Article:
Erikainen, S., Friesen, P., Rand, L. et al. (2021) Public involvement in the governance of population-level biomedical research: unresolved questions and future directions. Journal of Medical Ethics, 47 (7). pp. 522-525. ISSN: 0306-6800

https://doi.org/10.1136/medethics-2020-106530

OPEN ACCESS

Public involvement in the governance of population-level biomedical research: unresolved questions and future directions

Sonja Erikainen ,[1] Phoebe Friesen,[2] Leah Rand,[3] Karin Jongsma ,[4] Michael Dunn ,[5] Annie Sorbie,[6] Matthew McCoy ,[7] Jessica Bell,[8] Michael Burgess,[9] Haidan Chen,[10] Vicky Chico,[11] Sarah Cunningham-Burley ,[1] Julie Darbyshire ,[12] Rebecca Dawson,[13] Andrew Evans,[14] Nick Fahy,[15] Teresa Finlay,[15] Lucy Frith ,[16] Aaron Goldenberg,[17] Lisa Hinton,[18] Nils Hoppe,[19] Nigel Hughes,[20] Barbara Koenig,[21] Sapfo Lignou,[22] Michelle McGowan,[23] Michael Parker,[5] Barbara Prainsack,[21] Mahsa Shabani,[24] Ciara Staunton,[25] Rachel Thompson,[26] Kinga Varnai,[27] Effy Vayena,[28] Oli Williams,[29] Max Williamson,[30] Sarah Chan,[1] Mark Sheehan[5]

For numbered affiliations see end of article.

Correspondence to
Dr Sonja Erikainen, Centre for Biomedicine, Self and Society, Usher Institute, The University of Edinburgh, Edinburgh EH8 9YL, UK; sonja.erikainen@ed.ac.uk

Received 29 May 2020
Revised 17 August 2020
Accepted 20 August 2020

To cite: Erikainen S, Friesen P, Rand L, *et al*. *J Med Ethics* Epub ahead of print: [please include Day Month Year]. doi:10.1136/medethics-2020-106530

ABSTRACT

Population-level biomedical research offers new opportunities to improve population health, but also raises new challenges to traditional systems of research governance and ethical oversight. Partly in response to these challenges, various models of public involvement in research are being introduced. Yet, the ways in which public involvement should meet governance challenges are not well understood. We conducted a qualitative study with 36 experts and stakeholders using the World Café method to identify key governance challenges and explore how public involvement can meet these challenges. This brief report discusses four cross-cutting themes from the study: the need to move beyond individual consent; issues in benefit and data sharing; the challenge of delineating and understanding publics; and the goal of clarifying justifications for public involvement. The report aims to provide a starting point for making sense of the relationship between public involvement and the governance of population-level biomedical research, showing connections, potential solutions and issues arising at their intersection. We suggest that, in population-level biomedical research, there is a pressing need for a shift away from conventional governance frameworks focused on the individual and towards a focus on collectives, as well as to foreground ethical issues around social justice and develop ways to address cultural diversity, value pluralism and competing stakeholder interests. There are many unresolved questions around how this shift could be realised, but these unresolved questions should form the basis for developing justificatory accounts and frameworks for suitable collective models of public involvement in population-level biomedical research governance.

Population-level biomedical research involving large-scale biobanks, genetic data repositories and digital records offers new opportunities to improve population health, but also raises new challenges to traditional systems of research governance and ethical oversight. This is in particular because conventional models of informed consent and ethics review are unable to account for long-term storage, sharing and linkage, and use of data for future unspecified research.[1–4] Public concerns regarding the extent to which research institutions can be trusted to handle data ethically have contributed to a 'crisis in trust' in biomedical research.[5 6]

Partly in response to these concerns, various models of public involvement in population-level biomedical research initiatives are being introduced.[2 7–11] However, while public involvement is generally seen as a way to foster public trust, overcome barriers to individual consent, encourage accountable and responsible research, and address other ethical issues, the ways in which public involvement can or should meet these challenges are often unarticulated and poorly defined. Although it is increasingly agreed that we have reasons to support public involvement in population-level biomedical research, it is often unclear *what* those reasons are and how they connect with the challenges that involvement is intended to address. This lack of a normative framework for public involvement in governance poses a further barrier to addressing the concerns identified. It is clear that population-level biomedical research requires a shift in ethical thinking, compared with other forms of biomedical research that have focused more on the individual, towards new approaches to governance that can better account for the collective concerns and interests involved.

To explore these issues and areas of uncertainty, we convened the Public Involvement and Governance in Population-Level Biomedical Research workshop in Oxford, UK in January 2020. The workshop brought together an international, interdisciplinary and cross-sectoral group of 36 participants with expertise and stakes in relevant issues from different perspectives, including senior and emerging academic specialists, patient and public involvement professionals and patient advocates. Invitations were sent directly to several stakeholders and information about the event was also circulated online, so that those interested, but unknown to us,

could register and attend. As part of the workshop, we conducted a study using the World Café method, which is a group dialogue method that facilitates knowledge synthesis via group conversation and exchange of information and experiences, enabling the generation of a great deal of data in a relatively short amount of time.[12] Our World Café consisted of several rounds of small group table discussions, each focused on a different but related topic and involving a facilitator who also took notes on spreadsheets. Following small group discussions, all participants came together for a general discussion where overarching themes across the discussions were identified and evaluated.

The participants were asked to identify and discuss, first, key governance challenges in biomedical research involving biobanks, genetic repositories and digital databases, and, second, possibilities of public involvement in this mode of research and how involvement should be fostered, if at all.

The World Café generated 36 sheets of notes, which were subjected to qualitative thematic analysis, where the data were first analysed independently by four members of the research team, who then came together to merge the analyses and identify the most prominent cross-cutting themes. The findings and conclusions were then circulated to, and confirmed and approved by, all participants. This brief report discusses these four cross-cutting themes: the need for (1) moving beyond individual consent; (2) issues in benefit and data sharing; (3) delineating and understanding publics; and (4) clarifying justifications for public involvement.

MOVING BEYOND INDIVIDUAL CONSENT

A major theme in the discussions was informed consent, which has arguably been the dominant ethical doctrine in research involving humans.[13] Participants expressed concern about the 'consent obsession' suggesting that the traditional focus on consent is inappropriate in the context of population-level research. Participants highlighted the fact that existing consent models generally focus on individual rather than community or group consent. Ethical and governance challenges in population-level research, however, go beyond individual concerns and single research initiatives, raising the possibility of community or group harm.[14 15]

Many participants agreed that, as research gets bigger, involving larger amounts of linked population-level data, individual consent for population-level research is neither sufficient nor central to appropriate governance. A shift is needed to refocus both the theory and practice of research ethics oversight to suit the contemporary landscape of population-level research. Participants suggested that it is no longer sufficient to conceptualise research subjects as individuals taking part in research; instead, the subjects should be understood to be groups and communities, and attention should be paid to the social contexts in which the members of these groups live and the diverse interests they may express in relation to population-level biomedical research.

ISSUES IN BENEFIT AND DATA SHARING

Population-level biomedical research raises tensions, not just between individual versus collective interests, but also between different collective interests: for example, a community that has donated samples to a biobank versus the commercial entity that runs the biobank. In the context of enduring systemic national and global health inequities, participants repeatedly highlighted the importance of benefit sharing and priority setting. Some participants suggested that because biomedical ethics has focused on protecting research participants (ie, mitigating the risks of research), the focus should now be redirected more towards what can be done for communities that engage in research. The mechanisms to enable benefit sharing were widely debated, especially in contexts where commercial interests are closely tied to research outputs: one role for public involvement could be to provide input into how benefits ought to be shared with particular communities.

Social justice and data sharing were also tied to practical questions around resources, management and curation. Population-level biomedical research initiatives often rely on data sharing including across national and regional borders, but the regulatory frameworks in different national and regional contexts are diverse and requirements for accessing different databases, depositories and biobanks are inconsistent, presenting barriers to data linkage and collaborations. Some participants noted that there can be 'gatekeeping' behaviours around databases that result, among other things, from the conflicting interests and priorities of the various stakeholders in these research initiatives. In this context, it is unclear who should decide on data access and for what purposes it can be used.

DELINEATING AND UNDERSTANDING PUBLICS

Although there has been a shift away from singular conceptualisations of 'the public' towards recognition of publics as plural,[16 17] some participants observed that further scrutiny of the notion of 'publics' is also needed: which public(s) matter, when and why? This is connected with the question of the meaning and nature of 'public interest' and 'public benefit'. While these notions are often invoked and circulate in discussions and debates around research governance and public involvement, they are often poorly defined and rarely conceptually unpacked: as different individuals and social groups have different interests, diverse needs and perspectives on biomedical research and how it should be governed; it is unclear whose interests should count as 'public' interests, who benefits when 'public benefits' are distributed and who can legitimately answer these questions.

Participants observed that there is a need to generate better mechanisms for public involvement in population-level health research. Although such mechanisms have been developed at the level of individual research initiatives (eg, public deliberation methods), there are currently few *governance* mechanisms for making decisions collectively at a wider population level. Some participants observed that there are unresolved tensions between the centralisation of governance and local needs: high-level governance mechanisms aim to be impartial and universal, whereas local priorities and needs often require partial rather than equal benefit sharing.

DISTINGUISHING JUSTIFICATIONS FOR PUBLIC INVOLVEMENT

Despite a clear trend toward public involvement in the governance of population-level research, there remains a great deal of uncertainty and opacity about why public involvement should be undertaken.

In particular, 'legitimacy' was a noteworthy focus of discussion. Participants distinguished between descriptive and normative uses of the concept[18]: in practice, public involvement may be undertaken to increase public acceptance or to promote a more favourable perception of research, which would provide 'descriptive legitimacy', but does not necessarily ensure 'normative legitimacy'. The general feeling of the discussion was that

public involvement strategies should aim at normative legitimacy: involving the public for reasons other than the management of perception. Participants proposed a variety of justifying reasons, including: to understand public expectations, inform research initiatives or to ensure that research is conducted 'in the right way'. All of these were contested, and no single justification prevailed, and each raised further questions.

Participants also discussed transparency and public trust in research as an important justification for public involvement. In this context, trust was distinguished from trustworthiness[19–21]: public trust may be gained through public involvement, but the research institution may not be trustworthy. This was importantly seen as mapping onto the distinction between the descriptive and the normative senses of legitimacy: by ensuring that they were trustworthy, research institutions were in this sense demonstrating their normative legitimacy.

Another relevant distinction, within the idea of normative legitimacy, was between intrinsic and extrinsic (or outcome-oriented) justifications for public involvement. Extrinsic justifications focus on the ways in which public involvement can facilitate 'better' research outcomes, whereas intrinsic justifications focus on the research process and the role of public involvement in making the process 'right'.[22] However, while this distinction was seen as useful by many participants, their separation in practice looked difficult because the two rationales are often intertwined: public involvement in the research process may be valuable in its own right and enable better outcomes.

With this set of rationales for public involvement in play, it was apparent across the conversations that there is a need for more conceptual clarity about them and their application to specific publics, as well as how different involvement methods should map onto those justifications and how such methods should be evaluated.

CONCLUSION

Our aim in this report has been to provide a starting point for developing a more systematic ethical approach to the relationship between public involvement and governance in population-level biomedical research, showing connections, potential solutions and issues arising at their intersection. We acknowledge that there are vast literature on public involvement in health research, democratic deliberation and participation, and public engagement in decision-making, and we do not claim that our findings provide a comprehensive account of the governance challenges and justifications for public involvement. They do, however, indicate that embedding involvement in the governance of population-level biomedical research may require a novel approach. Our findings revealed how, in population-level research in particular, there is a pressing need for a shift away from conventional governance frameworks that are focused on the individual as the subject of rights, risks and benefits, and towards a focus on the collective, both in terms of foregrounding ethical issues connected with social justice, and in terms of developing ways to address cultural diversity, value pluralism and competing stakeholder interests. Existing perspectives from political theory and social science on public involvement and democratic participation could be usefully drawn on to inform further analysis on these issues. There are many unresolved questions around how this shift can or should be realised in theory and practice, including how consent should be re-conceptualised, the justifications for public involvement in research and how they map onto distinct mechanisms of involvement, how benefits should be distributed and different interests prioritised, and who should decide how these questions are answered. These questions should be taken up by those seeking to develop accounts and frameworks of public involvement in the governance of population-level biomedical research.

Author affiliations
[1]Centre for Biomedicine, Self and Society, Usher Institute, The University of Edinburgh, Edinburgh, UK
[2]Biomedical Ethics Unit, McGill University, Montreal, Quebec, Canada
[3]Harvard Medical School and PORTAL, Brigham and Women's Hospital, Boston, Massachusetts, USA
[4]Department of Medical Humanities, Julius Center, University Medical Center Utrecht, Utrecht University, Utrecht, The Netherlands
[5]The Ethox Centre and Wellcome Centre for Ethics and Humanities, University of Oxford, Oxford, UK
[6]Mason Institute for Medicine, Life Sciences and the Law, Edinburgh Law School, University of Edinburgh, Edinburgh, UK
[7]Department of Medical Ethics and Health Policy, University of Pennsylvania Perelman, Philadelphia, Pennsylvania, USA
[8]HeLEX, University of Oxford, Oxford, UK
[9]School of Population and Public Health, The University of British Columbia, Vancouver, British Columbia, Canada
[10]School of Health Humanities, Peking University, Beijing, China
[11]School of Law, University of Sheffield, Sheffield, UK
[12]Department of Primary Care Health Sciences, University of Oxford, Oxford, UK
[13]Usher Institute, University of Edinburgh, Edinburgh, UK
[14]Melanoma Patient Network Europe, N/A, UK
[15]Primary Care Health Sciences, University of Oxford, Oxford, UK
[16]Institute of Population Health Sciences, University of Liverpool, Liverpool, UK
[17]Department of Bioethics, Case Western Reserve University, Cleveland, Ohio, USA
[18]THIS Institute, University of Cambridge, Cambridge, UK
[19]Centre for Ethics and Law in the Life Sciences, Leibniz University, Hannover, Germany
[20]Observational Health Data Analytics/Epidemiology, Janssen Research and Development, Raritan, New Jersey, USA
[21]Department of Political Science, University of Vienna, Vienna, Austria
[22]NeuroSec and Wellcome Centre for Ethics and Humanities, University of Oxford, Oxford, UK
[23]Ethics Center, Cincinnati Children's Hospital Medical Center Department of Pediatrics, University of Cincinnati, Cincinnati, Ohio, USA
[24]Faculty of Criminology, Criminal Law and Social Law, University of Ghent, Ghent, Belgium
[25]Middlesex University School of Law, Middlesex University London, London, UK
[26]Research Institute for Ethics and Law, Swansea University, Swansea, UK
[27]OUH NHS FT and NIHR Oxford Biomedical Research Centre, Oxford, UK
[28]The Health Ethics and Policy Lab, University of Zurich, Zurich, Switzerland
[29]King's College London and THIS Institute, University of Cambridge, Cambridge, UK
[30]University of Oxford, Oxford, UK

Twitter Sonja Erikainen @SonjaErikainen, Sarah Cunningham-Burley @Sarah_C_Burley and Lucy Frith @lucy_frith

Contributors SE and KRJ designed and lead the study. Chan, MD, SE, PF, KRJ and LR collected the data, and Chan, SE, PF and MD conducted the data analysis. SE drafted the manuscript, and Chan, MD, PF, KRJ, MM, LR, Sheehan and AS all contributed substantially to the writing. Sheehan and Chan were the senior leads on the wider project from which this paper derives.This paper reports the findings and conclusions from a collaborative, participatory World Café study. All authors participated in and collaborated on the study, and in so doing contributed substantially to the intellectual content of this paper. All authors approved the final version.

Funding This research was principally funded by the Wellcome Trust small grant 'Public Involvement in the Governance of Population Level Research' (219383/Z/19/Z). The authors would also like to acknowledge the stimulus of the Wellcome Trust Centre for Biomedicine, Self and Society Award (209519/Z/17/Z), the Wellcome Centre for Ethics and Humanities Award (203132/Z/16/Z), and the Oxford NIHR Biomedical Research Centre, all of which facilitated this work.

Competing interests None declared.

Patient consent for publication Not required.

Ethics approval Ethical approval was granted by the MS IDREC University of Oxford, under reference R64628/RE001.

Provenance and peer review Not commissioned; externally peer reviewed.

Data availability statement No data are available.

ORCID iDs
Sonja Erikainen http://orcid.org/0000-0002-1442-3050
Karin Jongsma http://orcid.org/0000-0001-8135-6786
Michael Dunn http://orcid.org/0000-0002-5603-6200
Matthew McCoy http://orcid.org/0000-0002-5273-3877
Sarah Cunningham-Burley http://orcid.org/0000-0002-0009-7653
Julie Darbyshire http://orcid.org/0000-0002-7655-1963
Lucy Frith http://orcid.org/0000-0002-8506-0699

REFERENCES

1 Knoppers BM, Leroux T, Doucet H, *et al*. Framing genomics, public health research and policy: points to consider. *Public Health Genomics* 2010;13(4):224–34.
2 Joly Y, Dalpé G, So D, *et al*. Fair shares and sharing fairly: a survey of public views on open science, informed consent and participatory research in biobanking. *PLoS One* 2015;10(7):e0129893.
3 Sanderson SC, Brothers KB, Mercaldo ND, *et al*. Public attitudes toward consent and data sharing in Biobank research: a large multi-site experimental survey in the US. *Am J Hum Genet* 2017;100(3):414–27.
4 Mostert M, Bredenoord AL, Biesaart MCIH, *et al*. Big data in medical research and Eu data protection law: challenges to the consent or anonymise approach. *Eur J Hum Genet* 2016;24(7):956–60.
5 Haddow G, Cunningham-Burley S, Bruce A, *et al*. Generation Scotland: consulting publics and specialists at an early stage in a genetic database's development. *Crit Public Health* 2008;18(2):139–49.
6 McGuire AL, Majumder MA, Villanueva AG, *et al*. Importance of participant-centricity and trust for a sustainable medical information commons. *J Law Med Ethics* 2019;47(1):12–20.
7 Beaton A, Hudson M, Milne M, *et al*. Engaging Māori in biobanking and genomic research: a model for biobanks to guide culturally informed governance, operational, and community engagement activities. *Genet Med* 2017;19(3):345–51.
8 Boers SN, Bredenoord AL. Consent for governance in the ethical use of organoids. *Nat Cell Biol* 2018;20(6):642–5.
9 Burgess MM. From 'trust us' to participatory governance: Deliberative publics and science policy. *Public Underst Sci* 2014;23(1):48–52.
10 Chalmers D, McWhirter RE, Nicol D, *et al*. New avenues within community engagement: addressing the ingenuity gap in our approach to health research and future provision of health care. *J Responsible Innov* 2014;1(3):321–8.
11 Buyx A, Del Savio L, Prainsack B, *et al*. Every participant is a Pi. citizen science and participatory governance in population studies. *Int J Epidemiol* 2017;46(2):dyw204–384.
12 Brown J, Isaacs D. *The world Café: a resource guide for hosting conversations that matter*. Berrett-Koehler Publishers, 2005.
13 Manson NC, O'Neill O. *Rethinking informed consent in bioethics*. Cambridge Univeristy Press, 2007.
14 Koenig BA. Have we asked too much of consent? *Hastings Cent Rep* 2014;44(4):33–4.
15 Grande D, Mitra N, Shah A, *et al*. The importance of purpose: moving beyond consent in the societal use of personal health information. *Ann Intern Med* 2014;161(12):855–62.
16 Felt U, Fochler M. Machineries for making publics: inscribing and de-scribing publics in public engagement. *Minerva* 2010;48(3):219–38.
17 Hull SC, Wilson Diné DR. Beyond Belmont: ensuring respect for AI/AN communities through tribal IRBs, laws, and policies. *Am J Bioeth* 2017;17(7):60–2.
18 Buchanan A, Keohane RO. The legitimacy of global governance institutions. *Ethics Int Aff* 2006;20(4):405–37.
19 O'Neill O. *Autonomy and trust in bioethics*. Cambridge University Press, 2002.
20 O'Neill O. Linking trust to Trustworthiness. *Int J Philos Stud* 2018;26(2):293–300.
21 Hawley K. Trustworthy Groups and Organisations. In: Faulkner P, Simpson T, eds. *The philosophy of trust*. Oxford University Press, 2017: 230–50.
22 McCoy MS, Warsh J, Rand L, *et al*. Patient and public involvement: two sides of the same coin or different coins altogether? *Bioethics* 2019;33(6):708–15.