



Beyond One Million Genomes

# D1.6

## Citizen engagement and public trust in genomic data sharing.

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03/05/2023	0v3	Carla Van El (VUMC), Eric Vermeulen (VSOP)	Final edits including incorporating comments and changes from stakeholders.
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# 1. Executive Summary

The '1+ Million Genomes' (1+MG) initiative, coordinated by the Beyond 1 Million Genomes (B1MG) project, aims to enable secure access to genomic and corresponding clinical data across Europe for research, personalised healthcare, and policy making. It is a joint initiative of 24 EU countries, the UK, and Norway. Citizen engagement and public trust have been raised as critical components in the B1MG stakeholder forum meetings and country visits. To discuss these components, an online expert workshop was facilitated on January 18, 2023.

14 Participants were invited to give a short presentation on their lessons learned regarding citizen engagement. They were selected for their expertise in bioethics, ELSI, governance, human genetics, patient representation and citizen engagement. After discussion they formulated recommendations to participating 1+MG countries.

Lessons learned from various engagement activities were shared under three subthemes: “When an ELSI framework is in place, what is the (additional) role of citizen engagement in fostering data sharing and public trust?”; “How does citizen engagement relate to (interests of) other stakeholders?”; “When and how to engage and at what level?”.

It was discussed that citizen engagement can have a diversity of aims that may help create preconditions for accountable research and trust. ELSI or trustworthiness frameworks don't necessarily lead to participants having trust in an institution. The addition of citizen engagement can help build and contribute to trust. Engagement activities are situated in context with potentially conflicting interests, power differences and priorities, for instance between publicly accountable researchers and private companies. While data sharing involves international collaboration, trust is relational and context specific and dependent on many factors including the relation to the professionals or institutions asking for data sharing and trust. The tools used for citizen engagement vary for different contexts and goals. If engagement starts at the beginning of a project, there are more options to influence choices and policies.

Recommendations (See D1.7) stress the need for dedicated resources; acknowledging different views and interests; the need for capacity building to enable engagement; creating meaningful participation via selected tools through early engagement with a careful choice of engagement strategy; having good and transparent governance for enabling trustworthiness to contribute to accountability; and the need to find ways to connect the relational trust established at the national level to the EU level. Health care professionals and professional organisations, particularly in the field of medical genetics, can play an important part in informing and engaging with citizens and patients and building trust.

Citizen engagement needs sustained resources and attention across projects and national and EU initiatives. Trust depends on citizens and patients, so a trustworthy data sharing infrastructure needs transparent governance to consider and incorporate citizen views.



## 2. Contribution towards project objectives

With this deliverable, the project has reached or the deliverable has contributed to the following objectives/key results:

	Key Result No and description	Contributed
<b>Objective 1</b>  Engage local, regional, national and European stakeholders to define the requirements for cross-border access to genomics and personalised medicine data	1. B1MG assembles key local, national, European and global actors in the field of Personalised Medicine within a B1MG Stakeholder Coordination Group (WP1) by M6.	Yes
	2. B1MG drives broad engagement around European access to personalised medicine data via the B1MG Stakeholder Coordination Portal (WP1) following the B1MG Communication Strategy (WP6) by M12.	Yes
	3. B1MG establishes awareness and dialogue with a broad set of societal actors via a continuously monitored and refined communications strategy (WP1, WP6) by M12, M18, M24 & M30.	Yes
	4. The open B1MG Summit (M18) engages and ensures that the views of all relevant stakeholders are captured in B1MG requirements and guidelines (WP1, WP6).	Yes
<b>Objective 2</b>  Translate requirements for data quality, standards, technical infrastructure, and ELSI into technical specifications and implementation guidelines that captures European best practice	<b>Legal &amp; Ethical Key Results</b>	
	1. Establish relevant best practice in ethics of cross-border access to genome and phenotypic data (WP2) by M36	Yes
	2. Analysis of legal framework and development of common minimum standard (WP2) by M36.	No
	3. Cross-border Data Access and Use Governance Toolkit Framework (WP2) by M36.	No
	<b>Technical Key Results</b>	
	4. Quality metrics for sequencing (WP3) by M12.	No
	5. Best practices for Next Generation Sequencing (WP3) by M24.	No
6. Phenotypic and clinical metadata framework (WP3) by M12, M24 & M36.	Yes	
7. Best practices in sharing and linking phenotypic and genetic data (WP3) by M12 & M24.	Yes	



	<b>8.</b> Data analysis challenge (WP3) by M36.	No
	<b>Infrastructure Key Results</b>	
	<b>9.</b> Secure cross-border data access roadmap (WP4) by M12 & M36.	No
	<b>10.</b> Secure cross-border data access demonstrator (WP4) by M24.	Yes
<b>Objective 3</b>	<b>1.</b> The B1MG maturity level model ( WP5) by M24.	No
Drive adoption and support long-term operation by organisations at local, regional, national and European level by providing guidance on phased development (via the B1MG maturity level model), and a methodology for economic evaluation	<b>2.</b> Roadmap and guidance tools for countries for effective implementation of Personalised Medicine (WP5) by M36.	No
	<b>3.</b> Economic evaluation models for Personalised Medicine and case studies (WP5) by M30.	No
	<b>4.</b> Guidance principles for national mirror groups and cross-border Personalised Medicine governance (WP6) by M30.	No
	<b>5.</b> Long-term sustainability design and funding routes for cross-border Personalised Medicine delivery (WP6) by M34.	No

### 3. Methods

An expert workshop was organised for which scholars and professionals were invited to discuss citizen and patient engagement. 14 Experts (expertise in bioethics, ELSI, governance, human genetics, patient representation and citizen engagement), were asked to give a short presentation in one of three thematic subgroups.

Lessons learned were gathered for three subthemes: “When an ELSI framework is in place, what is the (additional) role of citizen engagement in fostering data sharing and public trust”; “How does citizen engagement relate to (interests of) other stakeholders?” ; “When and how to engage and at what level?”

In a concluding session participants formulated recommendations on the role of engagement in genomic data sharing and public trust. These recommendations will be published separately in D1.7.



Based on the presentations and discussions key points from these subthemes were formulated. A draft report was distributed to the participating experts and comments were integrated into this final report.

## 4. Description of work accomplished

This report has been made based on the notes and recording of presentations and discussions at the expert workshop on 18 January 2023.

This deliverable falls under the heading of B1MG WP1, but input was asked from WP2. A short summary of the B1MG ELSI framework was presented at the beginning of the workshop, while the participants were asked about the relevance of citizen engagement in addition to ELSI frameworks, such as elaborated under WP2. A representative of WP2 attended the expert workshop and provided input in the discussions.

An abstract was submitted to the ESHG Conference in Glasgow, June 10-13, which was selected for presentation as a hybrid poster.

## 5. Results

### Introduction:

### Miriam Beusink, ELSI program officer Health-RI

#### A brief introduction to B1MGs WP2 ELSI framework

The B1MG ELSI framework is based on the following principles:

1. Autonomy: Different countries have different legislation, ethics principles, subjects' wishes.
2. Flexibility: IT enables tagging of data with ELSI relevant metadata.
3. Minimum requirements for participation: these are determined by EU norms and implemented for national norms through procedures
4. Single portal and access procedure.

Several documents have been developed specifying policies on specific areas:

Data governance policy for research access:





Policies were developed on how to include data into 1+MG, pertaining to e.g. the responsibilities of the data holders, 1+MG, member countries et cetera); how to make data accessible in 1+MG (e.g. procedures to ensure efficient but compliant data access); and how to use data (specifying user and data subjects' rights and measures for protection. This policy is currently under review by governmental stakeholders in the GDI project.

The incidental findings policy:

In line with the autonomy principle there is no obligation from 1+MG for data providers on how to handle Incidental findings. Reporting to subjects is decided locally in line with policies or subject's wishes. Data users are required to feedback (only) valid, serious and actionable IFs to data submitters.<sup>1</sup>

The inclusion of special subjects' policy:

Special subjects, e.g. minors, persons not able to consent, minorities, vulnerable groups and deceased persons should not be abused, exploited or discriminated. Recommendations are to include them only if research can't be carried out without them, but they should also not be unnecessarily excluded. Capacity to understand and to give consent differs, but, when possible, consent should be obtained.<sup>2</sup>

The communication of research results policy:

Feedback is organized by national nodes. Content should be anonymous. Data subjects should only be actively informed when they indicated that e.g., their e-mail address can be used for this. Recommendations were made to provide a list of publications or examples of studies on websites.<sup>3</sup>

Transparency and Consent policy:

This document indicates what *must*, *should* and *may* be done in terms of information provided to people whose data are to be used in 1+MG and, where applicable, consent obtained. E.g.: the cultural, linguistic, and socio-economic context SHOULD be considered when preparing information sheets and consent forms. Community groups MAY be involved to ensure such issues are appropriately communicated in the documents. This policy was translated into a guidance which is currently being tested by the members of the ELSI Working Group in 1+MG.

## Session 1: Chair - Carla van El, Amsterdam UMC

"Given that a solid B1MG ELSI framework (responsible consent, storage, privacy, data sharing policies etc.) would be in place, what is the (additional) role of citizen engagement in fostering

<sup>1</sup> <https://zenodo.org/record/6362131>

<sup>2</sup> <https://zenodo.org/record/6362184>

<sup>3</sup> <https://zenodo.org/record/6362164>



data sharing and public trust? What goals should citizen engagement aim to achieve: communication, participation, transparency, trust, etc.”

## Panelists:

### 1. Christine Patch - Need for citizen/participant engagement; 100.000 genomes project

In recent years the United Kingdom has implemented policies for patient and participant involvement (PPI), stressing that research should be done ‘with’ or ‘by’ the public, not ‘to’, ‘about’ or ‘for’ them, contributing to how research is designed, conducted and disseminated.<sup>4</sup>

Research involving patients is considered better because they are more relevant, acceptable and understandable to participants, and provide better experience of research and communication of results.

The 100.000 Genomes Project, a partnership between NHS England and Genomics England – sequencing 100,000 genomes from around 85,000 NHS patients affected by rare disease or cancer – is leading to groundbreaking insights and continued findings into the role genomics can play in healthcare.<sup>5</sup>

The project established a participant panel to be active in various committees at Genomics England. As is elaborated in an article<sup>6</sup> the panel acted not merely as ‘critical friends’, being ‘invited to the party’ while giving advice on decision making and governance, communications, implementation and new ethical issues such as reconsenting. They also acted as ‘Gatecrashers’ when making recommendations. The panel advised to recontact all participants about consent, learn if they still consented to return of results, and pushed for return of positive and ‘nothing found’ results, and not only positive findings.

Lessons learnt were: Patient voices should be heard throughout the whole organisation, the leadership teams should routinely seek input, there should be clear terms of reference and clarity of expectations, participants need to keep legitimacy as ‘lay people’, and it is important to allow ‘new’ voices into the room, such as minority populations. Engagement should not be tokenistic.

Reflecting on the panel: it engendered trust and increased the effectiveness of implementation. Challenge: given that stewardship of genomic data is a long-term commitment, how can this be maintained when an organisation changes in function, leadership etc. Is such an intensive model

<sup>4</sup> <https://www.gov.uk/government/publications/best-research-for-best-health-a-new-national-health-research-strategy>; <https://www.hra.nhs.uk/planning-and-improving-research/policies-standards-legislation/uk-policy-framework-health-social-care-research/>

<sup>5</sup> Turnbull C, Scott RH, Thomas E, et al. [The 100 000 Genomes Project: bringing whole genome sequencing to the NHS](#) [published correction appears in BMJ]. 2018 May 2;361:k1952. Devereaux A [corrected to Devereau A]]. BMJ. 2018;361:k1687. Published 2018 Apr 24. doi:10.1136/bmj.k1687

<sup>6</sup> Hastings Ward J, Middleton R, McCormick D, et al. [Research participants: critical friends, agents for change](#). Eur J Hum Genet. 2022;30(12):1309-1313. doi:10.1038/s41431-022-01199-3



transferable across multiple projects and jurisdictions, how does data federation affect potential aims of engagement. As presented by the regulatory and ethics working group of The Global Alliance for Genomics and Health (GA4GH), a policy-framing and technical standards-setting organisation, it is crucial to establish in advance what any citizen involvement/engagement activity is 'for' and what are hoped for outcomes?

The framework for participant and public engagement produced by this group and adopted by GA4GH outlines 4 principles<sup>7</sup>: Fairness, ensuring research and health care are undertaken responsibly and do not produce or exacerbate inequity or inequality; Context, ensuring that the purpose of engaging stakeholders aligns with the context being considered; Heterogeneity, referring to diversity and inclusivity; Recognising tensions and conflict. As stated earlier fundamental questions are: what are the aims and desired outcomes of the activity and how will outcomes be assessed?

## 2. Gabby Samuel - Striving for trust and trustworthiness

ELSI or trustworthiness frameworks don't necessarily lead to participants having trust in an institution.

Trust is complex and can be based on desires and hopes, personality, socio-political issues, class, etc. and is contextual (e.g., can be based on ethnic or religious diversity). Trust can be based on knowledge and experiences. The example of the Estonian Biobank shows that trust can be compromised for different reasons: some Russian speaking participants withdrew their samples from the biobank after political decisions that had nothing to do with the biobank actions and/or policy. Previous research<sup>8</sup> exploring investigative genetic genealogy showed that those who had more positive experiences with the criminal justice system were more trusting of the technology.

Citizen engagement is one way of helping promote trust because it can build hopes as well as positive interactions and experiences between participants and the biobank. In some focus groups we have conducted with a biobank, trust over time increased for many participants due to the communication the biobank provided about the biobank being a trustworthy organization, as well as showcasing the research they were conducting (forthcoming). For any project that includes large scale data sharing engagement and communication is crucial.

Who is representing the project, and their perceived virtue and expertise e.g., in keeping data safe is crucial to trust, is also important e.g. also the ability to relate to various cultural contexts.

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<sup>7</sup> Murtagh MJ, Machirori M, Gaff CL, Blell MT, de Vries J, Doerr M, et al. [Engaged genomic science produces better and fairer outcomes](#): an engagement framework for engaging and involving participants, patients and publics in genomics research and healthcare implementation. Wellcome Open Res. 2021;6:311. doi:10.12688/wellcomeopenres.17233.1

<sup>8</sup> Samuel G, Kennett D. [The impact of investigative genetic genealogy](#): perceptions of UK professional and public stakeholders, Forensic Sci Int Genet. 2020;48:102366. doi:10.1016/j.fsigen.2020.102366



Finally, it is important to remember that citizen engagement is good in itself – not merely in relation to building trust. Striving for trust must not lead to ‘trustworthiness washing’<sup>9</sup>, merely aiming to appear trustworthy.

### 3. Johannes van Delden - Principles of citizen engagement and trust

Informed consent, often broad consent, can lead to ‘disentanglement’ if it is a mere ‘one time consent’ without further communication or contact with donors. With disentanglement I mean that the link between the donor and the sample is lost: donors may get the feeling that they have no connection, no influence and no say in what is happening to the sample after they signed a broad consent form.<sup>10</sup> A project or biobank needs to find ways for ‘re-entanglement’, asking for consent at the moment of data collection is not enough. The emphasis should be on governance, institutions should organise governance and live by it. This should not amount to window dressing.

Broad consent should always be ‘consent for governance’<sup>10</sup>, most notably participatory governance. PPI should be embedded in the governance structure. This may take several forms, for instance individual influence, access to information or participation in a data access committee (DAC). All of these can be forms of shaping PPI.

Principles for this type of infrastructure for research or health care improvement include<sup>11</sup>:

- Legitimate legal ground (e.g. informed consent);
- Earn a ‘social license’ which goes beyond legitimacy principles<sup>12</sup>;
- Whatever you do with material or data should have social value;
- Participatory governance;
- Accountability - ‘learning accountability’ (develop and listen, improve structures, over the course of a programme).<sup>13</sup>

Conclusions: Don’t ask participants to sign at the X and then disengage because this causes disentanglement. Create a social license for data-intensive health research through participation, co-creation and public value.<sup>12</sup>

<sup>9</sup> Samuel G, Broekstra R, Gille F, Lucassen A. [Public Trust and Trustworthiness in Biobanking: The Need for More Reflexivity](#). *Biopreserv Biobank*. 2022;20(3):291-296. doi:10.1089/bio.2021.0109

<sup>10</sup> Boers SN, van Delden JJ, Bredenoord AL. [Broad Consent Is Consent for Governance](#). *Am J Bioeth*. 2015;15(9):53-5. doi:10.1080/15265161.2015.1062165

<sup>11</sup> Kalkman S, Mostert M, Gerlinger C, van Delden JJM, van Thiel G. [Responsible data sharing in international health research: a systematic review of principles and norms](#). *BMC Med Ethics*. 2019;20(1):21. <https://doi.org/10.1186/s12910-019-0359-9>

<sup>12</sup> Muller SHA, Kalkman S, van Thiel G, Mostert M, van Delden JJM. [The social licence for data-intensive health research: towards co-creation, public value and trust](#). *BMC Med Ethics*. 2021;22(1):110. <https://doi.org/10.1186/s12910-021-00677-5>

<sup>13</sup> Muller SH, Mostert M, van Delden JJ, Schillemans T, van Thiel GJ. [Learning accountable governance: Challenges and perspectives for data-intensive health research networks](#). *Big Data & Society*. 2022;9(2). <https://doi.org/10.1177/20539517221136078>



#### 4. Barbara Prainsack - Promises and pitfalls of data sharing and personalised medicine

Precision Medicine – as the data rich iteration of - Personalized Medicine aspires to replace symptom-based taxonomies at different stages of disease with data-rich characterisations of individuals, making (digital) data a currency of personalisation. The data driven dimension of personalisation in both health and non-health related applications needs more attention, as it comes with new benefits and challenges. Also, the political economy that data practices are embedded in has changed. The big commercial players in the field of health are no longer only big pharma, but big tech. The latter are pushing further into the health domain, setting research agendas, funding research, providing tools and software, and sometimes owning data. In this situation, talking about social license in the context of engagement needs specification as it can mean different things.

The 'Your DNA, Your Say' study found that a lot of people are happy for their data to be used if there is evidence of it benefiting themselves or others, they are more hesitant for their data to be used by companies, rather than their doctor. Other work, on COVID contact tracing apps, showed people are skeptical concerning their own privacy but also over-surveillance of others. The two concerns shade into one another. We should not think of benefits and risk for others versus people themselves as a binary, but understand how people weigh these aspects in making good choices and thinking about what society they want to live in. For instance, they may not be against commercial profit per se but this needs to be balanced and proportional and seen as fair. Such considerations are relevant when thinking about a social license.

Trust can be seen as relational: a study found that for many people trust was based on the specific researchers carrying out the work, which has implications for secondary uses and collaborations with other researchers.

We published a framework on solidarity-based data governance<sup>14</sup> and are now developing an online tool for the assessment of the public value of data use.

We need a better understanding of what public value is so it may be the basis of a social license – in the past this has been abused by corporations, and therefore we need an assessment weighing the benefits and risks for both individuals and groups.

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<sup>14</sup> <https://www.governinghealthfutures2030.org/wp-content/uploads/2022/12/DataSolidarity.pdf>



## Discussion themes session 1

This workshop addresses (genomic) data sharing and trust in a general sense, however, there are specific contexts such as the clinic, biobank or screening, including other aspects such as data storage. How do these different contexts matter? Trust may depend on and will differ in various contexts. ELSI frameworks and legal arrangements are relevant for all these contexts (e.g., thorough consent procedures) but beyond that foundation patient and public involvement in all contexts are needed to make sure there is a social license for data use or public value is established. Institutions need to show their effort to be trustworthy and try to earn trust. Also reflection on national contexts is relevant e.g. having a national biobank collaborating internationally while international conflicts and foreign policies regarding human rights may challenge existing data sharing infrastructures and trust.

At the level of single projects where individuals give access to their data, you rely on the relational quality of trust, but this is not enough and poses a challenge for the 1+MG initiative, where trust needs to be established in this large monolithic European organisation.

An important question is how to ensure trustworthiness has teeth, how can people enforce trustworthiness, who holds organisations and corporate players accountable?

### Key points Session 1: “Given that a solid B1MG ELSI framework would be in place, what is the (additional) role of citizen engagement in fostering data sharing and public trust? What goals should citizen engagement aim to achieve?”

- It is important to determine in advance what the goals of citizen engagement or public involvement are or should be.
- The B1MG ELSI framework provides a necessary legal foundation for data access but citizen engagement is a crucial element in addition to such a framework.<sup>15 16</sup> **Error! Bookmark not defined.**
- ELSI or trustworthiness frameworks don't necessarily lead to participants having trust in an institution. The addition of citizen engagement can help build and contribute to trust.<sup>9</sup>

<sup>15</sup> Middleton A, Milne R, Almarri MA, Anwer S, Atutornu J, Baranova EE, et al. [Global Public Perceptions of Genomic Data Sharing: What Shapes the Willingness to Donate DNA and Health Data?](#) Am J Hum Genet. 2020;107(4):743-52.

<sup>16</sup> Milne R, Morley KI, Almarri MA, Anwer S, Atutornu J, Baranova EE, et al. [Demonstrating trustworthiness when collecting and sharing genomic data: public views across 22 countries.](#) Genome Med. 2021;13(1):92.



- Trust is relational and context specific and dependent on many factors such as desires and hopes, socio-political factors and developments, knowledge and experience, the relation to the professionals or institutions asking for data sharing and trust.
- Involvement/engagement is crucial for the social license and to establish the public value of a project and the data use.<sup>11 12 17 18</sup>
- Involving citizens in the governance of a project is important because otherwise citizens are disentangled from their data and having a say in data use.<sup>10</sup>
- The challenge for the coming years is to find ways to bridge specific, national and international contexts for engagement and trust: engagement takes place on a project or national level in the member states where the data are gathered and stored, but 1+MG, B1MG and GDI are international endeavors.
- Citizen engagement can have a diversity of aims that may help create preconditions for accountable research and trust:<sup>19 20 21 22 23 24</sup>
  - Communication about the project;
  - To improve research by identifying research questions and concerns relevant for patients and citizens, ranging from consultation to collaboration or collaboration or co creation;
  - Capacity building;
  - Transparency and accountability;
  - It can create data-solidarity;
  - Help building and maintaining trust;
  - Engagement can contribute to being a ‘trustworthy organisation’;
  - Engagement can contribute to the governance of a project.

<sup>17</sup> Amorim M. [Benefits and Risks of Sharing Genomic Data for Research](#): Comparing the Views of Rare Disease Patients, Informal Carers and Healthcare Professionals. *International Journal of Environmental Research and Public Health*. 2022;19(8788).

<sup>18</sup> Allen J, Adams C, Flack F. [The role of data custodians in establishing and maintaining social licence for health research](#). *Bioethics*. 2019;33(4):502-10.

<sup>19</sup> Erikainen S, Friesen P, Rand L, Jongsma K, Dunn M, Sorbie A, et al. [Public involvement in the governance of population-level biomedical research: unresolved questions and future directions](#). *J Med Ethics*. 2020.

<sup>20</sup> McMahon A, Buyx A, Prainsack B. [Big Data Governance Needs More Collective Responsibility: The Role of Harm Mitigation in the Governance of Data Use in Medicine and Beyond](#). *Med Law Rev*. 2020;28(1):155-82.

<sup>21</sup> O'Doherty KC, Shabani M, Dove ES, Bentzen HB, Borry P, Burgess MM, et al. [Toward better governance of human genomic data](#). *Nat Genet*. 2021;53(1):2-8.

<sup>22</sup> Voigt TH, Holtz V, Niemiec E, Howard HC, Middleton A, Prainsack B. [Willingness to donate genomic and other medical data: results from Germany](#). *Eur J Hum Genet*. 2020;28(8):1000-9.

<sup>23</sup> Prainsack B, El-Sayed S, Forgo N, Szoszkiewicz L, Baumer P. [Data solidarity: a blueprint for governing health futures](#). *Lancet Digit Health*. 2022;4(11):e773-e4.

<sup>24</sup> Laux J, Wachter, S., Mittelstadt, B. [Trustworthy artificial intelligence and the European Union AI act: On the conflation of trustworthiness and acceptability of risk](#). *Regulation & Governance* (2023). 2023.





## Session 2: Chair - Eric Vermeulen, Patient Alliance for Rare and Genetic Diseases

“How does citizen engagement relate to (interests of) other stakeholders?”

### Panelists:

#### 1. Edith Sky Gross - Focus on patient; experiences from EURORDIS

EURORDIS<sup>25</sup> is a non-profit alliance of over 1000 rare disease patient organisations from 74 countries which takes part in the Screen4Care<sup>26</sup> project. This is a European Project funded under the IMI2 (Innovative Medicine Initiative) aimed at shortening the pathway to diagnosis using advanced technologies. Patient engagement is transversal throughout the project, and patients are represented in the Patient Advisory Board that provides recommendations, guidance and advice to all project activities. One arm is devoted to genomic newborn screening. It investigates screening by two different panels, differentiating conditions that are ‘treatable’ or ‘actionable’. A multi-stakeholder NBS working group driven by patients and including experts is closely involved in monitoring and assessment. Questionnaires are distributed via the EURORDIS Rare Barometer Programme, to gather the input from 20,000 participants (patients with rare diseases, family members, patient advocates) to enable input on the newborn screening research from a wider range of patients in 23 languages.

#### 2. Michaela Mayrhofer (BBMRI-ERIC) - Engaging patients and researchers

BBMRI-ERIC<sup>27</sup> is a European research infrastructure for biobanking, aiming to facilitate access to samples and data in over 20 countries in Europe. It incorporates ELSI activities ranging from doing research, providing training and advice through an ELSI help desk. Patients and citizens advocacy groups are involved through the Stakeholder forum, that aims for participatory governance. The SF has three pillars: patients, industry and societies such as the EMA. The chair of the SF is always a patient who also sits on the Scientific and Ethical Advisory Board.

When reflecting on the relation between citizen engagement and other stakeholders it is important to acknowledge that every stakeholder has their own interest and outlook, these can be found out and explored via dialogue. Everyone goes in with the best intentions, but, for instance, researchers tend to just think about publishing papers. We need to find ways to allow for dialogue and also have potentially uncomfortable conversations.

<sup>25</sup> <https://www.eurordis.org/>

<sup>26</sup> <https://screen4care.eu/>

<sup>27</sup> <https://www.bbmri-eric.eu/>





We always need dialogue and be aware of the relationship with participants, which needs to be built and reinforced. Keywords such as trust or data altruism should be defined clearly and be supported by governance rules. Engagement and governance need teeth. We need to live and scrutinize the 'trust' everyday and not assume to speak on behalf of patients and citizens. We should be aware there is a power relation as researchers are often closer to the funding.

### 3. Vera Frankova - Communicating with patients and public

Insights from two projects are shared on patient and public engagement. Solve-RD<sup>28</sup> is a project funded by the European Commission to help patients who are long-term undiagnosed. Patients and their relatives had already experience with genomics, research and registries, they had in comparison to general public relatively high awareness, trust and interest to participate in research and share their data, on the basis of altruistic motives. They receive relatively solid and reliable information from health care professionals and patient organisations and expect benefits from their participation.

The ACGT<sup>29</sup> project was looking for 1000 healthy donors to contribute to the analysis of Czech genomes for theranostics. It was established in the general population there is no or low experience with genomics, and though the public is heterogeneous there was overall relatively lower awareness, interest and trust in comparison to group of patients and their relatives. People get the information about genomics from media and potentially unreliable resources. This raises questions about their motivation to participate and what benefits they expect. For this project lessons were learned about targeting different age groups through different media (social media 20-40, newspapers 40-60, and television 40-80 years old), and that the message should be focused to their interests. The project website was visited more often after each media exposure (written article in newspaper, on social media, television interviews, flyers etc.) raising interest in the project. Using pages of trustworthy institutions and prevention of inappropriate comments is important.

Lessons learned for the project with rare disease patients were that their previous experiences with the healthcare services can influence participation and trust, that achievable benefits help to maintain trust, and that collaboration with patient organisations can play an important role.

### 4. Borut Peterlin - Genetic professionals and ESHG

Involvement of genetic professionals and professional societies contributes to engagement and building trust by informing member states and inviting citizens to participate in genome sharing. The purpose of the sharing is important. From a pilot on neonatal screening, we learned participants would more gladly share data if it would help others in a similar situation, to develop better services and medical applications rather than sharing with a general international community.

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<sup>28</sup> <https://solve-rd.eu/>

<sup>29</sup> [www.acgt.cz/en/](http://www.acgt.cz/en/)



In a survey on national genome projects in the world we found much heterogeneity in the purposes of national genome projects, impacting on type of data to be shared<sup>30</sup>

For instance, the Slovenian genome project is highly focused on health care applications, on the improvement and diagnosis of rare diseases - health applications are seen as very important.

Responsibility for the welfare of the patients is key.

Determinants of trust are among others, patient-centered care, access to and continuity of care after the genomic data have become available, reliability and dependability, building a partnership.

If we want to provide trustworthy governance of genomic data collection and sharing at the level of member states, their purpose and output should be clearly stated. Genetic professionals and national and European societies are important in building trust and advancing health and research.

## 5. Helena Machado - Medical/Research field in relation to other domains, e.g. forensics

Instead of treating ethical issues in medical and forensic biobanks separately, both need democratic control by multiple publics. For public engagement to be a meaningful and effective tool to open genomic research and decision making to debate and democratic control we need to recognise 3 crucial aspects:

1. Tension on the role and goals of public engagement exercises – transparency is needed to clarify the role of the participants and the aims of the public engagement activities – e.g., is the role of participants to be data subjects, co- developers or decision makers? Is the goal robustness of policy choices, to reach consensus and minimise controversy, a means to acceptance and trust for a decision already made? Social scientists have expressed fear that outcome of engagement may be used by more powerful actors. Unfortunately, engagement activities may be used to simply boost participation and not to discuss aspirations or directions of the projects.
2. Engagement activities are situated in a context with competing interests and priorities, e.g., in science, policy or business. Power structures, interests, inequalities shape the context of scientific and technological development and should be made explicit in engagement activities.
3. Impact measurement is difficult, so the desired impact should be defined in advance - ensure that those involved can actually make a difference in uptake of results of engagement. Output of engagement exercises may be wasted in the absence of a clear strategy for using the results of earlier engagement. For instance, a report by the Danish Board of Technology from 2021<sup>31</sup> provides relevant suggestions.

<sup>30</sup> Kovanda A, Zimani AN, Peterlin B. [How to design a national genomic project-a systematic review of active projects. Human genomics](https://doi.org/10.1186/s40246-021-00315-6), 2021;15(1), 20. <https://doi.org/10.1186/s40246-021-00315-6>

<sup>31</sup> [https://tekno.dk/app/uploads/2021/02/Public-engagement-with-ethical-issues-in-science\\_FINAL.pdf](https://tekno.dk/app/uploads/2021/02/Public-engagement-with-ethical-issues-in-science_FINAL.pdf)



## Discussion themes session 2:

Speakers have similar comments e.g., along the lines of the relevance of recognizing tensions between interests of stakeholders. The data-driven dimension needs more attention, the different interests and scrutiny towards public research and private companies. It is mentioned that conflicts or differences of opinion may not only be present between but also within stakeholder communities. In a democratic dialogue it is okay to disagree or say that we do not know. Sometimes success is just having ongoing honest (and sometimes challenging) dialogue with/between stakeholders, without necessarily getting positive 'results' or outcomes.

When organising engagement stakeholders need to be able to join, e.g., time (e.g. meeting during or outside office hours) for citizens may be crucial in allowing people to participate.

## Key points Session 2: "How does citizen engagement relate to (interests of) other stakeholders?"

- Transparency is needed to clarify the role of the participants, what is expected of them, and the aims of the public engagement activities.
- Engagement activities are situated in context with potentially conflicting interests, power differences and priorities, for instance between publicly accountable researchers and private companies, which should be made explicit.
- If differences in interests occur, that need not be a problem. In a democratic dialogue it is okay to disagree or say that we do not know. Engagement is a learning process in which various sides can learn, from each other's perspective and interest.<sup>32</sup>
- Researchers may have their own interest and should not automatically be assumed to speak on behalf of patients and citizens.
- Differing interests may also exist or appear within stakeholder communities.
- Citizen engagement can relate to the interests of researchers in the sense that it may help define research problems, secure trust, help enhance data sharing.<sup>33 34 35 36</sup>

<sup>32</sup> Russell J, Fudge N, Greenhalgh T. [The impact of public involvement in health research: what are we measuring? Why are we measuring it? Should we stop measuring it?](https://doi.org/10.1186/s40900-020-00239-w) Res Involv Engagem. 2020;6:63. <https://doi.org/10.1186/s40900-020-00239-w>

<sup>33</sup> Haug CJ. [Whose Data Are They Anyway? Can a Patient Perspective Advance the Data-Sharing Debate?](https://doi.org/10.1056/NEJMp1704485) N Engl J Med. 2017;376:2203-2205. DOI: 10.1056/NEJMp1704485

<sup>34</sup> Bitsch, L. [The landscape of science, ethics and public engagement and their potential for the future](#). 2021

<sup>35</sup> Abels G. [Next level citizen participation in the EU](#). 2022.

<sup>36</sup> CIOMS. [Patient involvement in the development, regulation and safe use of medicines](#). 2022.



- Health care professionals and professional organisations, particularly in the field of medical genetics, play an important part in informing and engaging with citizens and patients and building trust.
- Impact measurement is difficult, so the desired impact should be defined in advance - ensure that those involved can actually make a difference. Engagement and governance need teeth.

## Session 3: Chair - Denis Horgan, Executive Director, EAPM

“When (continuously) and how (formats/tools) to engage and at what level (national-European)?”

### Panelists:

#### 1. Heidi Howard - Surveys and beyond

The tools used for citizen engagement need to be appropriate for the different contexts and goals. In general, there may be tensions between academic requirements for publishing and e.g. public health authorities wanting to raise awareness via engagement. Quantitative surveys as a tool are good if you have simple(ish) questions - not so well with high-tech topics. In the ‘Your DNA, Your Say survey’ this was addressed by providing **informational videos**<sup>37</sup>. Multiple choice questions are giving options that the experts think are important (not necessarily what publics think is important), there is no room for interaction, pressure to publish positive results may lead to overblowing results, and minority voices may be missing. Qualitative methods such as interviews and focus groups are interactive and can give deeper insights, but are difficult to interpret accurately, and results may be over-interpreted when perceived as similar to results from quantitative studies. What would we ideally do if we would approach engagement research and activities really seriously? Give engagement enough money, time, resources, and the right expertise, it should be ongoing, finding appropriate tools for different sub-goals, and have spaces where the public can ask questions and raise their concerns. We should build a community, not see it as an event, have more education in high schools.

We should be building trustworthiness and trustworthy places, rather than expect trust.

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<sup>37</sup><https://societyandethicsresearch.wellcomeconnectingscience.org/project/your-dna-your-say/>



## 2. Carina Pittens - Learnings from national examples - Austrian-Dutch focus group study:

The Austrian Dutch “GARiB-LIFELINE, Issues, and Publics Project” (GLIPP) studied attitudes towards participating in biobanking and how to improve governance via focus groups with mixed lay publics and participants of Lifelines biobank.

The general public had little prior knowledge of biobanks and their function but focus group participants (after a bit of explanation) were happy and willing to discuss. They mentioned public interest as advantage, and threats to privacy as a disadvantage. Biobanks participants considered personal benefit an advantage and had concerns for possible negative findings.

Lay publics mentioned consent to research goals and scope, and commercialisation as most important issues. Biobank participants were also concerned about commercialisation

The respondents had no clear suggestions for regulation or control, or who should be responsible for these, although they felt that this regulation is very important.

## 3. Wannes van Hoof - Learnings from national examples: Belgium

Since 2017 there was continuing public engagement in genomics via Sciensano. For instance, a Citizen forum was organised on ‘My DNA’, followed by a larger online engagement ‘DNA Debat’<sup>38</sup> and recently a contribution to TEHDAS Healthy Data e-consultation<sup>39</sup>.

Lessons learnt from these engagement activities are that people are willing to contribute, and see benefits, but there is also fear. Education and engagement may even result in people getting more worried. Uncertainty is inherent in genomics, and due to the individual and societal nature of genomics, we should be cautious and have safeguards in place. People feel vulnerable donating and learning about uses of their genome, they may feel ‘prisoner’ of their genome. Engagement may allow for a dialogue which is important to respect diverging values and views. Organisations or projects should aim for being trustworthy and trust could be a by-product of being trustworthy. Via engagement citizens and patients can make meaningful contributions to the development of an ethical, legal and societal framework for genomic technologies.

## 4. Lise Bitsch - How to connect national and international engagement initiatives

The Danish Board of Technology Foundation<sup>40</sup> has engaged publics and citizens on science and policy in Denmark and internationally for many years.

We need to focus on the ‘why’ of engaging citizens as this has consequences for the tool of engagement. Why engage with citizens? Trust is not a goal of citizen engagement, but it could result from engagement. Goals of engagement could be to learn about the knowledge from

<sup>38</sup> Mayeur C, Saelaert M, Van Hoof W. [The Belgian DNA Debate: An Online Deliberative Platform on the Ethical, Legal, and Social Issues of Genomics](#). *Public Health Genomics*. 2021;24(3-4):149-159. doi:10.1159/000515356

<sup>39</sup> <https://tehdas.eu/results/tehdas-consultation-people-support-health-data-use-with-solid-safeguards/>

<sup>40</sup> [The Danish Board of Technology \(tekno.dk\)](#)



citizens, potential conflicts, new ideas for solutions, issues that you didn't even realise were issues, which questions are most important across a diverse population. Will this increase trust? Maybe....

Trust is about beliefs that an organisation wants to do something good, and about the willingness of people to depend on your organisation also in risky situations. Engagement activities could actually result in losing trust if you are not perceived to do good or be reliable. You need engagement because otherwise you do not know if you live up to citizens' expectations.

When and how should we engage? Any time during the developmental process, with various tools, but at the beginning the possibility to change elements is bigger. Most important is to allow citizens to influence the processes, priorities and choices.

## 5. Mark Bale - How to connect national and EU level; policies and tools

The Council of Europe (46 MSs) organised a Public dialogue on Genomic medicine<sup>41</sup>, the video is available and the report is finalised<sup>42</sup>. The aim of that workshop was to discuss how you can engage the public in developing genomic medicine. It addressed among others the duties of policy makers and funders to take part in dialogue as a process of continuous engagement, open to feedback and co-production, also open to being challenged by publics.

During the seminar several examples were presented from UK biobank and Genomics England, the German cancer biobank, the dialogue on genome editing in the Netherlands, and IPPOSI<sup>43</sup> in Ireland.

These national examples showed elaborate processes underscoring the need to have funds and resources for dialogue and engagement. Such processes need to be transparent and open.

At the workshop there was room for interesting perspectives from younger people, such as undergraduates/postgraduates – it is important to stress the relevance of engaging them as they will increasingly be confronted with genomics. For this group, tools such as online engagement are useful. A public dialogue may also help to educate and empower people to discuss mis/dis-information online and how to challenge this. There was also input from Industry, getting their perspective is relevant, as well as the perspective of the public on industry involvement, to allow for discussions on fairness and commercial uses.

Public dialogue is also important in policy making and the education of policy makers.

Public engagement should be a properly funded part of genomic medicine in order to help build trust as a part of good governance and transparency.

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<sup>41</sup><https://www.coe.int/en/web/bioethics/workshop-public-dialogue-genomic-medicine>

<sup>42</sup><https://rm.coe.int/cdbio-2023-1-rapporteur-report-on-public-dialogue-genomic-medicine-e/1680aae608>

<sup>43</sup><https://www.ipposi.ie/>



## Discussion

In the Discussion it was added that also human rights are an important driver for the Council of Europe. The CoE also published a **Guide to public debate**<sup>44</sup> on human rights and biomedicine which aims to assist Member States in raising public awareness about biomedical developments by encouraging the circulation of information, views and opinions. The Guide also aims to help to develop, plan and promote exchanges between different people and actors with a view, where appropriate, to inform policy making based on shared values and respect for human rights. The Guide does this with reference to a selection of good practices and experiences in member states which illustrate aspects of public debate in action.

Discussion focused on: how to reach specific publics with e.g., surveys and disseminate the findings to specific audiences? Different tools may be necessary to reach specific publics and goals.

### Key points Session 3: “When (continuously) and how (formats/tools) to engage and at what level (national-European)?”

- Engagement can be done at various moments, and preferably continuously<sup>45 46 47 48 49 50</sup> Specific consultations on topics are necessary and possible.<sup>34 35 36</sup>
- If engagement starts at the beginning of a project, there are more options to influence choices and policies.
- The tools used for citizen engagement need to be appropriate for the different contexts and goals:
  - By a citizen board, or patient forum, which allow for interaction and influence.<sup>6</sup>
  - By consultations with several methods: questionnaires, focus groups, interviews.

<sup>44</sup> <https://www.coe.int/en/web/bioethics/public-debate>

<sup>45</sup> EURORDIS. [Community Advisory Boards - connecting patients with clinical research](#) 2018.

<sup>46</sup> Kubicek K, M. Robles. [Community Advisory Board Toolkit](#) 2016

<sup>47</sup> Marinello D. [Patient engagement in healthcare: a preliminary set of measures to evaluate patient engagement in the European Reference Networks](#). Rare Disease and Orphan Drugs Journal. 2021;1(2).

<sup>48</sup> Paradigm. [Paradigm tools Public consultation](#) 2020.

<sup>49</sup> Paradigm. [Community Advisory Boards Toolkit](#) 2021.

<sup>50</sup> PFMD. [Patient Engagement in Digital Health and Data](#) 2022



Social media and online tools become more relevant, and may especially be useful to engage youth <sup>51</sup>

- Engagement often takes place at a project or national level. The 1+MG initiative and the B1MG project has 24 participating MS and they are each respectively working towards their own genomic medicine programmes and public outreach and engagement programmes. Storing genomes takes place on a national level. Sharing data across borders demands special attention. This international dimension should be made explicit to be transparent about data use across borders.
- Education and engagement may not automatically result in more trust or cooperation in data sharing, people may sometimes become more worried and feel vulnerable.

## 6. Next steps: Recommendations (See Deliverable 1.7)

After the presentations and discussions, the participants were asked to list their three key Recommendations or key messages from the meeting. These can be grouped under 6 headings, listed below.

The content of the Recommendations will be listed in Deliverable 1.7.

Resources and infrastructure

Accountability

Connect engagement across national initiatives

Specify goals and instruments:

Interests and diverse viewpoints

Capacity building

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<sup>51</sup> Wong BLH, Gray W, Holly L. [The future of health governance needs youth voices at the forefront](#). Lancet. 2021;398(10312):1669-70.

