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Preferences for the research use of electronic health records among young adults with fragile X syndrome or autism spectrum disorder

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Abstract

Background.—Health researchers are increasingly using electronic health records (EHRs) to study the health care needs of people with neurodevelopmental disorders (NDDs). However, little is known about the preferences of people with NDDs for sharing EHRs for research.

Objective.—To explore preferences for sharing EHRs for research among young adults ages 18–40 who make their own legal decisions and who have autism spectrum disorder (ASD), fragile X syndrome (FXS), or no NDDs.

Methods.—We conducted a qualitative study with seven focus groups: 2 ASD groups, 3 FXS groups, and 2 no-NDD groups. We asked participants about factors that could affect their willingness to share their EHRs for research: type of organization, type of information, study purpose, duration, contact frequency, return of results, benefits, and risks. We analyzed the qualitative data using directed content analysis.

Results.—Participants with NDDs valued personally relevant and directly beneficial EHR research. Participants with NDDs expressed willingness to share sensitive data if the study was personally relevant. Most participants wanted to receive results, but only participants with FXS indicated it would affect their willingness to participate. Participants were concerned about privacy risks, discrimination, researcher misconduct, and financial conflicts of interest.

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Conclusion.—This study provides initial evidence suggesting that young adults with NDDs prefer EHR research that is personally relevant, benefits themselves and their communities, and is conducted in the context of trusting, reciprocal participant-researcher relationships. The findings point to the need for researchers to improve the informed consent process and to better engage individuals with NDDs in research.

Previous presentation of abstracts at meetings regarding the research:

Andrews S, Raspa M, Wheeler AC, et al. Including individuals with IDD in EHR research: What factors inform consent? Presented at the 2018 American Public Health Association Annual Meeting; 2018; San Diego, CA.

Edwards A, Raspa M, Andrews S, et al. Perspectives of young adults with an autism spectrum disorder on the use of electronic health records in research: The balance between privacy and genetic discovery. Presented at the 2018 International Society for Autism Research Conference; 2018; Rotterdam, Netherlands.

Edwards AM, Turner-Brown L, Andrews SM, et al. The decision to participate in EHR research: Factors important to adults with ASD. Poster presented at the 2019 International Society for Autism Research Conference; 2019; Montreal, Canada.

Wagner L, Frisch M, Edwards A, et al. Perspectives of young adults with neurodevelopmental disorders on the research use of electronic health records. Poster presented at the 2019 American Association on Intellectual and Developmental Disabilities Annual Meeting; 2019; St. Paul-Minneapolis, MN.

Keywords

Electronic health records; patient preference; neurodevelopmental disorders; autism spectrum disorder; fragile X syndrome

Introduction

Neurodevelopmental disorders (NDDs) are conditions characterized by developmental differences that lead to impairments in personal, social, academic, or occupational functioning (1). Individuals with NDDs have higher rates of health care utilization but still experience health care disparities (2–4). Although electronic health records (EHRs) are primarily used to document health care visits, they are increasingly being used for research to understand causes, symptoms, and comorbidities of NDDs, as well as to evaluate treatments and therapies (5, 6). This method provides valuable information that is difficult to obtain through other methodologies and promises to shed light on the health and health care needs of individuals with NDDs.

Several studies among the general population have examined patient views toward the use of EHRs for research. They have identified patients' concerns about trust, privacy, and security (7–11). Patients have reported concerns about sharing data with health insurers, pharmaceutical companies, and government agencies (10, 11). They tend to prefer data to remain anonymous and favor limiting the sharing of sensitive information to maintain

privacy. Despite this, the public generally supports the research use of EHRs, and most people perceive the benefits to outweigh the risks (8–10).

However, little is known about the views of individuals with NDDs on this topic. Some studies have examined the decision-making ability of individuals with NDDs and their desire to participate in research (12–14). Studies have shown that some individuals with NDDs may need supports to make informed health care decisions (15–17). Other work has shown that those with NDDs want to be active participants in research, starting with the development of research questions and study design (18, 19). We must hear directly from individuals themselves to sufficiently understand their preferences, as their values and goals may differ from those of families, caregivers, surrogates, or those with more severe impairments (20). Individuals with NDDs face important challenges during young adulthood, i.e., the late teens, twenties, and thirties (21). They increasingly develop more independence and gain more self-reliance in making their own medical and legal decisions, while typically being in good health, so exploration of the preferences of this age group is particularly important.

In this qualitative study, we explored factors that young adults with NDDs consider when deciding to consent to the research use of their EHRs. As part of a larger study of the preferences of individuals with NDDs regarding the research use of EHRs, this study aimed to identify attributes for a subsequent quantitative preference study (manuscript forthcoming). Gathering qualitative evidence prior to developing a preference study is important to strengthen the quality, relevance, and validity of evidence (22).

We focused on two conditions that fall under the umbrella of NDDs: fragile X syndrome (FXS), a known genetic condition, and autism spectrum disorder (ASD), a suspected genetic condition. Individuals with either condition can have intellectual disability or co-occurring conditions like anxiety, attention problems, seizures, or aggressive behavior. Although FXS and ASD do not represent all individuals with NDDs, they are excellent examples because of their heritability patterns and wide spectrum of involvement (i.e., some individuals with FXS or ASD are very severely impacted, while others are only mildly impacted). To explore how their views compared to individuals who did not report significant health or genetic conditions, we included a group without NDDs for comparison.

Our research questions were informed by a literature review and input from an advisory panel of experts on NDD research. Our research question was: What are the preferences, interests, and needs of individuals with NDDs when deciding whether to participate in EHR research when compared to those without NDDs? We highlight the themes that emerged from focus groups and discuss implications for researchers.

Methods

Participants

We conducted seven focus groups in various parts of the United States: Northeast (Baltimore, MD; Newark, NJ), Midwest (Chicago, IL), and Southeast (Raleigh-Durham, NC; Huntington, WV). Three groups were conducted with young adults with FXS (n=13),

two groups with young adults with ASD (n=15), and two groups with young adults without NDDs (n=16). Young adults ages 18–40 who were their own legal guardian were eligible to participate. Individuals with FXS or ASD were eligible if they had graduated high school with an academic degree, were living or working independently, and were capable of participating in the study. We excluded individuals without NDDs if they had genetic disorders, major health concerns, or intellectual disabilities. To achieve four to nine participants per group, we recruited a convenience sample of participants with NDDs from research registries based at the authors' institutions, university programs for students with ASD, and advocacy organization listservs. We used a research firm to recruit young adults without NDDs. Many of the participants or their parents had taken part in previous research studies.

During each focus group, we collected demographic information and measured participants' level of functioning using the Waisman Daily Living (WDL) Scale (23). We noted some key demographic differences among the groups (Table 1). Groups with NDDs significantly differed by sex ($p<.0001$); all participants with FXS were women, while most participants with ASD were men. Groups differed by race with fewer racial and ethnic minorities in the groups with NDDs ($p=0.0023$). Fewer participants with NDDs had 4-year college degrees ($p=0.0013$) or were employed full-time ($p=0.0001$) than those without NDDs. Over half of participants without NDDs were married; all participants with NDDs were single ($p<.0001$). The mean age was 26.8 years, and on average, participants without NDDs were older than those with NDDs ($F=6.72$, $p=0.003$). The age range was 23–38 for participants without NDDs, 19–32 for participants with ASD, and 18–38 for participants with FXS. Mean WDL scores differed between groups also ($F=6.98$, $p=0.0025$), with mean scores for FXS (30.2) and ASD groups (31.0) being slightly lower than the group without NDDs (34.0). Mean WDL scores for each group with NDDs were all above 24.9 (the cut-off point considered to indicate a mild level of intellectual disability).

Data collection

A moderator and one or two notetakers conducted the focus groups. The moderator used a semi-structured guide (Table 2) to facilitate the discussion. Moderators of the NDD groups had experience working with individuals with ASD and FXS. To reduce the potential for social anxiety, the FXS groups were smaller than the others, and parent transportation costs were covered by the study (many FXS participants were unable to travel alone). In keeping with recommended practices for focus groups, moderators asked if participants needed additional information, checked for understanding, redirected off-topic conversation, encouraged different opinions, and directly engaged quieter participants. The moderator presented an overview of EHRs (e.g., type of information in EHRs, who uses EHRs and how, privacy protections of EHRs) and their use in research. Discussion questions focused on factors that could affect participants' willingness to allow their EHRs to be used for research, including (a) familiarity with EHRs, (b) general attitudes towards sharing EHRs for research, (c) type of organization that could conduct the research, (d) type of information, (e) study purpose, (f) study duration, (g) frequency of contact about use of EHRs, (h) return of results, (i) benefits, and (j) risks.

The Institutional Review Board of the lead author's institution approved the study. We obtained written consent from participants. Each focus group lasted 90 minutes, and participants were given a monetary gift for their time.

Analysis

Focus group recordings were transcribed verbatim, de-identified by members of the research team, and imported into NVivo 11 qualitative analysis software (24). Coded data were analyzed using directed content analysis (25). The study team drafted a codebook based on the moderator guide and notes from the focus groups. Eight coding categories structured the codebook: 1) type of organization, 2) type of information, 3) study purpose, 4) study duration, 5) frequency of contact, 6) return of results, 7) benefits, and 8) risks. Two team members coded a transcript using the draft codebook, calculated inter-rater reliability, compared coding, and refined the codebook. After two rounds of refinement with three transcripts, Kappa coefficients for each code fell between 0.51–0.89, which is considered moderate to excellent agreement (26, 27). The remaining four transcripts were coded independently.

The team held debriefing meetings throughout the analysis to synthesize the data. Coding categories were classified into a framework matrix to compare similarities and differences across the three groups and identify themes regarding factors that affected willingness to share their EHRs for research (28). We have identified relevant quotations to illustrate findings within each category and have presented them verbatim, including speech dysfluencies.

Results

Type of organization

In general, participants were willing to share information from their EHRs for research with non-profit organizations, universities, and hospitals and felt most academic and clinical researchers were credible and trustworthy. Groups with NDDs emphasized the importance of a trusting relationship and said they were willing to share information if their physician endorsed or conducted the study. As one participant with FXS said, “like, I, if my doctor wanted to perform a research on something, I would be totally game for it because I trust them and I know that they would be—they wouldn't give the information to the wrong people.” If they did not know the researcher personally, some individuals with ASD and FXS said they would be willing to share information if they felt the researchers had robust and appropriate credentials.

Willingness to share information with government researchers varied based on how participants interpreted the term “government.” Participants trusted specific, well-known, health-related government organizations, like the National Institutes of Health, the Food and Drug Administration, and the Centers for Disease Control and Prevention. However, participants were worried about “the government” writ large, mentioning concerns about invasion of privacy and politician involvement.

Most participants across all groups expressed limited willingness to share their EHRs with for-profits (e.g. pharmaceutical companies, insurance companies) than with non-profits. Participants strongly distrusted health insurers because of potential financial conflicts of interest. They felt ambivalent toward pharmaceutical companies. As one participant with ASD said, “the researching drugs is, is beneficial in every way, but there’s—the profit part is where it gets tricky.”

Type of information

Most participants were willing to share de-identified data but were hesitant or unwilling to share identifiable data. Typically, participants considered data “sensitive” if the data held identifiers. Other types of data, even if de-identified, were seen as sensitive, including mental health history, medical diagnoses (including ASD and FXS), and reproductive and sexual health history.

Participants with FXS felt that they would be willing to share information they might otherwise consider sensitive, as long as the study was related to FXS.

“I feel like it kind of goes back to that whole thing, like, about if it’s just about the fragile X part, then that’s fine, but, like, if it’s—because having fragile X sometimes can, I mean, for me, like, I’ve had, like, some depression and some anxiety here and there, but if they wanted to know more about that part, maybe I wouldn’t, I mean, I wouldn’t mind sharing it.”

(FXS participant)

Similarly, participants with ASD were willing to share sensitive information, such as autism diagnoses, treatments, and therapies, for autism research. Participants with NDDs were reluctant to provide information that they saw as irrelevant to research on their condition, including demographics, income, education level, and insurance information.

Purpose of study

Most participants were willing to share EHRs for research if the study could benefit others or improve health care. They had positive attitudes towards research that aimed to prevent or cure illness, develop new medications, create more affordable health care, or advance genetic research. Many participants with ASD or FXS said they were especially willing to share EHRs on topics of personal importance, i.e., topics that they were passionate about or that mattered to them, such as conditions they or a family member had.

“I’d be more than happy to share my records just because I don’t want someone to go through what I went through and not have any answers because they should have answers by now. No kid should have to go through what I went through.”

(FXS participant)

Duration of study

Participants preferred to share their data for a set amount of time rather than indefinitely, often due to uncertainty about what would happen if data access were unlimited.

“If I say you can have free rein of my information for the next ten years, are they going to actually tell me that I’m now part of this study, or are they going to just say, ‘he gave us permission, let’s go ahead with it?’ That’s primarily why I favor shorter times.”

(ASD participant)

Frequency of contact

Participants preferred to give consent for studies on a case-by-case basis and did not feel comfortable with a blanket consent model that could enroll them in any studies at the institution. They were open to researchers re-contacting them and preferred more contact rather than less, feeling this would give them more autonomy over how their EHRs were used. As one participant with ASD stated:

“Let’s say I want to share something with this company and the Department of Health and Human Services for about five years or so, that’s the term of consent. They can use such info beyond that if I sign an extension of that consent and if I don’t sign, then they would need to get rid of such records, and they wouldn’t be allowed to use my records again unless I renew such consent. That’s where the freedom thing kicks in.”

While participants’ preferences about how often they wanted to be re-contacted varied widely (e.g. weekly, every 6 months, every 5 years), the common factor was their desire for greater control over their data, which they valued over possible trade-offs such as increased cost or length of research.

Return of results

Participants in most groups expressed interest in receiving a summary of research results. They were interested in learning how their data had contributed to research, how the findings were relevant to them, and how the study had helped others. The relative importance of receiving results varied between groups. Participants with ASD or without NDDs said results were “nice to know,” but that they would not be a “deciding factor” in their willingness to share EHRs. Some were concerned about not understanding the results or about their privacy. In contrast, receiving results was important to participants with FXS. These participants were interested in receiving results both for increasing their own knowledge and for sharing with their health care providers. Several said they would only share their EHRs if researchers promised to share results.

“It’s very important. Being at this stage, I would like to know what they’ve come up with and with regard to what I have. But in, in general, I would like to know what they’ve come up with—it’s very important to me. Because I didn’t just give you that for kicks, I want to know what you’ve come up with.”

(FXS participant)

Benefits

Participants in all groups cited the altruistic desire to “help a good cause” or improve the quality of life of others through research participation. They discussed how EHR research

could benefit others by improving health care, helping to find cures for diseases, and increasing scientific knowledge. Participants with NDDs also saw EHR research as providing both personal and community benefit. They felt it directly benefited them and their families, helped to improve the quality of life of those like themselves, and raised awareness about their condition.

“Since the personal interest is tied in this, like, you could find a little bit more about yourself, depending on what the study is about, and just gain, like, more knowledge of, like, how you can just better yourself and maybe someone that, like, has the same thing as you.”

(FXS participant)

Risks

The major risks of sharing EHRs for research that all groups identified were loss of privacy and confidentiality, stigma and discrimination, and research misconduct. Participants saw sharing identifiable information as risky, and they were concerned about data breaches and their data getting “in the wrong hands.” If identifiable data were leaked, participants were concerned about possible consequences, including research being done without consent, identity theft, social stigma, job discrimination, and higher insurance costs. Most participants agreed that de-identification of data could mitigate these risks. Participants disagreed about how much they trusted that legal measures could mitigate risks, with some more cynical than others.

Participants across groups voiced concerns about the risk of research misconduct. Concerned that a researcher could mislead them about the purpose of a study or withhold information from them, participants feared EHRs would be used against a group of individuals to exploit them for profit or personal gain. Participants were concerned that that they might not understand what they were agreeing to because of the complexity or length of consent forms. Participants without NDDs also alluded to past human subject violations, suggesting that historical instances of misconduct continue to affect current-day perceptions about research. All groups were concerned about profit motives negatively affecting patients, but it seemed particularly salient for groups with NDDs.

“There are a lot of, frankly, ethical considerations. I mean, say that one day they do come up with a cure for autism, say the company is run by this Shkreli guy and he wants to charge a million dollars a patient, could you support that ethically? That would be really tough. And say they have a patent on it for thirty years. And frankly, a lot of autistic people or disabled people are broke.”

(ASD participant)

Participants mentioned ways their concerns could be allayed. Some said that seeing the researchers' credentials would help. Others felt that they would benefit from clear, short consent forms and suggested patients be given more options about what to share. Some participants with FXS preferred one-on-one verbal communication instead of reading documents. Others wanted to talk with researchers, previous participants, peers, or family before consenting.

Discussion

Here, we primarily discuss findings from the two groups with NDDs because previous work has described the views of the general population in detail, while the views of those with NDDs have been under-represented. We found in our study that the two groups with NDDs valued research that is personally relevant, is directly beneficial, helps others with NDDs, and is conducted in the context of trusting, reciprocal participant-researcher relationships. Key concerns centered on risks to privacy and confidentiality, stigma and discrimination, researcher misconduct, and financial conflicts of interest. Participants reported that their perceptions of trust and transparency could affect their willingness to participate. Many participants with NDDs expressed an interest in engaging in a dialogue with researchers.

Maximizing benefit to research participants with NDDs

Our study provides qualitative evidence that young adults with NDDs may perceive the relevance and importance of potential benefits of EHR research differently than those without NDDs. The results suggest that young adults with NDDs highly value personally relevant research that may directly benefit them or their communities. How the potential direct benefits affect willingness to participate, even despite potential risks, is an area for exploration in future research.

One way to ensure relevance and impact of a study on the population of interest is to take a community-based participatory research approach or employ other participatory research methodologies (29). These approaches gather meaningful input from the target population to help shape research questions, aims, and methods; translate findings into accessible formats; address barriers to participation; and conduct research that helps improve the lives of the people being studied (30). Building patient engagement into research is an important step. For example, organizations can follow the lead of institutions such as the Patient-Centered Outcomes Research Institute, which requires that all funded studies have an engagement plan that details how patients and stakeholders will be meaningfully involved in the study (13, 31).

Another way to benefit young adults with NDDs who participate in research is to share study results. In our study, participants with FXS reported that return of results was important and could affect their willingness to participate. Other studies have also found that participants are more eager to participate in research when they know they will receive results (32). For example, people who have participated in studies that investigated genetic conditions have also been in favor of receiving results, especially if they suspect they may be at risk for a genetic condition (33). Unfortunately, in current practice, researchers who study NDDs often do not take the time or have the resources to disseminate results in a participant-friendly way (34). Some individuals with disabilities have said that research results are not usually in a form they can understand (35). A potential solution to this challenge is building in participant-friendly dissemination into research practice.

Minimizing risk to participants with NDDs

Clearly identifying risks is paramount to informed consent. In our study, participants were particularly concerned about how loss of privacy could result in stigma or discrimination; they saw these risks as a direct consequence of a data breach. This finding provides additional evidence that—despite laws such as the Genetics Information Non-discrimination Act—individuals with known or suspected genetic NDDs are still concerned about privacy of their information, as a data breach could not only impact the individual, but their family members as well (36). Who has access to participant data (and for how long) should be carefully considered by researchers and conveyed to participants during the consent process (37).

Many of our findings speak to the issue of trust more broadly. This initial evidence suggests that, to consider sharing EHRs for research, young adults with NDDs need to trust that their data will be safe with the researcher. They prefer having control over who accesses their data and how, and they favor having an interpersonal connection or relationship with the researchers. Our findings align with other studies on preferences for research use of EHRs, which have also reported participants' need to trust the individuals who use their data (38–40). With the movement towards precision medicine and the use of biomedical data, researchers will continue to need to develop trusting relationships with participants (41).

Limitations

This was a qualitative study that relied on convenience sampling. The groups had significant demographic differences, such as age, which were not controlled for in the qualitative analysis, which could have contributed to differences in responses between groups. Because many of the participants or their parents had participated in research before, they may have had different views of EHR research than individuals not connected to research registries or recruitment firms. We also recognize that the moderators' roles as researchers may have introduced bias into focus group discussions. Thus, inferences about the transferability of our results to other settings or groups should be made cautiously, and the findings reported here are not meant to indicate statistically significant differences in opinions towards EHR research among respondent groups. Rather, the findings identify areas to explore in future research with a broader sample of individuals.

Conclusions

With the continued advancement of health information technologies for research, we must direct our attention to carefully considering young adults' preferences about data sharing for research. This is especially true for young adults with NDDs who may benefit the most from this type of research, but whose data may also need special protections.

This study has highlighted conditional support for EHR research among those with ASD or FXS. While young adults with NDDs in our study raised concerns about privacy, confidentiality, discrimination, and stigma, they were generally willing to participate in EHR research when they perceived it to be personally relevant, directly beneficial, beneficial to their community, and conducted in the context of a trusting, reciprocal relationship with

researchers. This study also points to the need for researchers to improve the consent process to raise awareness about current research practices, support patients in making informed decisions, and promote transparency. EHR-based research must fulfill both participants' values and researchers' goals in seeking to end health disparities and to improve the health and health care of those with NDDs.

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References

1. American Psychiatric Association. Neurodevelopmental Disorders Diagnostic and Statistical Manual of Mental Disorders. 5th ed. Arlington, VA, 2013.
2. Wheeler AC, Wylie A, Villagomez A, Bishop E, Raspa M. Health care for individuals with fragile X syndrome: Understanding access and quality. *Disabil Health J.* 2019;12(2):269–77. [PubMed: 30442576]
3. Krahn GL, Walker DK, Correa-De-Araujo R. Persons with disabilities as an unrecognized health disparity population. *Am J Public Health.* 2015;105 Suppl 2:S198–206. [PubMed: 25689212]
4. Vogan V, Lake JK, Tint A, Weiss JA, Lunsy Y. Tracking health care service use and the experiences of adults with autism spectrum disorder without intellectual disability: A longitudinal study of service rates, barriers and satisfaction. *Disabil Health J.* 2017;10(2):264–70. [PubMed: 27899267]
5. Tyler C, Schramm S, Karafa M, Tang AS, Jain A. Electronic health record analysis of the primary care of adults with intellectual and other developmental disabilities. *J Policy Pract Intellect Disabil.* 2010;7(3):204–10. [PubMed: 26113866]
6. Doshi-Velez F, Ge Y, Kohane I. Comorbidity clusters in autism spectrum disorders: An electronic health record time-series analysis. *Pediatrics.* 2014;133(1):e54–63. [PubMed: 24323995]
7. Kim KK, Sankar P, Wilson MD, Haynes SC. Factors affecting willingness to share electronic health data among California consumers. *BMC Med Ethics.* 2017;18.
8. Trinidad SB, Fullerton SM, Bares JM, et al. Genomic research and wide data sharing: Views of prospective participants. *Genet Med.* 2010;12(8):486–95. [PubMed: 20535021]
9. Clerkin P, Buckley BS, Murphy AW, MacFarlane AE. Patients' views about the use of their personal information from general practice medical records in health research: a qualitative study in Ireland. *Fam Pract.* 2013;30(1):105–12. [PubMed: 22850249]
10. Willison DJ, Keshavjee K, Nair K, et al. Patients' consent preferences for research uses of information in electronic medical records: Interview and survey data. *BMJ.* 2003;326(7385):373. [PubMed: 12586673]
11. Spencer K, Sanders C, Whitley EA, et al. Patient perspectives on sharing anonymized personal health data using a digital system for dynamic consent and research feedback: A qualitative study. *J Med Internet Res.* 2016;18(4).
12. Nicolaidis C, Raymaker D, McDonald K, et al. Collaboration strategies in nontraditional community-based participatory research partnerships: Lessons from an academic-community partnership with autistic self-advocates. *Prog Community Health Partnersh.* 2011;5(2):143–50. [PubMed: 21623016]

13. McDonald KE, Kidney CA, Patka M. 'You need to let your voice be heard': Research participants' views on research. *J Intellect Disabil Res.* 2013;57(3):216–25. [PubMed: 22292970]
14. Wheeler AC, Wylie A, Raspa M, et al. Decisional capacity for informed consent in males and females with fragile x syndrome. *J Autism Dev Disord.* 2019.
15. 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):260–5.
16. Fisher CB, Cea CD, Davidson PW, Fried AL. Capacity of persons with mental retardation to consent to participate in randomized clinical trials. *Am J Psychiatry.* 2006;163(10):1813–20. [PubMed: 17012694]
17. Dye L, Hare DJ, Hendy S. Capacity of people with intellectual disabilities to consent to take part in a research study. *J Appl Res Intellect Disabil.* 2007;20(2):168–74.
18. Pellicano E, Dinsmore A, Charman T. What should autism research focus upon? Community views and priorities from the United Kingdom. *Autism.* 2014;18(7):756–70. [PubMed: 24789871]
19. Pellicano E, Dinsmore A, Charman T. Views on researcher-community engagement in autism research in the United Kingdom: A mixed-methods study. *PLoS One.* 2014;9(10):e109946. [PubMed: 25303222]
20. McDonald KE, Conroy NE, Olick RS, Project ETHICS Expert Panel. A quantitative study of attitudes toward the research participation of adults with intellectual disability: Do stakeholders agree? *Disabil Health J.* 2018;11(3):345–50. [PubMed: 29292211]
21. Hollin IL, Craig BM, Coast J, et al. Reporting formative qualitative research to support the development of quantitative preference study protocols and corresponding survey instruments: guidelines for authors and reviewers. *Patient.* 2019.
22. Racine E, Bell E, Yan A, et al. Ethics challenges of transition from paediatric to adult health care services for young adults with neurodevelopmental disabilities. *Paediatr Child Health.* 2014;19(2):65–8. [PubMed: 24596475]
23. Maenner MJ, Smith LE, Hong J, et al. Evaluation of an activities of daily living scale for adolescents and adults with developmental disabilities. *Disabil Health J.* 2013;6(1):8–17. [PubMed: 23260606]
24. NVivo 11. 11th ed. Melbourne, Australia: QSR International; 2011.
25. Hsieh HF, Shannon SE. Three approaches to qualitative content analysis. *Qual Health Res.* 2005;15(9):1277–88. [PubMed: 16204405]
26. Cicchetti DV. Multiple comparison methods: Establishing guidelines for their valid application in neuropsychological research. *J Clin Exp Neuropsychol.* 1994;16(1):155–61. [PubMed: 8150886]
27. Landis JR, Koch GG. The measurement of observer agreement for categorical data. *Biometrics.* 1977;33(1):159–74. [PubMed: 843571]
28. Averill JB. Matrix analysis as a complementary analytic strategy in qualitative inquiry. *Qual Health Res.* 2002;12(6):855–66. [PubMed: 12109729]
29. Viswanathan M, Ammerman A, Eng E, et al. Community-Based Participatory Research: Assessing the Evidence. Rockville, MD: Agency for Healthcare Research and Quality (US); 2004 Contract No.: 99.
30. Fletcher-Watson S, Adams J, Brook K, et al. Making the future together: Shaping autism research through meaningful participation. *Autism.* 2019;23(4):943–53. [PubMed: 30095277]
31. The Value of Engagement: Patient-Centered Outcomes Research Institute; 2018 [updated October 30, 2018]. Available from: <https://www.pcori.org/about-us/our-programs/engagement/public-and-patient-engagement/value-engagement>.
32. Purdy S, Finkelstein JA, Fletcher R, Christiansen C, Inui TS. Patient participation in research in the managed care environment: Key perceptions of members in an HMO. *J Gen Intern Med.* 2000;15(7):492–5. [PubMed: 10940136]
33. Ruiz-Canela M, Valle-Mansilla JI, Sulmasy DP. What research participants want to know about genetic research results: The impact of "genetic exceptionalism". *J Empir Res Hum Res Ethics.* 2011;6(3):39–46.
34. McElfish PA, Long CR, James LP, et al. Characterizing health researcher barriers to sharing results with study participants. *J Clin Transl Sci.* 2019;3(6):295–301. [PubMed: 31827902]

35. Smith JG. Self-advocacy online: Making online information accessible for all American Association on Intellectual and Developmental Disabilities; 2019; St. Paul, MN.
36. Stowe MJ, Turnbull HR, Schrandt S, Rack J. Looking to the future: Intellectual and developmental disabilities in the genetics era. *J Dev Disabl*. 2007;13(2).
37. Beskow LM, Burke W, Merz JF, et al. Informed consent for population-based research involving genetics. *JAMA*. 2001;286(18):2315–21. [PubMed: 11710898]
38. Kerasidou A Trust me, I'm a researcher! The role of trust in biomedical research. *Med Health Care Philos*. 2017;20(1):43–50. [PubMed: 27638832]
39. Perera G, Holbrook A, Thabane L, Foster G, Willison DJ. Views on health information sharing and privacy from primary care practices using electronic medical records. *Int J Med Inform*. 2011;80(2):94–101. [PubMed: 21167771]
40. Aitken M, de St Jorre J, Pagliari C, Jepson R, Cunningham-Burley S. Public responses to the sharing and linkage of health data for research purposes: A systematic review and thematic synthesis of qualitative studies. *BMC Med Ethics*. 2016;17(1):73. [PubMed: 27832780]
41. Kraft SA, Cho MK, Gillespie K, et al. Beyond consent: Building trusting relationships with diverse populations in precision medicine research. *Am J Bioethics*. 2018;18(4):3–20.

Table 1.

Demographic characteristics of focus group participants *

	No NDDs (n=16)		ASD (n=15)		FXS (n=13)		Total (n=44)		
	n	%	n	%	n	%	n	%	p
Gender									<.0001 [†]
Male	8	50%	13	87%	0	0%	21	48%	
Female	8	50%	2	13%	13	100%	23	52%	
Race									0.0023 [†]
White	10	63%	14	93%	12	92%	36	82%	
Black or African American	6	38%	0	0%	0	0%	6	14%	
Other race	0	0%	1	7%	1	8%	2	5%	
Ethnicity									0.1866
Hispanic or Latinx	0	0%	1	7%	2	15%	3	7%	
Not Hispanic or Latinx	16	100%	14	93%	11	85%	41	93%	
Marital status									<.0001 [†]
Single or never married	7	44%	15	100%	13	100%	35	80%	
Married	9	56%	0	0%	0	0%	9	20%	
Education									0.0013 [†]
< High school	0	0%	0	0%	2	15%	2	5%	
High school graduate or equivalent	0	0%	3	20%	4	31%	7	16%	
Some college	3	19%	10	67%	3	23%	16	36%	
2-yr degree or trade certificate	1	6%	0	0%	0	0%	1	2%	
4-yr degree	9	56%	2	13%	4	31%	15	34%	
Graduate or professional degree	3	19%	0	0%	0	0%	3	7%	
Employment									0.0001 [†]
Full-time	13	81%	1	7%	3	23%	17	39%	
Part-time	1	6%	5	33%	7	54%	13	30%	
Not employed	2	13%	8	53%	3	23%	13	30%	
No response	0	0%	1	7%	0	0%	1	2%	
	Mean	SD	Mean	SD	Mean	SD	Mean	SD	P
Age (years)	30.3	4.9	23.7	3.6	26.2	6.7	26.8	5.7	0.0030 [†]
Waisman Daily Living score	34.0	0.0	30.2	3.2	31.0	4.3	31.8	3.4	0.0025 [†]

* Gender, race, ethnicity, marital status, education, and employment were tested for differences using Fisher's Exact Test. Age and Waisman Daily Living score were tested for differences using Analysis of Variance. Acronyms used in this table: NDD: neurodevelopmental disorder, ASD: autism spectrum disorder, FXS: fragile X syndrome, SD: standard deviation. Percentages in table columns may not sum to 100% due to rounding error (at most 1%).

[†]
p < .05.

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Table 2.

Focus group discussion topics and questions

Topic	Question(s)
Familiarity with Medical Records	To get us started, what comes to mind when you think of the term “medical records?”
Attitudes	In general, are you in favor or against sharing your medical record for research?
Type of Organization	Who <u>would</u> you be OK with being able to use your medical record for research? Who <u>wouldn't</u> you be OK with using your medical record for research purposes? Why or why not? How do you feel about people or groups <u>who you know personally</u> being able to use it, such as your primary care doctor or other health care providers? How do you feel about people or groups <u>who you don't know personally</u> being able to use it (such as researchers in general, insurance companies, pharmaceutical companies, government agencies)?
Type of Information	What kind of information from your medical record would you view as <u>OK to share</u> ? What kind of information would you <u>not be OK</u> with sharing? Why or why not?
Purpose of Study	What kinds of research would you be OK with researchers using your medical record for? What purposes could they use it for? Why or why not?
Duration of Study	For <u>how long</u> would you want to let researchers use your medical record? Would you want it to be for a set amount of time such as 1 year? Or, would you let them use it for as long as they wanted to? What are some of the reasons you'd think about in making that decision?
Frequency of Contact	How <u>often</u> would you want researchers to ask you to let them use your medical record? Would you want to be asked each time they wanted to use your medical record? Or, would you let them use it as many times as they wanted without asking again?
Return of Results	How important is it for you to learn about the results of the research that is being conducted with your medical record? What makes you say that?
Perceived Benefits	Overall, what do you think are the <u>benefits</u> , or things that could help, from sharing your medical record for research? What makes you say that?
Perceived Risks	Overall, what would be the <u>risks</u> , or chances of something bad happening, from sharing your medical record for research? What makes you concerned or worried about that?