et al., 2019), being excluded based on language (Smith et al., 2018), to communication, consent, and other research processes that are culturally inappropriate or that fail to resonate with diverse groups (Bonevski et al., 2014; Halbert et al., 2016; Hughson et al., 2016; Kelley et al., 2020).

Writing about the participation of minority ancestry groups in biobanking research, Kim et al. (2019) argue that the most significant barrier to taking part is never having been asked to. The ability to participate in research that will be of benefit to one's community relies first and foremost on knowing about the research. Groups can be excluded because they are not linked into networks or part of the communication channels through which information is usually distributed (Davis et al., 2019). People also need to know why participation in research is important, and increasing the representation of minority ancestry groups in genomics research will entail addressing the low levels of knowledge about genomic research that is known to be a barrier to participation (Middleton et al., 2020; Scherr et al., 2019).

The way in which project materials and processes are designed plays an important role in how inclusive research is of Australia's minority ancestry groups. The presentation of information and development of consent materials and processes that do not take into account community needs have been shown to be barriers to participation (Hughson et al., 2016). In Australia, research indicates that a major reason that CALD people are less likely to take part in health research is that fluency in English is often a prerequisite for participation (Low et al., 2019; Smith et al., 2018). Similarly, different groups may be prevented from participation by failures to cater to different literacy levels, or to consider communities' knowledge and understandings of health and illness (Bonevski et al., 2014; Buseh et al., 2017).

Researchers may also unwittingly erect further barriers for minority ancestry groups to take part in research through inflexibility around how people are able to participate. With socioeconomic and ethnic minority status closely related, challenges around the logistics of attendance can be significant for some groups. Community members may face barriers around the financial cost of attending appointments as well as the time commitment needed (Bonevski et al., 2014; George et al., 2014; Hughson et al., 2016). As well, researchers can overlook how opportunities for participation may be limited for particular subgroups (e.g. women) by not designing research processes that incorporate a community's mores and that are sensitive to cultural taboos (Bonevski et al., 2014; Hughson et al., 2016).

Finally, mistrust is frequently cited as a central barrier to the participation of ethnic minority groups in research (Bonevski et al., 2014; George et al., 2014; Hughson et al., 2016). Levels of trust will vary from community to community, based on historical experiences of abuse in medical research (Garrison et al., 2019; Nooruddin et al., 2020), experiences in countries of origin (Buseh et al., 2014), and may also be influenced by how included members of particular communities feel in Australian society (Polonsky et al., 2011). Establishing trusting relationships and specifically addressing mistrust are seen to be crucial to greater inclusion of minority ancestry groups in medical research.

Wendler and colleagues (2006) suggest that efforts to include minority ethnic groups in health and medical research should focus on making research accessible to communities rather than assuming a reluctance to participate. The Australian Clinical Trials Alliance (2020) notes that a cornerstone of research that includes minority ethnic groups is the recognition of diversity, requiring efforts on the part of researchers to cater to this diversity. Community-based approaches and other participatory methods are acknowledged to be an important part of developing inclusive research designs (De las Nueces et al., 2012; Holzer et al., 2014; Jones et al., 2018; Skinner et al., 2015). Such approaches are considered to be important not only in building trusting relationships with communities (McElfish et al., 2017) but also in designing research approaches that are relevant to communities and that facilitate their participation (Culhane-Pera, Straka et al., 2017; Skinner et al., 2015).

This document reviews the literature relevant to designing an inclusive approach to genomics research. The aim of this review is to inform project design decisions for CPG's reference database project that will support the development of culturally salient pathways for the participation of Australians of non-European ancestry.[1] Our initial engagement work with communities will seek to understand the needs, perspectives, and preferences of participating groups through community-based research methods, including co-design. This literature review will inform initial project design decisions on which our in-depth work with communities will build. It will also inform the identification of research questions that we will need to answer in our initial engagement work with communities.

Section 1 of this document discusses the all-important issue of establishing trust with minority ancestry communities. It covers the nature of mistrust in research amongst different groups and discusses strategies for trust building. Sections 2-6 discuss different aspects of genomic research design and consider the ways in which CPG can ensure that its design of the reference database project does not create barriers to the participation of community members. Section 2 covers the development of project materials generally to ensure their cultural and linguistic relevance to communities, while Section 3 specifically looks at communication strategies and education materials. Section 4 deals with consent, particularly dynamic consent, and the things that should be considered to make the consenting process inclusive. Section 5 covers the collection of biospecimens, particularly focusing on the barriers to the collection of venous blood samples. Section 6 reviews the use of incentives in research with ethnic minority groups to support and encourage participation in genomic research. Section 7 discusses community-based participatory research approaches and in particular, codesign, providing examples from genomic research studies. Finally, Section 8 provides an overview of CPG's approach and its commitment to an inclusive genomics. The section concludes with research questions that CPG will explore in our research and engagement work with communities.

[1] Australian Indigenous groups will be included in the reference database via collaboration with Indigenous genomic researchers working in Aboriginal and Torres Strait Islander communities. For this reason, our literature review focuses on literature relevant to the recruitment of non-European CALD groups.

1. Building trust in work with minority ancestry groups

Key points from the literature:

- Mistrust has been shown to be a barrier to genomic research participation of ethnic minority groups internationally
- Fears around privacy, use of data, safety of procedures, and exploitation are common
- Community-based participatory methods are recommended as a way of building trusting relationships with communities
- Researchers can demonstrate trustworthiness by being transparent and prepared to address community concerns in a straightforward, upfront way
- Institutional reputations and the involvement of community members in the research can play a role in trust

Recommendations for CPG:

- Our community research and engagement work should seek to understand the specific nature of mistrust in different communities
- We should adopt a community-based participatory approach to facilitate trust-building with communities
- CPG can demonstrate trustworthiness by being upfront about risks to participants and clear about the extent and limit to which individuals and communities will benefit from participation
- CPG should make its affiliation to Garvan and MCRI clear to communities
- In some communities, it may be helpful to employ bicultural workers to support CPG's work

Mistrust is known to be a factor in African Americans' and Native Americans' decision-making on genetics and genomics research participation based on well documented historical abuses (Bussey-Jones et al., 2009, 2010; Dang et al., 2014; Garrison et al., 2019; Nooruddin et al., 2020; Rosas et al., 2020; Scherr et al., 2019). It has also been found to be a salient factor in Indigenous Australians' receptivity to genomics research (Kowal, 2012; Kowal et al., 2012). The extent to which mistrust plays a role in the underrepresentation of CALD Australians of non-European ancestry in research is less clear.

A small number of Australian studies describe differences in trust in the health care system amongst ethnic minority groups. Qualitative research on acceptance of a government screening colorectal cancer program that included Iranian Australians found comparable levels of trust to Anglo- and Greek Australians in contrast to low levels of trust amongst Indigenous Australians (Ward et al., 2015). On the other hand, another qualitative study of help-seeking for mental health issues amongst Somali Australians found mistrust of the health system to be a major barrier (Said et al., 2021). Fisher and colleagues note that while medical mistrust is sometimes used as a proxy for understanding underrepresentation in medical research, it does not fully account for it. The authors argue that researchers should seek to understand the specific barriers for different groups and design approaches with communities that work for them (Fisher et al., 2020).

The specific nature of genomics research makes building trust a key factor in promoting greater participation of diverse groups. In Fisher et al.'s (2020) systematic review of studies investigating ethnic minorities' participation in genomics research, trust was a recurring theme in the barriers the authors identified in the literature. These included concerns about privacy, unauthorised use of the information, concerns about potential harm from access to one's genetic information, and the safety of procedures involved in participation. Mistrust in whether the research will be used for the benefit of the community has also been shown to play a role in the willingness of minority ancestry groups to take part in research (Bussey-Jones et al., 2009; Yeh et al., 2020), as has the fear of being exploited in the research process (Buseh et al., 2017).

CPG's approach to recruiting large numbers of Australians of minority ancestry groups into the reference database will centre on building respectful, trusting relationships with communities. Community-based approaches to research and engagement have been shown to be an effective way of building such relationships and are frequently cited as being crucial to addressing mistrust (Buseh et al., 2014; Holzer et al., 2014; Jones et al., 2018; Scherr et al., 2019). As well as working to establish trust through engagement, researchers can also demonstrate their trustworthiness to participants in other ways. Kraft et al. (2018) argue that being able to demonstrate trustworthiness to participants is not just a matter of intrinsic ethical value. It is also a matter of instrumental value since being seen by communities as trustworthy would support the participation of diverse groups in genomics research and address the inequity of their underrepresentation.

Researchers investigating trust in medical research in ethnic minority communities emphasise the need to understand the nature of mistrust in different communities and to be willing to address concerns in a clear and transparent way. Griffith et al. (2020) note that based on the well-documented instances of mistreatment of ethnic minority groups at various times in history, mistrust is an entirely rational response to invitations to take part in medical research. The authors argue that it is incumbent on researchers to take these fears seriously and to be prepared to address them. Even in groups with high levels of mistrust, Griffith et al. found that people were open to taking part in research once their particular concerns and fears were acknowledged and addressed.

As well, researchers have investigated how participants make assessments about trustworthiness and how these factor into decision making about whether or not to participate (Bussey-Jones et al., 2010; Guillemin et al., 2018; Kraft et al., 2018; Passmore et al., 2020). Through an understanding of what matters to communities, researchers can design project communications, processes, and structures that support trust in the research. Kraft et al. (2018) note for example that incorporating governance structures that reflect the values of communities is one way that researchers can help demonstrate their trustworthiness to communities. This has been especially important in Australian genomics work with Aboriginal and Torres Strait Islander peoples (NCIG, 2018), and indigenous communities internationally (Beaton et al., 2015; Claw et al., 2018; Garrison et al., 2019; James et al., 2014).

Concerns about privacy and potential harms are prominent in the literature on ethnic minorities' participation in genomic research (Fisher et al., 2020), and suspicion of research for profit is a recurrent theme, with ethnic minority participants often being less trustful of

research conducted by pharmaceutical companies (Buseh et al., 2014; Griffith et al., 2020; Kraft et al., 2018; Rosas et al., 2020), though this has also been found to be the case amongst the general public across many countries worldwide (Middleton et al., 2020). In the US, reference to Henrietta Lacks, the African American woman whose cancer cells were used to develop a profitable cell line without her or her family's knowledge or consent, is regularly made by African American participants (Kraft et al., 2018; Lee et al., 2019; Rosas et al., 2020). Yet these well-known stories about exploitation and harm also figure in the concerns of people of other communities. Lee et al. (2019) found that participants of Hispanic, Chinese, and South Asian backgrounds also raised the example of Lacks as an explanation for why they were wary of participating in genomics research.

In their focus groups with Americans of mixed ancestries on trustworthiness in precision medicine, Kraft et al. (2018) found that participants were less trusting of government and insurance companies. Some participants expressed fears about their genomic information being used by the government to identify their ethnicity and target them. Some participants feared the possibility of data being used for surveillance purposes, or that the data might be used by groups outside of the US where there was not the same oversight. There were fears that findings about the health of their community might be used to discriminate against them.

In a large comparative study conducted across 22 countries in 15 languages about attitudes towards genomic data sharing, Middleton et al. (2020) found that the majority of participants were unwilling to have their genomic information shared with multiple users. Participants were most likely to trust medical doctors and least likely to trust research for profit, though willingness to have data shared across multiple users differed from country to country. Willingness to donate DNA was associated with familiarity with genomics and with trust. Middleton et al. (2020) argue that for greater numbers of people to take part in genomics research, there is a need to familiarise the public with the purpose of genomics research and to emphasise the necessity of collaborative research across different categories of users for the full benefits of genomic medicine to be realised.

Participants consider a range of factors in making assessments about whether researchers are trustworthy. Studies have found that African Americans are likely to be less trustful of white researchers who are conducting research in African American communities, both in medical research generally (Griffith et al., 2020), and in precision medicine research in particular (Kraft et al., 2018). Having people of the same background involved in the project has been shown to be able to overcome some of this mistrust. Rosas et al. (2020) found that African American participants were much more predisposed to take part in precision medicine research if the research was led by an African American researcher. Buseh similarly found that their participants who were US immigrants of African background had more favourable attitudes towards research where African immigrant scholars were on the genomics research team (Buseh et al., 2017).

In groups with high levels of mistrust in research, having people from the community involved in the project may be especially important in establishing trust. Employing bicultural workers who are able to be liaisons between communities and researchers is a common practice in research seeking to engage ethnic minority communities and has been recommended as a strategy for ensuring greater representation of diverse groups in health research (Brijnath et al. 2020). Bicultural workers are people employed to work with

communities with whom they share not only a common language but also a similar cultural background, serving as conduits between the community and the organisation they work for (Centre for Multicultural Youth, 2011). Having team members of the same cultural background is not however essential for all groups by any means. Rather, demonstrating respect for communities by engaging in a way that is based on an appreciation of community norms and expectations can also engender trust (Kelley et al., 2020; Passmore et al., 2020).

Noting that debates around trust have identified aspects of social trustworthiness as barriers, and that the solutions proposed have largely focused on personal trust through approaches like community-based research, Passmore et al. (2020) investigated the characteristics that give rise to social trustworthiness in the absence of a personal connection. The researchers found in their research with African American participants about their decisions to take part in genomics research that familiarity with the institution and perceiving the institution to be reputable were key factors in their assessment of trustworthiness. Griffith et al. (2020) similarly found in their review that ethnic minority communities were more likely to trust research conducted by institutions that were well known in the community, while Australian research found people made judgements about whether to participate or not first and foremost based on the reputation of the institution (Guillemin et al., 2018). Passmore et al. (2020) also found that participants looked for evidence that the institution shared the same goals that they did, for example, whether they were interested in addressing diseases that the individual cared about (Passmore et al., 2020).

The way in which information is communicated can also have an impact on how trustworthy researchers appear to community members. The ethnically diverse participants in Griffith's et al.'s (2020) study said that they were suspicious of long consent forms written in 'legalese', while other studies have found dense consent documents to be a barrier to participation (Hughson et al., 2016). Passmore et al. (2020) similarly found that ambiguous communication had an impact on African American participants' decisions to take part in genomic research as this was seen to convey that researchers had hidden agendas.

Other research has also emphasised the importance of transparency in building trust with communities. Kraft et al. (2018) found that participants from different ethnic minority groups in the US wanted information about all potential uses of the genomic data, especially uses that might lead to profit. They also wanted regular updates about how the data was being used and who was accessing the data, as well as information on any changes that were made to the study. Similarly, other studies have shown that African American participants especially want complete transparency around data sharing and study procedures (Hagiwara et al., 2014; Skinner et al., 2015).

Transparency in how research design decisions are made may also be valued by communities. The Alabama Genomic Health Initiative adopted a transparent decision-making approach to building trust with African American communities. The study used a facilitated deliberative decision-making process as a way of fostering a genuine partnership between community members and researchers in the design of the project (May et al., 2020). This is described in more detail in Sub-section 7.2 which provides examples of community-based approaches in genomic research.

In their review of studies investigating the participation of African Americans in precision medicine and pharmacogenetics research, Scherr et al. (2019) make suggestions for how high levels of mistrust can be overcome. The authors recommend genuine collaboration with communities rather than merely making contact with community leaders to help with recruitment once the study is ready to collect data. Scherr et al. suggest that collaboration that begins before the study starts, and engagement that goes beyond the completion of data collection, demonstrates respect for communities and fosters trust. Scherr et al. summarise opportunities for designing messages that build trust. These strategies include: enlisting trusted community members to deliver recruitment messages; being transparent about risks and addressing fears directly; and including details of data privacy processes in the education resources provided to communities. Scherr et al. note that studies where procedures and privacy safeguards were clearly explained were able to address concerns and overcome these barriers to participation. The authors emphasise the importance of greater research on understanding what strategies work to help build trust with ethnic minority communities.

Lee et al. (2019) argue the association in many communities of biospecimens with self can raise the bar on the levels of trust needed for participation. In their investigation comparing US ethnic minorities' perspectives on biospecimen donation versus the collection of other personal health information, Lee et al. found that the act of donation can leave participants feeling vulnerable in a way that access to their electronic health records did not. Lee et al. recommend open, public discussion with communities in particular around biospecimen collection. They note that assurances about the procedures may be insufficient to address concerns because biospecimens are tied up with identity[2]. Lee et al. suggest that understanding how institutional policies around storage have an impact on trust is important for genomics researchers seeking to engage ethnic minority communities, and that investigation of community concerns about biospecimens must go beyond concerns about privacy.

[2] What the collection of biospecimen types can mean in different groups is discussed in Section 5.

2. Developing culturally and linguistically relevant project materials and platforms

Key points from the literature:
- Requiring proficiency in English is a key barrier that has limited the participation of culturally and linguistically diverse groups in research in Australia
- Researchers should cater to different general and health literacy needs
- Questionnaires and other tools may need to be adapted for and validated with different groups
- Design of online portals must also take into consideration cultural differences in how users interact with digital interfaces

Recommendations for CPG:
- CPG must cater to different language needs as a bare minimum, and design materials that will suit different literacy levels
- Depending on the information being collected, CPG should consider whether the health questionnaires that participants will be asked to fill out need to be validated for different groups
- The design of CPG's online participant portal should consider principles of cross-cultural web design to ensure inclusivity

Australian research on CALD Australians' participation in clinical trials indicates that the requirement that participants be proficient in English is a significant barrier to inclusion. In their retrospective analysis of cancer clinical trial participation over ten years in the South Western Sydney Local Health District, Smith et al. (2018) found that CALD people (including those of European ancestry) were far less likely to take part, but this difference disappeared once English language proficiency was controlled for. This suggests that the underrepresentation of CALD groups in research in Australia is in a large part due to a failure of researchers to cater to diverse language needs. Indeed, in their search of clinical trial registries, Low et al. (2019) found that almost half of dementia clinical trials required participants to be fluent in English.

Providing materials in the languages of the communities that we work with is just the first step to an inclusive communication and education strategy for CPG's reference database project. This will involve working closely with translators and interpreters to accurately translate genomics and related terms that may not have obvious equivalents in different languages. As well as catering to varying levels of English proficiency, materials will also need to cater to a range of general and health literacy levels. For example, in their community-based participatory research project to promote the participation of Hmong Americans in genomics research, Culhane-Pera et al. provided two possible routes for informed consent, catering to Hmong community members with no high school education and low English proficiency, and those with high school education and a high level of English proficiency. (Culhane-Pera, Straka et al., 2017). We discuss literacy further in Section 4 on consent.

In addition to developing culturally salient information resources and consent materials, CPG will also need to consider the suitability of the various survey tools that might be used, as

well as the appropriateness of the digital platforms through which participants will be expected to engage with the Centre and the project. Health questionnaires and other self-report measurement tools to be used may not only need to be translated accurately but also culturally adapted and validated for use with different groups (Beaton et al., 2000; Ortiz-Gutiérrez and Cruz-Avelar, 2018). This will ensure that the questionnaires are measuring what they are intended to measure.

Beaton et al. (2000), drawing on work by Guillemin et al., note that the extent to which cross-cultural adaptation is needed for health questionnaires will depend on a number of factors relating to the source of the questionnaire and its target. These include whether the questionnaire is used in the same country in which it was developed, with the same cultural group that it was developed for, whether it will be administered in the same language, and whether it will be used with established or new migrant groups. For example, Scott et al. (2000) found that the commonly used SF-36 health survey had good construct validity for New Zealanders of European background, as well as for Maori under the age of 45 who, the authors reasoned, had relatively high levels of urbanisation. Construct validity was weaker for Maori aged 45 and over, as well as for New Zealanders of Pasifika background. In Australia, standardised instruments measuring well-being have been found to be unsuitable for use amongst Indigenous populations (Le Grande et al., 2017).

To gather information about participants' health, in addition to health record linkage and physical measurement, the US's All of Us project protocol states that participant-provided information will be collected via a self-report questionnaire. The survey will be a collection of modified questions based on health surveys used in several large cohort studies and will be translated and tested to ensure their suitability for use with diverse groups (National Institutes of Health (NIH), 2018). The UK's Genes and Health project similarly collects self-reported health information via a questionnaire that has been designed to be linguistically and culturally relevant to British Bangladeshis and Pakistanis (Finer et al., 2020).

Participation in CPG's reference database project (including consenting and filling out self-report questionnaires) will occur via an online portal. This portal will also need to be developed in a way that will not present barriers to Australians belonging to minority ancestry groups. While the design of the portal will need to be inclusive of a broad range of Australians of many backgrounds, key elements of cross-cultural web design will need to be taken into consideration so that the design does not exclude particular groups. In addition to ensuring that the content of the portal is culturally appropriate and presented in the languages relevant to the groups we seek to engage, we will also need to attend to the different needs around the way in which information is presented and laid out; the suitability of the imagery used in the portal; the use of colours which represent particular things in different cultures; as well as how people of different cultures are likely to interact with and navigate websites (Mushtaha and De Troyer, 2012).

3. Communication and development of education materials

Key points from the literature:

- Education about the importance of genomics research is central to recruiting underrepresented populations
- To be successful in improving recruitment, education needs to be tailored to meet the specific information needs of the target population
- Taking a participatory approach that involves communities in the design of materials will require upskilling community members to be able to make meaningful contributions
- Starting with existing understandings about heredity can be a good way to build information on genetics and genomics that is culturally relevant
- Researchers must consider how the way they communicate the project may be at odds with how different groups understand health and illness
- Preferred styles of communication, terminology, and sources of information need to be taken into account

Recommendations for CPG:

- A central aim of CPG's community-based approach should be to develop relevant education materials and plan suitable communication strategies with communities
- CPG's education and communication strategy should be based on sound understanding of communities' knowledges and perspectives
- Consider what resources community members will need to be able to meaningfully engage in designing the project

Communication with communities will be central to CPG's relationship building work with different minority ancestry groups, and the development of suitable and effective education materials will be a key part of this communication. Knowledge about genetics and genomics is known to be low in the general population (Condit, 2010; Molster et al., 2009) as well as in minority ancestry groups (Dang et al., 2014; Rosas et al., 2020; Scherr et al., 2019; Simon et al., 2017), though this appears to be improving (Dar-Nimrod et al., 2018). Researchers have shown how initial reluctance or indifference to taking part in research that is associated with a lack of awareness about genomics and the importance of genomics research can quickly shift once people are informed. In their research with US ethnic minority groups on precision medicine, Rosas et al. (2020) found that participants had little knowledge to start with, but that members of some ethnic groups (namely Chinese, Vietnamese, and Latino participants) expressed enthusiasm for participating once they understood the purpose of the research[3].

Providing relevant education has also been shown to be a facilitator to participation even in groups that may be more hesitant to take part in genomics research. This education must, however, be developed with the needs of different communities in mind. Scherr et al. (2019) found in their review of literature on personalised medicine research amongst African Americans that general education was ineffective in increasing participation. Instead,

[3] African Americans and Native American participants were more hesitant, as is consistent with the literature (Rosas et al., 2020).

education needed to be targeted and to be developed to address the specific concerns of communities.

CPG's central strategy for developing educational and other informational materials that resonate with the minority ancestry groups we work with will be through co-designing materials with community members. A key principle of a community-based participatory approach is that community members need to be provided with the necessary information and tools for meaningful engagement in the design process (Israel et al., 1998). CPG's initial engagement work will thus require us to ensure that community members are equipped to contribute to decision-making about educational materials for the broader community that will effectively communicate complex concepts about genomics, disease, data, privacy, and so on. Helping community members to get up to speed on issues relating to genomics research participation will also support them to participate in other design decisions about the pathways for community participation.

In the GTEx (Genotype Tissue Expression) initiative carried out in a Hispanic American community, Mosavel et al. (2019) engaged in an extensive process of education for community advisory board members who then became active participants in developing educational resources for the community and in talking to community members about genomic biobanking. Advisory board members participated in regular meetings that initially focused on improving knowledge of genomics. These meetings then increasingly focused on developing resources for the community as board members became more equipped to make these decisions. Further details about Mosavel et al.'s community-based approach are described in Sub-section 7.2.

For information about genomics to be taken on board by different communities, researchers emphasise that education must incorporate and build on community values, beliefs, and knowledge (Barlow-Stewart et al., 2006; Christensen et al., 2010; Mosavel et al., 2019). Christensen et al. (2010) recommend identifying community concepts about heredity that are scientifically sound and building on these in developing education materials. This was the approach taken by Culhane-Pera et al. (2017) in their work with Hmong Americans. The researchers conducted semi-structured interviews and focus groups to investigate community understandings of heredity, genetics, disease, and medicine. While participants were largely unaware of genetics, they were well-versed in Hmong concepts of heredity. Culhane-Pera et al. found that many ideas around the heritability of different diseases and the relative contributions of men and women to their offsprings' physiology were at odds with modern genetics. The authors identified, however, that the Hmong idea of a child's characteristics arising from a combination of a mother's egg and a father's seed was an opportunity that could be built on to provide the community with education about genetic inheritance, disease, and responses to medication.

One of the first steps in our work with communities will be to learn about the ways in which different ancestry groups understand health, illness, and heredity. In their work with African immigrants in the US, Buseh et al. (2017). found that traditional understandings of the body, causes of disease, and how illness was dealt with in families, was often at odds with the Western biomedical approach of genetics and genomics researchers. Buseh et al. argue that because concepts relating to genetics and health touch on important cultural beliefs about the self, kinship, and the spiritual, researchers must be prepared to discuss these openly

and respectfully. It will thus be important to understand what the introduction of information about genetics and genomics means in different communities.

Commonly held views about the aetiology of disease will be one area that will be especially important for the CPG team to investigate. For example, Saleh et al. (2011) found in their research of the understandings of genetics and cancer amongst Arab Australians, that participants commonly saw disease to be the inevitable consequence of trauma and war. For the African immigrants in Buseh et al.'s study, disease was attributed to witchcraft or ill fortune rather than genes, and participants suspected that fellow community members would have little inclination to take part in genetic research because of this (Buseh et al., 2017). Amongst the Yu'pik in Alaska, to anticipate disease is considered to invite it, and thus West et al. suggest that the way in which geneticists often talk about the predictive power of genetics was off-putting to the community (West et al., 2013).

Within any community, there are likely to be a broad range of perspectives and varying degrees of health and science literacy. Building education approaches that are respectful of community worldviews while introducing ideas that may run contrary to widely held beliefs should not be seen as a campaign to supplant mistaken beliefs with correct ones. Rather, it is helpful to recognise that both traditional and biomedical worldviews are cognitive frames that shape the way people experience and act in the world (Good, 1993). Indeed, biomedical understandings often sit alongside traditional beliefs about disease causation, as Eiesenbruch et al. (2004) found in their interviews with Chinese Australian families about cancer genetics. For example, while cancer might be understood as the result of 'faulty genes', the antecedent may be thought to lie in past actions. The attribution of faulty genes to families can thus be experienced as deeply stigmatising. Ideas of kinship, heredity, and disease causation, thus need to be taken into account when communicating genetics and disease to different groups (Barlow-Stewart et al., 2006).

Respectful and effective engagement with communities will also require understanding communities' preferred modes of communication, the terminology that makes sense to them, and the channels through which they access information that they trust. Recruitment can be influenced by styles of communication, such as the preference for storytelling in the provision of information and *personalismo* or one-on-one friendly interactions with researchers amongst Hispanic Americans (Kelley et al., 2020). Initial face-to-face engagement has been shown to have a positive impact on the recruitment of African Americans in genetics and genomics research (Johnson et al., 2011). Recruitment is also influenced by the information source. Davis et al. (2019) found in focus groups with underserved populations in the US that participants were more likely to report willingness to participate if the information had come from a health provider that was trusted versus if they had seen the information in a paper, on TV, or advertised at a supermarket.

Appreciation of the differences in the meaning of commonly used metaphors in science, as well as the way in which community members understand different terms is also important. Cho et al. (2017) investigated the 'bank' metaphor used by large genomics projects, finding that, amongst diverse groups including native English, Spanish, and Mandarin speakers, the term 'library' was preferred. The authors found that the use of this term over 'biobank' had more positive connotations, engendered greater trust, and gave rise to fewer misunderstandings. Cho et al. recommend more careful consideration and investigation of

how commonly used metaphors in science are perceived amongst minority ancestry groups. Similarly, in focus groups about genetic research with African, Hispanic, and Native Americans, Ridgeway et al. (2019) found differences in the language used by focus group facilitators and participants and the way in which terms were understood. The authors suggest that researchers especially explore community concepts of risk and future benefit and incorporate these into communication.

4. Consent in genomic research with diverse ancestral groups

Key points from the literature:

- Broad consent is considered beneficial from a scientific perspective but requiring it can be a barrier to the participation of diverse groups
- A tiered approach to consent that provides participants with options demonstrates transparency and can help build trust with communities
- Attitudes towards broad consent can shift with education and dialogue
- Openness to broad consent is associated with higher health information efficacy
- Building community health and science literacy may help support greater data sharing
- Traditional participant information sheets that overwhelm participants with detail are ineffective in supporting informed decision making
- Innovative digital education and consent processes can help to present information in an accessible way, at times when participants need it
- These digital approaches should not replace face-to-face engagement
- Differences in literacy and access should be considered (e.g. providing information at a 5th grade reading level; making portals compatible with mobile devices)
- In some communities, individual consent alone may not be sufficient

Recommendations for CPG:

- Promote the benefit of broad consent to communities but offer participants options for how their data is shared
- Maximise the potential of the planned online portal for effective information sharing and ongoing connection with participants
- The online portal should be integrated with CPG's Inclusive Genomics team's face-to-face engagement (e.g. maintaining communication with communities in between in-person meetings)

Developments in genomics, biobanking in particular, have led researchers to rethink and adapt traditional approaches to consent because of the potential for broader and longer-term use of participants' genetic samples. The recognition of the need for greater diversity in genomic datasets has further necessitated consideration of the suitability of these approaches for culturally and linguistically diverse groups. This section reviews the literature on consent in genomic research, explicitly focusing on diverse ancestral groups.

The document covers three key themes, each with relevant findings:

1) **The continued relevance of broad consent to research with diverse and historically marginalised groups**
    a) Broad consent presents both ethical and practical challenges to work with diverse communities
    b) It continues to be considered to be an important ideal that is thought to maximise the scientific benefit of research participation and allow communities to reap the full benefits of genomics
    c) It must occur within a framework that builds trust and prioritises transparency

2) **The centrality of ongoing engagement and dialogue with communities**
    a) The long-term and indeterminate nature of genomic research, as well as the complexity of information that needs to be conveyed to participants, requires a commitment to ongoing engagement and education
    b) Investment in building science and health literacy of communities may support communities to participate in ways that best facilitate data sharing

3) **The importance of considering context and tailoring approaches to particular communities**
    a) Communication strategies must consider the role of intersecting variables such as socioeconomic status and education, and aim to communicate to the broadest possible audience within a community
    b) Innovation in modes of communication in genomics (e.g. digital consenting platforms) are important ways of maintaining ongoing engagement
    c) Use of these should to occur alongside face-to-face engagement, and in line with particular community needs and preferences
    d) Individualised approaches to consent may be insufficient for some communities

These points are elaborated in the sections below with reference to the literature. We conclude the document with implications for the CTRL redevelopment for CPG's reference database project.

4.1 Broad consent and alternatives

Broad consent has been adopted in genomics as the most practical approach to consent for biobanking and data sharing. Broad consent means that participants grant researchers consent to use their samples for research purposes beyond a single study and, often, over a long period. It can also include permission to share the samples with other researchers and across public and private sectors. Alternatives to broad consent, involving choices to opt in or out of specific research, have been developed to address the concerns about the lack of autonomy for participants in research (Haas et al., 2021; Kaye et al., 2014). The requirement of broad consent for research participation raises additional issues with diverse and often marginalised communities with experiences of colonisation, dispossession, and harm in medical research who may reasonably be less inclined to trust researchers to make broad decisions about the use of samples without specific consent (Garrison et al., 2020; Nordling, 2021)

These are not only ethical problems but also practical ones. In a systematic review of research on consent in genomics, Garrison et al. (2016) found that ethnic minorities are less likely to prefer broad consent. This suggests that the requirement of broad consent for participation in research is likely to be a barrier to the inclusion of diverse peoples in genome reference databases.

On the other hand, others argue that overprotectiveness can deny participants opportunities to be involved in research that will benefit themselves and their communities. Faden et al. (cited in Bromley et al. 2020) argue for consent processes that instead focus on

transparency and trust-building rather than on being overly protective of participants (Bromley et al., 2020). Consent processes with diverse communities thus involve balancing the benefits of a framework that facilitates data sharing and one that addresses historic mistrust of medical research in a way that would enable large numbers of people from diverse communities to participate in research.

In a recent news article for *Nature*, Nordling (Feb 2021) describes the debates around broad vs tiered consent approaches for populations in Africa. A recently released report by the African Academy of Sciences and the African Union Development Agency recommends a tiered approach to consent to protect participants from exploitation (AESA, 2020). The tiered approach involves offering participants options for how their data can be used. These can include consenting to research for a specific project only, for a range of projects related to a particular disease, or for unspecified future medical research. Despite concerns about broad consent, major genomics projects such as H3Africa and the UK's 100,000 Genomes projects continue to promote broad consent. Nordling cites Jennifer Troyer, director of the H3Africa project, who argues that broad consent is crucial for data from underrepresented populations to be included in global analyses. Similarly, Nigerian-born NIH scientist Charles Rotimi argues that overprotectiveness over data is counterproductive as it will prevent communities from reaping the benefits of open science.

Based on their work on H3Africa, Tindana et al. (2017) write that broad consent can be an acceptable approach that makes sense for future global genomic research if it occurs within the right framework. The authors suggest that broad consent must be considered within a framework of 'entrustment'. Within such a framework, participants entrust their biospecimens to an institution. The establishment of this relationship, where participants' biospecimens are held in trust, requires clearly outlining the institution's obligations to participants and involves ongoing engagement with communities.

Nevertheless, the provision of options other than broad consent to participants can itself be part of trust-building, even if obtaining broad consent is the ideal. Ambroise Wonkam (cited in Nordling 2021), co-director of H3Africa, argues that a tiered approach need not necessarily lead to data restriction. Wonkam suggests that most people will opt for broad consent when they are presented with a range of options. Wonkam emphasises that a tiered consent framework in the context of genomic research on the African continent is about creating structures that support transparency and promote greater trust between participants and scientists.

4.2 Engagement and education

Increasingly, the informing and consenting processes are seen as an opportunity to build and maintain ongoing relationships with participants (Bromley et al., 2020; Wilbanks, 2018). With diverse communities, it can be an opportunity to develop a culturally salient approach to engagement with participants that can facilitate the involvement of underrepresented groups (see Dickert in Kraft and Doerr 2018).

Facilitating participation

An approach that emphasises ongoing engagement can contribute to greater openness to data sharing. In their systematic review, Garrison et al. (2016) found that, even though ethnic minorities were less inclined to provide broad consent, attitudes shifted with education and dialogue. Other research indicates that participants' confidence in their ability to find and understand health info was associated with their need for control over data. Hong et al. compared the biobanking consent preferences of African Americans and European Americans (Hong et al., 2020). The authors found that high health information efficacy was associated with a lower need for control and a greater openness to broad consent, an effect that was especially significant amongst African American participants. Hong et al. argue that building community health information efficacy can help to support broad consent. This suggests that investment in building the health and science literacy of communities may allow communities to participate in ways that align more closely with genomic researchers' ideals.

Informing to support decision making

The complex and fast-changing nature of genomics research has raised questions about the feasibility of genuine 'informed' consent, and on the suitability of the modes in which informed consent is achieved. Traditional informed consent processes have been shown to be inadequate in facilitating meaningful decision-making for participants (see Flory et al., Montalvano et al., cited in Kraft and Doerr 2018). Wilbanks (2018) points out that pages-long consent documents that cover all the legal bases, but that overwhelm participants with more information than they can reasonably take in, does not constitute meaningful informed consent.

Some have even questioned the feasibility of informed consent in genomics per se. Based on interviews with researchers and ethicists working in the area of biobanking, Bromley et al. (2020) suggest that there is a move away from the notion of 'informed' consent to one of 'engaged' consent. With engaged consent, the impracticality of participants being fully informed and comprehending the full extent and implications of the research prior to consenting is acknowledged. The focus is instead on maintaining an ongoing conversation with participants that allows them to be as informed as they need to be at different points in the process of participation.

Innovation in modes of information delivery

Dynamic consent has emerged as a model that aims for more engaged participation and mechanisms to build science literacy in participants (Kaye et al., 2014). The All of Us project in the US, which aims to recruit participants with a broad range of backgrounds, makes use of an interactive, multi-media platform that combines limited amounts of text, images, and video (Kraft and Doerr, 2018; Wilbanks, 2018). Key concepts are reiterated over the course of a participant's interaction with the platform, and participants can test their understanding through quizzes. Kraft and Doerr (2018) also review the CHARM study's (Cancer Health Assessments Reaching Many) consent processes which include a similar use of multimedia optimised for smartphones. The study designed its platform with the guidance of a patient advisory group, and the content was developed by health communications experts. The material was translated into relevant languages by bilingual staff who were skilled in communicating health information in language.

The inadequacy of traditional informed consent processes is even more apparent in research with diverse groups (see Kutner cited in Kraft and Doerr 2018). In their study of consent processes in a genetics study in a resource-poor setting, Tekola et al. found that traditional approaches to informed consent, such as the provision of participant information sheets, were ineffective in helping participants make informed decisions about participation (Tekola et al., 2009). The authors found that participants were more likely to gain an understanding of the research through conversation.

4.3 Tailoring approaches

Genomics research requires communication of complex ideas with implications beyond the individual, requiring innovative approaches to delivery to ensure meaningful informed consent. These approaches require tailoring if they are to be effective with diverse groups with a range of cultural backgrounds, and intersecting socioeconomic, educational, factors.

Different levels of literacy

Catering to a broad range of literacy levels is important for communicating with participants in general. It is especially important when working with diverse groups where migratory trajectories (including fleeing from war), and other experiences of social disadvantage, can disrupt education. Multimedia approaches have been demonstrated to be effective with people with low levels of literacy (Afolabi et al., 2015). To reach the broadest section of society and to include people with low levels of literacy, it is normally recommended that communication of health information be pitched at a grade 4-6 level (See Centre for Culture, Ethnicity & Health recommendations). Both the All of Us and CHARM projects catered to people of a range of literacy levels by producing written material at the 5th grade reading level (Kraft and Doerr, 2018). The All of Us platform was particularly evaluated with people with low levels of literacy, while the CHARM study provided an audio option for participants.

Low levels of literacy need not hinder the communication of complex ideas about genomics and the implications of participating in research. Bukini et al. (2020) explored the perspectives of genomic research participants in Tanzania, investigating the perceived relation between literacy and comprehension. Participants felt that the mode of delivery of the information and the time the researchers took to explain the research were more important than levels of literacy in determining their understanding.

Other modes of engagement in addition to digital

Bukini et al. (2020) also found that there was a preference for group information sessions where participants felt more comfortable asking questions, and face-to-face information sessions were favoured over learning from informational videos. In their assessment of the suitability of electronic dynamic consent platforms for engaging with Indigenous Australian communities, Prictor et al. (2020) emphasise that these cannot replace face-to-face engagement with communities. The authors note that digital dynamic consent processes provide innovative means to present information in accessible ways, for example, by embedding informational videos alongside the electronic forms used to document decisions,

videos that participants can revisit. These approaches are useful as a way of building literacy and maintaining engagement with communities in between face-to-face interactions.

The reliance on digital platforms also raises the issue of equity of access. While acknowledging the value of e-consenting platforms for work with Indigenous communities, there remain issues around different levels of digital literacy and access that require consideration, particularly in work with Indigenous populations (Garrison et al., 2020; Prictor et al., 2020). Ensuring that digital platforms are optimised for use on smartphones can increase access (see Hausen cited in Kraft and Doerr 2018). Prictor et al. (2020) note that in remote Indigenous communities with limited access to information technology, people are much more likely to have access to smartphones than a desktop computer.

Who needs to consent

The nature of genomics research has given rise to considerations of whose consent is needed, requiring researchers to consider models that go beyond individual consent. Because an individual's genetic information has implications for biological relatives, models of familial consent are important (Minari et al., 2014). The need to broaden out the focus on individual consent is especially relevant in research with diverse groups. Tsosie et al. (2019) argue that individualised consent does not take into account risks to small groups such as tribes. The authors cite the example of the All of Us project which has been successful in recruiting Indigenous participants from urban centres. As large numbers of Indigenous people live in urban settings, recruitment through urban centres has been much more efficient than the slow work of building trusting relationships with tribes. Tsosie et al. argue that while these approaches are successful in recruiting large numbers of Indigenous people, they undermine the work that has gone into setting up protections for Indigenous communities by working with tribes. Similarly, Garrison et al. (2020) note that individuals are not equipped to make assessments about group risk.

Prictor et al. (2020) note that dynamic e-consenting platforms can help support models of collective decision-making where there is the need to track the decisions of multiple individuals within a community. The authors emphasise the need to develop digital consenting tools with communities. While dynamic digital consent is a good way of building trust with Indigenous communities, partnership with communities must be the foundation of these approaches (Prictor et al., 2020) and these must be used in the context of Indigenous control of the research (Garrison et al., 2020)

4.4 Implications for CTRL redevelopment for CPG's reference database project

- The digital consenting platform can simultaneously provide participants with autonomy and support their ability to provide broad consent. It can be a useful way of building communities' health literacy to support communities to participate in ways that will be of greatest benefit to them. This potential should be maximised in the redesign of CTRL.
- Provision of information should be focused on what supports genuine decision making. This may mean that less is more.

- Consider engaging health communication experts to help develop informational content to effectively maximise the potential of the digital consenting platform to build literacy.
- Consider gathering input and evaluating the platform with a diverse range of community members to ensure that the CTRL platform is suitable for the broadest possible range of people.
- Redevelopment of the CTRL platform will need to be integrated with the Inclusive Genomics team's community engagement approach. The platform should support the engagement with communities in between face-to-face meetings.
- A function within CTRL that allows for dialogue and Q&A with communities may be beneficial.
- The nature of the consenting process will be different between communities. Functions that support the tracking of decision making in communities where collective consenting is necessary will need to be considered.

5. Biospecimen collection methods

Key points from the literature:

- Requiring a venous blood sample for participation is likely to have an impact on recruitment to the reference database project
- Apart from common concerns relating to the procedure of donating blood (e.g. fear of pain, needles etc.), differences in the meaning of blood between different communities will likely play a role in willingness to give blood samples
- Willingness is also likely to vary within communities along other demographic lines (e.g. age, time in country of settlement etc.)
- Recruitment challenges related to requiring blood samples can be addressed through a culturally salient community-based approach and more extensive recruitment efforts
- Research indicates that recruitment through community events is more effective than recruitment through education sessions

Recommendations for CPG:

- Understanding communities' concerns about donation of blood samples, and what it means for members to donate blood, should be a first step in CPG's research and engagement with communities
- CPG should incorporate these concerns into the design of participation pathways (e.g. education, processes of donation etc.) through IG's community-based approach.
- Additional resources should be put into lowering other barriers to participation (e.g. logistical issues, convenience etc.) and boosting facilitators to participation

This document provides a review of relevant literature to inform CPG's decision-making about sample collection type for the centre's reference database project. To address low levels of participation of diverse ancestral groups in medical research, researchers have employed a number of strategies. These have included ensuring cultural and linguistic appropriateness and accessibility of the research, providing incentives to facilitate participation, addressing mistrust of research, as well as lowering the burden of participation to make participation as easy and convenient as possible (Bonevski et al., 2014; Fisher et al., 2020; Hughson et al., 2016; Kim and Milliken, 2019; Smith et al., 2018).

A venous blood draw is considered to be best for testing and analysis, but being more invasive, is thought to raise the bar for participation compared to the collection of other biospecimen types. Reluctance to have blood drawn was found to be the most common reason in one US study for nonparticipants' unwillingness to take part in genetic biobanking (Sanner and Frazier, 2007). In this document, we look at the evidence that suggests that this is likely to also be the case in many of our communities of interest. We discuss some of the possible reasons for this and present some examples of how these barriers have been

addressed in research. Finally, we discuss the implications for the reference database project.

The literature specifically looking at comparisons of biospecimen types for willingness to participate in genomic research in diverse ancestral groups is limited. We have supplemented this literature with studies that focus on biospecimen collection for biobanking generally, as well as the literature on blood donation. These should be considered with a number of caveats in mind. First, people may have particular concerns about the extraction of genomic information (Lee et al., 2019) which may not apply to biobanking or blood donation generally. Second, the processes of blood donation, frequency, and the volume of blood collected are different. Nevertheless, the literature in these areas provides helpful background for considering some of the challenges that the project is likely to encounter if venous blood samples are required for participation.

5.1 Will a venous blood sample requirement have an impact on recruitment rates?

While we were able to find only a small number of studies that compared willingness by biospecimen type (particularly comparing blood to saliva), these studies suggest that, in many communities, willingness to donate saliva is likely to be higher than the donation of blood, as Kwan et al. (2020) found in their work with Pacific Islanders in the US. The project involved education about biospecimen research which culminated in the collection of saliva samples from willing participants. Participants watched an educational video explaining the purpose of biospecimen research, which covered what biospecimens were used for and what was done with them after collection. Of 219 participants in the study, 214 donated a saliva sample. Donors completed a questionnaire after donating. They were asked how likely they were to donate various biospecimen types in the future. Over 90% said they would donate saliva or hair, while just under half (48.6%) were willing to donate blood. Forty-five percent (45.3%) said they were willing to donate urine, and 28% toenails.

Tong et al. (2014) found similar attitudes towards biospecimen types in a study conducted with Chinese Americans. The researchers found that, before the delivery of a culturally tailored education seminar, 73% of participants reported being willing to donate saliva, while only 40% were willing to donate blood. The willingness for other biospecimen types were 62% for urine, 55% for hair, and 43% for toenails.

A strong preference for donating saliva over blood is not a given. A study among Native Americans in Seattle found that while less than half of participants were willing to donate biospecimens of any type, participants reported being most willing to donate blood. The study assessed participant attitudes to biobanking after they had been provided information about biospecimen research and what would happen to the samples that they donated. Forty-eight percent of participants said they were willing to donate blood; approximately 40% were willing to donate saliva and hair; 30-35 % would donate urine or nails, 24% skin, and 18% tumours (Sinclair et al., 2021). Similarly, amongst older African American participants who were recruited through a research registry, Hagiwara et al. (2014) found that while participants were most willing to donate saliva, almost as many said they would donate blood, with skin being the least popular option.

The literature suggests that willingness to donate particular biological materials will differ between communities depending on the meanings of those materials for communities. For example, while the process of collecting some biospecimen types such as toenails may seem far less intrusive than the collection of venous blood, these materials may be seen to be unclean or embarrassing (Tong et al., 2014), or may be considered materials that could be used to harm the individual (e.g. via sorcery) if not securely stored (Dang and Chen, 2018; Grassineau et al., 2007; Kowal et al., 2015).

Within-community differences are also likely to be associated with willingness to donate blood samples. A study among Korean Americans found that participants who were older, had lived in the US longer, had higher incomes, and who regularly saw a general practitioner were more likely to be willing to donate (Yen et al., 2015). The authors suggest that acculturation can explain the relation between demographic variables and attitudes to biospecimen research. Other studies have found that community members who hold more traditional beliefs (Gao, Ma, Tan, Fang, Weaver, Jin and Lai, 2014), and older people (Simon et al., 2017) were less willing to donate blood samples.

These examples suggest that there are likely to be differences between and within communities in terms of how much requiring a venous blood sample will have an impact on recruitment. Further, the extent to which particular communities and subsections of communities have been socially excluded is likely to play a role. In the literature on blood donation amongst ethnic minorities, discrimination and social exclusion have been identified as key barriers (Wittock et al., 2021). A study of blood donation among Australians of Sub-Saharan African background showed that willingness to donate blood was mediated by how included participants felt in Australian society (Polonsky et al., 2011). Many community members had the perception that their blood was not wanted.

Historical and ongoing exclusion is thus likely to be an important factor in CPG's work with communities. The impacts of social exclusion on genomics research participation are most pronounced in the area of trust in research (Kraft et al., 2018). Trust has been demonstrated to be an important mitigating factor in biospecimen donation. In a study comparing actual blood sample donation in different groups (as distinct from reported willingness to donate), Bussey-Jones et al. (2010) found that while African American participants were less likely than white participants to donate, this difference disappeared when trust was controlled for.

While diverse communities may be initially hesitant to take part in genomic research, researchers have found that once people understand the purpose and importance of the research for their community (Dang and Chen, 2018), and have their concerns addressed (Dirks et al., 2019), they are likely to be just as willing to take part as the majority population. However, ethnic minorities are less likely than the majority population to think that research will benefit them (Kraft et al., 2018). In a systematic review of the literature on the participation of underserved populations in medical research, Bonevski et al. (2014) found that a perceived lack of benefit to self or community was a key reason for low rates of participation. Reddy et al. (2020) found that while African American participants had positive attitudes towards the value of biobanking and research on biospecimens, fear of exploitation and unintended use of the biospecimens prevented participants from ultimately donating.

5.2 What might some of the challenges of requiring a venous blood sample be?

Blood has different meanings in diverse communities that relate to self, relationships with others, and one's well-being. Understanding the symbolism of blood in different communities will be an important part of CPG's work in communities. In the blood donation literature, the failure to recognise these differences, and incorporate them into donation processes and recruitment strategies, plays a role in low levels of blood donation amongst migrant communities (Wittock et al., 2021).

Writing about genomic research in Indigenous communities, Aramoana et al. (2020) have argued that researchers must be aware of what exactly they are asking of people when they ask for blood samples. For some Native Americans, donating blood is seen as giving up part of one's connection to one's ancestors (Sahota, 2014). For Indigenous Australians, bodily parts are representative of connections with land, ancestors and one's culture, and the donation of these materials have implications for the integrity of the human person, as well as the safety of both self and community (e.g. through risks of sorcery using bodily materials). These have implications for the storage of materials and for what happens to samples once the genomic information has been extracted (Kowal et al., 2015).

Similar questions can be asked when requesting the donation of tissue from other cultural groups. In the context of the meanings of blood and other bodily materials for different communities, what are individuals being asked to give? What does donation risk for them? For example, in traditional Chinese, Vietnamese, and Hmong belief systems, blood is seen to be a source of vital energy (Dang and Chen, 2018; Tison et al., 2007). Donations of blood would thus be seen to have an impact on health through the loss of this energy or *qi* (in the case of traditional Chinese medicine) (Tison et al., 2007). In a qualitative study of Chinese American women's attitudes towards the donation of biospecimens, Simon (2017) found that while participants expressed positive attitudes towards biospecimen research generally, donations of hair or nails were preferred because they were seen to be renewable, whereas blood was not, and was viewed as being particularly precious.

What happens to the blood once it is collected could also be a concern for some communities. In a study aimed at increasing blood donation amongst migrants from the Comoros living in France, Grassineau et al. (2007) found that blood was considered symbolic of family relationships, and donation was considered appropriate only if given to family members in need of blood. This meant that for some Comorian participants, donation of blood for research was considered to be far more acceptable than donation to a blood bank for a stranger to receive a transfusion. On the other hand, the researchers also found that participants expressed fears about the existence of bodily materials outside the body. The authors note that while this fear has roots in traditional Comorian beliefs about the potential for evil spirits to use body parts to harm people, these fears had shifted in the context of the migration to fears of harm from the French state.

Grassineau et al. (2007) found that Comorian community members were in disagreement over whether Islam prohibited or encouraged blood donation. To supplement their qualitative data, the researchers interviewed community religious leaders. Some religious leaders advised against donation as the body was considered to be on loan from Allah, while others

emphasised the Koran's instruction on the value of preserving life. Overall, the community religious leaders concluded that donations were permitted if the recipient's life was in danger, the donor was in good health, and if the procedure involved a Muslim physician (Grassineau et al., 2007). Boenignk et al. (2015) argue that while individuals may often cite religious reasons for reluctance to participate, religion is seldom a barrier in and of itself. Rather, the inclusion of diverse communities requires consideration of how the usual blood donation processes will need to be adapted in line with religious sensitivities.

The blood donation literature also indicates that concerns about the safety of the blood collection process may also prevent members of some ethnic minority communities from donating; these include negative perceptions of blood banks in their countries of origin and fears about possible infection (Gahan et al., 2021; Klinkenberg et al., 2019). Simon et al. (2017) found similar concerns in relation to biospecimen collection, including fears of health professionals making mistakes in the process. In all cases, the researchers recommend transparency about the donation procedures.

Apart from the fear of needles and pain from the blood collection process as a barrier to donation (Dang et al., 2014; Sanderson et al., 2013), mainstream approaches to the collection of blood (that assume that blood is a biological substance with no particular cultural meaning) have failed to engage ethnic minority communities (Charbonneau and Tran, 2013). In the next section, we discuss some of the strategies that have been used with diverse communities that take into consideration their concerns surrounding blood by implementing culturally grounded education programs, including those that address the logistical barriers to participation and facilitate greater inclusion of diverse groups in research.

5.3 Possible strategies to facilitate the collection of blood from diverse ancestral groups

Researchers have addressed particular community concerns about blood by providing culturally appropriate avenues for participation. A good example of this is the work of the National Centre for Indigenous Genomics (NCIG) where the blood byproducts after DNA extraction were returned to the community for burial on country or other appropriate disposal (Hermes et al., 2021; Lewis, 2019).

Other studies have provided education that addresses community concerns. In their study trialling an education seminar for Chinese Americans, taking into consideration the communities' concerns about loss of *qi*, Tong et al. (2014) emphasised to participants that only two vials of blood were needed to donate a blood sample since the amount of *qi* lost would correlate with the volume of blood that they might be asked to donate. Further, information about how quickly blood was replenished was provided.

Culturally grounded education can improve the receptiveness of communities to biospecimen research not only by addressing specific concerns about the collection of blood, but also by building trust. Reddy et al.(2020) found that levels of trust, and consequently willingness to take part in research, was associated with a lack of knowledge. Knowledge

about biobanking has been shown to be low in ethnic minority communities in the US (Dang et al., 2014), and research suggests that knowledge about genomics is similarly low among these groups in Australia (Uebergang et al., 2021). This suggests that there may be high levels of initial reluctance and mistrust when starting our work in communities, but that it is possible to address this through culturally salient education (Reddy et al., 2020), as well as through relationship building, and demonstration of institutional trustworthiness (Bussey-Jones et al., 2010; Kraft et al., 2018).

Based on a systematic review of ethnic minorities participation in personalised medicine research, Fisher et al. (2020) recommend providing alternative options to participation apart from the donation of blood samples as one way of increasing the participation of diverse groups. If this is not possible, other strategies used in medical research to increase the participation of diverse groups could be emphasised. These include lowering the barriers to participation in other ways by making participation as convenient as possible (e.g. through the provision of childcare or flexible scheduling), or engaging project staff from the community in a way that makes participation more accessible to community members (Bonevski et al., 2014; Hughson et al., 2016).

People are more likely to be willing to donate biospecimens when they feel that they or people close to them will benefit directly. Sinclair et al. (2021) found that willingness to donate biospecimens increased dramatically for Alaskan Native participants when asked about donations for cancer research when a family member had cancer (from 43% to 76 %). Tong et al. (2014) similarly found among Chinese American participants a diminishing willingness to donate blood samples according to how distant the relationship was, from 85% being willing if an immediate family member would benefit, to 22% for strangers.

Providing incentives for participation is an important strategy in increasing the participation of underserved groups. Possibilities for these will need to be investigated further and will be especially important in considering how to make participation more attractive for communities. Fisher et al. (2020) found in their systematic review that apart from having their information needs and privacy concerns addressed, the other key facilitators of ethnic minority participation in personalised medicine research were receiving direct benefits and getting individual results. Examples of direct benefits include monetary compensation[4], provision of healthcare, and receiving results (Fisher et al., 2020).

Examples of project strategies to improve diverse communities' donation of blood samples for research

A number of studies amongst Chinese Americans have investigated the impact of education interventions on willingness to donate biospecimens. Gao et al. (2014) took a community-based approach to developing an intervention to increase donation of blood samples for Hepatitis B research. The researchers developed the intervention with community leaders. The intervention involved small-group education sessions that emphasised the severity of Hepatitis B in the Chinese community, the benefit of the research

[4] There are taboos in some cultural groups regarding the exchange of biospecimens for money.(Kraft et al., 2018)

to the community, and that addressed community concerns about the impacts of donating blood. A control group was provided with education about health and healthy living. Donations of blood samples were successfully collected from 83% of participants in the intervention group compared to just 1.1% of the control group.

Tong et al. (2014), on the other hand, had less success in influencing willingness to donate blood. The researchers conducted a randomised-control trial of a culturally tailored information seminar developed through focus groups with community members. The researchers found that the seminar had an impact on willingness to donate biospecimens like urine, toenails, hair, and saliva, and this was associated with recognising the benefit of participation for future generations. This intervention effect was not observed for willingness to donate blood, however.

Other papers have reported on examples of a combination of education efforts and convenient enrollment and recruitment models. The Asian American Cancer Education Study (AACES) aimed to increase Asian American blood biospecimen donations to cancer research (Dang and Chen 2018). Part of the study involved the recruitment of healthy adults through community blood biospecimen drives. These blood biospecimen drives occurred at community events and venues, in partnership with community health organisations and with the involvement of community health educators. Potential participants were offered the option of having their blood tested for Hepatitis B or donating to future cancer research, or both, and were able to donate on the spot. Ten drives were conducted, 5 in the Hmong community, and 5 in Chinese and Vietnamese communities, and 359 community members donated biospecimens to cancer research.

Taking a similar approach, the Hoy y Mañana study aimed to recruit African and Hispanic American participants into cancer research involving the donation of blood samples. Recruitment was either through a culturally appropriate education session of 45 min to an hour after which participants could enrol and donate straight away, or via open events with onsite mobile blood labs staffed by phlebotomists. Both events were staffed also by health educators of similar cultural and linguistic backgrounds. Fifty percent of participants in the education program reported willingness to donate following the seminar, while only 36% eventually donated. The donation rate was 55% for open events (Rodriguez et al., 2016). Other research has found recruitment to be more effective at community events compared to education sessions (Kiviniemi et al., 2013).

In a genomics specific example, the Alabama Genomic Health Initiative successfully improved the recruitment of African Americans into their study through a community-based recruitment effort that was designed in partnership with communities. The researchers engaged the community through a democratic deliberative process to help design education, communication, and recruitment strategies. This involved facilitated deliberative process meetings where researchers and community members discussed the results people wanted to receive from whole genome sequencing, how they wanted to receive results, and how the community could be engaged. All participants in the discussion voted at the end, including the researchers. This resulted in a transparent decision-making process that the authors argue helped to build trust with the community (May et al., 2020).

The initiative involved a number of events where participants were able to enrol and donate on the spot. These included having a visible presence at a major clinic, holding a town hall meeting followed immediately by an enrolment event, as well as mobile pop-up clinics. The study achieved 20% African American enrollment which is higher than the proportion of the population nationally who are African American (14%), though lower than Alabama's 26% (May et al., 2020).

5.4 Implications for the reference database project

This review suggests that requiring a venous blood sample for participation in the reference database will be a deterrent for many members of the communities we seek to recruit from if compared to other options such as saliva collection. Encouragingly, the literature on blood sample donation for biospecimen research (not necessarily genomic research) indicates a willingness of about 40-50% (Kwan et al., 2020; Sinclair et al., 2021; Tong et al., 2014), though as Bussey Jones et al. (2010) note, there is a difference in reported willingness and actual donations. While offering other biospecimen donation options such as saliva rather than blood is likely to yield a higher recruitment rate, it is nevertheless possible to overcome this challenge through a culturally salient community-based approach, and more extensive recruitment efforts. Furthermore, there is intriguing evidence that education about the purposes of the research, while useful, plays a secondary role to culturally-aligned community connectedness in the decision to participate and provide a sample for people who identify strongly with a cultural community.

The issues and barriers presented in this summary will not apply to all Australians descended from the ancestry groups of interest to our reference database. They may be particularly relevant for those people who are more recently arrived in Australia or who maintain a belief system from their ancestry of origin. However, we can expect these issues to be important in a number of our communities of interest and should therefore be prepared to encounter them.

6. Using incentives to facilitate recruitment and ongoing participation of diverse ancestral groups in CPG's reference database project

Key points from the literature:

- The future benefits of participation in genomic research can be motivators, but these need to be made tangible to prospective participants
- Appeals to altruism may be successful in some communities and not others
- Incentives should be reasonable and commensurate with the burden/benefit of participation so as not to constitute undue inducement
- Incentives in the form of reimbursements can be used to support participation where there are financial barriers to taking part
- Being provided with information has been shown to be a strong incentive to participation
- Incentives need to be considered in the context of existing motivations for taking part so that payment does not interfere with intrinsic motivation
- Providing incentives that address communities' expectations can be an important way of showing respect and building trust

Recommendations for CPG:

- Design context-specific incentive structures based on an understanding of the barriers that incentives may need to be mobilised to overcome
- Consider whether incentives already exist in any given community or whether providing other tangible, more immediate benefits may be useful
- Seek to understand the nature of gift-giving in the community and whether appeals to altruism are likely to be successful
- Capitalise on the potential for information to be an incentive to participate

This document provides an overview of relevant literature to assist in decision-making around incentives for CPG's reference database project. It covers:

- Motivations for taking part in genomic research
- Why incentives might be needed
- The kinds of incentives that might be considered and
- What should be considered in the design of incentives

NOTE: The literature on incentivising participation in genomics comes mainly from US studies that are especially focused on increasing the representation of African Americans in research. This literature is useful in outlining the elements that should be considered in designing incentive structures that support the participation of ethnic minorities communities, though historical and contextual specifics will be different in Australia.

6.1 Definitions

The NHMRC guidelines on payment of research participants uses the overarching term '**payment**' to refer to monetary or in-kind support given to participants. The document distinguishes between **incentives** and other kinds of payment such as **reimbursement**, **remuneration**, or **compensation**. The term 'incentive', however, is used elsewhere to include all kinds of payment to participants (Parkinson et al., 2019). In this document, we have used the term 'incentive' in the more inclusive sense, but have referred to the NHMRC's distinctions as these will be important in making the case for what kinds of incentives are needed and appropriate.

NHMRC classification of types of payments to research participants

Remuneration	A monetary payment, similar to a wage, paid in recognition of time spent, or inconvenience experienced.
Compensation	Monetary or in-kind support to make up for loss of wages.
Reimbursement	Monetary or in-kind support for expenses incurred including transport, food, accommodation.
Incentive/inducement	Other monetary or in-kind support beyond those listed above that would encourage participation.

Incentives will need to be considered in relation to the benefit to participants in taking part in the research. The Human Genomic Organization (HUGO) Ethics Committee defines a 'benefit' as:

> "a good that contributes to the well-being of an individual and/or a given community (e.g. by region, tribe, disease-group...). Benefits transcend avoidance of harm (non-maleficence) in so far as they promote the welfare of an individual and/or of a community. Thus, a benefit is not identical with profit in the monetary or economic sense. Determining a benefit depends on needs, values, priorities and cultural expectations." (HUGO Ethics Committee., 2000)

6.2 What motivates people to take part in genomic research?

Research in the US with African and Hispanic/Latin Americans (Streicher et al., 2011) and other diverse ancestry groups (Sanderson et al., 2013) suggest that motivations for taking part in genetics and genomics research are similar to motivations in the general population. Streichler et al. (2011) conducted research with participants in the Mount Sinai Biobank, a large DNA biobanking study that enrols diverse patients attending the Mount Sinai Medical Centre. Participants reported being motivated by feelings of altruism and the desire to participate in research that they thought would be of benefit to family, friends, and future generations.

In some cases, there may be a stronger imperative to take part. In research on the perceptions of genomics research amongst African Americans and Afro-Carribeans. Lewis et al. (2019, 2021) found that despite high levels of mistrust in participants in the ClinSeq project, participants reported being motivated by a strong desire to be represented in

medical research. Similarly, Yu et al. (2013) compared the motivations of African American and non-African Americans for taking part in exome and whole genome sequencing research, finding that African Americans were more likely to be motivated by racial justice and by contributing to research that would be of benefit to their community, rather than for individual benefit.

This research indicates that, depending on the community, there may already be high levels of intrinsic motivation to take part in research. On the other hand, research has also shown that ethnic minority groups may be less confident that research will actually benefit their communities, as George et al.(2014) found to be the case amongst African Americans and Native Hawaiians in their systematic review of attitudes towards research generally.

A similar lack of trust has been found in personalised medicine research. Kraft et al. (2018) found through focus groups with South Asian, Hispanic/Latino, Chinese, African, and European Americans about precision medicine research, that while most participants could see the value of this, people of European background were much more confident in the promised future benefit. Non-European participants on the other hand were more likely to question whether the research would genuinely be of benefit to their communities. These participants had favourable attitudes towards research that addressed diseases that were priorities for their community. Thus, while people may appreciate the benefit of taking part in genomics research, participation requires a leap of faith on their part to trust that these benefits will be delivered. Participants thus make assessments of the trustworthiness of the institution carrying out the research (Kraft et al., 2018).

Future benefits are less tangible to prospective participants, and while they can be motivators to participate, providing participants with immediate, direct benefits is likely to support greater numbers of community members to participate. Fisher et al. (2020) conducted a systematic review of the barriers and facilitators to US ethnic minority groups participation in precision medicine research. Their review of 27 quantitative and qualitative studies including African American, Asian American, and Hispanic American groups found that direct benefits such as access to health services and monetary compensation were practical facilitators for participation.

From the literature, the prospect of gaining information appears to be a salient motivating factor. Fisher and colleagues found that apart from benefiting future generations, the possibility that they might gain information relevant to their health motivated people from ethnic minorities to participate in personalised medicine research (Fisher et al., 2020). This interest in information appears not to be confined to the return of individual results. Martinez et al. (2017) found in their review of Latinos' attitudes to participation in alcohol-related genetic research that information was considered to be a benefit of research participation. This included not only the personal health information that might be returned to them, but also the opportunity to gain general information relevant to their health and to learn about the outcomes and progress of the study. Sanderson et al. (2013) similarly found in their research with multiethnic participants that interviewees expressed an interest in learning more about genetics as well as an interest in receiving information about their health. The authors were investigating attitudes to participating in genomic research on four complex diseases - obesity, heart disease, type 2 diabetes, and cancer - that were prevalent in the population attending a New York outpatient clinic.

6.3 To incentivise or not incentivise?

A key ethical concern about incentivising research participation through payment (monetary or in-kind) is that it may result in 'undue inducement' through influencing potential participants decision-making in a way that leads them to overlook the risks involved in taking part (Vellinga et al., 2020; Wertheimer and Miller, 2008). The NHMRC guidelines state that payment to participants should ideally be limited to reimbursement or compensation for time, expenses, and inconvenience. However, incentives and inducements are acceptable when recruitment is likely to be difficult and participation offers no direct benefit to participants. This is providing that the payment is commensurate with the burden of participation, and is not of a magnitude that would cause participants to ignore the risk or provide false information in order to take part (National Health and Medical Research Council (NHMRC), 2019).

The NHMRC guidelines further note that while overpayment is considered ethically unsound, non-payment and underpayment could also be considered to be unethical, and might constitute exploitation. This is especially so when participants are burdened in a way where benefits fall overwhelmingly on the side of researchers, or where non-payment results in the underrepresentation of certain groups in the research (National Health and Medical Research Council (NHMRC), 2019). Some kind of payment that reimburses participants is especially important when research is conducted with socially disadvantaged groups (Bonevski et al., 2014). Transportation and other participation costs have been found to be barriers to participation in some studies with ethnic minority groups (Brown et al., 2000; Nooruddin et al., 2020; Woods et al., 2002), and reimbursement of these expenses can provide support for participation.

Lack of a perceived direct benefit has been found to be a disincentive to ethnic minority groups taking part in genomics research (Fisher et al., 2020), and designing incentives that provide a sense that participants are gaining a direct benefit in some way can facilitate inclusion. In their systematic review of facilitators to African, Hispanic, Asian, and Pacific Islander Americans' participation in research, George et al. (2014) found that perceived benefits of research participation included small monetary incentives, being provided with refreshments, or being able to access free health assessments, while having transport costs reimbursed supported participation. In focus groups with African Americans about participation in psychiatric genetic research, Murphy and Thompson (2009) found that participants said that they were inclined to participate if they received direct benefits from taking part. These benefits included monetary compensation as well benefits coming from the process of participation such as assessment, treatment, and education.

Moore et al. (2017) notes that to facilitate greater recruitment of people of diverse ancestral backgrounds, it is important for researchers to understand the expectations of communities and to design incentives based on these. For example, in their systematic review of African Americans' perceptions of personalised medicine research, Scherr et al. (2019) found that participants expected that they would be compensated for the time that they spent taking part in the research. Participants perceived suitable compensation to include cash and fuel cards, as well as other in-kind incentives such as food, healthcare and medication.

Other expectations that participants have include receiving information from sequencing. In their work that took a community-based participatory approach to engaging Hmong Americans in genomic research, Culhane-Pera and colleagues found that the community had a strong preference for having individual results returned, and when the researchers were unable to offer this due to lack of IRB approval, this was a disincentive for some members of the community. When individual results were not able to be returned, some participants perceived there to be no benefit to them in participating (Culhane-Pera, Straka et al., 2017; Culhane-Pera, Moua et al., 2017).

This preference for individual results has been demonstrated in research with other groups. In their survey of a national sample of African Americans on willingness to participate in precision medicine research, Halbert et al. (2016) used vignettes that explicitly indicated whether results would or would not be provided. The authors found that participants reported being less likely to take part when results were not provided. Sanderson et al. (2013) found a similar result in a study of a mixed group of patients from an hospital outpatient clinic which oversampled African American and Hispanic American participants. When participants were presented with the possibilities of personal results being offered through genomic research participation, 89% reported being willing to participate when results would be provided, compared to 62% if they were not. However, the authors found as well that when the possibility of results being returned was not raised at all, 82% said that they would take part.

6.4 What kind of incentives might be considered?

In their review of incentivisation in clinical trials for both participants and recruiters, Parkinson et al. (2019) note that incentives, (under which they include also remuneration and reimbursement), can take the form of:

1. Cash or cash-like rewards (e.g. vouchers)
2. Social, emotional, or tokenistic rewards (e.g. small gifts; donations to charity)
3. Reputational rewards (e.g. recognition, authorship for recruiters)

In their population-based survey of US adults that oversampled Black and Hispanic participants, the NIH's All of Us project presented participants with six possible incentives for participation. Participants were asked how important each was to their decision to take part in the All of Us program. The most compelling reported motivator for participation was gaining information about their health (90% somewhat/very important). Being paid for their time was the next most important incentive for participants (80%), followed by receiving healthcare (77%). Other options were gifts of free internet connections (56%), activity trackers (55%), and smartphones and data plans (52%). The results varied by demographics with younger people and those of lower socio-economic status being more interested in gifts of technology (Kaufman et al., 2016). The table below presents some examples of incentives that have been used in genetics and genomics research, as well as in other research studies.

Examples of incentives offered in research with diverse ancestry groups

Project/org	**Participants/ communities**	**Incentives**
Genetics and genomic research		
All of Us (US)	Diverse ancestry groups	Option to receive individual results including WGS data (Crawford et al., 2019); opportunity to participate as 'citizen scientists' and be involved in oversight, design, implementation, evaluation (National Institutes of Health (NIH), 2018)
Genes and Health (UK)	Bangladeshi and Pakistani communities	Community benefits through prioritising research on conditions that community members were concerned about: diabetes, cardiovascular disease, dementia and mental health (Finer et al., 2020)
VariantBio (US)	Diverse ancestry groups	Benefit sharing through 4% yearly revenue donation to organisation/initiative of community's choice until IPO complete, followed by one-time distribution of net proceeds (Variant Bio, n.d.)
Genetic testing to Understand and Address Renal Disease Disparities (GUARDD) Trial (US)	People of African ancestry	$40 gift cards per visit for time spent (Horowitz et al., 2019)
Other research		
DC-Healthy Outcomes of Pregnancy Education (DC-HOPE) (US)	Low-income African American and Latina women	Cash incentives at different stages: $5 for screening; a 30-minute telephone card for consenting; $15 per telephone interview. Intervention group: $10 per intervention session; $15 and $25 gift vouchers for 2 postpartum sessions. (El-Khorazaty et al., 2007)
Group or Individual Diabetes Self Management Education (DSME) study (US)	Rural African Americans with Type 2 diabetes	Gifts, door prizes, and other incentives including: pedometers, light refreshments, diabetic cookbooks, foot care products; exercise videotapes; certificate of completion at end of intervention; $25 compensation to ensure continued attendance at each session. (Loftin et al., 2005)
SeniorWISE study (US)	Hispanic, African American, European older adults	$180 for attending five training and testing sessions over two years (McDougall et al., 2015)

A structure on ongoing incentivisation is of particular importance with disadvantaged groups, and especially when ongoing engagement and retention are necessary for the success of the project (Bonevski et al., 2014). While research on the use of incentives in clinical trials has found monetary incentives to be the most effective in the general US population, interestingly, a small number of studies that have compared lottery-based rewards to definite payments have found no difference in impacts on recruitment and retention (Parkinson et al., 2019).

Benefit-sharing

Benefit-sharing models for genetics and genomics research are sometimes recommended to address the public's concerns about commercialisation. Acknowledging the important role of commercial and pharmaceutical companies in translating the outcomes of genetics research into tangible, therapeutic realities, Haddow et al. (2007) argue that the benefit-sharing model is a way of addressing concerns about the exploitation of the public for private gain.

Focus groups with ethnically diverse groups in the US found lower levels of trust in research for profit (Griffith et al., 2020). Benefit-sharing is thought to be especially important with groups that have low levels of trust in research, and for whom commercialisation is likely to be a barrier to participation (Buseh et al., 2013; Halbert et al., 2016; Sanderson et al., 2013). Based on their consultations with African migrant community leaders in the US as part of a community-based participatory research study, Buseh et al. (2013) recommend benefit-sharing as a model for addressing high levels of mistrust among the African immigrant population in the US, stemming from the history of colonialism in participants' countries of origin. In Buseh et al.'s consultations with community leaders, research that was not seen to be clearly benefiting the community was considered exploitative, and hypothetical research where benefits were shared were thought to be likely to facilitate participation.

Variant Bio, a biotech startup based in Seattle, employs a benefit-sharing model in its work with diverse ancestral groups. A key tenet of Variant Bio's ethical framework is 'Benefit – providing a research process that maximizes the potential benefits and minimizes potential harms for participating individuals and communities." (Variant Bio, n.d.) An incentive plan is developed with communities that meets communities' expectations, needs, and priorities. For example, Variant Bio contributed to the SING Consortium in New Zealand to build Indigenous capacity in genomics (Variant Bio, 2021).

On the other hand, other projects have been explicit about there being no benefit beyond the expected benefit to the community, framing participation in the language of a disinterested, altruistic gift. Genes & Health, the UK population genomics and health study of people of Bangladeshi and Pakistani descent, refers to participants as 'volunteers' and explicitly states in its volunteer information sheet that volunteers will receive no compensation for any discoveries made:

What are the benefits of joining East London Genes & Health?

There will be no direct benefit to you by joining (at Stage 1) but you will be making a contribution to science and future improvements in NHS care. The information we will gain from studies using East London Genes & Health will help lead to a better understanding of the links between genes and environmental factors in causing disease and will contribute to improving healthcare and the long term prevention and treatment of a number of common diseases.

The benefits to the Bangladeshi and Pakistani communities will include better understanding and treatments for disease, for example diabetes and heart disease. We will also understand more about how genes work, which will help the development of new treatments. We will understand better the normal variation in Bangladeshi and Pakistani genomes, which will help the diagnosis of inherited rare diseases

...

What happens if a discovery is made using the donated sample?

The samples donated to East London Genes & Health are given as an "absolute and non-returnable gift". For example if results from the research undertaken with the donated samples are used to develop a new test to improve diagnosis or better medicines for treatment, then you will not receive any compensation nor will funds be forthcoming to you. East London Genes & Health will work in partnership and share your samples and data in anonymous format with others in the public (e.g. Universities) and the private sector (e.g. pharmaceutical or biotechnology industry, who may be UK based or overseas) to develop any discoveries for the benefit of patients.

In research with Indian communities in Houston, conducted to inform the collection of samples from Gujaratis for the HapMap project, Reddy (2007) found that the donation of blood for research was thought about in the first instance in terms of *dāna*, disinterested giving in service of the greater good. In her ethnographic work, Reddy was surprised to find outright commitment to this abstract promised good of genomic research. The concept of *dāna* as a particular kind of gift where there is no expectation of return, is distinct from the usual way in which gifts are framed across different cultures, and indeed from different modes of gifting within South Asian cultures. In other contexts, appealing to the logic of gifting to encourage donations, implies engaging the giver and receiver in a reciprocal relationship involving mutual exchange (Mauss, 2002). Because of the relational nature of gift giving and receiving and the obligations associated with gifts, in some contexts, the concept of altruistic giving may only make sense when applied to kin, as researchers studying the use of the language of gifting in blood donation have found (Simpson, 2014; Wittock et al., 2021). Thus, while the approach taken by Genes & Health in the UK may be

suitable in South Asian communities where there is a conceptualisation of a de-personalised, altruistic gift (Reddy, 2007), it may be less appropriate in other contexts.

Return of individual results

In genomics, return of individual results can be a way of providing direct benefit to participants (Crawford et al., 2019; Fisher et al., 2020; Lewis et al., 2021). Lewis et al. (2021) suggest that return of results is an important way of engaging underserved groups such as people of African descent. The All of Us project uses return of results as an incentive for people to take part, citing evidence from US studies that indicate that the general population have an interest in receiving genetic results (Crawford et al., 2019), including raw data (Middleton et al., 2015). While not returning results has been found to be a disincentive to taking part (Fisher et al., 2020), the return of results comes with substantial logistical and ethical challenges which are beyond the scope of this document (Crawford et al., 2019; Jarvik et al., 2014).

A survey of the general US population conducted for the All of Us project found that participants were most interested in receiving laboratory results such as blood sugar or cholesterol levels, and 74% were interested in receiving genetic results.

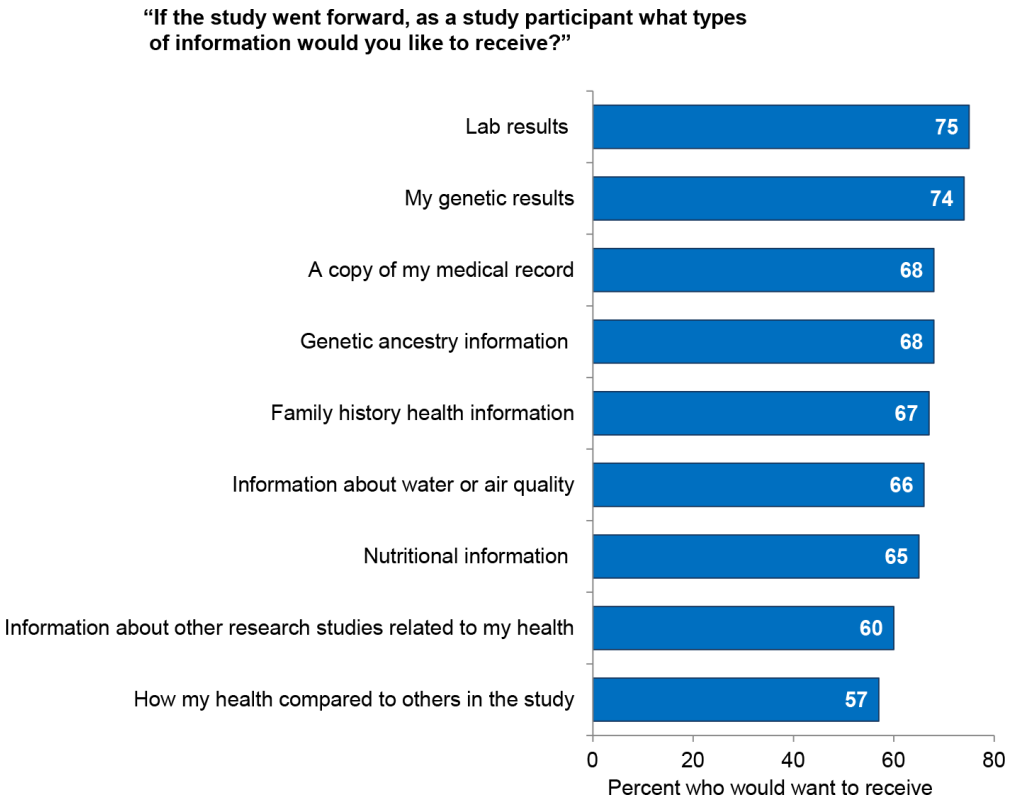


Table from Kaufman et al. 2016.

A challenge with using return of results as an incentive is deciding what results to return. In a study comparing African American and non-African Americans attitudes to the kinds of results that were returned in exome and whole genome sequencing studies, Yu et al. (2013) found that African Americans were less likely to be interested in results that could be

relevant to their health in the future. Participants were more concerned about information that would be helpful in understanding their or their family members' present health. Yu and colleagues argue that this present-orientation can be at odds with much genomic research which is future-orientated, and that communities must be consulted ahead of time about the kind of results they would like to receive.

In another study on the type of results people of Black or African American descent were interested in receiving, Lewis et al. found that receiving information about one's genetic ancestry was a low priority (Lewis et al., 2021). This is in contrast to the 68% of participants in Kaufman et al.'s (2016) study who were interested in learning about their genetic ancestry. (Kaufman et al.'s study consisted of a weighted percentage of 66% non-Hispanic White participants).

Conducting focus group discussions with self-identified Black and African American participants in the ClinSeq project, Lewis et al. (2021) asked participants about the kinds of results they were most interested in receiving and participants first ranked their choices individually. Participants then engaged in a process of consensus ranking of their priorities for return of results in which they were able to discuss and change their initial individual decisions. Information on life threatening diseases was ranked most highly, followed by information relating to mental health, physical health, then treatable conditions. Despite other research indicating that people want all results returned, Lewis et al. found that participants were open to the prospect of some results being withheld, with the group ranking for preventable conditions dropping significantly following discussion. The authors suggest that in the case of genomics, people are often forming an initial opinion based on information that is new to them, and that with discussion and deeper understanding of the complexities of return of results, attitudes may shift (Lewis et al., 2021).

Australian attitudes to return of research results

Vears et al. (2021) have found through a systematic review of 221 international studies (including 5 Australian) that stakeholders are overwhelmingly in favour of the return of individual results from genomics research. Thus far, there has been one Australian study that gauged the attitudes of the general Australian population (as opposed to patient populations and their families). The general Australian population appears to be well disposed to receiving individual results from research. Fleming et al. (2015) conducted a nationwide telephone survey of a representative sample of Australians, where 87% percent of the sample self-identified as Australian. Participants were asked what sort of information they would want to receive if they donated tissue for research. The overwhelming majority of participants (94.4%) wanted to receive results that were relevant to their health and treatment; 83.4% also wanted information of their genetic risk of inherited disease. Fewer, though still a majority (70%), wanted other incidental findings not related to a potential diagnosed condition. The authors note that these results are consistent with the data on the attitudes of the general population in Scandinavia, the US, and the UK.

Rasmussen et al. (2018) similarly found high levels of interest in research results amongst Australians, in their study comparing Australian and French attitudes to receiving incidental findings for the International Sarcoma Kindred Study (ISKS). Participants were presented with four scenarios, one where prevention or treatment was not possible, and three involving

conditions of varying severity in which prevention, risk reduction, or treatment were possible. Preference for receiving incidental findings was higher amongst Australians across all scenarios with between 68 and 94% wanting findings returned. The authors conclude that interest in receiving incidental findings is likely to be high in Australia.

To date, there do not appear to be studies specifically addressing the attitudes of culturally and linguistically diverse Australians to individual research results though research has been conducted on attitudes to carrier testing amongst culturally diverse consanguineous couples. Josephi-Taylor et al. (2019) conducted interviews with twenty couples attending a genetic clinic at Liverpool hospital in Sydney. Participants included 7 Iraqi, 6 Lebanese, 2 Syrian, 1 each Kenyan, Sudanese, Indian, Pakistani, Bangladeshi, and Iranian couples. Nine couples from Lebanon, Iraq, Syria, and Iran referred to routine premarital screening programs in their countries of origin. Two-thirds of the couples from a range of religious backgrounds said they would use information from carrier testing in making reproductive choices.

6.5 Implications for CPG's reference database project

This overview of the literature indicates that future benefits to the community may be motivators for participation, but that more tangible, direct benefits may be helpful in supporting greater recruitment and ongoing engagement. Based on reviews of the use of incentives in clinical trials and literature on the economics of incentives, Parkinson et al. (2019) make a number of recommendations for the design of incentive structures that will be useful for considering for CPG's reference database:

- **Design context-specific incentive structures based on an understanding of the barriers that incentives may need to be mobilised to overcome.**

  In the case of the CPG's work with diverse communities, these barriers might include different levels of trust in research that could have an impact on the extent to which a promised, future benefit to the community may serve as a motivating force. They might include the perceived burden of donating venous blood, for example, or could relate to the relative overall economic disadvantage of particular communities.

- **Consider what incentives already exist in any given community**

  It is possible that in some communities that CPG engages with, there could be a strong collectivist culture with an emphasis on altruism that may already provide a strong incentive for community members to participate. Research has found that in such contexts, additional incentives, over and above reimbursement, can have a de-motivating effect (Wenemark et al., 2010). Monetary incentives in particular may undermine intrinsic motivation by setting up a market relation between participants and researchers (Burgess and Ratto, 2003). The NHMRC guidelines recommend that decisions about whether or not to provide incentives should take into account whether participants are patients who could potentially benefit from treatment, family members of patients, or are healthy participants who do not stand to gain any direct benefit (National Health and Medical Research Council (NHMRC), 2019).

The literature indicates that information can be a way of providing direct, individual benefit to participants, and one that has been shown to have resonance amongst ethnic minority

communities in the US. This may mean that emphasising that clinically actionable results will be returned may be perceived as a benefit to participation in some groups. It is also possible that in CPG's work with different communities, providing ongoing information, whether education about genomics or regular updates about the project, may serve as an incentive and be an important way of maintaining community engagement.

In assessing whether providing incentives will be necessary in any given community, it will be important for CPG to understand whether appeals to altruism for the benefit of the community are likely to be successful. This will involve coming to an understanding of the conceptualisation of gift-giving in the different communities. Finally, it will be important to understand how community members perceive benefit to themselves and their communities, how this may change over time through CPG's engagement with them, and how CPG's engagement strategies can be designed to support the recognition of benefit in participating in the reference database project.

7. Community-based participatory research and engagement

Key points from literature:

- Public and patient involvement in research is increasingly encouraged by funders and publishers to ensure relevance of research and efficacy of interventions
- Participatory methodologies have particularly been used with marginalised groups to address power imbalances that have historically led to harm by researchers
- A fundamental principle of such approaches is that participants are experts in their own lives and in the issues that affect them
- Approaches run along a continuum of levels of participant involvement
- Co-design is a type of participatory method that involves end users in the design of solutions to problems
- Co-design tools are aimed at supporting a diverse group of people to make meaningful contributions
- These tools can facilitate access to aspects of users' experience that the traditional research interview cannot

Recommendations for CPG:

- Adopt a co-design approach to development of CPG's reference database project
- Consider what needs to be known at each stage of the research process and consider the methods and tools that can best access this information

7.1 Why is a participatory approach important for CPG's reference database project?

In the last decade, there has been a growing push for greater public and patient involvement in research, including in genomics (Nunn et al., 2019, 2021). Some degree of public involvement is increasingly required by funding bodies and academic publishers (Greenhalgh et al., 2019; Wicks et al., 2018). This can range from the establishment of project advisory boards that include patients or consumers, up to and including co-design (Slattery et al., 2020). Public participation is seen to be an important way of ensuring the relevance of the research to the groups being researched and improving the efficacy of interventions (Edelman and Barron, 2016), as well as boosting study recruitment (Crocker et al., 2018).

Participatory methodologies have a long history in the public health and community sectors, and are often used with disadvantaged and marginalised communities. Having their roots in emancipatory philosophies seeking to empower marginalised communities (Freire, 2014), their methods seek to address power imbalances between researchers and participants that have historically resulted in harm to communities (Chambers, 1994, 2007). Participatory approaches are thus especially relevant in medical research with ethnic minority groups. Adopting a participatory paradigm in medical research is considered to be an important way of building trust with communities and of increasing the representation of diverse groups in research (Holzer et al., 2014). In genomics, participatory approaches have been crucial for

work with Indigenous communities where there are often high levels of mistrust of researchers (Garrison et al., 2019).

The variety of approaches that fall under participatory research share an emphasis on partnership with communities, and employ methods that facilitate power-sharing (Israel et al., 2012). Fundamental to participatory approaches in research is the recognition of the knowledge and expertise of members of the community. Non-academics, being experts in their own lives and the issues that affect them, are considered to have an equally important role in the production of knowledge as the researchers involved.

Participatory research occurs along a continuum with traditional, fully expert-led research sitting at one end, towards research that gives an increasingly active role to participants (Cornwall and Jewkes, 1995). At the other end of the continuum from traditional research approaches are community owned and led models where decision-making responsibilities reside fully with communities. (See Figs 1 & 2 for more detail on different degrees of engagement with stakeholders).

While co-design sits at the more participatory end of this continuum, it runs the gamut between forms where traditional researcher/participant distinctions are maintained, to more radical forms that significantly disrupt the traditional dynamic between expert and layperson (Burkett, 2012). Where a project sits on this continuum will depend on a number of factors. For example, community ownership of research is of particular importance in work with Aboriginal and Torres Straits Islander peoples in Australia (Claw et al., 2018; Prictor et al., 2020). In other situations, the purpose for which funding has been obtained may limit the degree to which community members can decide on what will be researched.

Power-sharing can be uncomfortable for researchers as it involves relinquishing some degree of control over the research process. In their review of self-identified participatory research in biomedicine, Kelty and Panofsky (2014) found that researchers were comfortable with some participatory elements, but less so with others. Incorporating education was a common participatory element in most studies, while involving participants in agenda-setting was less popular. The authors argue that while power-sharing has its risks, there are also opportunities for innovation in allowing a more active role for research participants. They cite the role of patient participation in the discovery of the genetic mutation responsible for PXE (pseudoxanthoma elasticum) as an example of how public and patient involvement can benefit research.

Fig 1. Levels of stakeholder engagement (Credit: Ken Knight, Understanding Co-Design seminar, MCRI, 21 April 2021)

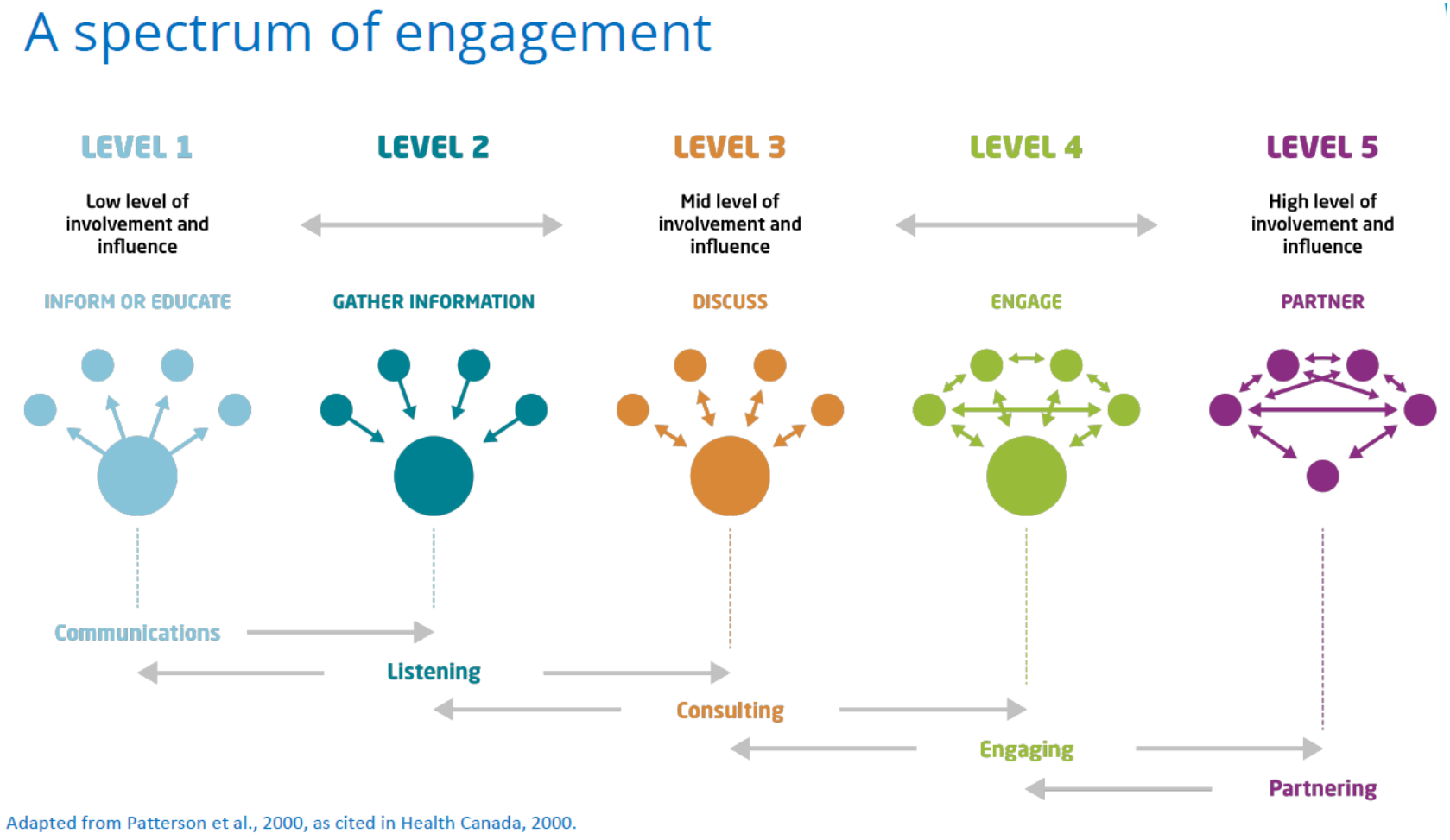


Fig 2: Spectrum of stakeholder engagement (Credit: Ken Knight, Understanding Co-Design seminar, MCRI, 21 April 2021)

SPECTRUM OF STAKEHOLDER ENGAGEMENT	Inform	Consult	Involve	Collaborate	Empower
	Information giving	*Information seeking*	*Information sharing and joint planning*	*Participatory decision making*	*Stakeholder leadership*
Purpose of Engaging Stakeholders	To provide information to consumers and stakeholders to assist them in understanding issues, alternatives and/or solutions.	To gather information from consumers, communities and stakeholders, including to capture lived experiences.	To involve communities and stakeholders in research, planning, policy development, delivery and evaluation of services.	To work in partnership with communities and stakeholders.	To place final decision-making in the hands of stakeholders.
When to Use	One-way exchange of information, e.g. to assist in access to and management of health issues.	Two-way exchange of information; share views, needs and interests.	To ensure community and stakeholder views are considered and reflected in decisions and outcomes.	To jointly develop solutions and initiatives.	To enable communities and stakeholders to decide and implement solutions/ outcomes (often with specified guidelines).
Commitment to Stakeholders	We will keep you informed.	We will keep you informed, listen to you, acknowledge your views and provide feedback.	We will work with you, consider your views and provide feedback on how your input influenced the outcomes.	We will look to you for advice and innovation in the formulation of solutions and incorporate your advice to the maximum extent.	We will implement your decisions and support and complement your actions.
Methods of Engagement	Including: • Public notices • Announcements • Websites • Fact sheets • Newsletters • Education programs.	Including: • Public meetings • Public hearings • Focus groups (e.g. CALD and communities in need) • Surveys.	Including: • Workshops • Forums • Deliberative polling • Panels • Task forces • Working parties.	Including: • Reference groups • Facilitated consensus building forums • Advisory committees • Policy round tables • Clinical networks.	Including: • Steering committees • Quality committees • Boards • Participatory governance • Standing strategic committees.
Level of Stakeholder Influence	Minimal influence	Low influence	Moderate influence	High involvement and influence	Community/Stakeholder control

Adapted from IAP2, 2014.

7.2 Examples of community-based approaches taken in genomics research

Project/organisation	Participants/ communities	Approach
The Alabama Genomic Health Initiative (AGHI) (May et al., 2020)	African Americans	• Advisory board of community representatives • Ran town hall meetings and outreach events • Use of democratic deliberation methods for decision making through facilitated deliberative process group meeting • Participants in the group meetings were provided with education to get them up to speed on the issues up for the discussion • Participants were able to ask questions of researchers and engage in open discussion with the group. • Participants (both community members and researchers) voted on aspects of the project design such as strategies for recruitment, the kinds of results to be returned following genome sequencing, and how results should be returned
All of Us (US) (National Institutes of Health (NIH), 2018)	Diverse ancestry groups	• Participant representatives on various committees and panels. Participants apply to be representatives for a two-year term. They are involved in all aspects of the research including design, operations, communications, and governance. • 'Citizen scientists' are involved in data collection and other research processes. • Network of participating community organisations, supporting community-specific engagement, recruitment, and retention.
GTEx (Genotype Tissue Expression) initiative (Mosavel et	Hispanic Americans	• Consulted community leaders to develop information sessions about genomics to recruit advisory board

al., 2019)		members with no prior knowledge of genomic biobanking; ● Community leaders were present at information sessions to answer questions pertaining to their area of expertise (e.g. religious leaders) ● Advisory board volunteers met every 2 months for 10 months, initially focused on building relationships between board members and researchers, and getting members up to speed on genomic biobanking ● Later meetings involved the development of consenting and education materials and a walkthrough exhibition for the broader community ● Advisory board members were provided with funding to host a social event with family and friends to discuss the GTEx project. ● Discussions were recorded, analysed by researchers, and used to refine materials ● Advisory board members served as hosts at walkthrough exhibitions and were on hand to answer the public's questions.
Heart Healthy Lenoir Genomics Study (Skinner et al., 2015)	African American	● Engaged community members to help develop consent documents and advise on recruitment processes ● Sought community guidance on interview schedule for focus group discussions ● Focus group facilitators were assisted by community members
West Side Hmong Genomics Board project (Culhane-Pera, Straka et al., 2017)	Hmong Americans	Established a Hmong Genomics Board including representatives from the Hmong community. Board members were involved in: ● Decisions on genomic questions that reflected the community's health concerns ● Appropriate informed consent approach that provided two streams for those without high school education (in Hmong language), those with high school education (in English, with Hmong as needed) ● Consent forms in English and Hmong written at 5th-grade level;

		Hmong/English form for those providing verbal consent ● Design of communication materials capitalising on scientifically accurate community understandings of heredity
Wisconsin Genomic Initiative (Buseh et al., 2014, 2017)	US African immigrant communities	● Partnered with a pan-African community organisation to conduct research into African immigrant communities attitudes to genomic research ● Community members involved in recruitment, data collection, analysis, and dissemination ● Conducted focus groups with community leaders and gatekeepers about perspectives on genomic biobanking ● Visited community leaders' constituencies and conducted in-depth interviews with community members ● Conducted population-based survey on knowledge, attitudes and beliefs about genomics

7.3 Existing frameworks for inclusion of ethnic minorities in research

Authors/Purpose	**Key elements of framework**
Woodland et al. 2021 Targeted research with CALD Australians	● Ensuring team is culturally competent and research is culturally informed ● Conducting research on community needs and health inequities ● Recognising difference in power between researchers and communities and rebalancing this ● Maximising the actionability of research ● Attending to safety of community (e.g. around the way results are treated)
Sharif et al. 2020 Competence framework for genomic research with ethnically diverse participants	Framework covers cultural competence, genomic competence, and research competence[5]. Core elements of cultural competence in genomic research Sharif et al. identify are: ● Striving for equity in representation of ethnic minority groups in genomic research:

[5] We expand only on the authors' recommendations for cultural competence

(developed through work with British Pakistanis)	○ Understand reasons for underrepresentation ○ Appreciate need for diversity in sampling ○ Involve diverse groups in development of protocol ○ Be aware of sensitivities relating to consent, storage, and dissemination, especially around unknown/unreported consanguinity, impact of info on family etc. ● Putting in place measures to overcome barriers to ethnic minorities' participation in genomic research ○ Understand community needs and preferences (e.g. cultural practices, language, beliefs etc.) ○ Consult healthcare providers working with communities who can advise on approach ○ Hire bilingual staff ○ Provide cultural competency training where needed ● Suitable communication ○ Provide translation if needed ○ Cater to different levels of literacy ● Demonstrate understanding of culture ○ Be aware of own values and beliefs ○ Recognise sociocultural factors involved in decision-making (e.g. family hierarchies) but support individual participant decision-making autonomy ○ Be respectful of practices (e.g. consanguineous marriage)
Claw et al. 2018 Genomics research with Indigenous communities	● Show respect for Indigenous data sovereignty and work with tribal governance structures ● Collaborate with communities in conducting genomic research ● Ensure cultural competency of research team ● Be transparent about the research ● Build the research capacity of community ● Disseminate findings to communities in an accessible form

7.3 What is co-design?

'Co-design' or 'collaborative design' is a type of participatory design methodology that involves end-users in coming up with solutions to problems. It is used increasingly in the health and public sectors to develop services, processes, systems, and materials that are relevant to the people who will be affected by them. Co-design involves a commitment to designing solutions *with* people as opposed to *for* them (Sanders, 2002).

The term 'co-design' has become a bit of a buzzword in the public sector and is sometimes used incorrectly to refer to any kind of consultative, collaborative process. This captures the

very important ‘co’ aspect, but the ‘design’ part is equally important. Genuine co-design is ‘design-led’ (Blomkamp, 2018). It draws on concepts from design thinking that foster creativity in developing real-world solutions to problems. Design thinking provides a structure that supports innovation and working towards concrete outcomes.

While design thinking is ‘human-centred’ and emphasises the needs of the end-user, it is not necessarily participatory. The early stages of a human-centred design process seek to understand end-users' experiences and points of view, but the subsequent steps are left to designers or researchers. In contrast, co-design brings stakeholders into the design process. End-users (or clients, customers, patients, and research participants, depending on the context) work alongside designers and researchers throughout.

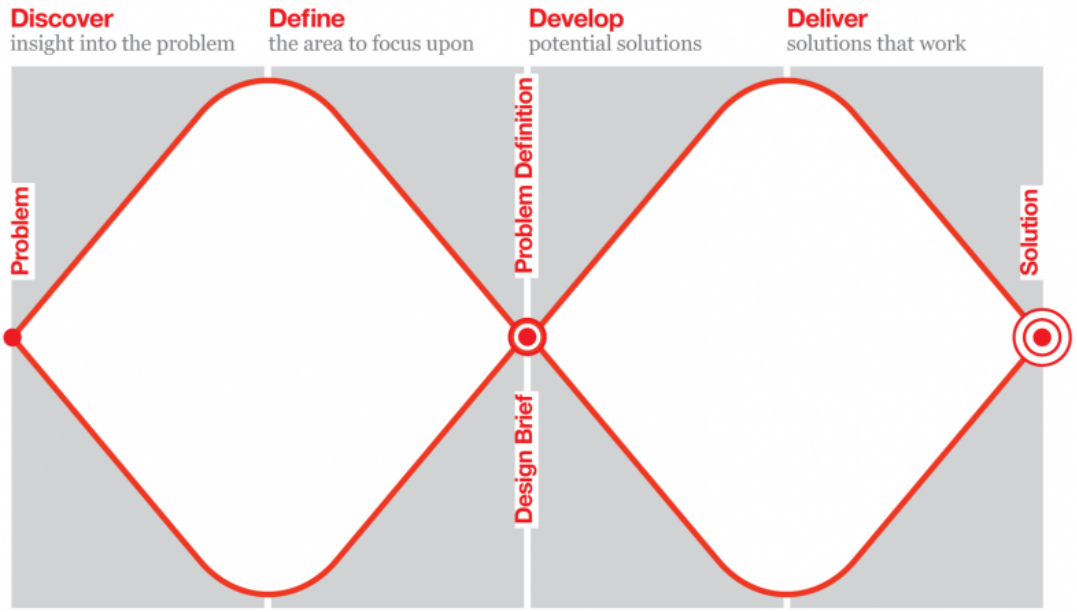
What is involved in co-design?

To understand what is distinctive about co-design, co-design researcher Emma Blomkamp (2018) recommends thinking about it in terms of its three components: process, principles, and practical tools.

Process

There are a number of frameworks that outline the design process. The most commonly cited framework is the UK Design Council’s double diamond model (Design Council 2022). The model depicts stages of the design process allowing for more exploratory work (divergence) followed by phases requiring focus (convergence). The stages in this framework outline an approach to problems and solutions. These are: discovery, defining, developing, and delivering (See Fig. 3). Another popular framework is the Stanford Institute of Design’s (d.school) model of the design thinking process which outlines five stages: empathise, define, ideate, prototype, and test (Standford d.school, n.d.).

Fig 3: UK Design Council’s Double Diamond Model (Credit: Adapted by Lipiec, 2019)



Tools

Co-design is often carried out through co-design workshops making use of a range of tools. A key characteristic of these tools are that they are designed to be inclusive, and are usually visual and tangible (Blomkamp, 2018; Burkett, 2012). Co-design tools allow designers and researchers to support a group of people from a broad range of backgrounds to access their own expertise and participate meaningfully in the design process (Sanders and Stappers, 2008). Some of these tools include collages, storyboards, roleplay, paper prototypes, games, maps, mockups, many of these involving the ubiquitous colourful Post-It note! These tools engage participants in what Brandt et. al. (2012) identify as the three key activities of co-design: telling, enacting, and making. Each of these activities helps participants to access relevant aspects of their experience that are not easily accessed with more traditional research methods such as interviews and surveys.

8. CPG's Approach to an Inclusive Genomics

Key points:

- CPG's approach to an inclusive genomics will be ethnographic, participatory, and design orientated
- Through such an approach, we aim to:
    - Privilege the perspectives and meanings of the communities we work with
    - Draw on community strengths and work with communities to develop an approach that works for them
    - Actively produce solutions to the challenge of recruiting large numbers of participants from Australia's minority ancestry groups
- Co-design will be a central feature of this work and we will be guided by its principles of being outcomes focussed, participative, inclusive, respectful, and iterative
- The reference database will eventually include all Australians currently not represented in genomic database though in-depth co-design will begin in a small number of priority groups

To achieve CPG's vision of a more inclusive future for genomic medicine in Australia, one of four goals of the CPG 2021-22 Strategic Plan is "Community partnerships - to collaborate with communities to co-design genomic health projects." By communities, we mean groups of people who perceive themselves to have a specific, shared ancestry.

8.1 Overarching approach to research and engagement

- **Ethnographic -** *This means we seek to understand a community we work with from the perspectives of its members.*

  Our work with communities will seek to understand the cultural worlds of communities in relation to genomics generally and to CPG's project in particular. Ethnography privileges the perspectives and meanings of community members, and seeks to understand these within the context of people's lives. In the case of CPG's reference database project, the context we seek to understand necessarily involves our own intervention in the community i.e. the introduction of the reference database project to communities and the promotion of ideas about the community's stake in the genomic medicine of the future. Our ethnographic approach will seek to understand what this means for communities and what matters most to them in CPG's call for their participation. Practically, this approach will inform culturally salient communication with participants and guide genuine collaborative design of participation pathways.

- **Participatory -** *We will involve community members throughout the process of developing pathways for community members to be part of the reference database*

  We draw on elements from community-based participatory research (CBPR) to inform an approach that brings together research with community engagement and

partnership building. A key principle of CBPR is that it draws on community strengths and assets (Israel et al., 1998). Our initial research with communities will identify the community leaders and structures that will facilitate CPG's work, to form partnerships between CPG and the community, as well as relevant external stakeholders who are able to support work with particular communities. These relationships will facilitate the recruitment of large numbers of community members into CPG's reference database.

- **Design-oriented -** *Our research and engagement with communities is focused on action. It is explicitly directed towards developing the solutions that will support large numbers of community members contributing samples to the reference database.*

  Co-design is a key component of how the IG team will work with communities to design pathways to involvement that work for community members. Such an approach is different from traditional ethnographic research that seeks to collect information about people fixed at a point in time; instead, a design-oriented approach is dynamic and brings researchers and their participants together in a collective act of future-making (Gatt and Ingold, 2013). Through co-design, we will actively produce solutions with communities to recruit their members, rather than merely collecting data that will inform design at a later stage.

8.2 Principles of co-design that will guide CPG's work with communities

The central feature of co-design are the participatory design philosophies that underpin it (Blomkamp, 2018). There are a number of documents that outline principles for co-design. These include work by the NSW Council of Social Services which has been further elaborated by Blomkamp, as well as those set out in the WA Council of Social Services toolkit (WACOSS, n.d). We expand on these here in relation to CPG's reference database project to consider what these principles mean for IG's co-design work with communities.

1. **Outcomes focussed**
   The goal of our co-design is to work towards greater health equity for Australia's multicultural population by ensuring that all communities are able to benefit from the genomic medicine of the future. The path towards getting to this outcome may look different in each community.

2. **Participative**
   The IG team aims to support genuine participation of community members. This means drawing on the strengths of communities. It will also require us to provide culturally appropriate education about the reference database project that will allow community members to meaningfully engage in co-design and decision-making.

3. **Inclusive**
   We recognise the diversity within each community and seek to draw on a range of knowledge and expertise in the co-design process. We aim to provide space for all

participants to feel comfortable and able to contribute.

4. **Respectful**
   Co-design aims for equal partnership with participants. We respect and value the expertise and contribution of all our partners equally.

5. **Iterative**
   Our co-design work with communities will not be linear. Co-design is a process of trial and error. It involves testing, evaluation, and fine-tuning to produce innovative solutions to problems.

8.3 What happens before the co-design starts?

As explained in CPG's strategic plan, before we begin the co-design process, we are prioritising communities for our initial focus. We are a small organisation and cannot work with dozens of different ancestry groups simultaneously, making prioritisation essential. However, we do not intend to leave out any group and expect that over time the reference database will grow to include participants from all of Australia's ancestry groups.

Prioritisation involves two steps. First, we are collecting information (via interviews, a survey and data analysis) regarding some important characteristics of ancestry groups. This will allow us to short-list 5-6 ancestry groups. Second, we will conduct "market research" to understand other characteristics of the short-listed groups, such as the geographic locations of major populations, languages spoken, religious and cultural gathering places, and so on. This will help us to then select 2-3 ancestry groups for our initial engagements.

Note that we do not expect to be working directly in Aboriginal or Torres Strait Islander or Maori communities. There are a number of Indigenous researchers in genomics already doing so and we intend to collaborate with them by invitation. We expect to be working with other Oceanian peoples and with culturally and linguistically diverse (CALD) communities that include many first and second generation immigrants to Australia.

Before we begin co-design work in community, we will develop a research protocol and obtain HREC approval for the community work. Other key components that will start before the co-design but will also be informed iteratively by our community engagement include:

- Communications – creating content and materials for communicating with communities
- Consenting process – plan and build consent mechanisms, including CTRL redevelopment
- Languages - identify language needs and contract NAATI-certified translators and interpreters
- Community engagement staff - recruit 1 to 2 staff (or consultants) with appropriate language and lived experience for each community

8.4 Will all our participants be recruited via a co-design approach?

There are dozens of ancestry groups in the Australian population and for our database to be truly useful we will need tens of thousands of individuals' samples. We cannot plan for detailed co-design across every ancestry group and are therefore exploring additional ways to recruit participants, such as collaborating with consulting groups working in CALD communities and contacting participants enrolled via other research platforms.

However, there is a lot of evidence that simply providing information about an initiative and how to participate is inadequate to achieving broad engagement across diverse communities. Recent campaigns by state governments regarding COVID, for example, found that information was not being read or used by CALD communities for a variety of reasons that had to do with cultural norms and behaviours. The kind of deep engagement that co-design processes offer will provide us with stronger connections and better two-way communication based on a deeper understanding of the cultures. This will improve our ability to explain genomics and the reference database to our communities, and therefore increase the number of people who decide they want to participate. Remember the problem that we are solving for: that significant numbers of individuals from the diverse communities referred to above on page 1 understand the purpose of the reference database and choose to participate. We therefore hope to apply the co-design principles as much as possible in all the ways we engage with participants for the reference database.

9. Conclusion

In this document, we have reviewed the literature on aspects of genomic research design that must be tailored to meet the needs of minority ancestry groups if their underrepresentation in genomic databases is to be addressed. We explored how a community-based participatory approach to recruitment can help to avoid recreating barriers that research-as-usual sets up for multicultural groups. Finally, we have outlined a framework for CPG's research and engagement work aimed at developing pathways that facilitate the participation of large numbers of community members in genomic research.

CPG will address the first barrier to participation identified at the start of this document, that ethnic minorities often do not hear about research and are not explicitly asked (Kim and Milliken, 2019), through targeted work inviting specific communities to be part of our reference database project. This work will seek to lower barriers and facilitate participation by first gaining an in-depth understanding of the needs of the community in taking part in the project. This will occur through a recognition of, and respect for, differences in community norms, knowledge, beliefs, language, literacy, motivations, fears, and concerns. Next, through co-design with communities, we will build on these understandings, drawing on the expertise of key community members to develop pathways to participation that meet these needs.

Some aspects of the reference database project design will be fixed prior to our engagement work and have limited amenability to co-design. Components of the project including method of biospecimen collection, a basic incentive structure, content of consent documents, and design of our online portal will, to some extent, need to be standardised across communities. The design of these components will be considered in light of the literature presented in this document to ensure that they are developed in a way that is as inclusive as possible. Particularly in the case of biospecimen collection and the consenting process, CPG will seek to understand what barriers, if any, these standard components may present for community members. We will work with communities to develop relevant supports and workarounds that address these barriers, recognising that any one community will itself be composed of a diverse membership.

Through an ethnographic, participatory, and design-centred approach that emphasises understanding, collaboration and action, CPG is working towards developing a methodology that will tackle the current challenge of the underrepresentation of minority ancestry groups in genomic databases and genomic research generally. This work will lay the groundwork for an inclusive genomics that secures a future where all Australians will be able to enjoy the benefit of developments in genomic medicine.

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