Toward Justice and Community Empowerment in Genomics Studies on Sensitive Traits

BY HEINI M. NATRI AND CAROLYN RILEY CHAPMAN

Genomics research has the potential to greatly benefit society through facilitating a better understanding of the etiology and management of health problems. Genetic tests developed through research can help establish a diagnosis for patients and identify individuals who have a higher genetic likelihood of developing traits in the future. Results about some disease- and non-disease-related genetically influenced traits, such as neurodevelopmental differences, mental health conditions, sexual orientation, gender identity, and addiction susceptibility, are sensitive because they relate to personal identity. They may also be associated with self and societal stigma. Thus, genomics studies, particularly on neuropsychiatric,[1] sociobehavioral,[2] and other sensitive traits,[3] pose notable risks. Since genetic information about traits could be used to discriminate against people, genomics research poses significant privacy concerns.[4] Genomics studies on sensitive traits may also reinforce existing stigma, biases, and prejudices, such as racism, ableism, and classism. Well-meaning studies could be misinterpreted or misused in ways that result in harm to individuals or communities in anticipated and unanticipated ways. The benefits of genetic research might be speculative, indirect, or realizable only in the long term, while possible harms, such as the stigmatization of affected communities, might be more immediate.[5]

Engagement with the groups and communities being studied has been proposed to help avoid any group harm in genomics research.[6] Calls for community partnership and collaboration in genetic research emerged in the mid-1990s, in part because of increased demand from participants but also because of greater awareness that genetic research could impact families and communities in addition to individual research participants.[7] Many scholars argued that affected communities should be involved in the research. Experts agree that responsible research must "engage the people with the most at stake in the study, at least if it is a novel line of research about which the relevant communities have not been engaged by other researchers."[8] Moreover, community engagement can also be employed to address the problem of underrepresentation in genomics studies, aiding in harnessing genomics for the well-being of all.[9] Indeed, the purpose for, as well as the approach to, community engagement in genomic research can vary greatly, possibly producing confusion

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There is increasing recognition that, for large-scale genomics studies to be sustainable, research must be responsive to the needs and concerns of the communities being researched.

among interested parties who are involved with or affected by genomic research.[10]

Over the past three decades, community partnerships and participation in research have expanded, but there is still a need for greater involvement and improvement.[11] Although this is true for all types of genetics research, change may be particularly important in genomics studies of sensitive traits; professional societies such as the Ethics Committee of the International Society of Psychiatric Genetics have noted that stakeholder engagement in psychiatric genetics is particularly important.[12] Without interacting with or consulting the relevant communities, genetics researchers may be unaware of valuable perspectives, underestimate potential harms, and assume that the benefit of generalizable knowledge outweighs any risks. Scientists may believe that research that can lead to therapeutic treatments is an unequivocal good, yet impacted communities may have more nuanced reactions.[13] Researchers may also underestimate the interest of various communities in engagement about research, for example, due to inaccurate assumptions or a lack of existing connections. For communities that are linked by traits that cause impairments to communication or intellectual ability, researchers may also presume that engagement is not possible. Many of these assumptions were recently challenged when members of the autistic community protested a large genomics autism study in the United Kingdom, and the project was paused for a formal consultation process.[14]

In this essay, we provide an overview of different approaches to community engagement in genomics studies and examine the challenges, opportunities, strengths, and weaknesses of past efforts. We explore how some large-scale genetic studies on sensitive traits approached community involvement and participation. Inspired by this special report's goal of setting out a bolder vision for a more equitable genomics, we challenge the genetics research community to take steps toward the realization of a more collaborative genomics research standard by which affected communities are included in all stages of the research as peers and collaborators, not solely as objects of study.

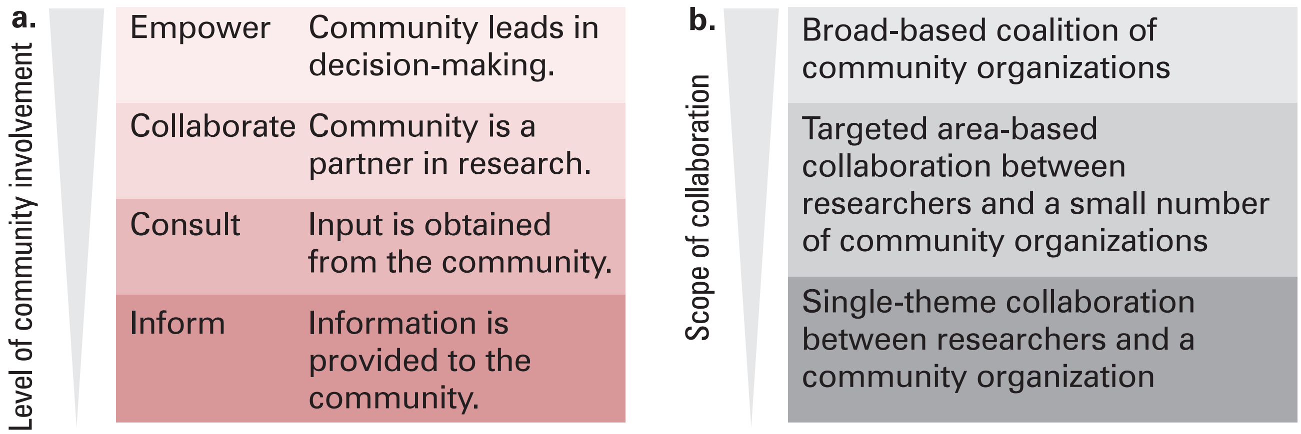
Community Involvement in Genomics Studies: Past and Present

Historically, genomics research has often been conducted on, rather than with, the communities it affects.[15] This approach has sometimes led to mistrust, misunderstanding, and inequities. Based in part on learning from past missteps, there is increasing recognition that, for large-scale genomics studies to be sustainable, research must be responsive to the needs and concerns of the communities being researched. In this section, we explore examples of genomics studies that have sought to engage with specific publics or communities.

To our knowledge, the first large-scale genomics study to conduct public consultation during the creation of a genetic database was CARTaGENE.[16] Focus groups and surveys assessed knowledge, incentives, and obstacles to study participation. The consultation identified concerns about privacy, transparency, communication, and participant involvement in governance. Since then, other studies have consulted communities using focus groups, advisory panels, and community sounding boards. For example, the Marshfield Clinic Personalized Medicine Research Project (a population-based biobank) assembled a nineteen-member Community Advisory Group to provide feedback to the study's principal investigator and the local institutional review board.[17] Focus groups elicited the perspectives of eligible community members, including those who elected not to participate in the study, and attendees expressed concern about potential discrimination, leading to an emphasis on data security. Although these efforts should be commended for seeking community input, the researchers retained complete control over study design and execution.

The National Institutes of Health's All of Us research program conducts outreach efforts to increase public awareness of the program and to create relationships with communities. Through the recruitment of "Engagement and Retention Innovators" and "Community Engagement Partners," this ongoing endeavor aims to increase enrollment and retention of communities historically underrepresented in biomedical research.[18] Moreover, All of Us aims to support "participants as partners" by encouraging its awardees to establish "Local Community and/or

Figure 1.
Community Involvement Varies in Breadth and Representativeness



Levels presented here are modified from the International Association for Public Participation's "Spectrum of Public Participation." See "Core Values, Ethics, Spectrum—the 3 Pillars of Public Participation," IAP2, International Association for Public Participation, https://www.iap2.org/page/pillars, and, from the same organization, "Public Participation Pillars: Internationally Recognized Principles for Making Better Decisions Together," accessed October 28, 2024, https://cdn.ymaws.com/www.iap2.org/resource/resmgr/communications/11x17_p2_pillars_brochure_20.pdf.

Participant Advisory Boards" to garner input from local communities.[19] By recruiting participant partners to serve in steering committees and advisory panels, the project seeks input on research priorities, participant retention, privacy and security, and the return of information to participants.[20] These efforts are coordinated by a designated "Engagement Core" that consists of experts on research, ethics, and engagement. The project continues to increase the number of participants engaged in the project and its governance and to monitor the engagement's impact on research.[21] These efforts can inform evidence-based engagement in future studies.

Researchers associated with the eMERGE network examined community engagement experiences and efforts undertaken by six U.S. biobanks.[22] The study identified a number of challenges in community engagement, particularly in eliciting participant views on topics that require domain expertise, identifying diverse and active participants for community advisory groups, and gauging the level of resource commitment needed. Moreover, genetics researchers may have different ideas about the purpose of community engagement.[23] Biobank personnel's interpretations of "community" and "community engagement" varied widely, affecting how engagement was practiced and what goals were achieved. Many viewed community engagement as an educational and recruitment tool, while others saw it as a process to monitor the community's views of the biobank or to address public perceptions about the ethics of biobanking. The goal of establishing trust between the study and the community was seen as a tool for improving recruitment. Overall, researchers' perceptions of community engagement are highly variable, and the way community engagement is approached is shaped by investigators' preexisting perceptions as well as by upstream decision-making regarding study goals and priorities.[24] Sometimes, community engagement is seen as necessary for the success of the research rather than as an obligation researchers have to ensure that research is designed to maximize benefits and minimize harms to the participating individuals and their communities.

Examples of Community Engagement in Genetic Studies of Sensitive Traits

There are examples of community engagement in genomics studies on social, behavioral, and other sensitive traits. Yet it is also unclear whether or at what level some larger studies have conducted community engagement. For example, the use of community engagement is not reported on the website for the iPSYCH project, initiated in 2012. This research consortium, which is managed by six research principal investigators and headed by a scientific director, aims to "find the causes and create the basis for better treatment and prevention of five of the most serious mental disorders": autism, attention deficit hyperactivity disorder (ADHD), schizophrenia, bipolar disorder, and depression.[25]

Although the Psychiatric Genomics Consortium describes itself as carrying out "open, inclusive, participatory and democratic science," it is also unclear whether and how the researched communities are engaged or consulted.[26] The consortium's language about inclusivity seems to refer to a commitment to scientific transparency and data sharing. While the potential benefits of psychiatric genomics

> *Building trust and sharing the table require humility, respect toward stakeholders, and the ability to question and modify the ways that genomics studies on sensitive traits have traditionally been conducted.*

for "understanding and predicting illness onset, course, and response to treatment, to implement personalized care" are described as "undeniable,"[27] potential risks are not emphasized.

Recognizing the controversial nature of genetics research on sexual orientation, investigators for a genome-wide association study (GWAS) on same-sex sexual behavior[28] pursued "community outreach" and engaged in conversations with community representatives in workshops.[29] Drawing on feedback from this outreach, they developed a website to justify and explain the results and limitations of their study. Yet, again, it is difficult to tell how far these efforts went; the available resources do not mention the potential risks of the project or how they were evaluated.

Another large-scale study, NeuroGAP, aims to increase the representation of African populations in genetic databases and in studies on neuro-psychiatric genetics in particular.[30] Consisting of two research arms, NeuroGAP focuses on the genetics of schizophrenia and bipolar disorder (in the NeuroGAP-Psychosis[31] arm) and childhood neurodevelopmental disorders such as autism, ADHD, intellectual disability, and other cognitive and developmental disorders (in the NeuroDev[32] arm). NeuroGAP is intended to advance research and genetic knowledge and to be beneficial "in the long term to the communities that contributed as participants."[33] NeuroGAP's Global Initiative for Neuropsychiatric Genetics Education in Research initiative aims to support the training of African researchers to develop genetics and genomics infrastructure in Africa.[34] The project has also established the Oxford Global Initiative in Neuropsychiatric GeneEthics (NeuroGenE), a bioethics research and capacity-building initiative for neuropsychiatric genetics research in Africa and around the world.[35]

The Autism Genetics Outcomes (AutGO) initiative, created as a "unique collaborative forum to facilitate the much-needed dialogue between genetics and outcomes researchers,"[36] sought to "support broad stakeholder partnerships and promote a new integrated concept" drawing from genetic and outcome research.[37] The initiative engaged various stakeholders, including researchers, clinicians, caregivers, and industry partners. Two of the twenty-eight AutGO working group members were autistic.

A more recent autism genomics study, Spectrum 10K, was paused because of protests and concerns from the community. The project investigators then tasked a consultation group, Hopkins van Mill, with consulting the community and assembling a coproduction group to guide the redesign of the study protocol and participant-facing materials. A report on these efforts, which are in some ways novel in the field, was expected in fall 2023, but, as of October 30, 2024, had not yet been published.[38]

Types of Engagement: From Consultation to Coproduction, Participatory Research, and Insider Experts

Consultation and engagement are essential steps toward a more just and equitable genomics research enterprise. Inclusive approaches are geared toward increasing the rigor, reach, and relevance of research, maximizing its benefits, and minimizing potential harms.[39] These approaches aspire to respect and empower communities by involving them as partners at every stage of the research process, from setting research questions to data collection, analysis, and dissemination. Although active community participation is increasingly recognized as an integral component of research aiming to improve health outcomes in clinical and translational research,[40] human genomics must also advance further toward coproduction and participatory research (see figure 1.a).

Coproduction. Coproduction involves collaborative research design, implementation, and analysis, with researchers and community members working together as equals. Research projects are planned and executed collaboratively, with all participants working together and coproducing research outcomes. This approach aims to maximize benefits and minimize harms by ensuring that research priorities align with community needs and values.

Participatory research. Participatory research goes a step further, with community members taking a lead role in driving the research agenda. This model fosters a sense of ownership and control over the research process. Community-based participatory research (CBPR) has the potential to empower stakeholders and build capacity within communities to address their needs through research. In CBPR, collaboration may vary in its area of focus and breadth of community involvement (see figure 1.b).[41]

Insider experts. Some genomic and biomedical studies are beginning to embrace "insider experts"—individuals who are deeply embedded in the communities being studied and who possess invaluable cultural and domain knowledge and research expertise. Including insider experts as full team members in "broadly engaged team science" complements other engagement efforts[42] and can help shape research questions, methods, and the interpretation of results. Important examples are found in genetics studies focusing on Indigenous communities.[43] In genomics studies on sensitive traits, insider experts can help ensure that research questions and methods align with stakeholder values. Insider experts can be invaluable in steering research toward the benefit of communities.[44]

Barriers to Community Engagement

Coproduction and participatory approaches have the potential to improve genomics research by making it more just, equitable, and inclusive for individuals and communities that are affected by it. However, several challenges and barriers hinder the wider adoption and employment of these strategies, especially for genomics studies on psychiatric, behavioral, and neurodevelopmental traits.[45]

Lack of identifiable group representation. In recent years, community calls for more meaningful inclusion have reshaped genomics research on Indigenous and Tribal communities.[46] Although many of the recommendations for more meaningful engagement and ethically sustainable genomics research on Indigenous communities can be applied across other marginalized communities, there are also significant differences between such research and genomics studies on sensitive traits. Engagement with Indigenous communities is guided by Tribal sovereignty and established processes for research review, yet communities sharing a sensitive trait often lack clear group organization, representation, and power. While some categories of people with disabilities may view themselves as part of groups with political or social recognition, other groups of people may have traits in common but lack political or cultural group organization.[47] Engagement with groups that are connected only by a shared trait can be more challenging than engagement with identifiable social groups, particularly if the trait or condition involves capacity or communication issues. In psychiatric genetics, for example, it has been suggested that the starting point of stakeholder involvement should consist not of general calls for equal partnership but of "careful explorations of dialogic possibilities and the implied power relations."[48]

Bias and lack of trust. Community leaders cite a lack of trust and bias, such as racism and classism, as the main barriers to CBPR.[49] Researchers engaging in genomics studies on sensitive traits, such as neurodevelopmental or psychiatric traits, may not think that community engagement is relevant. For example, researchers might incorrectly assume that individuals presenting certain differences are incapable of participating or contributing. Moreover, investigators might not even attempt to engage communities because of ableism and other forms of bias. At the same time, as the case of Spectrum 10k shows, community members might be hesitant to participate in conversations with researchers who may not have appropriately considered community values and concerns in prior projects. CBPR is a reciprocal relationship and learning process for all parties who should have a seat at the table. Building trust and sharing the table require humility, respect toward stakeholders, and the ability to question and modify the ways that genomics studies on sensitive traits have traditionally been conducted. Investigators may struggle to acknowledge and accept the ways in which their work may have disrespected stakeholders, and they may be reluctant to change or yield power.

Lack of competence. Investigators and community organizations alike often find themselves in unfamiliar territory when trying to engage in CBPR. Researchers may want to respectfully engage stakeholders in more meaningful ways, but many are not trained in these approaches and don't know how or where to start. Researchers may struggle to identify appropriate representation, to reach and connect with community organizations, and to put in place appropriate protocols for engagement and inclusion. They may encounter resistance or lack of support from institutional leadership, including institutional review boards. Difficult issues may arise in collaborations involving genomic research on sensitive traits, and researchers may find engagement on topics such as ableism, racism, classism, or other forms of oppression challenging and uncomfortable. Investigators may also encounter challenges in engaging communities due to accommodation needs.

Lack of resources and incentives. Researchers often cite a lack of resources, including funding, time, and infrastructure, as the main barrier to community engagement.[50] Participatory models are costlier and more time- and labor-intensive than traditional models because engaging communities, building infrastructure, and fostering collaborations require substantial investment, including dedicated staff. Effective and meaningful engagement also requires flexibility in timelines, as investigators must ensure that community perspectives are adequately heard and addressed at all stages. Another challenge is misalignment in researchers' incentives: in academia, the quality of research conduct is often not valued as highly as output quantity.

Power imbalances. The lack of trust between investigators and community partners is reinforced by inequitable power structures. Lack of representation at the leadership level, which may result from barriers such as lack of accommodations, may sustain inequities and hierarchies of tra-

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ditional research approaches. Further, insider experts who contribute to research may face additional barriers, such as increased scrutiny, criticism, or discrimination, due to their dual role and the stigma attached to sensitive traits.

Poor integration of engagement and decision-making. Whether engagement is integrated with research and decision-making from the start or happens in parallel has a critical impact on the success and outcomes of engagement.[51] Critically, upstream decisions, resource constraints, and opportunistic approaches to engagement often result in so-called pantomime community engagement, in which community involvement is not seen as a necessary part of research but as a parallel process where community partners have little to no power in actually shaping the research.

Toward Community Empowerment, Equity, and Justice

Enhancing the social relevance, rigor, and benefits of genomics and biomarker research on sensitive traits can be achieved only through long-term partnerships and increased trust. Coproduction and CBPR can serve as powerful tools for promoting equity, justice, and community empowerment in genomics research. Achieving these goals requires a comprehensive approach that encompasses changes at systemic, institutional, and personal levels. Here, we explore key strategies for addressing the barriers to CBPR. While some strategies may be particularly helpful in tackling specific barriers—for example, lack of resources and incentives is best addressed through funding and policy—all the approaches described below are essential for fruitful, long-term partnerships.

Addressing systemic issues through funding and policy. For real change to be possible, institutions and funding agencies should recognize community engagement and participatory approaches as valuable aspects of research and should incentivize researchers to engage communities in meaningful ways.[52] In academia, where tenure and promotion decisions are based largely on traditional scholarly activities, community engagement is often undervalued.[53] Indeed, some researchers view a lack of incentives as a major reason not to conduct participatory research.[54] Funding agencies must not only support the development of participatory research partnerships but also provide sustained, continuous funding and support to maintain these partnerships.[55] Funders can create incentives to raise the bar in community engagement by mandating it. Organizations such as the Patient-Centered Outcomes Research Institute fund studies that build an evidence base on engagement in research, in addition to supporting efforts in capacity building.[56] In another example, involving the large-scale Dutch research consortium Genetic Risk and Outcome of Psychosis, the main funder, the Netherlands Organization for Health Research and Development, required researchers to involve patients as a prerequisite for funding.[57]

On a federal level in the United States, while the National Institutes of Health (NIH) offers some funding opportunities for CBPR, these are limited, and, to our knowledge, no dedicated CBPR opportunities are available for genomics research.[58] The NHGRI Community Engagement in Genomics Working Group, established in 2017, aims to engage diverse communities, facilitate understanding, and inform the NHGRI leadership about issues relevant to its programs.[59] Additionally, the NHGRI Education and Community Involvement Branch aims to develop education and community involvement programs to engage the public and to increase the understanding of genomics and accompanying ethical, legal, and social issues.[60] However, it is unclear whether or how these efforts shape funding strategies. Encouragingly, in July 2024, the National Institute of Neurological Disorders and Stroke released a notice of special interest in receiving grant applications related to ethical issues arising in the context of brain and nervous system research, including genetic research; proposals could include "the direct engagement of stakeholders."[61] Realigning the federal, local, and institutional incentives—including material, social, and status incentives—with community goals and interests is essential for achieving the structural and systemic change required to increase justice and equity in genomics and biomedical research. In alignment with the principle that change must start from the top, community representatives should be included on panels that have the power to decide who and what gets funded. While the NIH "seeks advice from patient organizations and voluntary health associations"[62] when setting research priorities, the process lacks transparency, and gaps in funding have led to structural discrimination of individuals exhibiting some stigmatized traits.[63]

The NIH has been responsive to criticism for its "ableist mission statement,"[64] issuing a request for comments. In a subsequent report, the NIH concluded that more input is needed before finalizing the mission statement and announced plans to gather additional community feedback."[65]

Diverse and inclusive representation. To prevent tokenism and promote inclusivity, it is essential to have diverse representation at all levels of research, including in research leadership, advisory boards, and decision-making bodies. To avoid siloing and to ensure that all vital perspectives are included, geneticists, bioethicists, and members of affected communities should be included in planning and decision-making at all stages of research. Clearly defining community partner roles and engaging the right individuals is crucial for the success of CBPR. Further, when selecting researchers to participate in partnerships, priority should be placed on those who are prepared to challenge traditional approaches and willing to share power with stakeholders. Consistent with the principle "nothing about us without us," establishing processes for effective, accessible communication and power sharing ensures that community voices are heard and respected throughout the research process.

Build infrastructure for collaboration and researcher competence. To engage in ethical genomics research, it is imperative to build competence among researchers. Many researchers want to engage communities in more meaningful ways but lack the skills, knowledge, and resources to do so. Developing clear rubrics that researchers can follow and integrating ethical, legal, and social implications as an essential part of research on sensitive traits and vulnerable communities are critical steps. Education programs and ongoing training and oversight can help researchers and administrators navigate the complex ethical landscape of genomics research. Further, to facilitate effective collaboration, it is necessary to develop accessible platforms to connect researchers with communities and to facilitate continuous communication.[66] For example, the International Society for Autism Research (INSAR) Autistic Researchers Committee has recently launched the INSAR Community Collaborator Request to connect autism researchers with stakeholders who are interested in partnering with researchers in designing, conducting, and disseminating studies.[67] When applying these frameworks, it is recommended that investigators take a flexible approach: all groups may not have a stake or interest in every aspect of research, and group identities are not mutually exclusive.[68] Many researchers and clinicians are also directly affected by research as primary stakeholders.

Capacity building, community training, and benefits. Investing in education and training programs for communities is vital for meaningful engagement in genomics research. This should include supporting the valuable work of community members with fair compensation. Empowering communities with knowledge, skills, and leadership roles enhances their capacity to participate effectively in research and decision-making. Researchers must prioritize tangible benefits for the communities they work with by aligning their goals and efforts with the community's interests and priorities.

Transparency and accountability. Establishing and maintaining transparent communication channels between researchers and broader communities are essential for achieving the goals of CBPR. Establishing mechanisms to increase accountability is crucial for increasing the rigor and acceptability of research and tackling the ethical challenges in genomics research on sensitive traits. Building and maintaining trust between all affected and interested parties are parts of a continuous process that takes effort and serves as a foundation for productive community engagement.

Acknowledge and address stigma and oppression. Stigma is a social phenomenon that involves prejudice and discrimination against those who deviate from perceived norms. These prejudices have shaped research agendas in genomics. Stigma may lead to the underrepresentation of certain groups in genomics studies as well as to biased research agendas that could further stigmatize and pathologize those groups. Still, individuals exhibiting, for example, neurodevelopmental differences or experiencing psychiatric symptoms may benefit from genomics research. Safe and equitable research on and the use of genomics require awareness of the ways that societal stigma can shape research and how this can lead to injustice and oppression. Going forward, these issues must be addressed through additional safeguards, including participatory-based research approaches. Researchers must also acknowledge a responsibility in opposing misconceptions that may contribute to harmful societal attitudes.

Ultimately, addressing the negative impacts of ableism and other forms of systemic and interpersonal bias will require significant commitment and change at individual, institutional, and societal levels. However, cultural shifts and policy changes are possible. Some traits that were previously pathologized, such as certain sexual orientations and gender identities, are now recognized as normal and valuable aspects of human diversity. A similar evolution is already observable with respect to the neurodiversity community. Nonetheless, members of these communities still face discrimination and bias that must be acknowledged and addressed by genetic and biomedical researchers. To serve these communities, the scientific community must commit to fighting prejudice and one-sidedness at all levels. These efforts must push further forward for the better acceptance of exhibiting what are now highly stigmatized traits and disabilities, such as autism, bipolar disorder, and tendencies toward depression or anxiety.

Transforming Genomics for an Equitable Future

Achieving true community respect, empowerment, equity, and justice in genomics studies of behavioral, psychiatric, and neurodevelopmental traits is a bold vision that challenges current standards and norms. Making progress toward this vision requires a willingness to question the status quo and push ourselves toward a new, more equitable standard that can seem out of reach but that is attainable if we reflect on shortcomings and make needed changes. Ongoing commitment and collaboration are necessary to achieve any meaningful improvements, and support is needed from the highest levels of the research enterprise.

While some challenges are unique to studies on sensitive traits, the approaches presented in this essay are widely applicable across genomics and biomedicine. By addressing power imbalances, raising and broadening community engagement standards, and allocating the requisite resources in genomics studies, the field can move closer to a future where genomics research benefits all of humanity while respecting the rights and values of the diverse communities it aims to serve.

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Positionality Statement

Heini M. Natri is a geneticist who has contributed to autism-related research initiatives as an insider expert and as a member of participatory and c-production groups. Carolyn Riley Chapman is an academic bioethicist with training in genetics and social sciences research.

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