Published in final edited form as:

AJOB Empir Bioeth. 2018 ; 9(3): 128–142. doi:10.1080/23294515.2018.1505783.

Parents' Attitudes toward Consent and Data Sharing in Biobanks: A Multi-Site Experimental Survey

Armand H. Matheny Antommara, MD, PhD,

Ethics Center, Cincinnati Children's Hospital Medical Center and Department of Pediatrics, University of Cincinnati College of Medicine, 3333 Burnet Ave, ML 15006, Cincinnati, OH 45229, USA, armand.antommara@cchmc.org, www.linkedin.com/in/armand-antommara-3aa14b47

Kyle B. Brothers, MD, PhD,

Department of Pediatrics, University of Louisville, 231 E Chestnut St, N-97, Louisville, KY 40202, USA, kyle.brothers@louisville.edu, @kylebrothers

John A. Myers, PhD,

Department of Pediatrics, University of Louisville, 571 S Floyd St, Suite 432, Louisville, KY 40202, USA, john.myers@louisville.edu

Yana B. Feygin, MS,

Department of Pediatrics, University of Louisville, 401 E Chestnut St, Louisville, KY 40202, USA, yana.feygin@louisville.edu

Sharon A. Aufox, MS,

Center for Genetic Medicine, Northwestern University, 645 N Michigan Ave, Suite 630, Chicago, IL 60611, USA, s-aufox@northwestern.edu

Murray H. Brilliant, PhD,

Center for Human Genetics, Marshfield Clinic Research Institute, 1000 N Oak Ave, Marshfield, WI 54449, USA, brilliant.murray@mcrf.mfldclin.edu

Pat Conway, PhD, LCSW,

Essentia Institute of Rural Health, 502 E Second St, Maildrop 6AV-2, Duluth, MN 55805, USA, pat.conway@essentiahealth.org

Stephanie M. Fullerton, DPhil,

Corresponding Author: Armand H. Matheny Antommara, Ethics Center, Cincinnati Children's Hospital Medical Center, 3333 Burnet Ave, ML 15006, Cincinnati, OH 45208, Telephone (513) 636-4939, FAX (513) 636-4971, armand.antommara@cchmc.org.

¹Department of Humanities, Penn State College of Medicine, 500 University Dr, C1743, Hershey, PA 17033, USA

²Department of Radiology, Massachusetts General Hospital, 101 Merrimac St, Suite 1010, Boston, MA 02114, USA

³Department of Behavioural Science and Health, University College London, London WC1E 6BT, UK; and Great Ormond Street Hospital, London WC1N 3JH, UK

AUTHOR CONTRIBUTIONS: All authors conceived the study, designed the methods, coordinated and supervised the data collection, and/or analyzed and/or interpreted the data; Armand Antommara drafted the manuscript and all of the other authors revised the manuscript critically for important intellectual content; all authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

CONFLICTS OF INTEREST: None of the authors has any financial interest or benefit that has arisen from the direct applications of this research to disclose.

ETHICAL APPROVAL: This study was approved by the institutional review boards at Cincinnati Children's Hospital Medical Center, Boston Children's Hospital, Children's Hospital of Philadelphia, Essentia Health, Marshfield Clinic Health System, Geisinger Clinic, Kaiser Permanente Washington Research Institute, Mayo Clinic, Icahn School of Medicine at Mount Sinai, Northwestern University, and Vanderbilt University Medical Center.

Department of Bioethics and Humanities, University of Washington, Box 357120, Seattle, WA 98195, USA, smfullrtn@uw.edu, @smfullerton, www.linkedin.com/in/stephanie-malia-fullerton-b83a55a

Nanibaa' A. Garrison, PhD,

Treuman Katz Center for Pediatric Bioethics, Seattle Children's Hospital and Research Institute and Department of Pediatrics (Bioethics), University of Washington, 1900 Ninth Ave, M/S JMB-6, Seattle, WA 98101, USA, nanibaa@uw.edu, @NanibaaGarrison

Carol R. Horowitz, MD,

Department of Population Health Science and Policy, Icahn School of Medicine at Mount Sinai, 1425 Madison Ave, New York, NY 10029, USA, carol.horowitz@mountsinai.org

Gail P. Jarvik, MD, PhD,

Departments of Medicine (Medical Genetics) and Genome Sciences, University of Washington, Box 357720, Seattle, WA 98195, USA, gjarvik@medicine.washington.edu, @hail_CSER

Rongling Li, MD, PhD, MPH,

Division of Genomic Medicine, National Human Genome Research Institute, 5635 Fishers Ln, Suite 4076, MSC 9305, Bethesda, MD 20982, USA, lir2@mail.nih.gov

Evette J. Ludman, PhD,

Kaiser Permanente Washington Health Research Institute, 1730 Minor Ave, Suite 1600, Seattle, WA 98101, USA, ludman.e@ghc.org

Catherine A. McCarty, PhD, MPH,

University of Minnesota Medical School, 1035 University Dr, Duluth, MN 55812, USA, cmccarty@eirh.org, www.linkedin.com/in/cathy-mccarty-71671387

Jennifer B. McCormick, PhD, MPP,

Biomedical Ethics Program, Mayo Clinic¹, Rochester, MN 55905, USA, jmccormick@hmc.psu.edu

Nathaniel D. Mercaldo, PhD,

Department of Biostatistics, Vanderbilt University², Nashville, TN 32703, USA, nmercald@mgh-ita.org

Melanie F. Myers, PhD,

Division of Human Genetics, Cincinnati Children's Hospital Medical Center and Department of Pediatrics, University of Cincinnati College of Medicine, 3333 Burnet Ave, ML 4006, Cincinnati, OH 45229, USA, Melanie.Myers@cchmc.org, www.facebook.com/UCgeneticcounseling/

Saskia C. Sanderson, PhD,

Department of Genetics and Genomic Sciences, Icahn School of Medicine at Mount Sinai³, New York, NY 20029, USA, saskia.sanderson@ucl.ac.uk

Martha J. Shrubsole, PhD,

Vanderbilt Epidemiology Center, Vanderbilt University Medical Center, 2525 West End Ave, Suite 800, Nashville, TN 37203, USA, martha.shrubsole@Vanderbilt.Edu

Jonathan S. Schildcrout, PhD,

Department of Biostatistics, Vanderbilt University, 2525 West End Ave, Suite 1100, Nashville, TN 37203, USA, jonny.schild@Vanderbilt.Edu

Janet L. Williams, MS,

Genomic Medicine Institute, Geisinger, 100 N Academy Ave 26-20, Danville, PA 17822, USA, jlwilliams3@geisinger.edu, @janet_LCGC

Maureen E. Smith, MS,

Center for Genetic Medicine, Northwestern University, 645 N Michigan Ave, Suite 630-17, Chicago, IL 60611, USA, m-smith6@northwestern.edu

Ellen Wright Clayton, JD, MD, and

Center for Biomedical Ethics and Society, Vanderbilt University Medical Center, 2525 West End Ave, Suite 400, Nashville, TN 37203, USA, ellen.clayton@Vanderbilt.Edu

Ingrid A. Holm, MD, MPH

Division of Genetics and Genomics and the Manton Center for Orphan Diseases Research, Boston Children's Hospital and Department of Pediatrics, Harvard Medical School, 3 Blackfan Cir, CLSB 15022, Boston, MA 02115, USA, Ingrid.Holm@childrens.harvard.edu

Abstract

Background: The factors influencing parents' willingness to enroll their children in biobanks are poorly understood. This study sought to assess parents' willingness to enroll their children, and their perceived benefits, concerns, and information needs under different consent and data sharing scenarios; and to identify factors associated with willingness.

Methods: This large, experimental survey of patients at the 11 eMERGE Network sites used a disproportionate stratified sampling scheme to enrich the sample with historically underrepresented groups. Participants were randomized to receive one of three consent and data sharing scenarios.

Results: 90,000 surveys were mailed and 13,000 individuals responded (15.8% response rate). 5,737 respondents were parents of minor children. Overall, 55% (95% CI: 50-59%) of parents were willing to enroll their youngest minor child in a hypothetical biobank; willingness did not differ between consent and data sharing scenarios. Lower educational attainment, higher religiosity, lower trust, worries about privacy, and attitudes about benefits, concerns, and information needs were independently associated with less willingness to allow their child to participate. Of parents who were willing to participate themselves, 25% were not willing to allow their child to participate. Being willing to participate, but not willing to allow one's child to participate was independently associated with multiple factors, including race, lower educational attainment, lower annual household income, public healthcare insurance, and higher scores on a scale of religiosity.

Conclusions: Fifty-five percent of parents were willing to allow their youngest minor child to participate in a hypothetical biobank. Building trust, protecting privacy, and addressing attitudes may increase enrollment and diversity in pediatric biobanks.

Keywords

Biobank; Biorepository; Informed Consent; Broad Consent; Data Sharing; Pediatrics

INTRODUCTION

Increasingly, biomedical research relies on large collections of biological specimens and associated information, e.g., clinical and genomic data. Including pediatric participants in biobanks is important to study pediatric diseases, as well as pediatric antecedents of adult diseases (Brisson et al. 2012).

Seeking parental permission and participant assent for every study may be unduly burdensome for participants and logistically difficult for investigators. One alternative is “broad consent”—granting one-time permission for all future research with these specimens and data; another is “tiered consent”—granting one-time permission for categories of research, e.g., cancer research (Fullerton et al. 2010). Data sharing may be “controlled” or “open”; requiring approval of the data holder, and making data publicly available, respectively (Joly, Allen, and Knoppers 2012). Broad consent and open data-sharing facilitate research but reduce participants’ future control over their specimens and data (Kaye 2012).

A recent systematic review of studies of individuals’ perspectives on consent and data sharing found that the studies tend to be small, local, and non-representative (Garrison et al. 2016). Only one study identified by this systematic review examined parents’ willingness to enroll their children in a biobank (Neidich et al. 2008). When asked to choose among different consent models, individuals tend to favor models offering greater control (Garrison et al. 2016). Willingness to participate is associated with demographic factors; e.g., less willingness has been found among individuals affiliated with historically underrepresented racial or ethnic groups (Garrison et al. 2016). This may limit these populations’ enrollment in biobanks and the generalizability of their research findings (Fullerton 2011). Variation in willingness among racial and ethnic groups may be mediated in part by different levels of trust in the health care and medical research systems (Shavers, Lynch, and Burmeister 2000).

We therefore inquired about parental attitudes as part of a large, experimental survey of attitudes toward consent and data sharing in biobank research among a diverse group of participants (Smith et al. 2016; Sanderson et al. 2017). Our hypotheses included: parents would express less willingness to have their children participate in, and express more negative attitudes toward, a biobank that provided them with less control over their child’s specimens and data; willingness to participate would be lower among parents of lower socioeconomic status and members of underrepresented racial and ethnic groups; and differences in willingness to participate would be mediated by the parents’ trust in their healthcare system and medical researchers, concerns about privacy, and attitudes toward participating.

METHODS

Study Design and Procedures

This study is part of a larger study whose methods are fully described elsewhere (Smith et al. 2016; Sanderson et al. 2017).

Inclusion criteria for participants were that they or their minor child (1) were a patient at 1 of the 11 Electronic Medical Records and Genomics (eMERGE) Network sites (Gottesman et al. 2013) between October 1, 2013 and September 1, 2014, (2) had an address that could be geocoded, and (3) had their age and sex documented in the electronic medical record. Respondents were included in the present analysis if they also (4) reported that they were the parent or guardian of a child under 18 years old. To enrich the sample with socio-demographic groups that have previously been underrepresented in attitudinal research, we utilized a disproportionate stratified sampling scheme (Sanderson et al. 2017).

Individuals within each sampling strata were randomly assigned to 1 of 3 biobanking scenarios: broad consent and controlled data sharing, tiered consent and controlled data sharing, or broad consent and open data sharing. Offering a single option more accurately represents the situation individuals typically have when asked to participate in a biobank, as opposed to being asked to choose among several different consent and data sharing options. All the other details of the hypothetical scenarios were identical.

We mailed pre-notification postcards and optical scan surveys in April 2015, reminder letters in May 2015, and second surveys in July 2015. Participants could complete a paper survey and return it in a self-addressed stamped envelope, or an online survey through a secure REDCap database (Harris et al. 2009). Participants received a non-contingent \$2 pre-incentive. This study was approved by the institutional review board at each of the participating sites.

A multidisciplinary team developed the survey instrument based on the results of a systematic review of the literature (Garrison et al. 2016). The team conducted cognitive interviews to test and refine the instrument and a pilot study to evaluate the feasibility of study procedures (Smith et al. 2016; Sanderson et al. 2017).

Standard measures of demographic characteristics (see Table 1) were used. Rurality was determined from census data definitions. Religiosity, trust, concern about privacy, and willingness to participate in a biobank were assessed using items adapted from previous research. Questions about attitudes toward participation in a biobank were either generated specifically for this study or adapted from previous research. The team identified three sub-domains: (1) benefits of and (2) concerns about participating in the biobank, and (3) information needs about the biobank's governance. Perceived benefits and concerns were assessed for both the respondent and the respondent's youngest minor child. Items were measured using a five-point Likert-style scale; factor analysis confirmed that the sub-domains were distinct factors. Composite scale scores were created by calculating the mean of each sub-domain (Sanderson et al. 2017).

Data Analysis

The American Association for Public Opinion Research's (AAPOR's) criteria were used to calculate response rates (American Association for Public Opinion Research 2011). Differences in demographic and respondent characteristics among the three scenarios were tested using Pearson's chi-squared tests. For all other analyses, each respondent was assigned a post-stratified sampling weight to account for the stratified sampling design. Because historically understudied populations were intentionally oversampled, sampling weights varied within and across sites. Site-specific weight trimming and redistribution were conducted by combining two commonly used approaches (Sanderson et al. 2017).

Differences in trust and privacy among the three biobanking scenarios were examined using Pearson's chi-squared tests. Comparisons of willingness and attitudes among biobanking scenarios were performed via a Wald test from an ordinal logistic regression, proportional odds model (Sanderson et al. 2017).

To identify characteristics associated with respondents' willingness to have their youngest minor child participate in a biobank, willingness was dichotomized ("Yes probably" and "Yes definitely" as yes; all other responses as no) and regressed on covariates using unadjusted (marginal) and adjusted logistic regression analyses. Adjusted models were fitted hierarchically; sociodemographic variables were added first; trust and privacy items second; and attitudinal constructs third. Unadjusted estimates were summarized with percentages, and adjusted estimates were summarized with odds-ratios and 95% confidence intervals. Sub-domain analyses revealed little to no evidence that the relationship between survey type and willingness differed across socio-demographic variables (not shown). Responses to individual attitudinal items were described, and 95% confidence intervals computed (Sanderson et al. 2017).

Chi-squared techniques were used to test whether parents' willingness to participate was associated with their willingness to allow their child to participate. This test was performed overall and stratified by scenario. Wald tests were performed to assess differences between the biobanking scenarios. Willingness to have their child participate in the biobank was regressed on covariates using unadjusted (marginal) and adjusted logistic regression analyses, as described above. This second regression focused on the subset of parents who were themselves willing to participate in a biobank, comparing those who were willing versus those who were not willing to allow their child to participate.

Multiple imputation was conducted within each site to account for item non-response, which was generally less than 1% for all key variables, except income (6%). Socio-demographic variables, biobank participation willingness, attitudinal constructs, and deciles of post-stratified weights were used to impute all missing data. Five complete imputation datasets were created for each site. Survey weighted regression analyses were performed on each complete dataset and combined using standard "Rubin's (1987) rules." Site-specific estimates were then combined for every analysis to summarize characteristics of the entire eMERGE Network using multivariate random-effect, meta-analytic methods (Sanderson et al. 2017). All analyses were performed using R Version 3.2.2 (R Project for Statistical Computing) and SAS Version 9.4 (SAS Institute Inc.).

RESULTS

Respondent Characteristics

A total of 90,000 surveys were mailed; 7,672 individuals were ineligible due to an invalid address, death, or incapacity; 681 refused to participate; and 13,000 responded (AAPOR response rate 15.8%) (Sanderson et al. 2017). The responses of the 5,737 individuals who were parents of a minor child are the focus of this paper. A majority of the parents were female (73.6%); 36-50 years old (56.9%); white (52.5%); not Hispanic or Latino/a (81.4%); had attained at least a Bachelor's degree (64.8%); and had an annual household income of \$90,000 or more (50.9%) (Table 1). We were unable to detect sociodemographic differences among respondents receiving each of the three scenarios.

Fifty-nine percent agreed that they trusted their healthcare system, and 58% agreed that they trusted medical researchers. Ninety-two percent also agreed that health information privacy was important to them, and 55-68% expressed worry about privacy (Table 2 – available online). We were unable to detect differences in trust and privacy among respondents receiving each of the three scenarios.

Willingness and Attitudes Compared between Scenario Groups

Overall, 55% (95% CI: 50-59%) of respondents stated that they would be willing to let their youngest minor child participate in the biobank to which they were randomized (Table 3 – available online). We were unable to detect differences in willingness among the scenarios: Broad-Controlled 54% (50-59%), Broad-Open 53% (48-57%), and Tiered-Controlled 56% (51-65%) (p-value Broad-Controlled vs Broad-Open 0.32, and p-value Broad-Controlled vs. Tiered-Controlled 0.27).

Respondents generally perceived participation as beneficial and differentiated benefits to others from benefits to their child, i.e., 80% (95% CI 76-84%) of participants agreed or strongly agreed with the statement “I would feel that my child was helping other children,” and 58% (95% CI 55-61) agreed or strongly agreed with “I would feel that taking part could help my child.” The most frequently endorsed concern was “I would worry about my child's privacy” (63%, 95% CI 60-66%). The 2 least frequently endorsed concerns were “... some research would be done that I did not want to take part in” and “... someone might make money using my health information” (38%, 95% CI 34-42%). Greater than 63% of participants agreed or strongly agreed with each of the information need statements; e.g., 92% (95% CI 88-96) agreed or strongly agreed with the statement “I would want to know who makes sure that my health information is used in the right way” (Table 4 – available online).

A larger percentage perceived high benefits for themselves (76%; 95% CI 72-80%) than for their child (60%, 95% CI 57-66%). While 38% (95% CI 36-42%) of respondents had high concerns for themselves, 62% (95% CI 56-67%) had high concerns for their child. Seventy-seven percent (95% CI 72-82%) had high information needs (Table 3 – available online). We were unable to detect differences in any of the attitudes among respondents receiving each of the three scenarios.

Associations between Willingness to Permit Child to Participate and Respondent Characteristics

Because patterns of associations between sociodemographic variables and willingness were the same within each scenario, and because willingness did not differ significantly between groups, all subsequent analyses were conducted on the whole sample.

The following respondent characteristics were independently associated with willingness to allow their child to participate before attitudes were entered into the model: race, educational attainment, annual household income, healthcare insurance, rurality, religiosity, trust in my healthcare system, and privacy concerns. When attitudes toward the biobank were entered into the model, each of the five composite scale variables were independently associated with willingness. In this model, educational attainment, religiosity, trust in healthcare system, and privacy concerns remained associated with willingness. Respondents who reported educational attainment “up to some high school” (48%, OR 0.34, 95% CI 0.20-0.53), “high school graduate or GED” (50%, OR 0.54, 95% CI 0.33-0.86), or “some college” (52%, OR 0.61, 95% CI 0.44-0.87) were less willing to allow their child to participate than “PhD/MD/JD or equivalent” (59%, reference group). Respondents who were “very religious” were also less willing to allow their child to participate (45%) than those who were “not at all religious” (52%, OR 0.77, 95% CI 0.48-0.88) (Table 5).

Comparisons of Willing and Unwilling Parents

Overall, 25% of parents who were willing to participate were not willing to allow their youngest minor child to participate and only 10% of parents who were not willing to participate were willing to allow their child to participate ($p < 0.001$). We were unable to detect differences in either measure among the three scenarios (Table 6 – available online).

Associations between Willingness to Participate Themselves but Not Willing to Allow Their Child to Participate, and Respondent Characteristics

Among those parents who were themselves willing to participate, the following respondent characteristics were independently associated with willingness to allow their child to participate as well before attitudes were entered into the model: race, educational attainment, annual household income, healthcare insurance, religiosity, trusting my healthcare system, trusting medical researchers, and importance of health information privacy. When attitudes toward the biobank were entered into the model, all of the composite scale variables, except perceived benefits for youngest child, were independently associated with willingness. The previously identified socio-demographics, trust, and privacy remained associated with willingness. The largest odds ratios included “I trust medical researchers” (Disagree 41%, Agree 87%, OR 0.38, 95% CI 0.20-0.69), religiosity (“Somewhat religious” 72%, “Not at all religious” 85%, OR 0.51, 95% CI 0.49-0.55), and educational attainment (“Up to some high school” 72%, “PhD/MD/JD or equivalent” 81%, OR 0.57, 95% CI 0.52-0.67) (Table 7).

DISCUSSION

Fifty-five percent of participants in this study were willing to permit their youngest minor child to participate in biobank research. We were unable to detect associations between

willingness or attitudes, and the consent and data sharing scenario to which the participant was randomized. We were also unable to detect an independent association between willingness and socioeconomic status, race, or ethnicity. Less willingness was, however, independently associated with lower educational attainment, higher religiosity, lower trust, privacy worries, and perceived benefits, concerns, and information needs. Twenty-five percent of parents who were willing to participate themselves were not willing to let their child participate. Several differences exist between the analysis of the entire sample and this group: information needs was not independently associated with willingness to allow their child to participate, and Asian, American Indian, or Alaska Native, and other race; lower annual household income; public or no insurance; and less trust in medical researchers were negatively associated with willingness.

One additional study on parents' willingness to enroll their children in biobanks has been published since the systematic review (Kong et al. 2016). Parental willingness in our sample (55%) is closer to Neidich et al.'s (2008) (48%) than to Kong et al.'s (2016) (80%). These differences are likely due to differences in study populations. Neidich et al. surveyed postpartum adult women who delivered at the University of Chicago Hospitals and Clinics. Seventy-five percent of their participants' education level was less than college graduate which, based on our findings, would be expected to be associated with lower willingness. Kong et al. surveyed parents of patients at British Columbia Children's Hospital and Vancouver high schools and, unfortunately, did not provide detailed demographic data regarding their participants.

With the exception of Brothers and Clayton's (2012) work on opt-out models, studies have not generally inquired into parental preferences regarding consent and/or data sharing models (Kaufman et al. 2008; Salvaterra et al. 2014). Kong et al. (2016), for example, did not explicitly state the biobank's consent and data sharing model, and Neidich et al. (2008) specified broad consent and controlled data sharing. Our inability to detect an association between parental preference and the consent or data sharing model is novel.

Neither Kong et al. (2016) or Neidich et al. (2008) examined the association between willingness and sociodemographic factors. While Neidich et al. (2008) collected demographic data and attitudes regarding trust and justice, they did not perform logistic regression analyses to examine the association of these factors with willingness.

Although there is a substantial literature on the retention of blood samples from newborn screening and their use in research, this practice is not analogous to other forms of biobanking because, in the vast majority of states, informed consent is not obtained for the collection of samples, let alone research with them. However, in their survey of a nationally representative sample of parents, Tarini et al. (2010) did assess parents' willingness to permit research with newborn screening bloodspots. The percentage of participants who were very or somewhat willing was 76.4%. Willingness was associated with younger parental age and worse child health. The authors did not find a significant association with education and did not assess religiosity.

The broader literature on the association between parents' willingness to have their children participate in medical research and sociodemographic factors has substantial limitations; secondary analyses of actual recruitment rates often lack important sociodemographic and attitudinal data (Natale et al. 2017), and prospective studies are frequently inadequately powered to perform logistic regression analyses (Wendler and Jenkins 2008; Hoberman et al. 2013). Adequately designed studies have evaluated different factors and have found varying results. For example, Svensson et al. (2012) found, contrary to our results, that parents' willingness was associated with lower educational attainment. With respect to perceived benefits and concerns, Tait, Voepel-Lewis, and Malviya (2003) similarly found that perceived risk of the study, perceived benefits to the child, and perceived importance of the study were among the factors associated with parents' willingness to permit their child to participate in an actual, minimal or minor risk anesthesia or surgery clinical study. Our results augment and reinforce some of these findings.

We found that 25% of parents who were willing to participate were not willing for their child to participate. These parents' decisions are consistent with the best interest standard (Katz and Webb 2016). When making decisions for themselves, competent adults may assume significant risks in order to benefit others. When deciding for their children, parents are expected to focus primarily on their child's interests and to weigh the benefits and risks to the child. Consistent with this expectation, the parents who were not willing to give permission for their child to participate believed that the biobank had fewer benefits and greater risks than parents who were willing. Our findings did not support our speculation that parents would be more protective of younger children; parents' willingness was not associated with their child's age.

Our findings generally suggest that recruitment of children and the promotion of genetic diversity in biobanks might be improved by developing trust, protecting privacy, articulating benefits, addressing concerns, and meeting information needs. Additional research is needed to determine why parents with lower educational attainment and higher religiosity are less willing to allow their children to participate, and how to increase their participation.

Several limitations of this study should be acknowledged. The primary limitation is the low response rate and the possibility of non-responder bias. It is reasonable to assume that non-responders are less supportive of research and a higher response rate therefore might result in a lower willingness to allow their youngest child to participate in a biobank. Other limitations include our reliance on participants' self-reported intentions and the possibility that participants did not understand or appreciate the consent and/or data sharing model. We also did not assess all factors (e.g., child's perceived health) that may be associated with willingness. This study, nonetheless, has significant strengths including the diversity of sites, the rigorous sampling strategy, and the experimental design.

This study's results suggest that when parents are given the hypothetical choice whether or not to allow their child to participate in a biobank, the consent and/or data sharing model may not affect their decision. While racial differences in willingness to participate may be mediated by modifiable factors such as trust, privacy concerns, and attitudes, other sociodemographic differences independently predict willingness (e.g., educational

attainment and religiosity). Parents' greater willingness to participate themselves than to allow their children to participate appears consistent with the best interest standard. Individuals discriminate among various benefits, risks, and concerns. Investigators and policy makers may be able to increase enrollment and diversity through building trust, protecting privacy, and addressing benefits, concerns, and information needs.

ACKNOWLEDGMENTS:

We would like to acknowledge the work of the following: Melissa Basford, Meckenzie Behr, Jess Behrens, Laura Beskow, Diego Campos, David S. Carrell, Ariel Chandler, Beth Chau, Rosetta Chiavacci, Kurt Christensen, Beth Cobb, John Connolly, Natalia Connolly, Stephanie Devaney, Joe DeWalle, Steve Ellis, Alexander Fiksdal, John Harley, Diana Harris, Paul Hitz, David Kaufman, Yolanda Keppel, Terrie E. Kitchner, Nicole Lockhart, Keith Marsolo, Catherine Marx, Valerie D. McManus, Katherine Nowakowski, Jennifer Pacheco, Josh Pankratz, Lisa Price, Michelle Ramos, Marguerite Robinson, Aaron Scrol, Sarah Stallings, Allise Taran, Matt Verkemp, Gordon Willis, and Sonja Zinzel.

FUNDING: The Consent, Education, Regulation and Consultation (CERC) Survey project within the eMERGE Network was initiated and funded by NHGRI with additional funding by the NIH Office of the Director through the following grants: U01HG006828 (Cincinnati Children's Hospital Medical Center/Boston Children's Hospital), U01HG006830 (Children's Hospital of Philadelphia), U01HG006389 (Essentia Institute of Rural Health, Marshfield Clinic Research Foundation, and Pennsylvania State University), U01HG006382 (Geisinger Clinic), U01HG008657 (Kaiser Permanente/University of Washington), 3U01HG006379 (Mayo Clinic), U01HG006380 (Icahn School of Medicine at Mount Sinai), 3U01-HG006388 (Northwestern University), U01HG006378 (Vanderbilt University Medical Center), and 3U01HG006385 (Vanderbilt University Medical Center serving as the Coordinating Center).

REFERENCES:

- American Association for Public Opinion Research. 2011 Standard definitions: Final dispositions of case and outcome rates for surveys. Oakbrook Terrace, IL: American Association for Public Opinion Research.
- Brisson AR, Matsui D, Rieder MJ, and Fraser DD. 2012 Translational research in pediatrics: Tissue sampling and biobanking. *Pediatrics* 129(1):153–62. doi: 10.1542/peds.2011-0134. [PubMed: 22144705]
- Brothers KB, and Clayton EW. 2012 Parental perspectives on a pediatric human non-subjects biobank. *AJOB Prim Res* 3(3):21–29. doi: 10.1080/21507716.2012.662576. [PubMed: 23181193]
- Fullerton SM 2011 The input-output problem: whose DNA do we study, and why does it matter? In *Achieving justice in genomic translation: Re-thinking the pathway to benefit*, ed. Burke W, Edwards KA, Goering S, Holland S, and Trinidad SB. 40–55. New York: Oxford University Press.
- Fullerton SM, Anderson NR, Guzauskas G, Freeman D, and Fryer-Edwards K. 2010 Meeting the governance challenges of next-generation biorepository research. *Sci Transl Med* 2(15):15cm3. doi: 10.1126/scitranslmed.3000361.
- Garrison NA, Sathe NA, Antommaria AH, Holm IA, Sanderson SC, Smith ME, McPheeters ML, and Clayton EW. 2016 A systematic literature review of individuals' perspectives on broad consent and data sharing in the united states. *Genet Med* 18(7):663–71. doi: 10.1038/gim.2015.138. [PubMed: 26583683]
- Gottesman O, Kuivaniemi H, Tromp G, Faucett WA, LI R, Manolio TA, Sanderson SC, Kannry J, Zinberg R, Basford MA, et al. 2013 The Electronic Medical Records and Genomics (eMERGE) Network: past, present, and future. *Genet Med* 15(10):761–71. doi: 10.1038/gim.2013.72. [PubMed: 23743551]
- Harris PA, Taylor R, Thielke R, Payne J, Gonzalez N, and Conde JG. 2009 Research electronic data capture (redcap)-a metadata-driven methodology and workflow process for providing translational research informatics support. *J Biomed Inform* 42(2):377–81. doi: 10.1016/j.jbi.2008.08.010. [PubMed: 18929686]
- Hoberman A, Shaikh N, Bhatnagar S, Haralam MA, Kearney DH, Colborn DK, Kienholz ML, Wang L, Bunker CH, Keren R, et al. 2013 Factors that influence parental decisions to participate in

- clinical research: Consenters vs nonconsenters. *JAMA Pediatr* 167(6):561–6. doi: 10.1001/jamapediatrics.2013.1050. [PubMed: 23546617]
- Joly Y, Allen C, and Knoppers BM. 2012 Currents in contemporary bioethics. Open access as benefit sharing? The example of publicly funded large-scale genomic databases. *J Law Med Ethics* 40(1): 143–6. doi: 10.1111/j.1748-720X.2012.00652.x. [PubMed: 22458469]
- Kaye J 2012 The tension between data sharing and the protection of privacy in genomics research. *Annu Rev Genomics Hum Genet* 13:415–31. doi: 10.1146/annurev-genom-082410-101454. [PubMed: 22404490]
- Katz AL, Webb SA, and Committee on Bioethics. 2016 Informed consent in decision-making in pediatric practice. *Pediatrics* 138(2). doi: 10.1542/peds.2016-1485.
- Kaufman D, Geller G, Leroy L, Murphy J, Scott J, and Hudson K. 2008 Ethical implications of including children in a large biobank for genetic-epidemiologic research: A qualitative study of public opinion. *Am J Med Genet C Semin Med Genet* 148C(1):31–9. doi: 10.1002/ajmg.c.30159. [PubMed: 18189289]
- Kong CC, Tarling TE, Strahlendorf C, Ditttrick M, and Vercauteren SM. 2016 Opinions of adolescents and parents about pediatric biobanking. *J Adolesc Health* 58(4):474–480. doi: 10.1016/j.jadohealth.2015.12.015. [PubMed: 27013273]
- Natale JE, Lebet R, Joseph JG, Ulysse C, Ascenzi J, Wypij D, Curley MAQ, and Randomized Evaluation of Sedation Titration for Respiratory Failure Study Investigators. 2017 Racial and ethnic disparities in parental refusal of consent in a large, multisite pediatric critical care clinical trial. *J Pediatr* 184:204–208 e1. doi: 10.1016/j.jpeds.2017.02.006. [PubMed: 28410087]
- Neidich AB, Joseph JW, Ober C, and Ross LF. 2008 Empirical data about women's attitudes towards a hypothetical pediatric biobank. *Am J Med Genet A* 146A(3):297–304. doi: 10.1002/ajmg.a.32145. [PubMed: 18205141]
- Rubin DB 1987 Multiple imputation for nonresponse in surveys. New York: John Wiley & Sons.
- Salvatera E, Locatelli F, Strazzer S, Borgatti R, D'Angelo G, and Lenzi L. 2014 Paediatric biobanks: Opinions, feelings and attitudes of parents towards the specimen donation of their sick children to a hypothetical biobank. *Pathobiology* 81(5-6):304–8. doi: 10.1159/000362091. [PubMed: 25792219]
- Sanderson SC, Brothers KB, Mercaldo ND, Clayton EW, Antommaria AHM, Aufox SA, Brilliant MH, Campos D, Carrell DS, Connolly J, et al. 2017 Public attitudes toward consent and data sharing in biobank research: a large multi-site experimental survey in the US. *Am J Hum Genet* 100(3):414–27. doi: 10.1016/j.ajhg.2017.01.021. [PubMed: 28190457]
- Shavers VL, Lynch CF, and Burmeister LF. 2000 Knowledge of the Tuskegee study and its impact on the willingness to participate in medical research studies. *J Natl Med Assoc* 92(12):563–72. [PubMed: 11202759]
- Smith ME, Sanderson SC, Brothers KB, Myers MF, McCormick J, Aufox S, Shrubsole MJ, Garrison NA, Mercaldo ND, Schildcrout JS, et al. 2016 Conducting a large, multi-site survey about patients' views on broad consent: challenges and solutions. *BMC Med Res Methodol* 16(1):162. doi: 10.1186/s12874-016-0263-7. [PubMed: 27881091]
- Svensson K, Ramirez OF, Peres F, Barnett M, and Claudio L. 2012 Socioeconomic determinants associated with willingness to participate in medical research among a diverse population. *Contemp Clin Trials* 33(6):1197–205. doi: 10.1016/j.cct.2012.07.014. [PubMed: 22885788]
- Tait AR, Voepel-Lewis T, and Malviya S. 2003 Participation of children in clinical research: Factors that influence a parent's decision to consent. *Anesthesiology* 99(4):819–25. [PubMed: 14508312]
- Tarini BA, Goldenberg A, Singer D, Clark SJ, Butchart A, and Davis MM. Not without my permission: Parents' willingness to permit use of newborn screening samples for research." *Public Health Genomics* 13, no. 3 (2010): 125–30. doi: 10.1159/000228724. [PubMed: 19602864]
- Wendler D, and Jenkins T. 2008 Children's and their parents' views on facing research risks for the benefit of others. *Arch Pediatr Adolesc Med* 162(1):9–14. doi: 10.1001/archpediatrics.2007.3. [PubMed: 18180406]

TABLE 1

Sociodemographic and Health Characteristics of Survey Respondents by Consent and Data Sharing Model and Across Models

	Broad-controlled, <i>n</i> (%)	Broad-open, <i>n</i> (%)	Tiered-controlled, <i>n</i> (%)	Total, <i>n</i> (%)
<i>n</i>	1923 (33.5)	1923 (33.5)	1891 (33.0)	5737 (100)
Gender				
Female	1387 (73.2)	1425 (74.8)	1363 (72.9)	4175 (73.6)
Male	508 (26.8)	480 (25.2)	507 (27.1)	1495 (26.4)
Age (Years)				
18-35	470 (25.2)	489 (26.1)	501 (27.3)	1460 (26.2)
36-50	1069 (57.2)	1053 (56.1)	1053 (57.4)	3175 (56.9)
51-64	291 (15.6)	295 (15.7)	258 (14.1)	844 (15.1)
65 or older	38 (2.0)	39 (2.1)	23 (1.3)	100 (1.8)
Race				
White	991 (52.4)	993 (52.5)	974 (52.6)	2958 (52.5)
Asian	355 (18.8)	334 (17.7)	343 (18.5)	1032 (18.3)
Black or African American	214 (11.3)	227 (12.0)	218 (11.8)	659 (11.7)
Other	205 (10.8)	171 (9.0)	184 (9.9)	560 (9.9)
American Indian or Alaska Native	56 (3.0)	78 (4.1)	61 (3.3)	195 (3.5)
More than one race	58 (3.1)	67 (3.5)	52 (2.8)	177 (3.1)
Native Hawaiian or Pacific Islander	16 (0.8)	20 (1.1)	18 (1.0)	54 (1.0)
Ethnicity				
Not Hispanic or Latino/a	1550 (81.6)	1544 (81.8)	1500 (80.9)	4594 (81.4)
Hispanic or Latino/a	349 (18.4)	344 (18.2)	355 (19.1)	1048 (18.6)
Educational attainment				
Up to some high school (grades 9-12)	96 (5.1)	100 (5.4)	83 (4.5)	279 (5.0)
High school graduate or GED	150 (8.0)	149 (8.0)	161 (8.7)	460 (8.3)
Some college	399 (21.3)	423 (22.8)	399 (21.6)	1221 (21.9)
Bachelor's degree or equivalent	588 (31.4)	546 (29.5)	574 (31.1)	1708 (30.7)
Master's degree or equivalent	411 (22.0)	408 (22.0)	397 (21.5)	1216 (21.8)
PhD, MD, JD, or equivalent	227 (12.1)	227 (12.3)	232 (12.6)	686 (12.3)
Annual household income				
Less than \$30,000	303 (16.8)	356 (19.7)	329 (18.5)	988 (18.3)
\$30,000 - \$59,999	287 (15.9)	302 (16.7)	271 (15.3)	860 (16.0)
\$60,000 - \$89,999	262 (14.5)	247 (13.7)	287 (16.2)	796 (14.8)
\$90,000 to \$149,999	446 (24.7)	390 (21.6)	400 (22.5)	1236 (22.9)
More than \$150,000	505 (28.0)	512 (28.3)	489 (27.5)	1506 (28.0)
Total number of people in household				
1	19 (1.0)	24 (1.3)	18 (1.0)	61 (1.1)
2	108 (5.7)	113 (6.0)	106 (5.7)	327 (5.8)
3	489 (25.8)	528 (27.9)	479 (25.7)	1496 (26.5)
4 or more	1281 (67.5)	1228 (64.9)	1262 (67.7)	3771 (66.7)

	Broad-controlled, <i>n</i> (%)	Broad-open, <i>n</i> (%)	Tiered-controlled, <i>n</i> (%)	Total, <i>n</i> (%)
Work situation				
Working	1287 (67.4)	1239 (65.0)	1269 (67.6)	3795 (66.7)
Retired	44 (2.3)	47 (2.5)	26 (1.4)	117 (2.1)
Disabled / Unemployed	171 (9.0)	177 (9.3)	182 (9.7)	530 (9.3)
Other	408 (21.4)	444 (23.3)	399 (21.3)	1251 (22.0)
Healthcare insurance				
Private insurance	1503 (79.4)	1491 (79.3)	1490 (80.0)	4484 (79.6)
Public insurance	319 (16.9)	306 (16.3)	291 (15.6)	916 (16.3)
Other type of insurance	24 (1.3)	40 (2.1)	34 (1.8)	98 (1.7)
No insurance	47 (2.5)	43 (2.3)	47 (2.5)	137 (2.4)
Rurality (from census-level data)				
Suburban / Urban	1104 (57.4)	1114 (57.9)	1120 (59.2)	3338 (58.2)
Rural	819 (42.6)	809 (42.1)	771 (40.8)	2399 (41.8)
Marital status				
Married	1485 (79.0)	1434 (77.1)	1426 (77.3)	4345 (77.8)
Not married, living with someone	133 (7.1)	123 (6.6)	136 (7.4)	392 (7.0)
Not married, not living with someone	261 (13.9)	303 (16.3)	282 (15.3)	846 (15.2)
Age of youngest child (years)				
0-5	673 (37.4)	627 (35.0)	671 (37.4)	1971 (36.6)
6-11	539 (29.9)	585 (32.6)	583 (32.5)	1707 (31.7)
12-17	588 (32.7)	580 (32.4)	538 (30.1)	1706 (31.7)
Religiosity				
Not at all religious	240 (12.7)	262 (13.8)	252 (13.5)	754 (13.3)
Not very religious	292 (15.4)	255 (13.5)	290 (15.6)	837 (14.8)
Somewhat religious	856 (45.2)	848 (44.8)	856 (46.0)	2560 (45.3)
Very religious	505 (26.7)	529 (27.9)	464 (24.9)	1498 (26.5)

Observed frequencies and percentages are reported independent of sampling design. We performed Pearson's chi-square tests to assess differences between consent and data sharing models and each sociodemographic and health characteristic. No differences were detected. Test summaries are available from the authors.

TABLE 2

Trust in the Healthcare System and in Medical Researchers, and Privacy Concerns by Consent and Data Sharing Model and Across Models

	Broad-controlled, <i>n</i> (%)	Broad-open, <i>n</i> (%)	Tiered-controlled, <i>n</i> (%)	Total, <i>n</i> (%)
Trust				
I trust my healthcare system				
Disagree / Strongly disagree	268 (14)	258 (14)	258 (14)	784 (14)
Neither agree nor disagree	517 (27)	519 (27)	525 (28)	1561 (27)
Agree / Strongly agree	1126 (59)	1127 (59)	1090 (58)	3343 (59)
I trust medical researchers				
Disagree / Strongly disagree	152 (8)	150 (8)	140 (8)	442 (8)
Neither agree nor disagree	639 (34)	655 (35)	629 (34)	1923 (34)
Agree / Strongly agree	1111 (58)	1086 (57)	1091 (59)	3288 (58)
Privacy				
Health information privacy is important to me				
Disagree / Strongly disagree	22 (1)	32 (2)	30 (2)	84 (1)
Neither agree nor disagree	108 (6)	120 (6)	121 (6)	349 (6)
Agree / Strongly agree	1776 (93)	1749 (92)	1719 (92)	5244 (92)
I worry about the privacy of my health information				
Disagree / Strongly disagree	267 (14)	277 (15)	250 (13)	794 (14)
Neither agree nor disagree	331 (17)	319 (17)	349 (19)	999 (18)
Agree / Strongly agree	1305 (69)	1312 (69)	1266 (68)	3883 (68)
I worry about my privacy if participating in a biobank				
Disagree / Strongly disagree	545 (29)	496 (26)	538 (29)	1579 (28)
Neither agree nor disagree	325 (17)	325 (17)	308 (17)	958 (17)
Agree / Strongly agree	1022 (54)	1086 (57)	1020 (55)	3128 (55)
I worry about my child's privacy if participating in a biobank				
Disagree / Strongly disagree	391 (20)	349 (17)	380 (20)	1120 (20)
Neither agree nor disagree	377 (19)	378 (19)	303 (16)	858 (15)
Agree / Strongly agree	1223 (61)	1269 (64)	1174 (63)	3666 (65)

Observed frequencies and percentages are reported independent of sampling design. We performed Pearson's chi-square tests to assess differences between consent and data sharing models and each characteristic. No differences were detected. Test summaries are available from the authors.

TABLE 3

Willingness and Attitudes (Composite Perceived Benefits, Concerns, and Information Needs) Towards Participating in a Biobank by Consent and Data Sharing Model and Across Models

	Broad-controlled (BC)		Broad-open (BO)		Tiered-controlled (TC)		Total	p-value BC vs BO	p-value BC vs TC
	n	% (95% CI)	n	% (95% CI)	n	% (95% CI)	n	% (95% CI)	
Willingness to have youngest child participate in a biobank	1005	54 (50,59)	952	53 (48,57)	1019	56 (51,61)	2976	55 (50,59)	0.32
Benefits for self									
Low	55	4 (1,8)	56	5 (2,10)	57	4 (1,9)	168	5 (1,9)	0.38
Intermediate	416	23 (19,28)	439	17 (13,24)	447	25 (20,29)	1302	23 (17,27)	
High	1367	73 (68,78)	1346	78 (75,86)	1312	74 (68,79)	4025	76 (72,80)	0.41
Concerns for self									
Low	531	28 (24,33)	498	32 (27,34)	548	31 (27,37)	1577	30 (25,36)	0.37
Intermediate	555	32 (27,37)	550	31 (25,35)	540	34 (29,40)	1645	32 (27,37)	
High	725	41 (37,47)	779	37 (41,42)	707	38 (33,42)	2211	38 (36,42)	0.59
Information needs for self									
Low	64	5 (2,9)	68	6 (3,10)	64	3 (1,8)	196	5 (2,9)	0.63
Intermediate	343	22 (17,26)	281	15 (13,19)	322	17 (12,23)	946	18 (14,23)	
High	1413	73 (67,77)	1485	79 (73,83)	1431	80 (76,84)	4329	77 (72,82)	0.19
Benefits for youngest child									
Low	134	9 (5,15)	119	7 (3,11)	124	5 (1,10)	377	7 (4,11)	0.28
Intermediate	586	32 (28,37)	624	33 (27,38)	608	35 (31,39)	1818	33 (27,37)	
High	1154	59 (54,64)	1117	60 (58,64)	1106	60 (55,65)	3377	60 (57,66)	0.14
Concerns for youngest child									
Low	329	19 (14,24)	310	15 (12,21)	335	15 (10,220)	974	16 (14,23)	0.36
Intermediate	441	21 (16,27)	410	20 (16,24)	422	23 (18,27)	1273	22 (19,26)	
High	1088	60 (56,64)	1143	65 (56,70)	1068	63 (59,67)	3299	62 (56,67)	0.69

We dichotomized willingness (Yes = Yes probably and Yes definitely, No = All other responses).

Attitudes were defined as the average of the recorded survey items (1 = Strongly Disagree, ..., 5 = Strongly Agree) that comprised the outcomes. We characterized Low as <2.5, Intermediate 2.5-3.5, and High >3.5. Observed frequencies and survey-adjusted percentages (95% CI) are reported for all outcomes. Wald tests were performed to assess differences between data sharing models (BC vs BO) and between consent types (BC vs TC).

Attitudes (Specific Benefits, Concerns, and Information Needs) Towards Participating in a Biobank Categorized by Willingness for Self to Participate / Willingness for Child to Participate

TABLE 4

Agree or Strongly Agree						
Attitude	Total		Yes/Yes		Yes/No	
	n	% (95% CI)	n	% (95% CI)	n	% (95% CI)
Benefits of participating in a biobank						
I would feel that I was helping future generations.	4862	85 (82, 88)	2661	97 (94, 100)	230	89 (84, 94)
... that taking part could lead to better medical treatments.	4757	86 (82, 90)	2665	98 (94, 100)	234	93 (88, 98)
... that taking part would help doctors where I get my medical care take better care of patients.	4415	79 (75, 83)	2459	93 (89, 97)	207	82 (77, 87)
... that taking part could help my family.	3575	63 (58, 68)	2085	77 (72, 82)	170	66 (61, 71)
... that taking part could help me personally.	2286	39 (33, 44)	1358	49 (44, 54)	126	48 (44, 52)
Concerns about participating in a biobank						
I would worry about my privacy.	3128	56 (51, 61)	946	36 (32, 40)	167	63 (58, 68)
... about my medical record being shared.	2850	50 (46, 54)	781	29 (24, 34)	152	58 (53, 63)
... about how researchers would use my health information.	2596	47 (44, 50)	715	26 (21, 31)	135	55 (50, 59)
... about my genetic information being shared.	2370	43 (40, 46)	597	23 (18, 28)	121	49 (44, 54)
... that some research would be done that I did not want to take part in.	2195	38 (34, 42)	591	21 (16, 26)	115	44 (38, 49)
... that someone might make money using my health information.	2213	38 (34, 42)	695	25 (19, 31)	123	47 (43, 52)
Information needs regarding governance of a biobank						
I would want to know who makes sure that my health information is used in the right way.	5047	92 (88, 96)	2240	85 (81, 89)	234	94 (90, 98)
... what kind of knowledge would result from the use of my health information.	4970	89 (85, 93)	2333	88 (83, 93)	229	91 (86, 96)
... what would happen if a researcher misused the health information in the biobank.	4910	88 (84, 92)	2364	88 (83, 92)	232	91 (85, 96)
... if my health information might be used by insurance companies.	4654	86 (81, 90)	2118	82 (77, 86)	221	90 (85, 95)
... the types of research my health information would be used for.	4531	79 (75, 83)	2014	73 (69, 77)	220	84 (80, 89)
... who runs the biobank.	4302	73 (68, 78)	1931	69 (64, 74)	205	76 (71, 81)
... how the biobank covers costs.	3435	64 (60, 68)	1501	57 (52, 62)	174	71 (66, 76)
... if my health information might be used by drug companies that make money.	3444	63 (59, 67)	1406	54 (48, 59)	171	70 (65, 75)
Benefits of child participating in a biobank						
I would feel that my child was helping other children	4521	80 (76, 84)	2651	97 (92, 100)	115	46 (41, 51)

Agree or Strongly Agree					
Attitude	Total		Yes/Yes		Yes/No
	<i>n</i>	% (95% CI)	<i>n</i>	% (95% CI)	% (95% CI)
... that taking part could help my family.	2870	51 (48, 56)	1802	66 (61, 70)	31 (27, 36)
... that taking part could help my child	3189	58 (55, 61)	1982	76 (71, 80)	29 (25, 34)
Concerns about child participating in a biobank					
I would worry about my child's privacy.	3666	63 (60, 66)	1229	44 (40, 48)	80 (75, 85)
... about how researchers would use my child's health information.	3463	60 (55, 65)	1148	42 (38, 47)	72 (67, 77)
... that some research would be done that I did not want my child to take part in.	3062	53 (48, 58)	919	33 (28, 38)	73 (68, 78)

We dichotomized each item (1 = Strongly Agree or Agree, 0 = All other responses) and report the observed frequencies and ordered survey-adjusted percentages and 95% confidence intervals. Because we did not detect differences between data sharing and consent models on the construct level (Table 3), we only report overall summaries.

TABLE 5

Univariate and Multivariate Associations between Consent and Data Sharing Models, Socio-demographics, Trust and Privacy, and Attitudes, and Willingness for Youngest Child to Participate in a Biobank

Multivariable models, OR (95% CI)				
Independent variable	% (95% CI)	Socio-demographics	Socio-demographics, and, trust and privacy	Socio-demographics, trust and privacy, and attitudes
Consent and data sharing model				
Broad-controlled	52 (47, 57)	1	1	1
Broad-open	51 (47, 56)	0.95 (0.77-1.02)	0.89 (0.73-1.11)	0.89 (0.70-1.13)
Tiered-controlled	55 (50, 60)	1.13 (0.94-1.28)	1.00 (0.77-1.39)	1.00 (0.74-1.37)
Gender				
Female	48 (43, 53)	1	1	1
Male	52 (48, 57)	1.04 (0.84-1.24)	0.97 (0.80-1.20)	1.01 (0.81-1.26)
Age (Years)				
18-35	51 (46, 56)	1.11 (0.86-1.44)	0.98 (0.69-1.39)	1.03 (0.75-1.43)
36-50	50 (46, 55)	1	1	1
51-64	51 (46, 56)	1.05 (0.75-1.44)	0.99 (0.63-1.52)	0.91 (0.59-1.39)
65 or older	44 (39, 49)	1.14 (0.72-1.84)	0.91 (0.49-1.69)	0.89 (0.45-1.79)
Race				
White	57 (52, 62)	1	1	1
Black or African American	47 (42, 52)	0.64 (0.51-0.83)	0.73 (0.56-0.96)	0.81 (0.57-1.13)
Asian	50 (45, 54)	0.71 (0.54-1.03)	0.73 (0.58-0.92)	0.92 (0.65-1.13)
American Indian or Alaska Native	48 (43, 53)	0.72 (0.53-0.98)	0.80 (0.59-1.08)	0.95 (0.73-1.23)
Other *	48 (43, 53)	0.66 (0.49-0.94)	0.82 (0.46-1.44)	0.77 (0.61-1.61)
More than one race	59 (54, 64)	0.84 (0.54-1.28)	0.91 (0.60-1.35)	0.76 (0.65-1.48)
Ethnicity				
Hispanic or Latino/a	53 (48, 58)	1	1	1
Not Hispanic or Latino/a	48 (43, 53)	1.00 (0.75-1.33)	0.97 (0.63-1.49)	0.97 (0.64-1.37)
Educational attainment				
Up to some high school (grades 9-12)	48 (44, 53)	0.46 (0.33-0.64)	0.39 (0.27-0.56)	0.34 (0.20-0.53)
High school graduate or GED	50 (46, 55)	0.64 (0.43-0.94)	0.61 (0.39-0.91)	0.54 (0.33-0.86)
Some college	52 (48, 57)	0.76 (0.42-1.03)	0.77 (0.54-1.10)	0.61 (0.44-0.87)
Bachelor's degree or equivalent	57 (52, 62)	0.79 (0.57-1.10)	0.79 (0.55-1.14)	0.72 (0.50-1.03)
Master's degree or equivalent	52 (51, 57)	0.88 (0.68-1.15)	0.88 (0.72-1.15)	0.84 (0.59-1.19)
PhD, MD, JD, or equivalent	59 (54, 64)	1	1	1
Annual household income				
Less than \$30,000	49 (44, 54)	0.58 (0.43-0.79)	0.67 (0.47-0.98)	0.74 (0.48-1.13)
\$30,000 - \$59,999	51 (46, 56)	0.67 (0.44-0.98)	0.73 (0.53-0.99)	0.82 (0.58-1.15)
\$60,000 - \$89,999	51 (47, 56)	0.87 (0.62-1.19)	0.92 (0.65-1.31)	1.04 (0.70-1.50)
\$90,000 to \$149,999	55 (50, 60)	1.11 (0.82-1.49)	1.10 (0.84-1.45)	1.15 (0.83-1.62)
More than \$150,000	55 (51, 59)	1	1	1

Multivariable models, OR (95% CI)				
Independent variable	% (95% CI)	Socio-demographics	Socio-demographics, and trust and privacy	Socio-demographics, trust and privacy, and attitudes
Total number of people in household				
1	63 (58, 68)	2.37 (0.73-5.71)	2.96 (0.33-7.25)	2.22 (0.41-9.23)
2	48 (43, 53)	1.15 (0.91-1.47)	1.15 (0.83-1.60)	1.12 (0.79-1.57)
3	49 (4, 54)	1.17 (0.93-1.45)	1.26 (0.95-1.66)	1.24 (0.95-1.60)
4 or more	48 (43, 53)	1	1	1
Work situation				
Working	55 (50, 60)	1	1	1
Retired	56 (52, 61)	1.10 (0.84-1.47)	1.08 (0.80-1.49)	1.01 (0.66-1.38)
Disabled / Unemployed	50 (46, 59)	0.95 (0.74-1.22)	1.04 (0.81-1.34)	1.04 (0.74-1.45)
Other	50 (45, 54)	1.02 (0.79-1.32)	1.05 (0.69-1.62)	0.97 (0.68-1.40)
Healthcare insurance				
Private insurance	58 (53, 63)	1	1	1
Public insurance	55 (50, 60)	1.51 (1.08-2.12)	1.38 (1.00-1.91)	1.43 (0.93-2.21)
No insurance	51 (46, 56)	1.08 (0.66-1.73)	0.97 (0.58-1.62)	1.01 (0.64-1.92)
Rurality (from census-level data)				
Suburban / Urban	54 (48, 59)	1	1	1
Rural	52 (47, 56)	1.81 (1.09-2.99)	1.09 (1.02-3.26)	0.97 (0.82-3.40)
Marital status				
Married	57 (52, 62)	1	1	1
Not married, living with someone	60 (55, 65)	0.95 (0.80-1.11)	0.89 (0.81-1.13)	0.88 (0.79-1.30)
Not married, not living with someone	53 (48, 59)	1.03 (0.74-1.40)	0.80 (0.72-1.47)	0.65 (0.54-1.21)
Age of youngest child (Years)				
0-5	49 (45, 54)	0.96 (0.79, 1.10)	0.97 (0.82-1.13)	0.95 (0.84-1.08)
6-11	53 (48, 58)	1	1	1
12-17	57 (47, 62)	1.09 (0.91, 1.28)	1.10 (0.94-1.30)	1.12 (0.95-1.32)
Religiosity				
Not at all religious	52 (47, 57)	1	1	1
Not very religious	50 (46, 55)	0.94 (0.73-1.20)	0.75 (0.68-1.22)	0.64 (0.61-1.39)
Somewhat religious	44 (39, 49)	0.90 (0.69-1.18)	0.69 (0.65-1.15)	0.58 (0.48-1.92)
Very religious	45 (40, 49)	0.76 (0.61-0.95)	0.75 (0.58-0.98)	0.77 (0.48-0.88)
I trust my healthcare system				
Disagree / Strongly disagree	36 (29, 41)		0.91 (0.54-0.99)	1.08 (0.87-1.95)
Neither agree nor disagree	42 (37, 47)		0.18 (0.05-0.99)	0.53 (0.41-0.64)
Agree / Strongly Agree	65 (61, 70)		1	1
I trust medical researchers				
Disagree / Strongly disagree	35 (30, 40)		0.46 (0.33-1.14)	0.71 (0.60-1.52)
Neither agree nor disagree	40 (35, 45)		1.18 (0.80-1.21)	1.28 (0.82-1.37)
Agree / Strongly Agree	65 (60, 70)		1	1
Health information privacy is important to me		8.09		

Multivariable models, OR (95% CI)				
Independent variable	% (95% CI)	Socio-demographics	Socio-demographics, and trust and privacy	Socio-demographics, trust and privacy, and attitudes
Disagree / Strongly disagree	57 (52,62)	63.6	1.61 (1.26-2.42)	1.27 (1.17-1.45)
Neither agree nor disagree	57 (52,62)	.37	4.19 (2.77-5.21)	3.68 (3.38-3.86)
Agree / Strongly Agree	43 (38,48)	.72	1	1
I worry about the privacy of my health information				
Disagree / Strongly disagree	67 (63,72)		2.54 (1.08-3.32)	2.68 (1.19-3.47)
Neither agree nor disagree	57 (52, 62)		4.10 (1.82-6.17)	3.96 (1.68-6.03)
Agree / Strongly Agree	40 (35,45)		1	1
Benefits for self				
Low	4 (1, 9)			1
Intermediate	38 (33, 43)			6.83 (3.41-10.82)
High	76 (71, 81)			27.74 (13.71-55.79)
Concerns for self				
Low	61 (56, 66)			1
Intermediate	49 (44, 54)			0.34 (0.17-0.63)
High	19 (14, 23)			0.08 (0.05-0.17)
Information needs				
Low	61 (56, 66)			1
Intermediate	50 (45, 54)			1.39 (0.91-2.15)
High	42 (38, 46)			1.85 (1.16-2.54)
Benefits for youngest child				
Low	6 (2, 10)			1
Intermediate	47 (42, 51)			7.99 (3.64-12.15)
High	86 (81, 91)			61.37 (35.90-116.19)
Concerns for youngest child				
Low	69 (64, 74)			1
Intermediate	55 (51, 60)			0.39 (0.20-0.75)
High	24 (19, 291.0)			0.10 (0.04-0.19)

We transformed survey-adjusted logistic regression estimates from the univariate models to percentages (95% CI) and summarize multivariate models with odds ratios (95% CI).

* Due to small cell counts, we grouped Native Hawaiian/Pacific Islanders with the Other racial category.

Bold font denotes significant differences at the 0.05 level.

TABLE 6

Comparison between Non-parents and Parents Willingness to Participate, and between Willing and Unwilling Parents Willingness to Allow their Youngest Child to Participate by Consent and Data Sharing Model and Across Models

	Broad-controlled (BC)	Broad-open (BO)	Tiered-controlled (TC)	Total	p-value BC vs BO	p-value BC vs TC
Parents willing to participate (<i>n</i>)	1,261	1,183	1,209	3,653		
Willingness to have youngest child participate (<i>n</i> (%))	925 (73)	873 (74)	925 (77)	2723 (75)	0.638	0.293
Parents not willing to participate (<i>n</i>)	605	696	639	1,940		
Willingness to have youngest child participate (<i>n</i> (%))	57 (9)	63 (9)	71 (11)	191 (10)	0.818	0.484
p-value (Difference between Groups)	<0.001 *	<0.001 *	<0.001 *	<0.001 *		

Pearson's chi-squared tests were used to test whether parents' willingness to participate was correlated with their willingness to allow their child to participate.

Wald tests were performed to assess differences between data sharing models (BC vs BO) and between consent types (BC vs TC).

* Statistically significant

TABLE 7

Univariate and Multivariate Associations between Being Willing to Participate in a Biobank, Conditional on Being Willing to Let Your Youngest Child Participate, and Consent and Data Sharing Models, Socio-demographics, Trust and Privacy, and Attitudes, and Willingness for Youngest Child to Participate in a Biobank

Multivariable models, OR (95% CI)				
Independent variable	% (95% CI)	Socio-demographics	Socio-demographics, and trust and privacy	Socio-demographics, trust and privacy, and attitudes
Consent & data sharing model				
Broad-controlled	72 (67,77)	1	1	1
Broad-open	77 (72, 82)	1.07 (0.92-1.21)	1.09 (0.94-1.23)	1.10 (0.96-1.25)
Tiered-controlled	79 (74, 84)	0.91 (0.80-1.05)	0.91 (0.78-1.04)	0.92 (0.79-1.05)
Gender				
Female	77 (72,82)	1	1	1
Male	76 (71, 81)	0.97 (0.87-1.11)	0.96 (0.84-1.11)	1.01 (0.89-1.16)
Age (Years)				
18-35	79 (74,84)	0.92 (0.71-1.23)	0.94 (0.73-1.25)	0.98 (0.75-1.30)
36-50	75 (71,79)	1	1	1
51-64	72 (68,76)	0.88 (0.65-1.20)	0.98 (0.64-1.20)	1.00 (0.66-1.24)
65 or older	75 (71,79)	0.90 (0.62-1.48)	1.12 (0.61-1.50)	1.18 (0.68-1.36)
Race				
White	80 (75,85)	1	1	1
Black or African American	79 (73,84)	1.07 (0.93-1.21)	1.03 (0.95-1.13)	1.04 (0.96-1.14)
Asian	71 (66,76)	0.58 (0.54-0.62)	0.90 (0.84-0.97)	0.86 (0.79-0.93)
American Indian or Alaska Native	73 (68,78)	0.61 (0.54-0.70)	0.85 (0.78-0.91)	0.82 (0.76-0.89)
Other *	72 (68,77)	0.57 (0.50-0.63)	0.88 (0.79-0.99)	0.86 (0.77-0.97)
More than one race	79 (75,84)	1.07 (0.90-1.43)	1.07 (0.92-1.25)	1.09 (0.94-1.29)
Ethnicity				
Hispanic or Latino/a	71 (66,76)	1	1	1
Not Hispanic or Latino/a	74 (69,79)	0.97 (0.84-1.13)	0.99 (0.87-1.15)	1.02 (0.90-1.19)
Educational attainment				
Up to some high school (grades 9-12)	72 (67,76)	0.58 (0.52-0.66)	0.57 (0.51-0.66)	0.57 (0.52-0.67)
High school graduate or GED	73 (68,78)	0.65 (0.59-0.72)	0.67 (0.61-0.73)	0.65 (0.59-0.72)
Some college	82 (78,87)	0.68 (0.61-0.77)	0.66 (0.60-0.78)	0.68 (0.62-0.81)
Bachelor’s degree or equivalent	80 (75,84)	0.70 (0.64-0.77)	0.75 (0.69-0.82)	0.73 (0.67-0.79)
Master’s degree or equivalent	86 (82,91)	0.88 (0.79-1.00)	0.91 (0.81-1.05)	0.92 (0.82-1.06)
PhD, MD, JD, or equivalent	81 (76,87)	1	1	1
Annual household income				
Less than \$30,000	60 (55,65)	0.85 (0.78-0.94)	0.86 (0.79-0.88)	0.92 (0.83-1.00)
\$30,000 - \$59,999	79 (74,84)	0.86 (0.79-0.97)	0.87 (0.80-0.97)	0.88 (0.81-0.98)
\$60,000 - \$89,999	77 (72,82)	0.93 (0.84-1.04)	0.97 (0.88-1.09)	1.01 (0.90-1.12)

Multivariable models, OR (95% CI)				
Independent variable	% (95% CI)	Socio-demographics	Socio-demographics, and trust and privacy	Socio-demographics, trust and privacy, and attitudes
\$90,000 to \$149,999	75 (70,881)	0.97 (0.88-1.09)	1.01 (0.92-1.14)	1.04 (0.95-1.17)
More than \$150,000	83 (77,88)	1	1	1
Total number of people in household				
1	74 (69,79)	0.78 (0.63-1.15)	0.79 (0.64-1.18)	0.81 (0.66-1.21)
2	75 (70,80)	0.78 (0.63-1.09)	0.82 (0.66-1.15)	0.86 (0.69-1.20)
3	82 (77,87)	0.84 (0.70-1.06)	0.84 (0.71-1.07)	0.85 (0.73-1.08)
4 or more	77 (72,86)	1	1	1
Work situation				
Working	75 (70,81)	1	1	1
Retired	79 (74,83)	1.11 (0.96-1.27)	1.06 (0.91-1.20)	1.01 (0.87-1.15)
Disabled / Unemployed	75 (69,79)	1.05 (0.95-1.17)	1.02 (0.92-1.14)	0.99 (0.90-1.11)
Other	70 (65,75)	1.05 (0.94-1.21)	1.03 (0.92-1.18)	1.01 (0.90-1.15)
Healthcare insurance				
Private insurance	77 (72,82)	1	1	1
Public insurance	75 (70,79)	0.76 (0.64-0.95)	0.77 (0.65-0.96)	0.79 (0.66-0.98)
No insurance	71 (65,76)	0.78 (0.64-0.96)	0.80 (0.66-0.99)	0.83 (0.68-1.02)
Rurality (from census-level data)				
Suburban / Urban	74 (68,77)	1	1	1
Rural	72 (67,77)	0.92 (0.86-1.01)	0.93 (0.86-1.02)	0.94 (0.87-1.02)
Marital status				
Married	79 (74,84)	1	1	1
Not married, living with someone	79 (74,84)	1.16 (0.98-1.35)	1.13 (0.95-1.31)	1.11 (0.93-1.30)
Not married, not living with someone	75 (70,79)	0.88 (0.78-1.02)	0.91 (0.80-1.25)	0.93 (0.82-1.08)
Age of youngest child (Years)				
0-5	71 (66,76)	0.93 (0.77-1.20)	0.95 (0.79-1.21)	0.95 (0.81-1.20)
6-11	75 (70,80)	1	1	1
12-17	81 (76,86)	0.89 (0.74-1.16)	0.91 (0.76-1.22)	0.93 (0.78-1.23)
Religiosity				
Not at all religious	85 (80,89)	1	1	1
Not very religious	83 (78,88)	1.05 (0.91-1.22)	1.02 (0.89-1.19)	0.99 (0.86-1.16)
Somewhat religious	72 (67,77)	0.50 (0.47-0.54)	0.51 (0.48-0.54)	0.51 (0.49-0.55)
Very religious	75 (70,79)	0.52 (0.48-0.55)	0.55 (0.52-0.58)	0.58 (0.54-0.61)
I trust my healthcare system				
Disagree / Strongly disagree	33 (28,39)		0.64 (0.54-0.81)	0.65 (0.54-0.83)
Neither agree nor disagree	69 (64,74)		0.80 (0.65-1.05)	0.83 (0.67-1.09)
Agree / Strongly Agree	80 (75,85)		1	1
I trust medical researchers				
Disagree / Strongly disagree	41 (36,46)		0.36 (0.20-0.67)	0.38 (0.20-0.69)
Neither agree nor disagree	66 (61,71)		0.67 (0.54-0.88)	0.67 (0.53-0.87)

Multivariable models, OR (95% CI)				
Independent variable	% (95% CI)	Socio-demographics	Socio-demographics, and trust and privacy	Socio-demographics, trust and privacy, and attitudes
Agree / Strongly Agree	87 (82,92)		1	1
Health information privacy is important to me				
Disagree / Strongly disagree	65 (60,70)		0.82 (0.69-0.99)	0.80 (0.67-0.97)
Neither agree nor disagree	77 (72,82)		1.04 (0.92-1.20)	1.02 (0.90-1.17)
Agree / Strongly Agree	79 (74,84)		1	1
I worry about the privacy of my health information				
Disagree / Strongly disagree	55 (50,60)		0.64 (0.37-1.06)	0.65 (0.38-1.08)
Neither agree nor disagree	77 (72,82)		0.96 (0.84-1.12)	0.97 (0.85-1.12)
Agree / Strongly Agree	79 (74,84)		1	1
Perceived benefits for self				
Low	68 (63,73)			1
Intermediate	72 (68,77)			1.22 (0.98-1.60)
High	78 (74,82)			1.38 (1.09-1.60)
Concerns for self				
Low	81 (76,86)			1
Intermediate	75 (71,82)			0.68 (0.56-0.85)
High	68 (63,73)			0.67 (0.56-0.75)
Information needs				
Low	70 (65,75)			1
Intermediate	75 (71,79)			1.19 (0.96-1.55)
High	81 (76,86)			1.31 (1.03-1.52)
Perceived benefits for youngest child				
Low	71 (66,76)			1
Intermediate	76 (71,81)			1.15 (0.92-1.33)
High	82 (77,87)			1.16 (0.93-1.38)
Concerns for youngest child				
Low	86 (81,91)			1
Intermediate	76 (71,81)			0.58 (0.42-0.74)
High	67 (62,72)			0.51 (0.37-0.65)

We transformed survey-adjusted logistic regression estimates from the univariate models to percentages (95% CI) and summarize multivariate models with odds ratios (95% CI).

* Due to small cell counts, we grouped Native Hawaiian/Pacific Islanders with the Other racial category.

Bold font denotes significant differences at the 0.05 level.