Published in final edited form as:

J Empir Res Hum Res Ethics. 2013 October ; 8(4): 42–52. doi:10.1525/jer.2013.8.4.42.

Participants' recall and understanding of genomic research and large-scale data sharing

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Abstract

As genomic researchers are urged to openly share generated sequence data with other researchers, it is important to examine the utility of informed consent documents and processes, particularly as these relate to participants' engagement with and recall of the information presented to them, their objective or subjective understanding of the key elements of genomic research (e.g., data sharing), as well as how these factors influence or mediate the decisions they make. We conducted a randomized trial of three experimental informed consent documents (ICDs) with participants ($n = 229$) being recruited to genomic research studies; each document afforded varying control over breadth of release of genetic information. Recall and understanding, their impact on data sharing decisions, and comfort in decision making were assessed in a follow-up structured interview. Over 25% did not remember signing an ICD to participate in a genomic study, and the majority (54%) could not correctly identify with whom they had agreed to share their genomic data. However, participants felt that they understood enough to make an informed decision, and lack of recall did not impact final data sharing decisions or satisfaction with participation. These findings raise questions about the types of information participants need in order to provide valid informed consent, and whether subjective understanding and comfort with decision making are sufficient to satisfy the ethical principle of respect for persons.

Keywords

data sharing; genome research; informed consent; understanding; participant perspectives

To maximize data utility and foster collaboration, genomic researchers are urged to openly share generated sequence data with other researchers through public (open access) (The Wellcome Trust, 1996, 2003) and/or restricted (controlled access) databases (National Institutes of Health, 2007). Given this current culture and as more individuals are enrolled into genomic research, it is of vital importance to examine how key elements of genomic research (e.g., mandates for broad data sharing) are understood by participants, how participants' understanding influences their decisions, and how participants want this information conveyed to them during the informed consent process.

Valid informed consent generally requires: (1) *disclosure* of relevant information about the study, (2) in a way that can be *understood* by potential participants, (3) so that they can make a *voluntary* and informed decision to participate (Faden, Beauchamp, & King, 1986). Federal regulations for the protection of human subjects of research focus primarily on what information must be disclosed to participants in the informed consent document (ICD) (Code of Federal Regulations). However, studies suggest that participants often have difficulty recalling information and understanding basic scientific concepts included in research ICDs (Bergler et al., 1980; Joffe et al., 2001a). This issue is compounded in genomic research, where many concepts, like DNA sequencing and genomic data sharing, are complex, (McGuire & Beskow, 2010) and public understanding of the field is fairly limited (Lea et al., 2011). Largely in response to requests to provide more information, ICDs have grown in length and detail over time. With some ICDs now exceeding 20 pages in length, there are questions arising about how much information participants can retain and how much understanding is needed to provide valid, informed consent (Matsui et al., 2012).

Joffe et al. (2001b) suggest that there are two goals of the informed consent process: for subjects to *be* well informed (objective understanding) and for subjects to *feel* well informed (subjective understanding). Studies have shown that patients and participants who express satisfaction with the informed consent process, their medical decisions, or their decision to participate in research, and who *feel* well informed, may not *be* well informed according to objective measures of understanding (Joffe et al., 2001b; Sepucha et al., 2010). Authors of these studies conclude that patients and participants are not able to accurately assess how well informed they are and that better education is needed. In this paper, we assess research participants' objective and subjective understandings of genomic research, including their participation in genomic research, and examine the impact of each on their recall and data sharing decisions.

Method

We conducted a randomized trial of three experimental ICDs with participants being recruited to one of six ongoing genomic studies (pediatric autism, pediatric brain cancer, pediatric brain controls, adult/pediatric epilepsy, adult/pediatric liver cancer, and adult pancreas cancer) at Baylor College of Medicine (BCM) in Houston, Texas (McGuire et al., 2011). The experimental ICDs provided varying amounts of control over breadth of release of participants' genetic information.

The genomic study principal investigator (PI), a research nurse, or a medical resident obtained informed consent for participation in the genomic study using one of the randomly assigned experimental ICDs. The informed consent process took place in a face-to-face setting and, although the process varied by the underlying genomic study, each experimental consent type was implemented similarly. Participants were debriefed about the randomized consent study by a BCM Center for Medical Ethics and Health Policy research assistant either immediately following the informed consent process, during a follow-up clinic visit, or, for those who did not return to the clinic for a follow-up visit, by phone or by mail, and were given an opportunity to change their initial data sharing decisions. Details of the

randomization and debriefing processes have been described elsewhere (McGuire et al., 2011).

Participants who were debriefed in person were invited to a follow-up structured interview, which provided an opportunity to assess their understanding of and satisfaction with participation. Eligible participants were 18 years of age or older, English proficient, and included adult patients, parents/guardians of pediatric patients, and family members acting as matched case controls for the genomic study.

Interview participants were guided through a questionnaire using a laptop computer and an electronic interview data warehouse program, QDS (NOVA Research Company, Bethesda, MD). Verbal consent to participate in audio-recorded interviews was obtained. Interviews lasted approximately 45 minutes and were professionally transcribed verbatim. Those who participated in the interview were compensated with a \$25 gift card. All materials and methods were approved by the BCM Institutional Review Board.

Consent Documents

Three experimental ICDs were developed and modified for each genomic study (McGuire et al., 2008; McGuire et al., 2011). Each ICD varied in the data sharing option(s) offered and contained one or more of the following choices: (1) public data release, or the release of genetic and clinical information into both publicly accessible (open access through the Internet) and restricted scientific databases (accessible only to approved researchers); (2) restricted release, or the release of genetic and clinical information into restricted databases only; and (3) no release, or accessible only to the genomic study PI and staff. The *traditional* consent was an all-or-nothing form, whereby participants agreed to participate in the genomic study and also agreed to the public release of their genetic and clinical information. The *binary* consent allowed participants to enroll in the genomic study while opting out of data sharing altogether; participants were provided a choice between public data release and no release. *Tiered* consent allowed participants to choose from all three options; they could choose public data release, restricted release only, or no release (McGuire et al., 2011).

Structured Interview Questionnaire

The structured interview questionnaire assessed six general domains of inquiry: (1) recall and understanding, (2) comfort in decision making (adapted from the O'Connor scale [O'Connor, 1995]), (3) trust in medical researchers, (4) risk-benefit assessment, (5) preferences for and attitudes toward consent types and data sharing options, and (6) demographic characteristics.

Data Analysis

Participant characteristics were described with frequencies and means or medians. To explore relationships between variables and test differences between groups, chi-square tests, McNemar-Bower's tests, and nonparametric tests (Kruskal-Wallis test) were used as appropriate. Multivariate analyses were explored with binomial logistic regressions to examine the effects of possible confounding variables. Covariates included were randomized consent type, genomic study type or group, consentee relationship (either adult/self

consentee or parental consentee), the time interval between enrollment and debriefing, trust in medical researchers, and participant demographic characteristics. For all tests, a significance level of $p < 0.05$ was used. All analyses were conducted using SPSS 19 (IBM Corporation, Armonk, NY).

Of the six genomic studies, the two that enrolled primarily adult surgical patients were distinct in three ways: (1) participants were significantly older ($p < 0.001$), (2) acutely ill, and (3) because informed consent was usually obtained preoperatively and participants were often not well enough to debrief immediately after surgery, the time lapse from enrollment to debriefing was typically longer for these individuals (Table 1). Therefore, for analysis purposes, we split the genomic studies into two groups: Group A includes genomic studies that primarily enrolled parents of pediatric patients, adult patients with chronic conditions, and matched case controls (autism, brain cancer, brain controls, and epilepsy studies) and (2) Group B includes the remaining genomic studies which primarily enrolled adult surgical patients (liver and pancreas cancer studies).

Participants' trust in medical researchers was measured using a validated scale (Hall et al., 2006) which, as previously reported, was reduced to one question due to high ceiling effects during the study (Oliver et al., 2012). Five-point Likert-type scale response categories, anchored by strongly disagree (1) to strongly agree (5), were collapsed to a dichotomous variable of no to low trust (1–3) and some to high trust (4–5) for analysis. To assess participants' understanding of their data sharing decisions, a variable—coded as correct (1) or incorrect (0)—was created to compare responses identifying with whom participants thought they had agreed to share their genetic data to their actual initial data release selection as indicated on their signed ICD.

Content analysis (Hsieh & Shannon, 2005) of open-ended responses was conducted and data were managed using NVivo 8 (QSR International Inc., Cambridge, MA).

Results

Three hundred and thirty-six participants were enrolled into one of six genomic studies using one of the randomized ICDs: traditional, binary, or tiered. Of 285 eligible participants invited to a follow-up interview, 229 agreed (RR = 80%) (Oliver et al., 2012). The average time lapse from enrollment in the genomic study to debriefing was 24.6 days. The mean age of interview participants was 48 years; most were female, married, identified as non-Hispanic white, reported completing at least some college, and reported an annual household income over \$40,000 (Table 1). There were no significant differences in demographic characteristics between those who completed the interview and those who declined (data not shown).

As noted, there were significant differences between the two genomic study groups (Group A and Group B). Compared to genomic studies that primarily enrolled adult surgical patients (Group B), those in Group A were younger, predominantly female, more often identified as Hispanic, and, on average, experienced a shorter time lapse from enrollment to debriefing (Table 1).

Recall and Objective Understanding of Participation and Data Sharing Decisions

Over a quarter of participants did not remember signing an ICD to participate in a genomic study, and just over 40% reported they did not know they were enrolled in a genetic research study. A larger proportion (53%) responded *no/don't know* when asked whether they had already given DNA to their doctors, and 62% responded *no/don't know* when asked whether their DNA had been stored as part of a genetic research study. Those who responded *no/don't know* to these questions less often remembered signing an ICD (χ^2 , "Do you know if you are participating in a research study?" $p < 0.001$, 57% vs. 85% remembered respectively; "Do you know if you have already given DNA to your doctor?" $p = 0.042$, 67% vs. 81% remembered respectively; "Do you know if your DNA was stored as part of a study?" $p < 0.001$, 65% vs. 88% remembered respectively). Participants who reported knowing they were participating in a genetic research study (Kruskal-Wallis, $p = 0.020$) or remembered signing an ICD (Kruskal-Wallis, $p = 0.001$) experienced significantly shorter time lapse intervals between enrollment and debriefing. The time lapse interval and genomic study group were significant predictors of whether one remembered signing a consent form; in a binomial regression which controlled for covariates, participants who experienced shorter time lapses ($p < 0.001$) and were in a Group A genomic study ($p = 0.046$) were significantly more likely to remember signing the ICD (Table 2). Recall of participation in a research study and of signing an ICD was not associated with final data sharing decisions.

After asking objective understanding questions related to their research participation, participants were debriefed, shown their signed ICD, and reminded that they had agreed to participate in a genomic research study and to which data sharing option they had consented. Participants were then asked to choose from the following options, all of which were included in the tiered ICD, as to who they thought could access and use their genetic information: (1) anyone in the public with access to the Internet, (2) only approved researchers, or (3) only researchers at this institution and their staff. Fifty-five percent of participants incorrectly identified who could access and use their genetic information, and participants generally tended to overestimate the level of restriction of their initial data release selection (Table 3). For example, before debriefing, 53% of participants who initially agreed to public release, either by choice or by enrolling in the study using traditional consent, thought only approved researchers could access and use their genetic information. During the study, the questionnaire was revised to ask participants ($n = 123$) again, after explicitly reviewing all of the data sharing options provided in each experimental ICD, who they thought could access and use their genetic information. Over a third of participants (34%) asked this question a second time incorrectly identified who could access and use their genetic information, choosing a more restrictive option than they had initially selected. All but four of these participants had also incorrectly identified this information when asked the first time.

Hispanic and older participants were significantly less likely to correctly identify who could access and use their genetic information. In particular, participants 60 years and older most often made an incorrect identification of their data sharing selection, both prior to and following the full debriefing (Table 2). Although we observed some differences in recall and objective understanding between genomic studies in Group A and Group B, these

differences did not remain significant after controlling for confounding variables, such as the difference in time lapse between the groups and consentee relationship status. Participants randomized to the tiered ICD were able to correctly identify who could access and use their genetic information more frequently than those randomized to the other experimental ICDs (χ^2 , $p = 0.003$) (Table 2). However, this association did not remain significant (binomial regression, $p = 0.083$) after controlling for other participant study and demographic characteristics.

Subjective Understanding of Genetic Research and Comfort in Decision Making

While a large number of participants did not correctly identify with whom they agreed to share their genetic information, their self-reported (i.e., subjective) understanding of genetic studies and their purpose was strong. The majority (72%) of participants reported they had heard of genetic studies, 89% reported they understood the purpose of genetic studies, and 59% felt they understood the role of DNA in their bodies. When asked what genetic information was most similar to, 72% selected fingerprint; other options provided were medical diagnosis (19%), Social Security number (6%), credit card number (1%), lottery number (1%), and name (1%). When asked to explain their selection, those who chose “fingerprint” felt it was, similar to DNA, *unique* or *identifying*.

Although all groups reported strong subjective understanding, after controlling for potential confounding factors in a binomial regression, participants in a Group B genomic study ($p = 0.033$), Hispanics ($p = 0.006$) and non-Hispanic minorities ($p = 0.007$), men ($p = 0.027$), and participants who had not completed high school ($p = 0.033$) or completed high school or the GED ($p = 0.027$) were less likely to report having heard of genetic studies (Table 2). Hispanics ($p = 0.005$) and older participants ($p = 0.020$) were less likely to report understanding the purpose of genetic studies (Table 2). No other participant study or demographic characteristics significantly influenced responses to these items and there was no significant association between subjective understanding and one’s final data release decisions.

Participants not only reported feeling that they subjectively understood the purpose of genetic studies, they also expressed comfort or ease with the decisions they made concerning sharing their genetic information. After reviewing with them their signed ICD and the data sharing option they chose, most participants agreed or strongly agreed with the statements: “I feel sure about what to choose with regard to sharing my genetic information” (86%), “this decision about sharing my genetic information is easy for me to make” (87%), and “I feel I have made an informed decision about sharing my genetic information” (91%). Compared to participants exhibiting no to low levels of trust in medical researchers, those exhibiting some to high levels of trust more often reported the decision was easy to make (χ^2 , $p = 0.001$, Table 4). They also more often reported they made an informed decision (χ^2 , $p = 0.001$, Table 4), and after controlling for covariates in a binomial regression, trust remained a significant predictor of whether one felt the decision was easy to make ($p = 0.001$). Participants’ responses to questions rating their comfort in decision making were otherwise not significantly associated with participant study or demographic characteristics.

and there were no significant associations between comfort in decision making and final data release decisions.

Discussion

Many participants we interviewed were not fully aware that they were participating in a genomic research study or what that participation entailed. These findings are consistent with previous studies that report poor recall and objective understanding of informed consent in biobanking and other areas of research (Bergler et al., 1980; Dixon-Woods et al., 2007; Hoeyer et al., 2005; Joffe et al., 2001a; Stolt et al., 2005). Participants were less likely to know this information, or remember signing the ICD, as the time lapse between enrollment and debriefing increased, suggesting this was an issue of recall. Also, the majority of participants in this study were not able to correctly identify who they agreed could access and use their genetic information during the informed consent process. Even though understanding improved for some participants as a result of the debriefing process, a substantial number of participants answered this question incorrectly even after a thorough review of all potential data sharing options. This may reflect the complexity of the concept, which is abstract and noted not to be well understood by the general public (Lea et al., 2011), or it may be due to poor communicability during the informed consent process or as it was explained in the ICD.

Another possible reason participants may display poor recall and understanding could actually reflect a conscious choice not to process all information presented, especially in modern society where “information overload” is becoming commonplace (Helgesson, Ludvigsson, & Gustafsson Stolt, 2005). Participants may be applying a voluntary filter to information in order to disregard what they deem irrelevant. This could be a productive mechanism for processing the growing volume of information that is communicated during the informed consent process since, arguably, individuals do not need to understand all that is disclosed during this process in order to make an autonomous decision to participate (Wendler & Grady, 2008). One study asked various groups of stakeholders to identify what information from a model biorepository ICD they felt was important to include and found that prospective research participants thought far less information is important than scientists or IRB members (Beskow et al., 2010). This is not to suggest that research subjects do not need to understand basic concepts, such as the risks and benefits of participating in a research study, that genetic analyses will be done, and their information will be shared, but providing too much information could interfere with the ability of subjects to decipher what information is important and what they can disregard.

Importantly, despite participants’ poor recall of study participation and baseline understanding of who could access and use their genetic information, neither significantly impacted their final data sharing decisions or their willingness to participate, and most participants felt that they sufficiently understood genetics and the purpose of genetic research. Participants also expressed comfort in their data sharing decisions, despite not having thoroughly considered the implications or risks involved. They articulated the importance of the ethical principle of respect for persons, and in response to open-ended questions expressed that the best way to respect research subjects was to be transparent,

provide them with information, and offer them choices through tiered consent (data not shown). This raises an important question about how much objective understanding is required to satisfy core ethical principles in research.

One influential factor in this study was the high degree of trust in researchers expressed by most of our participants. This may have been influenced by the fact that all of our study participants were patients, parents of pediatric patients, or relatives of patients who were recruited to participate in the genomic study by either their physician or a physician at the institution where they or their family member was being treated. Although trust in researchers was not associated with final data sharing decisions, it did have a significant impact on whether participants felt like they had made a well-informed, easy decision. Other participant populations (e.g., healthy controls or minority populations with historically lower trust in medical research) may feel differently.

One potential limitation of this study is that, although we manipulated the ICDs to similarly describe the procedures, risks, and benefits of genetic research and data sharing, and they had similar IRB-approved reading levels, we had no control over the actual informed consent process. We observed several consent processes in the studies; these varied in length from five minutes to one hour and were done either at a pre-operative or clinic visit, immediately prior to surgery, or in an in-patient hospital setting. While everyone administering the informed consent process was formally trained in human subjects research, the information and level of study details effectively communicated differed by who conveyed the information. In addition to the observed differences in the time lapse between enrollment and debriefing by genomic study, these differences in consent processes may help explain why we saw differences in recall between study groups. However, these differences in recall did not seem to impact participants' understanding of their data sharing choice or their final data sharing decisions.

Best Practices

This study raises concerns about research participants' ability to recall and understand information provided to them during the informed consent process for genomic research studies. ICDs for this type of research have become increasingly long and complex, making it difficult for study staff to accurately convey this information in an understandable way during the informed consent process. These concerns may be mitigated by abbreviated ICDs, as was recently proposed by the Department of Health and Human Services (2001) in their Advanced Notice of Proposed Rulemaking. They may also be addressed through the development of summary materials highlighting the most important information for subjects to consider.

Despite their lack of objective understanding, participants expressed a desire to be involved in the decision about data sharing. They felt that the best way to respect research participants was to be transparent, provide them information, and offer them choices. As genomic researchers continue to enroll participants with limited and imperfect understanding of the science, enhanced consent processes, including dynamic consent models that "enable individuals to change their mind and preferences over time, to have their choices revoked

where appropriate, to track and audit any changes made, and to choose when and how they are contacted” (Kaye, 2012), should be explored as a way to promote transparency and participant engagement in the research process.

Research Agenda

Future research should assess how much and what information research participants need to understand in order to provide valid informed consent for genomic research participation and large-scale data sharing. The effectiveness of abbreviated consent forms deserves careful consideration, and researchers should continue to explore the effect of trust on willingness to participate, as well as its impact on decision making. It will thus be important to conduct future research among diverse populations, especially individuals from vulnerable or underrepresented groups. Those conducting normative analyses of research ethics should assess the relative importance of subjective versus objective understanding for informed decision making and whether changing standards for informed consent satisfy ethical principles in research. Future research should also seek to explore how best to engage participants in research so that they feel respected and able to make informed decisions. Dynamic consent models should be evaluated for whether they satisfy participants’ desire for involvement and enhance understanding over time.

Educational Implications

Our findings demonstrate that the field of genomics is not well understood by those participating in genomics research. ICDs used for enrollment are becoming increasingly burdensome without providing the benefit of improved understanding. Increased emphasis should be placed on educating participants using innovative materials (e.g., summary documents, brochures, educational videos) and by focusing more on improving the informed consent process.

IRB members and investigators should also be educated about the ongoing issues and advances in the field (e.g., new privacy risks of genomics), which have implications for human subjects protection. Education could be delivered through the institution, online tools such as webinars, or other educational forums. It is also critically important to maintain open communication with research participants as new findings could influence their willingness to continue to participate. Dynamic models for engaging participants over time should be explored and study-related materials (findings, implications) should be made freely available to the public in lay terms. While resources are often scarce for these types of educational activities, it may help to increase understanding and willingness to participate.

Acknowledgments

This work was supported by grant NIH R01HG004333 (A. L. McGuire, S. G. Hilsenbeck, T. Wang, P. A. Kelly, R. A. Gibbs); The Greenwall Foundation Faculty Scholars Program in Bioethics (A. L. McGuire); and DLDCC P30CA125123 (S. G. Hilsenbeck, T. Wang). The content is solely the responsibility of the authors and does not necessarily represent the official views of the National Human Genome Research Institute or the National Institutes of Health. We are grateful for the commitment and unwavering support of William Fisher, Alica Goldman, John Goss, P. Adam Kelly, Ching Lau, Jeffrey Noebels, Mehmet Okcu, and Diane Treadwell-Deering, and sincerely appreciate all of the thoughtful and generous patients and research participants who participated in this study. We thank Laura Beskow, Wylie Burke, Mildred Cho, Rebecca Fisher, Gail Gellar, Laura Lyman Rodriguez, Louise Strong, and Richard Gibbs for their expert advice throughout this project, and Claudette Campbell, Sally E.

Hodges, Liz Hinojosa, Melissa Lambeth, Morgan Lasala, Melissa Pagaoa, Suzanne Wheeler, Tiffany Zgabay-Hunsucker, and Jennifer L. Graves for their valuable assistance and research coordination.

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References

- Bergler JH, Pennington AC, Metcalfe M, Freis ED. Informed consent: How much does the patient understand? *Clinical Pharmacology and Therapeutics*. 1980; 27(4):435–440. [PubMed: 6987027]
- Beskow LM, Friedman JY, Hardy NC, Lin L, Weinfurt KP. Simplifying informed consent for biorepositories: Stakeholder perspectives. *Genetics in Medicine*. 2010; 12(9):567–572. [PubMed: 20697289]
- Code of Federal Regulations. Protection of human subjects. Title 45, Part 46, Retrieved from <http://www.hhs.gov/ohrp/humansubjects/guidance/45cfr46.html>.
- Department of Health and Human Services. Human subjects research protections: Enhancing protections for research subjects and reducing burden, delay, and ambiguity for investigators. Advance notice of proposed rulemaking. *Federal Register*. 2011; 76(143):44512–44531. Retrieved from <http://www.gpo.gov/fdsys/pkg/FR-2011-07-26/html/2011-18792.htm>.
- Dixon-Woods M, Ashcroft RE, Jackson CJ, Tobin MD, Kivits J, Burton PR, et al. Beyond “misunderstanding” Written information and decisions about taking part in a genetic epidemiology study. *Social Science and Medicine*. 2007; 65(11):2212–2222. [PubMed: 17904716]
- Faden, RR.; Beauchamp, TL.; King, NMP. A history and theory of informed consent. New York: Oxford University Press; 1986.
- Hall MA, Camacho F, Lawlor JS, Depuy V, Sugarman J, Weinfurt K. Measuring trust in medical researchers. *Medical Care*. 2006; 44(11):1048–1053. [PubMed: 17063137]

- Helgesson G, Ludvigsson J, Gustafsson Stolt U. How to handle informed consent in longitudinal studies when participants have a limited understanding of the study. *Journal of Medical Ethics*. 2005; 31(11):670–673. [PubMed: 16269567]
- Hoeyer K, Olofsson BO, Mjorndal T, Lynoe N. The ethics of research using biobanks: Reason to question the importance attributed to informed consent. *Archives of Internal Medicine*. 2005; 165(1):97–100. [PubMed: 15642883]
- Hsieh HF, Shannon SE. Three approaches to qualitative content analysis. *Qualitative Health Research*. 2005; 15(9):1277–1288. [PubMed: 16204405]
- Joffe S, Cook EF, Cleary PD, Clark JW, Weeks JC. Quality of informed consent in cancer clinical trials: A cross-sectional survey. *Lancet*. 2001a; 358(9295):1772–1777. [PubMed: 11734235]
- Joffe S, Cook EF, Cleary PD, Clark JW, Weeks JC. Quality of informed consent: A new measure of understanding among research subjects. *Journal of National Cancer Institute*. 2001b; 93(2):139–147.
- Kaye J. The tension between data sharing and the protection of privacy in genomics research. *Annual Review of Genomics and Human Genetics*. 2012; 13:415–431.
- Lea DH, Kaphingst KA, Bowen D, Lipkus I, Hadley DW. Communicating genetic and genomic information: Health literacy and numeracy considerations. *Public Health Genomics*. 2011; 14(4–5):279–289. [PubMed: 20407217]
- Matsui K, Lie RK, Turin TC, Kita Y. A randomized controlled trial of short and standard-length consent forms for a genetic cohort study: Is longer better? *Journal of Epidemiology*. 2012; 22(4):308–316. [PubMed: 22447213]
- McGuire AL, Beskow LM. Informed consent in genomics and genetic research. *Annual Review of Genomics and Human Genetics*. 2010; 11:361–381.
- McGuire AL, Hamilton JA, Lunstroth R, McCullough LB, Goldman A. DNA data sharing: Research participants' perspectives. *Genetics in Medicine*. 2008; 10(1):46–53. [PubMed: 18197056]
- McGuire AL, Oliver JM, Slashinski MJ, Graves JL, Wang T, Kelly PA, et al. To share or not to share: A randomized trial of consent for data sharing in genome research. *Genetics in Medicine*. 2011; 13(10)
- National Institutes of Health. Policy for sharing of data obtained in NIH supported or conducted Genome-Wide Association Studies (GWAS). 2007. Retrieved from <http://grants.nih.gov/grants/guide/notice-files/NOT-OD-07-088.html>.
- O'Connor AM. Validation of a decisional conflict scale. *Medical Decision Making*. 1995; 15(1):25–30. [PubMed: 7898294]
- Oliver JM, Slashinski MJ, Wang T, Kelly PA, Hilsenbeck SG, McGuire AL. Balancing the risks and benefits of genomic data sharing: Genome research participants' perspectives. *Public Health Genomics*. 2012; 15(2):106–114. [PubMed: 22213783]
- Sepucha KR, Fagerlin A, Couper MP, Levin CA, Singer E, Zikmund-Fisher BJ. How does feeling informed relate to being informed? The DECISIONS survey. *Medical Decision Making*. 2010; 30(5 Suppl):77S–84S. [PubMed: 20881156]
- Stolt UG, Helgesson G, Liss PE, Svensson T, Ludvigsson J. Information and informed consent in a longitudinal screening involving children: A questionnaire survey. *European Journal of Human Genetics*. 2005; 13(3):376–383. [PubMed: 15657607]
- The Wellcome Trust. Summary of principles agreed at the International Strategy Meeting in Human Genome Sequencing, Bermuda. 1996. Retrieved from http://www.ornl.gov/sci/techresources/Human_Genome/research/bermuda.shtml#1.
- The Wellcome Trust. Sharing data from large-scale biological research projects: A system of tripartite responsibility, Fort Lauderdale. 2003. Retrieved from <http://www.genome.gov/Pages/Research/WellcomeReport0303.pdf>.
- Wendler D, Grady C. What should research participants understand to understand they are participants in research? *Bioethics*. 2008; 22(4):203–208. [PubMed: 18405318]

TABLE 1

Interview Participant Characteristics and Demographics.

	No.(%) ^a			P Value ^c
	All Genomic Studies	Group A ^b	Group B	
Genome study				
Autism (pediatric)	3(1)	3(3)		
Brain Cancer (pediatric)	55(24)	55(63)		
Brain Controls (pediatric)	12(5)	12(14)		
Epilepsy (adult/pediatric)	17(7)	17(20)		
Liver Cancer (adult/pediatric)	77(33)		77(55)	
Pancreas Cancer (adult)	65(28)		65(46)	
Consentee relationship				<0.001
Adult/self consentee	154(67)	23(26)	131(92)	
Parental consentee	75(33)	64(74)	11(8)	
Consent-to-debrief time lapsed				<0.001
Mean days (SD) ^e	24.6(38)	10.2(24)	33.5(42)	
Median (Range)	15(0–380)	3(0–168)	21.5(0–380)	
Age in years – Mean (SD)^d	47.8(15)	37.7(10)	54.2(15)	<0.001
Gender				<0.001
Male	95(42)	22(25)	73(51)	
Female	134(59)	65 (75)	69(49)	
Race/ethnicity				0.006
Hispanic or Latino	47(21)	27(31)	20(14)	
Minority, non-Hispanic ^f	48(21)	19(22)	29(21)	
White, non-Hispanic	132(58)	41(47)	91(65)	
Marital status				0.415
Married	142(64)	53(61)	89(65)	
Divorced, widowed, or separated	60(27)	23(26)	37(27)	
Never married	21(9)	11(13)	10(7)	
Education				0.862
<High school	16(7)	7(8)	9(7)	
High school/GED	55(25)	20(23)	35(26)	
Some college	79(35)	29(33)	50(37)	
Bachelor’s degree or higher	74(33)	31(36)	43(31)	
Annual household income				0.313
<\$20,000	31(16)	17(22)	14(12)	
\$20,000–\$40,000	49(25)	18(23)	31(26)	
\$40,000–\$60,000	26(13)	9(12)	17(14)	
>\$60,000	90(46)	34(44)	56(48)	
Trust in medical researchers				0.750

	No.(%) ^a			P Value ^c
	All Genomic Studies	Group A ^b	Group B	
Low to no trust	39(17)	14(16)	25(18)	
Some to high trust	189(83)	73(84)	116(82)	

^a Categories do not sum to 229 due to participant nonresponse; number (percent) of participants unless otherwise noted.

^b Group A includes autism, brain cancer and controls, and epilepsy genomic studies; Group B includes liver and pancreas cancer genomic studies.

^c P value based on chi-square test unless otherwise noted.

^d P value based on Kruskal-Wallis test.

^e Standard deviation (SD).

^f Minority, non-Hispanic includes: black/African American, Asian, Native American/Alaskan, Pacific Islander, and Other.

TABLE 2

Participants' Recall and Understanding by Study and Demographic Characteristics.^a

	Remember signing a consent form	Know if participating in a research study	Heard of genetic studies	Understand purpose of genetic studies	Correctly identify who could access data (before debriefing)	Correctly identify who could access data (after debriefing) ^b
Genomic study category						
Group A	74(89)	61(73)	63(75)	60(95)	45(52)	40(77)
Group B	90(64)	72(51)	97(69)	87(85)	56(40)	41(58)
Randomized consent type						
Traditional	49(70)	40(57)	44(62)	42(89)	24(33)	23(59)
Binary	58(78)	47(63)	54(73)	47(84)	29(39)	32(76)
Tiered	57(72)	46(58)	62(79)	58(94)	48(59)	26(62)
Time lapse interval (days)^c	18(0–150)	20(0–168)	25(0–233)	25(0–233)	23(0–233)	28(0–233)
Consentee relationship						
Adult/self consentee	102(69)	80(54)	105(71)	95(86)	62(40)	43(60)
Parental consentee	62(83)	53(71)	55(73)	52(95)	41(55)	38(75)
Age groups						
18–29	23(77)	20(67)	16(53)	14(88)	15(48)	14(82)
30–44	54(84)	42(66)	48(75)	45(90)	36(51)	27(71)
45–59	51(69)	45(61)	54(73)	53(95)	39(52)	27(66)
>60	36(69)	26(50)	41(78)	34(81)	11(22)	12(48)
Gender						
Male	65(71)	54(59)	60(65)	58(91)	38(41)	28(64)
Female	99(76)	79(60)	100(76)	89(88)	63(47)	53(67)
Race/ethnicity						
Hispanic or Latino	35(76)	29(63)	24(52)	21(75)	17(36)	12(50)
Minority, non-Hispanic	32(71)	25(56)	30(67)	27(90)	22(46)	16(70)
White, non-Hispanic	97(75)	79(61)	105(81)	98(93)	61(47)	52(69)
Education						
<High school	12(75)	6(38)	8(50)	6(75)	9(56)	3(50)
High school/GED	41(76)	30(56)	32(59)	30(86)	23(42)	16(62)

	Remember signing a consent form	Know if participating in a research study	Heard of genetic studies	Understand purpose of genetic studies	Correctly identify who could access data (before debriefing)	Correctly identify who could access data (after debriefing) ^b
Some college	48(63)	50(66)	52(68)	49(94)	38(48)	29(66)
Bachelor's degree or higher	61(85)	46(64)	66(92)	60(88)	30(41)	32(71)
Annual household income						
<\$20,000	21(70)	19(63)	17(57)	15(79)	13(42)	9(60)
\$20,000–\$40,000	33(69)	25(52)	34(71)	30(88)	20(41)	15(52)
\$40,000–\$60,000	18(72)	17(68)	16(64)	17(68)	13(50)	7(54)
>\$60,000	68(78)	54(62)	69(79)	63(89)	40(44)	38(81)

^aNot all categories sum to 229 due to participant nonresponse; number (percent) unless otherwise noted; percentages based on total number responded to questions and shown in table representing Yes and Agreed/Strongly Agreed responses or correctly identified information.

^bAfter debriefing, second assessment only asked of n=123 participants.

^cMedian days (range) of time lapse interval between enrollment and debriefing.

TABLE 3

Participants' Understanding of Their Data Release Selection.

	Participants' True Data Release Selection			
	Public release	Restricted release	No release	P value ^c
Before full debriefing				
Participants' reported data release selection ^a				<0.001
Anyone in the public (<i>Public release</i>)	69(36)	0(0)	0(0)	
Only approved researchers (<i>Restricted release</i>)	100(53)	16(89)	3(14)	
Only researchers at this institution (<i>No release</i>)	21(11)	2(11)	18(86)	
After full debriefing				
Participants' reported data release selection ^b				<0.001
Anyone in the public (<i>Public release</i>)	62(60)	0(0)	0(0)	
Only approved researchers (<i>Restricted release</i>)	33(32)	9(90)	0(0)	
Only researchers at this institution (<i>No release</i>)	8(8)	1(10)	10(100)	

^aN=229; number (percent) of participants response to "Who do you think can access and use your genetic information?" compared to their initial data release selection when asked before the full debriefing.

^bN=123; number (percent) of participants response to "Who do you think can access and use your genetic information?" compared to their initial data release selection when asked after the full debriefing.

^cMcNemar-Bower tests performed.

TABLE 4

Participants' Comfort in Decision Making by Study and Demographic Characteristic.

	Felt sure about what to choose with regard to sharing	Decision to share was easy to make	Felt decision to share was well informed
Genomic study category			
Group A	75(86)	71(83)	82(94)
Group B	120(86)	128(90)	127(89)
Randomized consent type			
Traditional	58(82)	63(88)	62(86)
Binary	63(84)	64(87)	70(93)
Tiered	74(93)	72(88)	77(94)
Time lapse interval (days)^b	25(0–380)	25(0–380)	24(0–380)
Consentee relationship			
Adult/self consentee	130(86)	140(91)	142(92)
Parental consentee	65(87)	59(80)	67(89)
Trust in medical researchers			
Low to no trust	30(79)	27(69)	30(77)
Some to high trust	164(88)	171(91)	178(94)
Age groups			
18–29	24(77)	22(73)	27(87)
30–44	61(90)	59(87)	64(94)
45–59	65(87)	68(91)	69(92)
>60	44(88)	47(90)	47(90)
Gender			
Male	82(88)	85(90)	88(93)
Female	113(85)	114(86)	121(90)
Race/ethnicity			
Hispanic or Latino	42(89)	38(83)	44(94)
Minority, non-Hispanic	39(81)	42(88)	47 (98)
White, non-Hispanic	113(87)	117(89)	117(89)
Education			
<High school	12(80)	12(75)	13(81)
High school/GED	47(86)	46(85)	49(89)
Some college	68(87)	71(90)	72(91)
Bachelor's degree or higher	64(87)	65(88)	71(96)
Annual household income			
<\$20,000	25(83)	25(81)	27(87)
\$20,000–\$40,000	41(85)	44(92)	46(94)
\$40,000–\$60,000	25(96)	23(89)	24(92)
>\$60,000	75(83)	78(87)	82(91)

^aNot all categories sum to 229 due to participant nonresponse; number (percent); percentages based on total number responded to questions and shown in table representing Agree/Strongly Agree responses.

^b Median days (range) of time lapse interval between enrollment and debriefing.