Ethical challenges in autism genomics: recommendations for researchers

Heini M Natri,[1]* Carolyn R Chapman,[2,3] Síofra Heraty,[4] Patrick Dwyer,[5,6,7] Nick Walker,[8] Steven K Kapp,[9] Heather A Dron,[10] Julian A Martinez-Agosto,[11] Lea Mikkola,[12] Mary Doherty[13]

1. Translational Genomics Research Institute (TGen), Phoenix, AZ 85004, USA
2. Department of Population Health (Division of Medical Ethics), NYU Grossman School of Medicine, New York, NY 10016, USA
3. Center for Human Genetics and Genomics, NYU Grossman School of Medicine, New York, NY 10016, USA
4. Centre for Brain and Cognitive Development, Department of Psychological Sciences, University of London, Birkbeck, London WC1E 7HX, UK
5. Center for Mind and Brain, UC Davis, Davis, CA 95618, USA
6. Department of Psychology, UC Davis, Davis, CA 95618, USA
7. MIND Institute, UC Davis Health, Sacramento, CA 95817, USA
8. California Institute of Integral Studies, San Francisco, CA 94103, USA
9. Department of Psychology, University of Portsmouth, Portsmouth, Hampshire PO1 2UP, UK
10. Sterilization and Social Justice Lab, UCLA Institute for Genetics and Society, Los Angeles, CA 90095, USA
11. Department of Human Genetics, University of California Los Angeles, Los Angeles, CA 90095, USA
12. Turku Bioscience Center, InFLAMES Research Flagship Center, University of Turku, Turku 20520, Finland
13. Department of Neuroscience, Brighton and Sussex Medical School, Brighton BN1 9PX, UK

Author contributions

H.M.N: Supervision, Project administration, Conceptualization, Investigation, Writing - Original Draft, Writing - Review & Editing

C.R.C: Conceptualization, Investigation, Writing - Original Draft, Writing - Review & Editing

S.H: Conceptualization, Investigation, Writing - Original Draft, Writing - Review & Editing

R.M: Conceptualization, Investigation, Writing - Original Draft, Writing - Review & Editing

P.D: Conceptualization, Investigation, Writing - Original Draft, Writing - Review & Editing

N.W: Conceptualization, Writing - Original Draft, Writing - Review & Editing

S.K.K: Conceptualization, Investigation, Writing - Original Draft, Writing - Review & Editing

H.D: Investigation, Writing - Original Draft, Writing - Review & Editing

J.A.M-A: Conceptualization, Writing - Review & Editing

L.M: Conceptualization, Writing - Review & Editing

M.D: Conceptualization, Writing - Review & Editing

Abbreviations

AAC: Augmentative and Alternative Communication

AIP: Affected and/or Interested Party

CBPR: Community-Based Participatory Research

ELSI: Ethical, Legal, and Societal Implications

GWAS: Genome-Wide Association Study

ID: Intellectual Disability

PGS: Polygenic Score

PGT: Pre-implantation Genetic Testing

Abstract

Equitable and just genetic research and clinical translation require an examination of the ethical questions pertaining to vulnerable and marginalized communities. Autism research and advocate communities have expressed concerns over current practices of genetics research, urging the field to shift towards paradigms and practices that ensure benefits and avoid harm to research participants and the wider autistic community. Building upon a framework of bioethical principles, we provide the background for the concerns and present recommendations for ethically sustainable and justice-oriented genetic and genomic autism research. With the primary goal of enhancing the health, well-being, and autonomy of autistic persons, we make recommendations to guide priority setting, responsible research conduct, and informed consent practices. Further, we discuss the ethical challenges particularly pertaining to research involving highly vulnerable individuals and groups, such as those with impaired cognitive or communication ability. Finally, we consider the clinical translation of autism genetics studies, including the use of genetic testing. These guidelines, developed by an interdisciplinary working group comprising autistic and non-autistic individuals, will aid in leveraging the potential of genetics research to enhance the quality of life of autistic individuals and are widely applicable across stigmatized traits and vulnerable communities.

Introduction

Genomics research has the potential to enhance well-being by increasing understanding of health and disease and facilitating the development of new or improved treatment and prevention methods. However, genetic research on historically marginalized and vulnerable communities[1,2] and stigmatized traits,[3] such as autism, raises complex ethical questions. Recently, researchers and numerous autistic community members have raised concerns regarding the use of genetic data in autism research and clinical settings. These critiques underscore the lack of overall benefit to autistic people, as well as risks related to future data

use and privacy, including the possibility that data or study results could be used in discrimination or prevention.[4–6] Such concerns are shared with the wider disability community[7] and other marginalized and vulnerable communities,[1,8] and parallel the broader conversation regarding the ethical, legal, and societal implications (ELSI) of genetics research and uses of genetic data.[9]

Autistic people are disproportionately affected by particular health conditions, such as epilepsy and gastrointestinal disorders,[10] and face barriers to accessing health care,[11] contributing to health disparities and inequities.[12] Some of these co-occurring health conditions may have biomedical remedies discoverable through genetic research.[13] Autistic people's priorities include research on health and well-being,[14,15] and despite reservations, some autistic people find potential value in genetic research, testing, and counseling.[6,16,17] In contrast, many do not endorse efforts to find causes of autism.[15,18–22] In a recent survey (n=6,004) with autistic (8.1%) and non-autistic respondents, studies on genetic markers and prenatal screening arose as an area to avoid.[18] Standing in contrast to traditional positions of non-autistic-led charities, the Autistic Self Advocacy Network has issued a position statement: "As genetic research continues to develop, autistic people must have our say in how it should be used, and not used."[23] While several studies have surveyed caregivers' and clinicians' views (e.g.,[19,24,25]), surveys on autistic perspectives and priorities regarding genetic research and testing are scarce.[6,16,18]

An ableist lens has shaped biomedical research, potentially biasing scientific development.[7,26] Ableism is common in healthcare[27] and autism research[28] and has shaped the contemporary research agenda: historically and currently, autism-related research has largely been built upon the assumption that autism needs to be prevented, eliminated, or treated. This has steered research toward discovering the causes of, prevention of, or minimization of autistic traits and away from other priorities, such as autistic well-being, tackling barriers to societal inclusion, or the societal benefits of neurodiversity. Indeed, standing in contrast with the affected parties' priorities, the majority of autism research funding is granted to biomedical and "risk factor research," as opposed to service research, and an increasing proportion of studies focus on prevention.[29] Most current genetic autism research focuses on the genetic architecture, networks, pathways, and "modifiable risk factors" underlying autism and its many presentations.[13] A recent systematic review utilizing a scientometric analysis identified major clusters of autism-related genetic studies, including studies focusing on causes and presentation; specific methods such as brain organoids and mouse models; and, due to the connection between autism and epilepsy, anti-seizure drugs.[30] Apart from epilepsy, research focusing on physical health or well-being was notably absent.

The growing criticism of current genetic and biomedical autism research suggests that existing ethical guidelines, regulations, and review processes are inadequate.[31] Researchers and advocates have urged the field to reform its ethical standards, for example, by requiring the meaningful involvement and leadership of autistic researchers or consultants at all stages of research.[4,32,33] Such reforms must grapple with the impact of current and past malpractice on marginalized, vulnerable communities, and acknowledge how prevailing societal biases continue to influence contemporary research and clinical implementations.[7] Importantly, equitable and ethical use of genetics requires the identification and mitigation of biases as well as careful consideration and balancing of possible harms and benefits (**Table 1**).

To address the autistic community's concerns, as a workgroup of autistic and non-autistic geneticists, bioethicists, autism researchers, clinicians, and advocates, we have produced a framework and recommendations to guide genetic autism research, clinical translation, and policy (Figure 1; **Table 2**). Our recommendations are geared towards fostering rigorous, safe, and beneficial research practices that have the potential to result in improvements in the quality of life of autistic people and their families. Specifically, we consider 1) the need to address health disparities and inequities while protecting study participants and the community, 2) informed consent and governance, 3) representations of autism in genetic testing and counseling, 4) research impacting those particularly vulnerable due to, e.g., age or disability, and 5) tackling power imbalances through inclusion and accountability. While these recommendations arise from the needs and concerns of the autistic community, they are widely applicable to genetics and genomics in the context of disability, complex social traits, and marginalized communities.

Table 1. Possible harms and benefits of the use of genetics in autism research and clinical implementations.

	Possible benefits	**Possible harms**
For individuals and families	Greater self-understanding; Membership in communities centered around genetic syndromes.	Lack of benefit; Genetic testing may not offer clear results or increase understanding; Increased genetic determinism, pathologization, stigma, and discrimination.
	The promise of "personalized medicine": individualized care informed by genetics, lifestyle, and environment; Increased autonomy over healthcare and reproductive decisions.	Loss of control over genetic data and personal health information; Reproductive coercion.
	Early identification: Possibly better quality of life[34]; Earlier recognition of some co-occurring health risks and conditions may enable amelioration; Increased awareness or preparedness within the family.	*Early identification*[35]: Possibly less positive ideas about autism[34]; Inflicted insight; Exposure to harmful interventions; Losing the right to choose whether and when to test or disclose; Harms and ethical challenges of prenatal testing.
For groups and society	Increased knowledge and a better understanding of some aspects of autism.	Increased genetic determinism, pathologization, stigma, and discrimination.
	Improved health and well-being and reduced health disparities through the prevention and amelioration of some co-occurring conditions.	Increased health disparities due to genetic testing, targeted interventions, etc., inequitably applied or only available to some; Erosion of disability and reproductive rights.

	Strengthen the formation of support/advocacy groups centering on shared genetic identity.	Potential for the emphasis of genetic or biological "subgroups" leading to division and additional harm, particularly to those most vulnerable.

Recommendations for ethical genetic and genomic autism research

Guiding principles

We hold it as a central guiding principle that the primary affected and interested parties (AIPs) in autism-related research are autistic people. While the perspectives of non-autistic family members, clinicians, researchers, and other professionals, and other interested parties are also essential, autistic persons are those most strongly and directly impacted by autism research. Still, in biomedical and genetic research, autistic perspectives are rarely elicited. In order to balance the diversity of perspectives and tackle the issue of partial representation in autism research,[36] we assert that the diverse community of autistic persons should have a central role in determining the objectives, directions, and methods of autism-related genetic research. Further, to be ethically justified, at a minimum, genetic research on autism needs to be designed and undertaken in a way that respects four principles that ground key ethical frameworks in biomedical research: respect for persons, beneficence, non-maleficence, and justice (Figure 1).

The principle of *respect for persons* encompasses 1) respect for autonomy, i.e. individuals' capacity for self-governance; and 2) the requirement to have additional protection in place for those with reduced autonomy.[37] In biomedical research, this principle requires us to obtain research participants' informed consent and to protect the rights of those with a reduced capacity to make decisions on their own behalf. We propose that, more fundamentally, respect for persons requires autism-related genetic research to align with the priorities, values, and best interests of its primary AIPs, including those with limited capacity to consent to participation. So far, priority setting for autism research has predominantly been determined by secondary AIPs, in part due to a paucity of methods to elicit the first-person perspectives of autistic individuals with learning disabilities or those facing the greatest communication challenges.[38] It is, therefore, essential to dedicate increased effort towards developing such methods.

Genetic autism research should be conducted for the *benefit* of autistic people. This could amount to, for example, developing interventions to address those characteristics or co-occurring conditions that some autistic people consider detrimental to their well-being. As autistic people's well-being is influenced by numerous and complex societal, environmental, and genetic factors, it is unlikely that genetic investigations alone will allow these benefits. It could, however, be an important *component* in achieving a better understanding of factors that support autistic well-being. Translating the findings of genetics studies to practice continues to be a challenge,[13] and in the context of socially complex traits, benefits are often unapparent. The principle of beneficence obliges us to systematically weigh the possible benefits of research against possible harms. However, calculating such a "risk/benefit" ratio, particularly when research is unlikely to benefit individual participants but may benefit future generations, requires a thorough assessment. Importantly, it is necessary to ensure that AIPs understand and agree with the purported value of such studies and are well-informed about these tradeoffs.

The principle of *non-maleficence* embodies the responsibility to prevent and minimize harm to research participants and others.[39] In biomedical research ethics, the focus is usually on protecting individual participants rather than on the broader societal implications of studies.[40] However, in genetic research, knowledge produced from participants' samples can be and often is used to make inferences about groups they represent. These indirect opportunities for third-party and group harm are increasingly recognized.[41] Some stakeholders are particularly concerned that genomic research will be used for the prevention of autism, for example, by calculating polygenic scores (PGSs) to discard embryos or terminate fetuses with an increased likelihood of developing autism.[42] While some scientists would not support research that aims at prevention, cure, or "normalizing" autistic people, genetic knowledge ostensibly pursued for the benefit of autistic people by some could be misunderstood or misused by others, with harmful consequences. As the investigation of the genetic determinants of the well-being of autistic people often involves the examination of the genetic architecture of autism, researchers should be cognizant of and carefully consider the potential risks and benefits of all research involving autistic participants (**Table 1**). Importantly, these possibilities for benefit and harm should be transparently acknowledged to allow informed decisions regarding participation.

Distributive *justice*, the fair and appropriate allocation of benefits and burdens, requires a fair selection of research participants in a way that 1) does not unfairly place the burdens of research on a particular group of participants who may be vulnerable and 2) aligns with the right to participate in and benefit from scientific progress. The principle emerged from considering historical examples of groups whose vulnerabilities scientists had exploited to conduct unethical high-risk studies.[43] However, excluding autistic individuals from research could also have negative consequences and exacerbate disparities. Autism often intersects with other marginalized characteristics (e.g., sex, gender, sexual orientation, ethnicity, disability, socio-economic status), and autistic individuals often face barriers to research participation or are explicitly excluded from it;[44] such differential treatment may constitute injustice. Moreover, when autistic individuals are excluded from research, beneficial findings may be limited in their generalizability, potentially exacerbating health disparities.

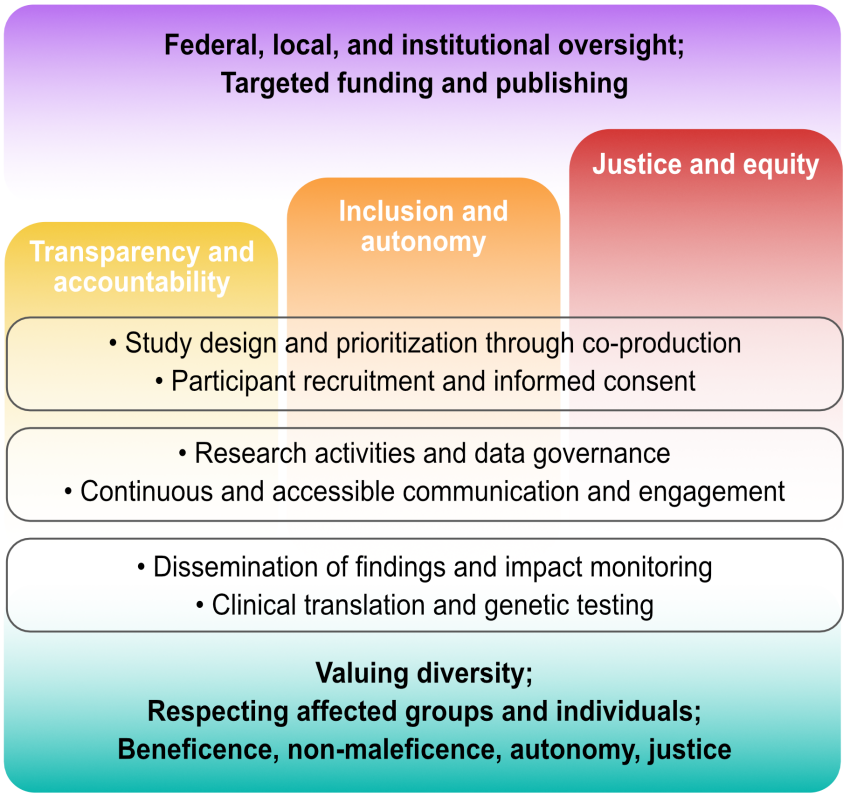

Figure 1. Steps towards justice and equity in autism-related genetics and genomics. *Increasing transparency, accountability, and inclusion is necessary for the development of a just and equitable genetic and biomedical research enterprise that respects the autonomy of affected groups. Transparent and inclusive practices ought to be applied at every stage of research, from study design and prioritization to clinical translation. Such practices are built upon a foundation of valuing diversity, respect towards affected groups and individuals, and the bioethical principles of beneficence, non-maleficence, autonomy, and justice. These practices can be promoted through federal, local, and institutional oversight and targeted funding and publishing.*

Recommendations for goal setting and responsible research conduct

The primary goal of genetic autism research should be improving the health, well-being, and autonomy of autistic persons, and it should reflect the priorities, values, and concerns of the autistic community. To better understand and meet the needs of the primary AIPs, large-scale studies to survey diverse cohorts of autistic individuals are urgently needed and should be prioritized. These goals can be achieved through targeted funding and publishing (Figure 1). We recommend that researchers ground their work on solid hypotheses for why and how studying differences at the genetic and molecular level could promote autistics' well-being. They should clearly communicate their hypotheses to the autistic community and other AIPs and demonstrate the scientific validity of their research, i.e., that their proposed methods to test those hypotheses are appropriate to their research objective.

Genetic and biomedical research must extend the appreciation of human diversity to neurodiversity, including autism (Figure 1).[45] We recommend that researchers avoid framing autism in itself as a negative outcome, but engage in a more nuanced examination of diversity,

disability, identity, and the contexts in which differences may become disabling. For example, researchers should avoid reductive framing that may obscure or misrepresent the factors underlying health disparities by emphasizing genetic factors while failing to adequately acknowledge and account for social determinants. Moreover, researchers should explicitly consider and acknowledge the contributions of ableism, stigma, discrimination, and lack of accommodations to health outcomes, and aim to use respectful and inclusive language concordant with current guidelines for unbiased, non-ableist terminology,[46,47] (e.g., "likelihood" instead of "risk," "non-autistic" instead of "healthy," and when appropriate, "trait" instead of "symptom").

In their grant proposals and participant-facing documents, investigators should clarify what opportunities for benefits their studies may produce and be realistic and transparent about the challenges that could hinder such opportunities. In consultation with autistic people, researchers must work to anticipate and address risks of harm, including unintended harms and harms to third persons, early on. For example, genome-wide association studies (GWAS) and other studies aiming to identify the genetic determinants of autism should be transparent about the possibility that findings may be later used in genetic testing in reproductive settings, even if the investigators conducting the original study may not aim to develop such implementations. Further, researchers should seek to disseminate their findings not only to study participants, but the wider autistic community, and continue to monitor the impact of their research after publication (Figure 1).[48]

Investigators involved with autism research may find navigating disagreements with AIPs challenging. Further, the highly diverse experiences and values of the autistic and autism communities create additional challenges. To balance conflicting perspectives, studies can employ principles and practices of coproduction[49]: to build consensus, researchers should solicit input on decisions, transparently communicate and rationalize these decisions, and establish processes for appealing and revising them. We further recommend that autism-related genetics studies move towards models of community-based participatory research (CBPR) and transparent, respectful engagement with the autistic community (Figure 1; Table 2).[50]

Informed consent and data governance

Voluntary informed consent rests upon providing comprehensive information, assessing understanding, and obtaining permission from a research volunteer who has the capacity in a context free from undue influence or coercion.[51] In the U.S., federal guidance suggests the inclusion of information that a "reasonable volunteer" would want to know when contemplating participation. Further, informed consent requires comprehension and voluntariness.[37] While consent procedures are directed at individual participants, decision-making is often interdependent, and genetic research may have implications for individuals and groups beyond research participants.[52] Some autism-related genetics studies have pursued *community consultation* to inquire about community members' views of the planned research. However, there are no standards or requirements on how such consultation should be conducted or how much community support is needed. Moreover, consultation does not necessarily produce consensus or *community consent*.[53]

A community consent approach is practiced in some research involving Indigenous communities and could be applied in other contexts.[54] While reaching full consensus and community consent may not always be feasible, studies should — at a minimum — invest in efforts to reliably scope and transparently communicate the degree of community support. CBPR and co-production methods can aid in determining AIPs values, priorities, and level of consent, however, caution must be taken not to conflate the use of CBPR methods with the achievement of their objectives: for example, researchers' perspectives on what constitutes adequate consultation might differ from community perspectives and the use of some methods common in CBPR does not necessarily constitute equal partnership.

A number of factors have influenced current standards for informed consent and data governance. Researchers may apply for waivers of consent under certain conditions (Common Rule 46.116.f), or if using de-identified data, some secondary biospecimen research may not even fall into the category of human subjects research. Broad consent for the storage, maintenance, and secondary research use of identifiable private information or biospecimens, collected either for studies other than the proposed research or non-research purposes, is permitted as an alternative to IRB informed consent requirements (46.116.b). Broad consent is a widely used alternative to study-specific informed consent in genomics studies.[55] Broad consent, however, makes participation difficult for those who consent to their information be used only for specific types of studies: for example, some might consent to studies investigating co-occurring conditions, but not the genetic basis of autism in itself. Further, because broad consent is not specific about future use, it is debatable how informed broad consent can be in terms of future risk of harm. For example, researchers may fail to inform participants about specific risks due to a lack of awareness, ostensibly unprecedented findings, or simply a lack of obligation to consider risks beyond the remit of local regulations. Moreover, as time passes, the direction of research tends to change such that it may be impossible for consent that was given at a single time point to be truly informed about future possibilities, particularly pertaining to social consequences and group harm.

Group harm, described as "damages or injury that impact the welfare interests of a group and its people",[56] in autism research can range from existential (e.g. prenatal testing and/or pregnancy termination) to issues concerning dignity and quality of life (e.g. stigma, discrimination). It may sometimes be assumed that individual informed consent implies tacit awareness of group harm, and thus assures individual acceptance of such issues.[56] However, current ethical infrastructure and standards are not set up to account for group harm (Chapman et al., in preparation). The lack of formal rubrics for assessing group harms in genetic research remains an ongoing issue; we recommend the development of such rubrics. We further recommend that, in their engagement, researchers consider the possibility of group harm and discuss how they seek to mitigate it.

As an alternative to broad consent, researchers should consider adopting more granular, dynamic, consent procedures.[57] Sufficient granularity should include offering participants a choice of which types of data to share with investigators, for which purposes it might be used, and by which parties. Importantly, participants need to be kept adequately informed about research progress and new potential risks and benefits; i.e., consent must *remain informed* across time.[58] For this purpose, we recommend the development of participant portals that allow

for changes to consent and frequent engagement and feedback. This offers a viable tool to address related challenges, for example, reconsenting transition-age participants. Dynamic consent has the potential to democratize genetic autism research, as it allows participants to more easily decide whether to share data, thereby shaping specific research pathways and future directions. However, care must be taken to ensure that dynamic consent does not facilitate the exploitation of vulnerable participants or proxies due to an uncontrolled consent environment.[59]

Ethical considerations regarding highly vulnerable groups

Vulnerability of research participants is generally linked to compromised autonomy or informed consent, i.e., inadequate comprehension or non-voluntariness. Regulations for human subject research have been developed since the 1970s in response to misconduct affecting vulnerable communities in cases involving violations of informed lack of meaningful consent, coerced or exploitative subject selection, and inattention to distribution of benefits and burdens.[43] Therefore, vulnerability may best be understood as an increased risk of being wronged or exploited.[60] As autistic individuals are underrepresented in decision-making affecting them, face marginalization, and are more likely to experience victimization,[61] many members of the autistic community can be characterized as vulnerable. Further, autistic individuals vary in their vulnerability with respect to age, cognitive and communication ability, co-occurring conditions, legal autonomy, and/or other intersectional characteristics. Thus, studies involving highly vulnerable autistic participants may require additional safeguards. While the vulnerability of research participants warrants caution, exclusion due to the lack of established protocols for the inclusion of vulnerable individuals may also further disempower these communities, contributing to disparities.

Many large-scale autism studies with a genetic component enroll minors, generating numerous ethical challenges.[62] Power imbalances, as well as different perspectives and priorities of adult researchers/caregivers and child participants, may increase the risk of violations of autonomy.[62] In participant recruitment, caregivers of autistic children often provide consent for enrollment and are likely to have a strong influence on the child's decision to assent (if even sought), thus impacting the actualization of their dependents' individual rights. Moreover, caregivers themselves may be vulnerable, further complicating the ethical recruitment of minors.[63] In the U.S., federal regulations include requirements for research involving children: in genetic autism research falling under a minimal risk categorization, these protections involve ensuring adequate provisions for soliciting the assent of the child and the permission of the guardians (Common Rule 46.404). Still, such protections may be inadequate, and many have proposed additional action, including reconsenting transition-age participants who were recruited to genetics studies as minors.[64] We recommend that such approaches are adopted as a part of the re-evaluation of informed consent practices in autism-related genetics studies.

According to recent estimates, approximately 33% of diagnosed autistic individuals have co-occurring intellectual disability (ID).[65] While people with ID face a number of health issues, they are often excluded from research studies.[66] Sometimes this may be due to the (perceived) inability to consent. However, it is often unclear how the capacity to consent is assessed, and current regulations do not provide adequate guidance regarding such assessment, third-party

consent, or responsibilities of investigators and IRBs in research involving participants with reduced consent capacity.[66,67] Studies indicate that with appropriate communication strategies, individuals with ID are often able to participate in the informed consent procedure.[68,69] Such strategies should be employed to reduce reliance on third-party consent. When integrating genetic information into the health care of individuals with ID, co-designed resources, as well as education of healthcare providers, are necessary.[70] Importantly, researchers should seek to document the first-person perspectives of people with intellectual and learning disabilities in order to better meet the needs of this underserved community and develop more inclusive practices for genetic healthcare.[70]

As many as 30% of diagnosed autistic individuals exhibit limitations in spoken communication,[71] creating barriers and challenges to study participation. Minimally and non-speaking autistic individuals are particularly vulnerable due to their limited ability to self-advocate, underlining the urgent need to develop and increase the availability of alternative and augmented communication methods (AAC).[72] Such methods are essential in enhancing autonomy in studies involving participants with limited vocal output. In a recent survey, parents of non-speaking autistic children (n=20) were supportive of some genomic research, as long as it was ethical, transparent, and designed to support autistic people.[73] Regarding consent, some parents suggested that if possible, they would choose to act according to the non-speaking child's wishes[73], further highlighting the need to seek the first-person perspectives of non-speaking autistic individuals instead of relying on secondary informants. Similarly, eliciting the perspectives and priorities of non-speaking AIPs is essential for the development of research programs for the enhancement of their well-being.

In the U.S., federal regulations note that if IRBs regularly review research involving subjects likely to face coercion or undue influence, consideration should be given to the inclusion of experts knowledgeable about and experienced in working with those groups (Common Rule 46.107). Further, IRBs should ensure that the selection of subjects in such research is equitable (46.111.a.3). Additional safeguards must be included to protect the rights and welfare of particular groups (46.111.b), and in research involving individuals with impaired decision-making capacity due to, e.g., developmental or intellectual disability or traumatic brain injury, the highest ethical standards to research and research oversight must be applied.[74] The Declaration of Helsinki states that research done with vulnerable groups should be responsive to the group's needs and priorities, aligning with our recommendations for AIP inclusion. We further recommend a wider adoption of existing guidance and the development of new detailed guidance tailored to genetics studies enrolling highly vulnerable autistic individuals. Such guidance should be developed in collaboration with diverse cohorts of autistic individuals, family members, and other AIPs, as well as bioethicists and disability scholars.

Considerations regarding clinical translation and genetic testing

Many of the potential benefits of genetic research on autism rely on the clinical translation of research findings. Currently, the leading application of autism-related genetics studies is genetic testing. While the availability and use of genetic testing in the context of autism and other neurodevelopmental and cognitive traits is to an extent driven by public demand for such testing, the advancement of genetic research in itself creates and reshapes such demand.[75] The

genetic architecture of autism and its many presentations includes inherited and de novo variation, and autism-associated loci exhibit highly variable penetrance and expressivity as well as pleiotropy.[13] Genetic testing is often sought out after a child has been diagnosed as autistic, in an effort to identify a genetic cause. Currently, the diagnostic yield of genetic testing of diagnosed toddlers is approximately 12%.[76] While genetic testing may sometimes lead to clinically actionable findings for autistic individuals, a substantial proportion of genetic testing in the context of autism focuses on guiding reproductive decisions.[77] For example, carrier and prenatal tests are currently available for Fragile X syndrome,[78] and in the near future, the development of PGS-based tests may open the door for broader identification of "carriers" across contexts, such as estimating the polygenic likelihood of autism in embryos created via *in vitro* fertilization through preimplantation genetic testing (PGT). . While the information obtained through prenatal or childhood genetic testing may provide valuable information supporting the preparedness of families (Table 1), wide adoption of genetic testing for stigmatized traits has numerous ethical and social challenges.[79]

Autism-related genetic testing is typically carried out in the context of clinical encounters and genetic counseling. While the literature on genetic counseling for autism focuses almost exclusively on parental perspectives on pediatric genetic testing, autistic individuals themselves may also consider genetic testing at different life stages for a variety of reasons in relation to reproductive planning, prenatal care, and/or predisposition to adult-onset conditions. The benefits of autism-related genetic testing are the most direct when genetic or molecular etiology may guide the treatment of specific health problems. For example, in specific cases identifying a molecular cause has informed pharmacotherapy resulting in reduced pain and fatigue and improved speech secondary to the treatment of mitochondrial dysfunction and acetylcholine synthesis.[80] On the other hand, genetic testing may result in harm through increased stigma, pathologization, or discrimination[81], and such possibilities of harm should be acknowledged in pre-testing counseling.

Although limited in number, sample size, and diversity, some studies have surveyed autistic individuals' experiences with genetic testing.[16,82] In a survey of 213 autistics and 868 caregivers, respondents wanted information about what testing can identify, test limitations, and potential risks and benefits of testing.[82] In a survey of 461 autistic adults, only 27% of respondents would have wanted genetic testing during childhood and 74% thought that testing should only be offered if the autistic individual was able to consent.[16] Respondents raised concerns about genetic testing, such as results impacting access to services, that genetic testing might lead to eugenics, and that societal consequences outweighed any potential personal benefits of testing.[16] We recommend further studies on diverse autistic perspectives and experiences to develop translational research and genetic counseling to better serve autistic clients. Studies of parental perspectives on genetic testing of their autistic offspring are more numerous and indicate that parents generally view genetic testing as benefiting families by establishing a cause for autism and informing preparedness or family planning (e.g., [24,83]). Parents did, however, also perceive potential harms, such as discrimination.[84]

Genetic testing in the context of autism involves ethical concerns that have long been voiced by disability rights activists and scholars.[85,86] Genetic counselors and other practitioners ought to aim to support patients in exercising reproductive autonomy, yet research shows that language

and approaches adopted by clinicians can be biased and directive.[87] Critiques of genetic testing and genetic counseling often relate to reproductive settings, and recent work has provided best practices for avoiding ableism in delivering genetic diagnoses.[88] While much of this work focuses on Down syndrome, recommendations can be applied in the context of autism and other neurodevelopmental traits and disabilities.[88] Importantly, as the use of genetic information continues to increasingly influence clinical and reproductive decision-making, researchers and clinicians must work in collaboration with the autistic community and other AIPs to develop an equitable research and health care enterprise. To this end, it is necessary to adopt a broader view of autism and autistic individuals' health care needs, with an increased focus on physical health problems that have so far received limited attention.[13,89,90]

Tackling power imbalances through accountability and autistic inclusion

Beneficial and just genomics research must be actively anti-ableist and conducting such research must be a shared agenda of investigators and stakeholders. Bias leading to inequities is often a reflection of systemic factors, and thus, tackling implicit and explicit bias in autism genomics requires not only individual-level interventions but system-level changes, accomplished through meaningful power-sharing with members of impacted communities.[91] So far, the vast majority of genetic and genomic autism studies have failed to consult or engage with the primary AIPs.[92] Moving forward, researchers should avoid approaches that are extractive or tokenizing and instead aim to facilitate collaboration and co-production by adopting participatory practices.[50,93,94] To tackle the underrepresentation of autistic people in autism research and the negative consequences of this disparity, institutions should seek to support the leadership of autistic investigators. Funders and editors may encourage studies with a strong representation of autistic contributors that make an adequate effort to include autistic community partners and self-advocacy groups in designing and conducting the research (Figure 1; Table 2).

Transparency in communication and decision-making is essential for rebuilding the trust which has been eroded. Thus, investigators, funders, and publishers should take measures to increase transparency and accountability in the field (Figure 1; Table 2).[95] Accountability may be promoted through, for example, open peer review that includes an evaluation of the level of autistic inclusion and transparency in community consultation. To facilitate partnership with the autistic community, the field must engage in an open dialogue with primary AIPs and commit to developing platforms to allow community feedback. This may include inviting self-advocacy groups to contribute to journals and conferences. Importantly, engagement should be accessible for autistic individuals with diverse communication needs and preferences. Institutions and funders may further promote autistic inclusion through fellowships, targeted funding, mentoring, and by establishing autistic review and advisory boards (Figure 1; Table 2).

Lastly, we urge institutions and professional societies to develop programs to educate the field about diversity, disability, and ableism and to incentivize socially responsible research. We encourage the field to welcome opportunities to participate in, reflect upon, and learn from the conversation about the topic, and to center the perspectives, autonomy, and well-being of the primary stakeholders. These efforts will aid in developing a more equitable research enterprise for more just genomics.

Table 2. Summary of the objectives, directives, and outcomes of these recommendations.

Objective	**Directives**	**Outcomes**
Acknowledging past and current abuses and ethical challenges in genetics, genomics, and autism research.	Education of researchers and clinicians.	Increased trust.
Aligning research goals to address the needs and concerns of the autistic community.	Surveying the diverse autistic community to identify research priorities and areas of concern; Co-production and participatory research; Targeted funding and publishing.	Increased trust, safety, and benefits to stakeholders.
Reducing implicit and explicit bias.	Eliminating framing bias; Adopting unbiased, anti-ableist language and research practices; Disability and neurodiversity training.	Reduced stigma; Increased understanding and acceptance; Increased trust; increased benefit to stakeholders.
Ensuring informed consent.	Developing tools to support diverse communications needs; Considering risks and benefits to research participants and the wider autistic community.	Increased trust, safety, and benefits to stakeholders.
Protecting highly vulnerable groups and individuals.	Employing additional safeguards: robust informed consent process, inclusion/exclusion criteria, ensuring that research is responsive to community needs.	Increased trust, safety, and benefits to stakeholders.
Tackling power-imbalances.	Meaningful inclusion; Empowering and supporting autistic investigators, trainees, and community partners through mentorship, funding, and accessibility; Equal partnership; Increasing transparency and accountability.	Increased trust, safety, and benefits to stakeholders; More equitable research enterprise.

Conclusions

There is an ongoing and increasing effort in genetics, bioethics, and biomedical research to examine the role of genetics in healthcare and society and to envision ways in which genetics and genomics can be leveraged for the well-being of all humans in an equitable manner. The development of new genetic technologies and prenatal screening methods has produced numerous ethical challenges that urgently require attention for appropriate resolution. These challenges are particularly notable in the context of neurodevelopmental, psychiatric, behavioral, and other stigmatized traits. Here, we have examined ongoing ethical challenges in autism-related genomics. Leveraging our expertise as geneticists, bioethicists, clinicians, and autism researchers, we have produced a framework and recommendations to enhance ethical autism genetics research for the well-being of autistic stakeholders. These recommendations are widely applicable across traits and communities.

Acknowledgments

We wish to thank Drs Arianna Manzini and Rebecca Mueller for their valuable input regarding this manuscript and the AIR-P for supporting this workgroup. J.A.M-A. is supported by the Health Resources and Services Administration (HRSA) of the U.S. Department of Health and Human Services (HHS) under the Autism Intervention Research Network on Physical Health (AIR-P) grant, UT2MC39440. We thank Drs Heather M. Brown, Maya Sabatello, and others who participated in the workgroup conversations at any stage of manuscript preparation.

Positionality statement

The recommendations presented in this manuscript are the result of numerous meetings of a wider workgroup. The authors are a group of autistic and non-autistic geneticists, bioethicists, autism researchers, clinicians, and advocates. Most authors have doctoral degrees, are affiliated with academic institutions, and are early career researchers. Some of the authors are parents to autistic children and some identify as disabled, multiply disabled, or multiply neurodivergent. Most are white European or North American. The group consists of individuals with diverse gender identities. While this workgroup is diverse, it is not a complete, direct representation of all affected and interested parties (e.g., none of the authors are non-speaking or have an intellectual disability).

Citations

1. Claw KG, Anderson MZ, Begay RL, et al. A framework for enhancing ethical genomic research with Indigenous communities. *Nat Commun*. 2018;9(1):2957.

2. Lipphardt V, Rappold GA, Surdu M. Representing vulnerable populations in genetic studies: The case of the Roma. *Sci Context*. 2021;34(1):69-100.

3. Millum J, Campbell M, Luna F, Malekzadeh A, Karim QA. Ethical challenges in global health-related stigma research. *BMC Med*. 2019;17(1):84.

4. Genetic Research & Autism - Autistic Self Advocacy Network. Accessed June 27, 2022. https://autisticadvocacy.org/genetic-research/

5. Kline W. How geneticists can gain greater buy-in from the autistic community. Spectrum | Autism Research News. Published September 9, 2022. Accessed October 19, 2022. https://www.spectrumnews.org/opinion/viewpoint/how-geneticists-can-gain-greater-buy-in-from-the-autistic-community/

6. Ellis R, Asbury K. AUTISTIC OPINIONS OF GENOMIC STUDIES 1 An investigation of autistic opinions about autism-related genomic research. Accessed July 7, 2023. https://psyarxiv.com/7x8cw/download?format=pdf

7. Miller PS, Levine RL. Avoiding genetic genocide: understanding good intentions and eugenics in the complex dialogue between the medical and disability communities. *Genet Med*. 2013;15(2):95-102.

8. Rajkovic A, Cirino AL, Berro T, Koeller DR, Zayhowski K. Transgender and gender-diverse (TGD) individuals' perspectives on research seeking genetic variants associated with TGD identities: a qualitative study. *J Community Genet*. 2022;13(1):31-48.

9. Berryessa CM, Cho MK. Ethical, legal, social, and policy implications of behavioral genetics. *Annu Rev Genomics Hum Genet*. 2013;14:515-534.

10. Ewelina Rydzewska, Laura A. Hughes-Mccormack, Christopher Gillberg, Angela Henderson, Cecilia MacIntyre, Julie Rintoul, Sally-Ann Cooper. General health of adults with autism spectrum disorders – a whole country population cross-sectional study. *Res Autism Spectr Disord*. 2019;60(April 2019):59-66.

11. Bishop-Fitzpatrick L, Kind AJH. A Scoping Review of Health Disparities in Autism Spectrum Disorder. *J Autism Dev Disord*. 2017;47(11):3380-3391.

12. Doherty M, Neilson S, O'Sullivan J, et al. Barriers to healthcare and self-reported adverse outcomes for autistic adults: a cross-sectional study. *BMJ Open*. 2022;12(2):e056904.

13. Vorstman JAS, Parr JR, Moreno-De-Luca D, Anney RJL, Nurnberger JI Jr, Hallmayer JF. Autism genetics: opportunities and challenges for clinical translation. *Nat Rev Genet*. 2017;18(6):362-376.

14. Roche L, Adams D, Clark M. Research priorities of the autism community: A systematic review of key stakeholder perspectives. *Autism*. 2021;25(2):336-348.

15. Warner G, Parr JR, Cusack J. Workshop Report: Establishing Priority Research Areas to Improve the Physical Health and Well-Being of Autistic Adults and Older People. *Autism in Adulthood*. 2019;1(1):20-26. doi:10.1089/aut.2018.0003

16. Byres L, Morris E, Austin J. Exploring Autistic adults' perspectives on genetic testing for autism. *Genet Med*. Published online January 19, 2023:100021.

17. Pellicano E, Dinsmore A, Charman T. What should autism research focus upon? Community views and priorities from the United Kingdom. *Autism*. 2014;18(7):756-770.

18. Frazier TW, Dawson G, Murray D, Shih A, Sachs JS, Geiger A. Brief Report: A Survey of Autism Research Priorities Across a Diverse Community of Stakeholders. *J Autism Dev Disord*. 2018;48(11):3965-3971.

19. Fletcher-Watson S, Apicella F, Auyeung B, et al. Attitudes of the autism community to early autism research. *Autism*. 2017;21(1):61-74.

20. Putnam OC, Eddy G, Goldblum J, Swisher M, Harrop C. How autistic adults' priorities for autism research differ by gender identity: A mixed-methods study. *Womens Health*. 2023;19:17455057231160342.

21. Dey I, Chakrabarty S, Nandi R, et al. Autism community priorities in diverse low-resource settings: A country-wide scoping exercise in India. *Autism*. Published online March 31, 2023:13623613231154067.

22. Cage E, Crompton CJ, Dantas S, et al. What are the autism research priorities of autistic adults in Scotland? *PsyArXiv*. Published online October 27, 2022. doi:10.31234/osf.io/nbq8h

23. Genetic Research & Autism - Autistic Self Advocacy Network. Accessed February 3, 2023. https://autisticadvocacy.org/genetic-research/

24. Johannessen J, Nærland T, Hope S, et al. Parents' Attitudes toward Clinical Genetic Testing for Autism Spectrum Disorder-Data from a Norwegian Sample. *Int J Mol Sci*. 2017;18(5). doi:10.3390/ijms18051078

25. Soda T, Pereira S, Small BJ, et al. Child and Adolescent Psychiatrists' Perceptions of Utility and Self-rated Knowledge of Genetic Testing Predict Usage for Autism Spectrum Disorder. *Journal of the American Academy of Child & Adolescent Psychiatry*. 2021;60(6):657-660. doi:10.1016/j.jaac.2021.01.022

26. Gewin V. NIH faces calls to change its "ableist" mission statement. Nature Publishing Group UK. doi:10.1038/d41586-023-00260-5

27. Iezzoni LI, Rao SR, Ressalam J, et al. Physicians' Perceptions Of People With Disability And Their Health Care. *Health Aff* . 2021;40(2):297-306.

28. Botha M, Cage E. "Autism research is in crisis": A mixed method study of researcher's constructions of autistic people and autism research. *Front Psychol*. 2022;13. doi:10.3389/fpsyg.2022.1050897

29. Cervantes PE, Matheis M, Estabillo J, et al. Trends Over a Decade in NIH Funding for Autism Spectrum Disorder Services Research. *J Autism Dev Disord*. 2021;51(8):2751-

2763.

30. Lim M, Carollo A, Dimitriou D, Esposito G. Recent Developments in Autism Genetic Research: A Scientometric Review from 2018 to 2022. *Genes* . 2022;13(9). doi:10.3390/genes13091646

31. Manzini A, Jones EJH, Charman T, Elsabbagh M, Johnson MH, Singh I. Ethical dimensions of translational developmental neuroscience research in autism. *J Child Psychol Psychiatry*. 2021;62(11):1363-1373.

32. Pukki H, Bettin J, Outlaw AG, et al. Autistic perspectives on the future of clinical autism research. *Autism Adulthood Knowl Pract Policy*. 2022;4(2):93-101.

33. Tan DW. Early-Career Autism Researchers Are Shifting Their Research Directions: Tragedy or Opportunity? *Autism in Adulthood*. Published online April 3, 2023. doi:10.1089/aut.2023.0021

34. Oredipe T, Kofner B, Riccio A, et al. Does learning you are autistic at a younger age lead to better adult outcomes? A participatory exploration of the perspectives of autistic university students. *Autism*. 2023;27(1):200-212. doi:10.1177/13623613221086700

35. Dawson G, Franz L, Brandsen S. At a Crossroads—Reconsidering the Goals of Autism Early Behavioral Intervention From a Neurodiversity Perspective. *JAMA Pediatrics*. 2022;176(9):839. doi:10.1001/jamapediatrics.2022.2299

36. McCoy MS, Liu EY, Lutz ASF, Sisti D. Ethical Advocacy Across the Autism Spectrum: Beyond Partial Representation. *Am J Bioeth*. 2020;20(4):13-24.

37. Department of Health, Education, and Welfare, National Commission for the Protection of Human Subjects of Biomedical and Behavioral Research. The Belmont Report. Ethical principles and guidelines for the protection of human subjects of research. *J Am Coll Dent*. 2014;81(3):4-13.

38. Tesfaye R, Courchesne V, Yusuf A, et al. Assuming ability of youth with autism: Synthesis of methods capturing the first-person perspectives of children and youth with disabilities. *Autism*. 2019;23(8):1882-1896. doi:10.1177/1362361319831487

39. Beauchamp TL, Childress JF. *Principles of Biomedical Ethics*. Oxford University Press, USA; 2019.

40. Ferretti A, Ienca M, Sheehan M, et al. Ethics review of big data research: What should stay and what should be reformed? *BMC Medical Ethics*. 2021;22(1). doi:10.1186/s12910-021-00616-4

41. Office for Human Research Protections (OHRP). The Protection of Non-Subjects from Research Harm. HHS.gov. Published March 11, 2022. Accessed February 4, 2023. https://www.hhs.gov/ohrp/sachrp-committee/recommendations/tab-c-the-protection-of-non-subjects-from-research-harm.html

42. Lencz T, Sabatello M, Docherty A, et al. Concerns about the use of polygenic embryo screening for psychiatric and cognitive traits. *The Lancet Psychiatry*. 2022;9(10):838-844. doi:10.1016/s2215-0366(22)00157-2

43. Emanuel E.J., Wendler D, Grady C. An ethical framework for biomedical research, in Emanuel E.J. et al. (eds.). The Oxford textbook of clinical research ethics, Oxford University Press, New York 2008, pp. 123-135.

44. Cascio MA, Weiss JA, Racine E. Making Autism Research Inclusive by Attending to Intersectionality: a Review of the Research Ethics Literature. *Review Journal of Autism and Developmental Disorders*. 2021;8(1):22-36. doi:10.1007/s40489-020-00204-z

45. Kopal J, Uddin LQ, Bzdok D. The end game: respecting major sources of population diversity. *Nat Methods*. Published online March 3, 2023. doi:10.1038/s41592-023-01812-3

46. Bottema-Beutel K, Kapp SK, Lester JN, Sasson NJ, Hand BN. Avoiding Ableist Language: Suggestions for Autism Researchers. *Autism Adulthood*. 2021;3(1):18-29.

47. Dwyer P, Ryan JG, Williams ZJ, Gassner DL. First Do No Harm: Suggestions Regarding Respectful Autism Language. *Pediatrics*. 2022;149(Suppl 4). doi:10.1542/peds.2020-049437N

48. Science must respect the dignity and rights of all humans. *Nature Human Behaviour*. 2022;6(8):1029-1031. doi:10.1038/s41562-022-01443-2

49. Iyer AA, Saade D, Bharucha-Goebel D, et al. Ethical challenges for a new generation of early-phase pediatric gene therapy trials. *Genet Med*. 2021;23(11):2057-2066.

50. Keating CT. Participatory Autism Research: How Consultation Benefits Everyone. *Front Psychol*. 2021;12:713982.

51. Faden RR, Philip Frankilin Wagley Professor of Biomedical Ethics and Director of the Bioethics Institute Ruth R Faden, Beauchamp TL, King NMP. *A History and Theory of Informed Consent*. Oxford University Press on Demand; 1986.

52. Farroni JS, Moore JA, Gallagher CM. An Ethical Framework for Disclosing Individual Genetic Findings to Patients, Research Participants, and Relatives. *Ethical Challenges in Oncology*. Published online 2017:275-296. doi:10.1016/b978-0-12-803831-4.00018-x

53. Tisherman SA. Defining “Community” and “Consultation” for Emergency Research that Requires an Exception from Informed Consent. *AMA J Ethics*. 2018;20(5):467-474.

54. Fitzpatrick EFM, Martiniuk ALC, D’Antoine H, Oscar J, Carter M, Elliott EJ. Seeking consent for research with indigenous communities: a systematic review. *BMC Med Ethics*. 2016;17(1):65.

55. Maloy JW, Bass PF. Understanding Broad Consent. *Ochsner Journal*. 2020;20(1):81-86. doi:10.31486/toj.19.0088

56. Lemke AA, Esplin ED, Goldenberg AJ, et al. Addressing underrepresentation in genomics research through community engagement. *Am J Hum Genet*. 2022;109(9):1563-1571.

57. Kaye J, Whitley EA, Lund D, Morrison M, Teare H, Melham K. Dynamic consent: a patient interface for twenty-first century research networks. *Eur J Hum Genet*. 2015;23(2):141-146.

58. Dankar FK, Gergely M, Dankar SK. Informed Consent in Biomedical Research. *Comput Struct Biotechnol J*. 2019;17:463-474.

59. Mikkelsen RB, Gjerris M, Waldemar G, Sandøe P. Broad consent for biobanks is best - provided it is also deep. *BMC Med Ethics*. 2019;20(1):71.

60. Bracken-Roche D, Bell E, Macdonald ME, Racine E. The concept of "vulnerability" in research ethics: an in-depth analysis of policies and guidelines. *Health Res Policy Syst*. 2017;15(1):8.

61. Griffiths S, Allison C, Kenny R, Holt R, Smith P, Baron-Cohen S. The Vulnerability Experiences Quotient (VEQ): A Study of Vulnerability, Mental Health and Life Satisfaction in Autistic Adults. *Autism Res*. 2019;12(10):1516-1528.

62. Thompson S, Cannon M, Wickenden M. *Exploring Critical Issues in the Ethical Involvement of Children with Disabilities in Evidence Generation and Use*.; 2020.

63. Pang RCC, Ho MSH, Wong PWC. A Review of the Literature on the Multiple Forms of Stigmatization of Caregivers of Children with Autism Among Ethnic Minority Groups. *J Racial Ethn Health Disparities*. Published online March 6, 2023. doi:10.1007/s40615-023-01540-6

64. Kasperbauer TJ, Halverson C. Adolescent Assent and Reconsent for Biobanking: Recent Developments and Emerging Ethical Issues. *Front Med*. 2021;8:686264.

65. Zeidan J, Fombonne E, Scorah J, et al. Global prevalence of autism: A systematic review update. *Autism Res*. 2022;15(5):778-790.

66. Lennox N, Taylor M, Rey-Conde T, Bain C, Purdie DM, Boyle F. Beating the barriers: recruitment of people with intellectual disability to participate in research. *J Intellect Disabil Res*. 2005;49(Pt 4):296-305.

67. Gupta UC, Kharawala S. Informed consent in psychiatry clinical research: A conceptual review of issues, challenges, and recommendations. *Perspect Clin Res*. 2012;3(1):8-15.

68. Horner-Johnson W, Bailey D. Assessing Understanding and Obtaining Consent from Adults with Intellectual Disabilities for a Health Promotion Study. *J Policy Pract Intellect Disabil*. 2013;10(3). doi:10.1111/jppi.12048

69. Ho P, Downs J, Bulsara C, Patman S, Hill AM. Addressing challenges in gaining informed consent for a research study investigating falls in people with intellectual disability. *British Journal of Learning Disabilities*. 2018;46(2):92-100. doi:10.1111/bld.12217

70. Strnadová I, Loblinzk J, Scully JL, et al. "I am not a number!" Opinions and preferences of people with intellectual disability about genetic healthcare. *European Journal of Human Genetics*. Published online 2023. doi:10.1038/s41431-023-01282-3

71. Wodka EL, Mathy P, Kalb L. Predictors of phrase and fluent speech in children with autism and severe language delay. *Pediatrics*. 2013;131(4):e1128-e1134.

72. Light J, McNaughton D, Caron J. New and emerging AAC technology supports for children with complex communication needs and their communication partners: State of the science and future research directions. *Augment Altern Commun*. 2019;35(1):26-41.

73. Asbury K, Toseeb U, Barrow N. What do parents of nonverbal and minimally verbal autistic children think about genomic autism research? Published online 2023.

https://psyarxiv.com/nj5yh/download?format=pdf

74. Office for Human Research Protections (OHRP). Attachment: Recommendations Regarding Research Involving. HHS.gov. Published July 15, 2009. Accessed July 2, 2023. https://www.hhs.gov/ohrp/sachrp-committee/recommendations/2009-july-15-letter-attachment/index.html

75. Nuffield Council on Bioethics (London). *Medical Profiling and Online Medicine: The Ethics of "Personalised Healthcare" in a Consumer Age*.; 2010.

76. Harris HK, Sideridis GD, Barbaresi WJ, Harstad E. Pathogenic Yield of Genetic Testing in Autism Spectrum Disorder. *Pediatrics*. 2020;146(4). doi:10.1542/peds.2019-3211

77. Griesi-Oliveira K, Sertié AL. Autism spectrum disorders: an updated guide for genetic counseling. *Einstein* . 2017;15(2):233-238.

78. Gutiérrez JF, Bajaj K, Klugman SD. Prenatal screening for fragile x: carriers, controversies, and counseling. *Rev Obstet Gynecol*. 2013;6(1):e1-e7.

79. Ethics Statements. Accessed April 5, 2023. https://ispg.net/ethics-statement/

80. Kreiman BL, Boles RG. State of the Art of Genetic Testing for Patients With Autism: A Practical Guide for Clinicians. *Semin Pediatr Neurol*. 2020;34:100804.

81. McMahon WM, Baty BJ, Botkin J. Genetic counseling and ethical issues for autism. *Am J Med Genet C Semin Med Genet*. 2006;142C(1):52-57.

82. Hellquist A, Tammimies K. Access, utilization, and awareness for clinical genetic testing in autism spectrum disorder in Sweden: A survey study. *Autism*. 2022;26(7):1795-1804.

83. Zhao S, Chen WJ, Dhar SU, Eble TN, Kwok OM, Chen LS. Genetic Testing Experiences Among Parents of Children with Autism Spectrum Disorder in the United States. *J Autism Dev Disord*. 2019;49(12):4821-4833.

84. Johannessen J, Nærland T, Hope S, et al. Attitudes among parents of persons with autism spectrum disorder towards information about genetic risk and future health. *Eur J Hum Genet*. 2022;30(10):1138-1146.

85. Parens E, Asch A. Disability rights critique of prenatal genetic testing: reflections and recommendations. *Ment Retard Dev Disabil Res Rev*. 2003;9(1):40-47.

86. Morris E, O'Donovan M, Virani A, Austin J. An ethical analysis of divergent clinical approaches to the application of genetic testing for autism and schizophrenia. *Hum Genet*. 2022;141(5):1069-1084.

87. Williams C, Alderson P, Farsides B. Is nondirectiveness possible within the context of antenatal screening and testing? *Soc Sci Med*. 2002;54(3):339-347.

88. Meredith S, Brackett S, Diaz KM, et al. Recommendations to improve the patient experience and avoid bias when prenatal screening/testing. *Disabil Health J*. Published online November 5, 2022:101401.

89. Croen LA, Zerbo O, Qian Y, et al. The health status of adults on the autism spectrum.

Autism. 2015;19(7):814-823.

90. McDonald TAM, Scudder A. Mind the NIH-Funding Gap: Structural Discrimination in Physical Health-Related Research for Cognitively Able Autistic Adults. *J Autism Dev Disord*. Published online January 13, 2023. doi:10.1007/s10803-022-05856-w

91. Vela MB, Erondu AI, Smith NA, Peek ME, Woodruff JN, Chin MH. Eliminating Explicit and Implicit Biases in Health Care: Evidence and Research Needs. *Annu Rev Public Health*. 2022;43:477-501.

92. Fletcher-Watson S. DART lab student outputs. Published online August 16, 2020. doi:10.17605/OSF.IO/HZEX2

93. Atutornu J, Milne R, Costa A, Patch C, Middleton A. Towards equitable and trustworthy genomics research. *EBioMedicine*. 2022;76:103879.

94. Talebizadeh Z, Shah A, AutGO Working Group. The AutGO Initiative: A Conceptual Framework for Developing Genetics-Outcomes Research Hypotheses. *Autism Research*. 2020;13(8):1286-1299. doi:10.1002/aur.2331

95. Devaney S. Rebuilding Trust in Health Research Among Marginalized Communities: Five Lessons from All of Us. Accessed March 9, 2023. https://www.wiley.com/en-us/network/publishing/research-publishing/trending-stories/rebuilding-trust-in-health-research-among-marginalized-communities-five-lessons-from-all-of-us