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The Role of Participants in a Medical Information Commons

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In 2015, Francis Collins and Harold Varmus highlighted the importance of "new models for doing science that emphasize engaged participants and open, responsible data sharing."[1] Others have explicitly tied the success of efforts to increase data sharing and create a large-scale, longitudinal, multi-purpose informational resource or medical information commons to participant engagement and participant-centricity.[2] As Charlotte Hess and Elinor Ostrom have observed, efforts to create common-pool or shared resources are most likely to endure and thrive if governance is "organized in a nested structure with multiple layers."[3] Thus, the term "medical information commons" or "MIC" may appropriately be used to describe both the networked space or ecosystem (singular) in which data sharing occurs and also the initiatives (plural) collecting and broadly sharing diverse kinds of data for research and other purposes, and attempts to engage participants and attend to their interests, values, and concerns may be assessed at multiple levels.[4]

Supporters of participant engagement in biomedical research cite a range of intrinsic and instrumental goals. They justify engagement in terms of respect for persons and autonomy, democratic norms, and considerations of social justice. They also mention benefits such as improved recruitment and retention and increased public buy-in.[5] Since an MIC involves longitudinal collection of sensitive personal information from large numbers of people, the potential for engagement to advance these goals may be particularly salient to MIC sponsors. However, some scholars have drawn attention to the potentially problematic implications of public engagement rhetoric, such as the contestable implication that there is a civic duty to participate in government-sponsored research initiatives and other initiatives promoted as serving the common good.[6]

While there is no consensus definition of "participant engagement" in the literature, there is broad agreement that the existence of channels for *bidirectional* communication between researchers and participant representatives is a necessary feature of participant engagement.[7] Conceptions of participant (or patient or community or public) engagement as a continuum from weak, limited forms of interaction to robust, comprehensive forms of interaction — such as community-based participatory research — are common.[8] Although there is typically an implicit normative assumption that movement along the continuum toward greater engagement is desirable, in some cases determination of the optimal degree of engagement is specified as requiring the input of those affected. The term "partnership" is often used to indicate the point on the continuum where leadership is truly shared between participant representatives and researchers or other experts.[9] Advocates and scholars such as Barbara J. Evans have identified a point beyond partnership where participants assume control over their own data, while perhaps enlisting experts and research institutions as consultants and allies.[10]

We present findings from interviews with diverse expert stakeholders involved in existing and emerging public and private data-sharing initiatives or the creation of relevant ethical and legal frameworks. Interviewees were selected to represent six sectors contributing to the creation of an MIC: academia (including investigators conducting biomedical research, law and policy research, and research on participant engagement), non-governmental organizations (including leaders of patient advocacy organizations and research foundations), technology companies, government, laboratories, and healthcare systems. The interviews began broadly, offering interviewees an opportunity to articulate their own vision of an MIC and assess the current landscape. The interviews then focused on specific areas of interest, including exploring interviewees' perspectives on the role of participants, meaning the people from whom data derive. While interviews were framed in terms of an MIC, much of this content is relevant to the participant role in biomedical research more generally. Building on our findings, we conclude by reflecting on the political aspects of participant engagement and efficiency concerns that we believe are worthy of further consideration by the bioethics and policy communities.

Methods

The research presented here is a sub-analysis of semi-structured interviews conducted with 41 expert stake-holders. Two interviews involved two interviewees; the remainder involved one interviewee. The strategies for sample selection and data collection are detailed in full elsewhere.[11] Interviews began with a question soliciting a definition of "medical information commons." Input was then sought on the following working definition: *Medical information commons are networked environments in which diverse sources of health, medical, and genomic data on large populations become broadly available for research use and clinical applications*. Interviewees were also asked for their views on the current landscape and barriers to MIC creation. The interviews then shifted to particular areas of interest. This manuscript presents a thematic content analysis of responses to the interview question, *What role should the people whose data populate the medical information commons play?* (and any reflection on participant role in responses to other questions). This open-ended question was sufficient to prompt an expansive response from many interviewees. The interview

guide did include a series of probes to draw out interviewees where necessary, including: *How, if at all, should they be involved? What are the challenges to engaging individuals or communities to carry out the role you envision for them?*

The initial question used neutral language such as "people" (the word "participant" is currently in favor in the research ethics literature precisely because it implies an active rather than passive role) and intentionally omitted the use of the term "participant engagement" in order to avoid steering interviewees to traditional conceptions of engagement. We use the terms "participant" and "participant engagement" in this paper because "people whose data populate the MIC" is an awkward construction and because the majority of interviewees envisioned an active role for the people whose data populate an MIC and themselves employed the terms "participant" and "participant engagement."

Coding and Analysis

Three of the authors (JB, MM, and AV) were responsible for coding and analysis of interview transcripts. The development of the codebook drew on a preliminary analysis of 11 transcripts. This initial review and assessment of the codebook led to the creation of additional codes that captured themes that emerged from holistic analysis of the fuller set of transcripts. Responses to the central question, plus additional related commentary on participant engagement stated in response to other questions, were coded by two coders using the NVivo 11 Pro software program. Coding differences were discussed by the coders until consensus was reached. Code reports were generated and reviewed for further analysis.

Results

The distribution of interviewees by sector is shown in Table 1. All interviewees favored expanding the role of participants beyond simply providing one-time informed consent, except for one interviewee who stated, "I think that once somebody agrees to participate, they want to be left alone." (P25) Here we explore further findings in two areas. First, while some interviewees focused on ways to increase engagement of participants as individuals, others focused on ways to increase engagement of participants as a collective. Second, many interviewees described challenges they had experienced or anticipated in seeking to expand the role of participants.

I. Individual Focus or Collective Focus

When prompted to reflect on the role of participants, some interviewees first or only discussed interactions with participants as individuals, while others first or only discussed the role of participants as a collective, particularly through representation in governance structures. The distrinction between participants as individuals versus a group is important to explore, as there are potential tensions between the two orientations. Illustrative quotes related to individual and collective focus are displayed in Table 2 and summarized in the following sections.

As shown in Table 2, interviewees who emphasized the role of participants as individuals identified a number of measures to enhance this role in the context of an MIC. A relationship between participants and an MIC was stated or assumed as a basis for MIC

interactions with participants; interviewees did not delve into details such as the precise role of intermediaries where data is flowing into an MIC through a number of primary research studies. Many interviewees noted opportunities for an MIC to return information to individuals, in some cases framing return of information to participants as an obligation. Several interviewees mentioned empowering individuals to remove data, or change their consent status over time consistent with a dynamic, granular consent paradigm (with the Platform for Engaging Everyone Responsibly as one example). One interviewee referenced an instrumental justification: the anticipated positive impact this would have on individuals' willingness to share data. A few interviewees talked about individual entry of phenotypic information or involvement in data curation, and one described how technological advances would allow individuals to take control over an increasingly large and important set of health-relevant data. Finally, several interviewees spoke of individuals having a right to access information about them and/or the desirability of having pathways for participants to suggest research questions or engage in citizen science using de-identified data. However, one interviewee objected to creating such pathways because of the additional work involved and the fact that participants may lack knowledge and expertise.

Other interviewees described measures to enhance the role of participants in collective terms. Several interviewees felt that it was important that participants as a collective be involved in governance, with some specifying a leadership role on the steering committee of an MIC. One interviewee went on to state that in the context of an MIC, this collective approach would be the most appropriate way of realizing respect for autonomy. A few interviewees outlined more modest forms of collective engagement as potentially acceptable, such as focus groups or opinion surveys, or stated that they preferred community advisory boards (with clear delineation of their non-decision-making role) to participant involvement in a steering committee or board. A few interviewees advocated for an involved-in-all-aspects role for participants, beginning with the involvement of representatives of potential participants as a collective (or particular communities) in shaping research priorities and research design and extending to representation of participants as a collective in governance and in the conduct of research, including opportunities to engage in citizen science (i.e., contribute research ideas or directly query databases), but also encompassing greater empowerment of participants as individuals.

II. Challenges

Interviewees explicitly or implicitly referenced challenges they experienced or foresaw in bringing reality into line with their view of the role participants should play in an MIC. Here we focus on five of those challenges and proposed strategies for addressing those challenges (where offered).

COST—Interviewees with experience in participant engagement noted increased recognition of the resources required. They cited initiatives such as the Patient-Centered Outcomes Research Institute (PCORI) as helping to normalize research expenditures on participant engagement. Yet they reported continuing resistance related to cost, for example: "I can have a budget that's approved by the funder and when I submit the invoice for my community partner to be paid I still get the 'Well, why are you paying them so much?' kind

of response." (P24) One strategy was simply to draw attention to the disjunction between rhetoric and budget: "How much are we spending on engagement versus technology systems and research protocols and that sort of stuff? I think we're going to find out 1% of the budget goes to these things and that is unconscionable and won't give us the results we want." (P13)

REPRESENTATION—Interviewees who considered or advocated for some representation of participants on governance bodies or other limited-membership groups described challenges related to selecting individuals who are truly representative of a cohort or population, and meaningfully engaging those representatives. Some linked the challenge of representative selection to scale: "The challenge is when you have 100,000 research participants… [, and] it's not going to be too far where it's going to be 10 million. How do three people…on a committee actually represent that?" (P19) Regarding representative selection, several interviewees noted that participants who volunteer or are selected as representatives are often atypical because they must have the time to participate and a likely unusual level of interest in research relative to other priorities. Related to this, some contrasted professional advocates with the "common (wo)man" or "person on the street." One interviewee considered selection of representatives from and for groups with schizophrenia or severe depression or severe cognitive impairment especially challenging. Another contrasted Native American nations, which have sovereignty and clear authority structures, with more fragmented communities lacking clear authority structures. A few interviewees mentioned the possibility of experimenting with some kind of election process.

Regarding meaningful engagement, institutional review boards (IRBs) were twice mentioned as examples of engagement that failed the meaningfulness test. One interviewee talked about the limited number of community members on IRBs, implying that critical mass in governance structures is necessary for meaningful engagement. The other brought up aspects of what has been called "epistemic inequity"[12]:

> In my experience, community members on IRBs are often overwhelmed by the other members of the IRBs with their greater expertise. And community members or representatives are not often given much credence. They're not listened to. They're just sort of tolerated.
>
> (P32)

Two interviewees reported success with a strategy that amounts to (in our words) multiple kinds of participant voices, adequately supported. This strategy embraces outreach to individuals who have been selected to lead community organizations and to those who put themselves forward as community leaders. It would also involve engagement with constituents outside the control (and presence) of formal and informal leaders. As to "adequately supported," these interviewees believed that meaningful engagement must include education, e.g.: "The typical individual doesn't walk in off the street…able to provide oversight in some sort of commons that also includes researchers and providers and whomever else is going to be a part of it. They need to be prepared and trained and sometimes they need to be compensated, especially if it's a significant amount of their time that's going to be involved." (P24)

PERCEIVED INEFFICIENCY—One interviewee reported running up against the view that it would be "unmanageably complicated" to give individuals more say in how their data are used, alongside complacency about the adequacy of traditional research consent: "There's sort of a puzzled look on people's [project leaders'] faces, and I don't know… they'll hold up a piece of paper and say, 'Well, we have the consent form right here.'" (P10) Another interviewee cited pressures to launch programs and do so under budget as working against a more robustly participant-centric vision. At the same time, several suggested that efficiency considerations had some legitimacy, expressing concerns about the feasibility of a framework that offers individuals more control. For example, an interviewee who expressed reservations about general or broad consent added: "On the other hand, it is a logistical nightmare to have all kinds of levels [of consent]… Not that the software can't handle it…, but it adds substantially to the information management challenge." (P31)

Desire/Ability to Engage: Several interviewees observed that people vary in both their desire and their ability to engage. For some, this was not a significant problem, so long as this variation was acknowledged and accommodated. For example:

> [T]here's like the concerned cohort that are just hyper on everything, right? They're your big wearables people, that kind of thing. Retirees who have a lot of time to answer emails, I don't know. If somebody started pinging me all the time they wanted to access my data at 23andMe, I would get on the phone and say, "Take me off your list. I don't want to be bothered anymore."
>
> (P36)

Others saw the potential to increase health disparities, insofar as only the relatively privileged would benefit (both directly and downstream) from a system designed to promote inclusion and deliver respect and rewards for participation via interactive mechanisms: "If we are starting a system in which it takes a high degree of sophistication and wherewithal [i.e., resources, including unencumbered time] and education and those sorts of other things to make full use of it, then we're going to have an aggravated chasm between the haves and the have-nots." (P32) Also, while one interviewee pointed to the success of patient advocacy organization-driven efforts as important and inspiring (e.g., multiple myeloma, cystic fibrosis), another suggested that the success of rare disease groups in particular could be misleading: "I think it's a model that works very well in the rare disease community because they're very savvy, very active, and rightly so, very concerned parents, and families, and extremely democratic in sharing their information… [But] to paint [MCI] participants with the same brush as having all those tools?…" (P1) In short, this interviewee questioned the generalizability of engagement strategies based on single-disease focused initiatives.

INAUTHENTIC ENGAGEMENT—A few interviewees were especially critical of efforts to engage participants as a means to an outcome unrelated to respect for individuals and communities. One interviewee contrasted conversations about engagement as a means to increase recruitment and retention (using words like "convincing," "selling," and getting "buy-in") versus conversations focused on how to achieve true "partnership" or true involvement through leadership roles or participation in governance. Several interviewees described programs that used desirable language, at least in their public-facing

communications, but in practice failed to live up to it. Finally, as noted above, several interviewees held up community-based participatory research as an ideal, with community needs and concerns a primary consideration even to the point of leading to a rethinking of research priorities, or to policy changes that could add costs or introduce delays.

Discussion

Some of the expert stakeholders we interviewed have written and spoken publicly about data sharing, the design of data-sharing initiatives, and MIC policy development, often touching on participant role; many of the themes we highlight here are already present in the literature and are also reflected in the emphasis on participant or community engagement in major grant-making initiatives like PCORI. Nonetheless, we believe political aspects of differing approaches to participant engagement and efficiency concerns have not received the attention they merit. In making this case, we build on the results reported above, but we also draw on our own experience with participant and public engagement.

Political Aspects of Participant Engagement

Some accounts of participant engagement focus on advances in *technology* and on participants as *individuals*.[13] Equal attention should be given to *relationships* and participants as a *collective*. While technology can support health-related "e-communities,"[14] our interviewees commented on the importance of off-line, face-to-face interactions among participants and between participants and researchers. Further, they recognized the importance of engaging participants as members of a collective despite the difficulties presented. We note that keeping participants isolated as individuals may serve to enhance the control of researchers and minimize participant voice and power – even if that is not the intention of those developing individual-focused engagement strategies. Yet, as Blassime and Vayena note, for large-scale "precision medicine" cohorts "the kind of communitarian bonds that a participatory ethos is supposed to capture and to promote are either yet to be formed or bound to compete for the opportunity to have a say regarding the governance of the cohort."[15] They suggest offering participants opportunities to become actively involved in governance structures at enrollment and leaving the composition and agendas of governance bodies open to reconfiguration by participants.

Blassime and Vayena's work is immensely valuable but stops short of confronting all the challenges associated with selecting and then empowering participant representatives in the context of an MIC, especially in the U.S. One challenge is the problem of representation. Sankar and Parker mention expertise, diversity, equity, inclusiveness, and convenience as possible criteria in the selection of participant representatives for cohorts such as All of Us. They then pose a question about the moral basis for decisions regarding selection criteria.[16] No pat answers emerged from our interviews, but we did hear from experts who were cognizant of the complexities and had practical experience confronting them. One clear message was rejection of what might be called a politics of scarcity (e.g., fixation on how to fill one or two designated "participant" slots on a steering committee). Multiple channels for two way communication should be created from the planning stage onward. Intrinsic and

instrumental ethical considerations underlie the importance of both diversity and equity in selecting individuals to represent collectives of participants or potential participants.[17]

As for expertise, we note that it is itself diverse and dynamic. A person who has learned important lessons from her experience advocating for the interests of her community in the face of numerous obstacles has expertise, as does a person who has learned as much as most clinicians or scientists about the particular health condition affecting him or his child. Lay people, especially those from groups that have experienced and continue to experience discrimination and exclusion, need support to develop and convey their expertise in a manner that will be recognized and valued by other stakeholders. For that matter, the ability and willingness of minority and underserved populations to engage at all will be contingent upon efforts to understand and address barriers to active participation (e.g., lack of resources), and to shape engagement to meet their needs rather than using them solely to advance researcher agendas.[18] Reliance on solicitations of interest for candidate identification, and convenience as a core selection principle, works against diversity and equity and narrows the kinds of expertise brought to the table.

Other pitfalls not explored in our interviews but relevant to engagement as a political project include ignoring intra- and inter-group differences and papering over tensions. For example, individuals rightly resent being asked to give voice to the perspective and concerns of an entire racial/ethnic group, and from the other side, question the ability of someone very unlike them in most respects to represent them based on a single commonality.[19] Some are skeptical about motivations and question whether anyone can truly represent them.[20] In addition, individuals who are directly affected by a health condition may have a different perspective than family members or caregivers. And patient advocates may have a different perspective than patients “on the street.” Patient advocates may differ in the degree to which they prioritize privacy versus open science, or developing new treatments versus comparative effectiveness research, or support services versus discovery research. Those who identify as “healthy” (or are recruited because others characterize them that way) may have concerns that differ from those affected by a specific condition or identifying as in “poor health.” Strategies such as building in repeated opportunities for face-to-face communication over time can help to create the bonds of trust within and across groups of participants and between participants and other stakeholders, including researchers and funders, that make uncomfortable but important conversations possible and productive. We are hopeful that some of those uncomfortable conversations will address the ways in which traditional research agendas and priorities have failed certain groups, such as American Indian and Alaskan Native communities and African Americans (especially as these identities intersect with sources of disadvantage such as poverty and powerful forces such as structural racism).[21]

Indeed, participant engagement cannot fulfill its quotidian or transformative promise without significant attention to process and to the promotion of epistemic equity. Ideally processes of engagement are designed to promote mutual understanding of the reasons or stories that underlie and explain positions, as in deliberative democracy approaches.[22] In studies in the IRB context, non-institutional members often report feeling intimidated and attribute this feeling to their unfamiliarity with technical terms and jargon. There is evidence that

professionals have a tendency to discount the input of others (lay people and professionals from other disciplines), which may be based in part on differing linguistic habits or privileging scientific paradigms of explanation. Early expressions of opinion from IRB chairs or other professionals ascribed special authority may preempt alternate points of view. As Wenner notes, "[t]he upshot is that a process…to bring multiple, divergent perspectives to bear on difficult and nuanced problems can very easily become one that instead overlooks or marginalizes precisely those outlier views to which it is intended to give voice."[23] Broadly, failed engagement reflects lack of communication and collaboration to address power differentials, not simply or primarily knowledge deficits. Nonetheless, education for all stakeholders may have value within a portfolio of strategies. Participant representatives would benefit from technical knowledge that reduces self- and third party-appraisals of incompetence.[24] In addition, knowledge about crucial conversations and enhancing storytelling as a mode of communication could equip participants to navigate tensions and relate their real-world knowledge to issues, while also providing them with skills of general value. Researchers and other stakeholder groups would benefit from education about team science as well as epistemic inequity and steps for minimizing it. The goal would be a long-term project of mutual learning.

We have highlighted complexities that may seem daunting to institutions and investigators who are receptive to the case for participant involvement but lack experience in this area. One possibility is to look for collaborators in schools of public health or community organizations with a health mission and a track record of successful engagement of constituents. Consultation with leaders from initiatives such as PCORI (or relevant component projects) or the Health Care Systems Research Network (HCSRN) Patient Engagement in Research Scientific Interest Group may also be valuable, especially as there is now a growing literature reporting "lessons learned" from these efforts.[25] Champions should be prepared to advocate with institutional leaders and funders for the resources necessary to do engagement well. As suggested by one of our interviewees, if budgets are a reflection of priorities, then these stakeholders should acknowledge that a financial investment in participant engagement is one marker for authentic commitment.

Efficiency Concerns

It is quixotic to expect researchers in the commercial sector to embrace engagement without considering its impact on efficiency (i.e., the amount of time and effort required to accomplish objectives), and efficiency is also a legitimate concern when scarce public resources are involved. Still, given the intrinsic and instrumental goals of participant engagement, efficiency as an excuse to avoid putting serious thought and effort into how best to engage participants should be resisted. It is certainly not a bad thing that attention is finally being paid to the long-term efficiency gains from meaningful participant engagement, including improved retention, better quality data, and a focus on the outcomes that are most important to patients/participants.[26] And yet to focus on such gains is to risk favoring the instrumental over the intrinsic and the pedestrian (i.e., business as usual, only a bit better) over the transformative. Adopting a more inclusive and experimental perspective, Kelty and Panofsky point out that even as engagement brings new risks such as "loss of control and

rise of contention in the research process," it offers "many intriguing possibilities for new forms of collaboration, data, knowledge production, funding, and serving public needs."[27]

Our results, including efficiency-focused reservations about dynamic, granular consent, do help make the case for beginning with experimentation rather than detailed mandates. (Innovative approaches can be tested before being promoted as best practices or required of all the initiatives that make up an MIC.) No one we interviewed articulated a goal of rapidly creating new layers of regulation and bureaucracy around participant engagement. Although models exist, there are no legal requirements for engagement, beyond requirements from funders, and rules that each IRB include a community representative – an ill-defined and problematic role (as noted by several interviewees).[28] There is also tension between the IRB mandate to protect individual participants and procedures that endorse attention to community concerns. For experimentation to yield maximal value, it will be important to develop appropriately nuanced approaches to evaluating the effectiveness of participant engagement efforts in achieving intrinsic and instrumental goals and systematically assess benefits and costs.[29] In the literature it is still uncommon to find evaluations of participant engagement, and published evaluations are usually limited to measurements of levels of engagement (such as the number of people initiating communication with researchers).[30] Given the importance of engagement and the resources at stake, it is time to build a more robust evidence base.[31]

Limitations of our research include our recruitment of individuals involved in data sharing, either directly through involvement in data-sharing initiatives or through their work on relevant ethical and legal frameworks. While a few of our interviewees harbor significant reservations about aspects of the data-sharing enterprise, most are committed to the goal of creating an MIC. Also, although we framed an MIC as a resource available for purposes other than biomedical research (e.g., clinical care), most interviewees discussed an MIC within the context of research rather than exploring connections to learning health care systems or public health.

Conclusion

While the challenges described by expert stakeholders are significant, enthusiasm for expanding the participant role in the context of an MIC remains. Each initiative within an MIC ecosystem is a *potential* laboratory of democracy and space for deliberation about how best to align medical, scientific, and social justice goals. Openness about successes and struggles will be critical to mutual learning and development of best practices for participant engagement.

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8. The classic work on engagement as a continuum is Carman KL et al., "Patient and Family Engagement: A Framework for Understanding the Elements and Developing Interventions and Policies," Health Affairs 32, no. 2 (2013): 223–229 (focus on health care context). An engagement "roadmap" building on this framework is available at <https://patient-familyengagement.org> (last visited January 10, 2019). [PubMed: 23381514] See also Ellis and Kass, *supra* note 5; Aungst, Fishman, and McGowan, *supra* note 6; Seid M, Margolis PA, and Opipari-Arrigan L, "Engagement, Peer Production, and the Learning Healthcare System," JAMA Pediatrics 168, no. 3 (2014): 201–202; [PubMed: 24446048] Nabatchi T, "Putting the 'Public' Back in Public Values Research: Designing Participation to Identify and Respond to Values," Public Administration Review 72, no. 5 (2012): 699–708. The continuum is critical to understanding the relationship between participant engagement and participant-centricity. While a project can employ limited participant engagement strategies without being participant-centric if these strategies in no way meaningfully empower participants, as one moves along the participant engagement continuum initiatives become increasingly participant-centric. It is hard to imagine an initiative that claims to be participant-centric having no participant engagement.
9. E.g., Blassime and Vayena, *supra* note 2.
10. E.g., Evans B, "Barbarians at the Gate: Consumer-Driven Health Data Commons and the Transformation of Citizen Science," American Journal of Law & Medicine 42, no. 4 (2016): 651–685. [PubMed: 29086656]
11. See Bollinger, *supra* note 2.
12. See, e.g., Jongsma K, Spaeth E, and Schicktanz S, "Epistemic Injustice in Dementia and Autism Patient Organizations – An Empirical Analysis," American Journal of Bioethics Empirical Bioethics 8, no. 4 (2017): 221–233.(Drawing on the work of Miranda Fricker, the authors define epistemic injustice as "a wrong done to someone in their capacity as a knower." It includes testimonial injustice, "the devaluation of the credibility of persons or groups by a hearer due to negative and culpable prejudices," and hermeneutical injustice, deprivation "of opportunities to contribute…social experiences to the collective understanding on an event or condition."). See also Wenner DM, "Barriers to Effective Deliberation in Clinical Research Oversight," HCE Committee Forum 28, no. 3 (2015): 245–259.
13. Kaye et al., *supra* note 5; Feeney O et al., "Genuine Participation in Participation-Centred Research Initiatives: The Rhetoric and the Potential Reality," Journal of Community Genetics 9, no. 2 (2018): 133–142; [PubMed: 29064073] Walker DM et al., "Information Technology to Support Patient Engagement: Where Do We Stand and Where Can We Go?" Journal of the American Medical Informatics Association 24, no. 6 (2017): 1088–1094; [PubMed: 28460042] Winickoff DE, Jamal L, and Anderson NR, "New Modes of Engagement for Big Data Research," Journal of Responsible Innovation 3, no. 2 (2016): 169–177; for a balanced overview of technology-enabled ("high-tech") and in-person ("high-touch") approaches to engagement, see Lavallee DC et al., "Stakeholder Engagement in Patient-Centered Outcomes Research: High-Touch or High-Tech?" Expert Reviews in Pharmacoeconomic Outcomes Research 14, no. 3 (2014): 335–344.
14. Walker et al. and Lavallee et al., *supra* note 13.
15. Blassime and Vayena, *supra* note 2. Other recent work that is attentive to the political dimensions of large assemblages of genomic and other data includes Kraft SA, Cho MK, Gillespie K et al., "Beyond Consent: Building Trusting Relationships With Diverse Populations in Precision Medicine Research," American Journal of Bioethics 18, no. 4 (2018): 3–20; Mamo LA, Browe DK, Logan HC, and Kim KK, "Patient Informed Governance of Distributed Research Networks: Results and Discussion from Six Patient Focus Groups," AMIA Annual Symposium Proceedings Archive (2013): 920–929; O'Doherty KC et al., "From Consent to Institutions: Designing Adaptive Governance for Genomic Biobanks," Social Science and Medicine 73, no. 3 (2011): 367–374; [PubMed: 21726926] Woolley et al, Reardon, and Prainsack, *supra* note 6.
16. Sankar and Parker, *supra* note 7.
17. Ellis and Kass, *supra* note 5.
18. Katsansis, *supra* note 7; see also Prainsack, *supra* note 6, especially pp. 198–201.
19. Regarding the general need to attend to intersectionality, including the interaction of race and gender in the experience of discrimination, see the work of Kimberlé Crenshaw, e.g., Crenshaw K, "Demarginalizing the Intersection of Race and Sex: A Black Feminist Critique of

Antidiscrimination Doctrine, Feminist Theory and Antiracist Politics" University of Chicago Legal Forum 1989, no. 1 (1989), Article 8, available at <http://chicagounbound.uchicago.edu/uclf/vol1989/iss1/8> (last visited January 10, 2019). Regarding intragroup differences and tensions in the context of research in particular, see Reardon, p. 195, Jongsma, Spaeth, and Schicktanz, *supra* note 12, Kaplan et al., *supra* note 7.

20. See S.A. Kraft, M.K. Cho, K. Gillespie et al., *supra* note 15.
21. See, e.g., Garrison NA, "Cases of How Tribes are Relating to Genetic Research," available at <http://genetics.ncai.org/what-do-tribes-think-about-genetics-research.cfm> (last visited January 10, 2019);Bitsóí (Diné) LL, "Enhancing Genomic Research Through a Native Lens," available at <http://genetics.ncai.org/enhancing_genomic_research.cfm> (last visited January 10, 2019);Washington H, Medical Apartheid: The Dark History of Medical Experimentation on Black Americans from Colonial Times to the Present (New York: Doubleday, 2006);Reardon, *supra* note 6.
22. Nabatchi, *supra* note 8.
23. See Wenner, *supra* note 12, at 253.
24. Ellis and Kass, *supra* note 5.
25. Related to PCORI, see Kim KK and Helfand M, "Engagement in PCORnet Research Networks," Medical Care 56, no. 10, Suppl 1, (2018): S1–S2; [PubMed: 30074945] Selby JV, Grossman C, Zirkle M, and Barbash S, "Multistakeholder Engagement in PCOR-net, the National Patient-Centered Clinical Research Network," Medical Care 56, no. 10, Suppl 1 (2018): S4–S5; [PubMed: 30074942] Wilkins CH, "Effective Engagement Requires Trust and Being Trustworthy," Medical Care 56, no. S6–S8 (2018); [PubMed: 30015725] Fagerlin A, "Learning from Others: Lessons for Improving Collaborations Between Stakeholders and Researchers," Medical Care 56, no. 10, Suppl 1 (2018): S9–S10; [PubMed: 30074944] Faulkner M et al., "Exploring Meaningful Patient Engagement in ADAPTABLE (Aspirin Dosing: A Patient-centric Trial Assessing Benefits and Long-Term Effectiveness)," Medical Care 56, no. 10, Suppl 1 (2018): S11–S15; [PubMed: 30074943] Nowell WB, Curtis JR, and Crow-Hercher R, "Patient Governance in a Patient-Powered Research Network for Adult Rheumatologic Conditions," Medical Care 56, no. 10, Suppl 1, (2018): S16–S21; [PubMed: 30074946] Boyer AP et al., "A Multilevel Approach to Stakeholder Engagement in the Formulation of a Clinical Data Research Network," Medical Care 56, no. 10, Suppl 1 (2018): S22–S26; [PubMed: 30074947] Haynes SC et al., "Engaging Stake-holders to Develop a Patient-centered Research Agenda: Lessons Learned from the Research Action for Health Network (REACHnet)," Medical Care 56, no. 10, Suppl 1 (2018): S27–S32; [PubMed: 30074948] Chung AE et al., "Crohn's and Colitis Foundation of America Partners Patient-Powered Research Network: Patient Perspectives on Facilitators and Barriers to Building on Impactful Patient-Powered Research Network," Medical Care 56, no. 10, Suppl 1 (2018): S33–S40; [PubMed: 30074949] Kim KK et al., "A Novel Stakeholder Engagement Approach for Patient-centered Outcomes Research," Medical Care 56, no. 10, Suppl 1 (2018): S41–S47; [PubMed: 30074950] Kimminau KS et al., "Patient vs. Community Engagement: Emerging Issues," Medical Care 56, no. 10, Suppl 1 (2018): S53–S57; [PubMed: 30074952] Warren NT et al., "Building Meaningful Patient Engagement in Research: Case Study form ADVANCE Clinical Data Research Network," Medical Care 56, no. 10, Suppl 1 (2018): S58–S63; [PubMed: 30074953] Arkind J et al., "Lessons Learned from Developing a Patient Engagement Panel: An OCHIN Report," Journal of the American Board of Family Medicine 28, no. 5 (2015): 632–638; [PubMed: 26355135] Ellis and Kass, *supra* note 5; Lavalee, *supra* note 13; PCORnet Engagement Assessment Project: Findings and Recommendations, 9 28, 2018, available at <https://pcornetcommons.org/resource_item/pcornet-engagement-assessment-project-findings-and-recommendations> (last visited January 10, 2019). Related to the HCSRN, see Madrid S and, Wright L, Patient Engagement Workbook, 10 2, 2014, available at <http://www.hcsrn.org/en/Tools%20&%20Materials/Plan_Field/HCSRNPatientEngagementWorkbook.pdf> (last visited January 10, 2019). For guidance developed for a different but related context, clinical trials, see National Health Council, Tackling Representativeness: A Roadmap and Rubric, available at <https://www.nationalhealthcouncil.org/sites/default/files/Representativeness%20in%20Patient%20Engagement.pdf> (last visited January 10, 2019); National Health Council, Lessons Learned and Pathways Forward: Practical Experiences in Patient Engagement, A Portfolio of Case

Examples, 9 19, 2017, available at <http://www.national-healthcouncil.org/sites/default/files/CM-SOAbstracts.pdf> (last visited January 10, 2019).

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27. Kelty C and Panofsky A, "Disentangling Public Participation in Science and Medicine," Genome Medicine 6, no. 1 (2014): 8. [PubMed: 24479693]
28. For information on one model that has been extensively tested, see Joosten YA et al., "Community Engagement Studios: A Structured Approach to Obtaining Meaningful Input from Stakeholders to Inform Research," Academic Medicine 90, no. 12 (2015): 1646–1650; [PubMed: 26107879] A. P. Boyer et al., *supra* note 25.
29. Reardon, *supra* note 6, at 196.
30. E.g., Ball MP et al., "Harvard Personal Genome Project: lessons from participatory public research," Genome Medicine 6, no. 10 (2014), available at <10.1186/gm527> (last visited January 10, 2019);Daugherty S, Wahba S, and Fleurence R, "Patient-Powered Research Networks: Building Capacity for Conducting Patient-centered Clinical Outcomes Research," Journal of the American Medical Informatics Association 21, no. 4 (2014): 583–586. [PubMed: 24821741]
31. Chen F, Anguiano B, Koenig BA, and Harris-Wai J, "Methods for the Evaluation of Deliberative Community Engagement Events: Mapping the Terrain" [Poster] ELSI Congress Farmington, CT, June 5–7, 2017.

Table 1

Sectors Represented by Respondents

	Count (%)
Academia	14 (34)
Non-Governmental Organizations	9 (22)
Technology Companies	8 (20)
Government	4 (10)
Laboratories	3 (7)
Healthcare Systems	3 (7)

Table 2

Individual Focus and Collective Focus

	Measure	Illustrative Quote(s)
INDIVIDUAL FOCUS	Dynamic consent	"[E]veryone should be empowered to…remove their data if they want or change the status of consent…because if you don't feel you're in control you're not willing to share, you lose interest." (P11)
	Information push to individuals	"I think at least people should know that their medical data is going to be used for research and is going to be shared, maybe used for research and maybe widely shared. By 'know,' I mean something more than one sentence at page 17 of a single-spaced consent form. I think they should get some information back, both crucial personally-relevant information like the ACMG-56 [now ACMG-59], but also information about the research. What have we got? Who's using this and what do they do? … [A] regular email newsletter or something that says: 'You may remember you are part of the Medical Information Commons and here's what we've been up to lately.'" (P7)
	Individuals contribute information	"[H]aving the patients fill out some of their phenotypic information is very powerful" (P34) "[S] ome editorial or input function" (P17) "The amount of data that's going to be generated that's relevant to our health that is generated outside of the healthcare system will massively [exceed] information within the healthcare system.We begin to aggregate that all in our personal cloud, 50 terabytes and $50….We will not only have control over it virtually but we will have more control over it physically as well." (P11)
	Opportunities to access and engage in citizen science	"I think they should be able to have access to information that's collected about them. As far as larger roles, maybe it might be nice to think about some of these conversations that have been taking place in the citizen science space and thinking about how they might be able to contribute ideas about what can be done with the research or maybe they have a section of the portal that people can look at or browse, though de-identified." (P18) But: "I think it's a very bad idea to give consumers the right to query these research databases…because they won't have the knowledge or the expertise to interpret the results." (P26)
COLLECTIVE FOCUS	Leadership in governance	"I think participant leadership is important in the governance structure, and I think that's the way…to the extent that we're concerned about questions of autonomy, it seems to me that's the way to realize the autonomy principle, more in a collective way than in an individual way." (P2) "The reason for ongoing interaction is, it's not just is this study okay, should the data access committee let this research have this particular set of data, but it's what is *our* research, how do *we* want these data to be used, in what ways can they be used to help solve the problems that *we* care about." (P5; emphasis added) "We need some other system where…the people whose data are involved can have a decision-making structure that lets them say, 'Yeah, we're going to contribute our data, and we're going to govern its use, and we will have a way to get benefits from doing so.' You need an institutional arrangement…that will let people have a say and let them make decisions that will then bind the group, and that's, I think, the place things have been hanging up is that…we're very uncomfortable in our ethical frameworks with ultimately binding people to a collective decision." (P22)
	Inclusion of perspectives	"I think it would be nice, it would be an aspirational goal to have some kind of representation…At the very least focus groups, if not participation in oversight and governance committees…where there would be actual research participants who are involved in a discussion of how the data is being used and actually a real-time kind of oversight." (P19)
	Comprehensive involvement	"I think that they should be involved in all aspects of the commons. They should be involved in establishing it, setting it up. They should have some role in the oversight and governance and monitoring of it. I think they should have access to the data if they want to do their own research. I think they should be involved in disseminating the data and helping interpret data or research or results. I think they should be able to individually download their own data and take it with them to their provider's office or for their own use if that's what they so choose to share with their faith healer or the medicine man or woman or give it to their Rabbi. I think they should be able to do whatever they want to with it." (P24)