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Motivations for data sharing – views of research participants from 4 European countries: A DIRECT study

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Conflict of Interest

The authors declare no conflicts of interest.

Abstract

The purpose of this study was to explore and compare different countries in what motivated research participants' decisions whether to share their de-identified data. We investigated European DIRECT (Diabetes Research on Patient Stratification) research project participants' desire for control over sharing different types of their de-identified data, and with who data could be shared in the future after the project ends. A cross-sectional survey was disseminated among DIRECT project participants. The results found that there was a significant association between country and attitudes towards advancing research, protecting privacy, and beliefs about risks and benefits to sharing data. When given the choice to have control, some participants (less than 50% overall) indicated that having control over what data is shared and with whom was important; and control over what data types are shared was less important than respondents deciding who data are shared with. Danish respondents indicated higher odds of desire to control data types shared, and Dutch respondents showed higher odds of desire to control who data will be shared with. Overall, what research participants expect in terms of control over data sharing needs to be considered and aligned with sharing for future research and re-use of data. Our findings show that even with de-identified data, respondents prioritise privacy above all else. This study argues to move research participants from passive participation in biomedical research to considering their opinions about data sharing and control of de-identified biomedical data.

Keywords: *ethics; participant engagement; data sharing; data stewardship; genetic research*

Introduction

International research consortia in the field of biomedicine collect large amounts of information consisting of different data types from participants that are often located in different countries. A key tenet that facilitates ongoing and future research is data sharing. Data sharing is viewed as good practice for advancing biomedical research, as it maximises the use of biological samples and other types of data, reduces participant burden, and stockpiling and pooling data helps to improve statistical power of research

{ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1016/j.ajhg.2009.10.002","abstract":"Concerns about privacy may deter people from participating in genetic research. Recruitment and retention of biobank participants requires understanding the nature and magnitude of these concerns. Potential participants in a proposed biobank were asked about their willingness to participate, their privacy concerns, informed consent, and data sharing. A representative survey of 4659 U.S. adults was conducted. Ninety percent of respondents would be concerned about privacy, 56% would be concerned about researchers having their information, and 37% would worry that study data could be used against them. However, 60% would participate in the biobank if asked. Nearly half (48%) would prefer to provide consent once for all research approved by an oversight panel, whereas 42% would prefer to provide consent for each project separately. Although 92% would allow academic researchers to use study data, 80% and 75%, respectively, would grant access to government and industry researchers. Concern about privacy was related to lower willingness to participate only when respondents were told that they would receive $50 for participation and would not receive individual research results back. Among respondents who were told that they would receive $200 or individual research results, privacy concerns were not related to willingness. Survey respondents valued both privacy and participation in biomedical research. Despite pervasive privacy concerns, 60% would participate in a biobank. Assuring research participants that their privacy will be protected to the best of researchers' abilities may increase participants' acceptance of consent for broad research uses of biobank data by a wide range of researchers.","author":[{"dropping-particle":"","family":"Kaufman","given":"David J","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Murphy-Bollinger","given":"Juli","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Scott","given":"Joan","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hudson","given":"Kathy L","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"American journal of human genetics","id":"ITEM-1","issue":"5","issued":{"date-parts":[["2009","11"]]},"language":"eng","page":"643-654","title":"Public opinion about the importance of privacy in biobank research.","type":"article-journal","volume":"85"},"uris":["http://www.mendeley.com/documents/?uuid=f47d8554-6ed8-3b81-bd8d-6c35ea4fa9d4"]},{"id":"ITEM-2","itemData":{"DOI":"10.1097/GIM.0b013e31818bb3ab","ISSN":"1530-0366 (Electronic)\\n1530-0366 (Linking)","abstract":"PURPOSE: Cohort studies investigating genes, environment, and lifestyle require large study populations. To recruit and retain participants, it is important to understand the relative significance of influences on people's motivation to participate. To this end, 4659 Americans were surveyed about support for and willingness to participate in a proposed large cohort study. METHODS: An online survey of US adults was conducted between December 2007 and January 2008. To measure the influence of study burden, compensation and receipt of individual research results on willingness to participate, respondents were randomized to one of eight different study scenarios. RESULTS: Most respondents (84%) supported the study, and 60% would participate. Returning research results (odds ratio = 1.6, 95% confidence interval 1.3-1.8) and increasing compensation from $50 to $200 (odds ratio = 1.5, 95% confidence interval 1.2-1.7) were associated with increased willingness to participate. Decreasing study burden was less important (odds ratio = 1.2, 95% confidence interval 1.0-1.4). Three in four respondents would be less likely to participate without the return of research results. Support and willingness varied little among demographic groups; variation in influences of the three factors on willingness was observed. CONCLUSION: Widespread support exists in the general public for a large national cohort study. Providing individual research results is a strong motivation to participate; compensating participants $200 may increase participation a similar amount. Incentives, recruitment, and return of results could be tailored to demographics groups' interests.","author":[{"dropping-particle":"","family":"Kaufman","given":"David","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Murphy","given":"Juli","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Scott","given":"Joan","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hudson","given":"Kathy","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Genetics in medicine : official journal of the American College of Medical Genetics","id":"ITEM-2","issue":"11","issued":{"date-parts":[["2008"]]},"page":"831-839","title":"Subjects matter: a survey of public opinions about a large genetic cohort study.","type":"article-journal","volume":"10"},"uris":["http://www.mendeley.com/documents/?uuid=da75dfdd-8e38-367b-9caa-4d2facdb6649"]},{"id":"ITEM-3","itemData":{"abstract":"PURPOSE: Sharing study data within the research community generates tension between two important goods: promoting scientific goals and protecting the privacy interests of study participants. This study was designed to explore the perceptions, beliefs, and attitudes of research participants and possible future participants regarding genome-wide association studies and repository-based research., METHODS: Focus group sessions with (1) current research participants, (2) surrogate decision-makers, and (3) three age-defined cohorts (18-34 years, 35-50, >50)., RESULTS: Participants expressed a variety of opinions about the acceptability of wide sharing of genetic and phenotypic information for research purposes through large, publicly accessible data repositories. Most believed that making de-identified study data available to the research community is a social good that should be pursued. Privacy and confidentiality concerns were common, although they would not necessarily preclude participation. Many participants voiced reservations about sharing data with for-profit organizations., CONCLUSIONS: Trust is central in participants' views regarding data sharing. Further research is needed to develop governance models that enact the values of stewardship.","author":[{"dropping-particle":"","family":"Trinidad","given":"Susan Brown","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Fullerton","given":"Stephanie M","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Bares","given":"Julie M","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Jarvik","given":"Gail

P","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Larson","given":"Eric B","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Burke","given":"Wylie","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Genetics in medicine : official journal of the American College of Medical Genetics","id":"ITEM-3","issue":"8","issued":{"date-parts":[["2010"]]},"page":"486-495","publisher":"Trinidad,Susan Brown. Department of Bioethics & Humanities, University of Washington, Seattle, WA 98195-7120, USA. sbtrini@uw.edu","publisher-place":"United States","title":"Genomic research and wide data sharing: views of prospective participants.","type":"article-journal","volume":"12"},"uris":["http://www.mendeley.com/documents/?uuid=c7443a0f-6e5d-39e8-a2a8-52bacaccb3fb"]}],"mendeley":{"formattedCitation":"^{1–3}","plainTextFormattedCitation":"1–3","previouslyFormattedCitation":"^{1–3}"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}**}**. Data sharing within research consortia and externally is encouraged and is increasingly being adopted and enabled through advanced storing and sharing technologies. Supported by research data governance strategies, the optimisation of data sharing has been an important focus for the Open Science Agenda**{**ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"abstract":"The parts of the Work Programme that relate to 2019 (topics, dates, budget) have, with this revised version, been updated. The changes relating to this revised part are explained on the Participant Portal. The parts that relate to 2020 are provided at this stage on an indicative basis. Such Work Programme parts will be decided during 2019.","author":[{"dropping-particle":"","family":"Commission","given":"European","non-dropping-particle":"","parse-names":false,"suffix":""}],"id":"ITEM-1","issued":{"date-parts":[["2018"]]},"title":"Horizon 2020 Work Programme 2018-2020, 16. Science with and for Society","type":"report"},"uris":["http://www.mendeley.com/documents/?uuid=4d1f2a71-af24-3a07-96b7-3fdfd671c44a"]}],"mendeley":{"formattedCitation":"⁴","plainTextFormattedCitation":"4","previouslyFormattedCitation":"⁴"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}**}**.

Empirical research to date has focused on three key areas: willingness of research participants and the public to share their data**{**ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1038/ejhg.2012.104","ISSN":"1018-4813","abstract":"Publics and biobanks: Pan-European diversity and the challenge of responsible innovation","author":[{"dropping-particle":"","family":"Gaskell","given":"George","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Gottweis","given":"Herbert","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Starkbaum","given":"Johannes","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Gerber","given":"Monica M","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Broerse","given":"Jacqueline","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Gottweis","given":"Ursula","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hobbs","given":"Abbi","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Helén","given":"Ilpo","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Paschou","given":"Maria","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Snell","given":"Karoliina","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Soulier","given":"Alexandra","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"European Journal of Human Genetics","id":"ITEM-1","issue":"1","issued":{"date-parts":[["2013","1","6"]]},"page":"14-20","publisher":"Nature Publishing Group","title":"Publics and biobanks: Pan-European diversity and the challenge of responsible innovation","type":"article-journal","volume":"21"},"uris":["http://www.mendeley.com/documents/?uuid=5888627e-48e0-395a-9ad4-f6c6945c0674"]},{"id":"ITEM-2","itemData":{"DOI":"10.1007/s12687-013-0146-0","ISSN":"1868-310X","abstract":"Population-based biobanks are a critical resource for genetic research. It is important to know what potential participants understand about the risks and benefits of providing samples in order to ensure adequate informed consent. Kaiser Permanente Colorado (KPCO) is currently planning a biobank where adult members would be asked to contribute an additional tube of blood during a routine blood draw. Adult KPCO members in clinic waiting rooms were asked to read an informational brochure and informed consent form. Respondents then completed a survey to evaluate their understanding of the materials, willingness to provide a blood sample to a biobank, and facilitators and barriers to participation. Two hundred three members participated in the survey, of whom 69 % indicated willingness to contribute to a biobank. Nearly all understood that they would not be paid for any products resulting from the use of their blood and would not receive results from their samples (91 and 84 %, respectively). Seventy-four percent would donate a sample because, \"it is important to contribute to research,\" and over half the participants (56 %) said they had no concerns about contributing to a biobank. Of those with concerns, 35 % said information security was a reason. In multivariate models, older age and trust in KPCO were significant predictors of willingness to participate (p = 0.03 and p < 0.0001, respectively). Data from this survey indicate an overall willingness to participate in a biobank, provide possible barriers to participation, and identify ways to improve informational materials to ensure adequate informed consent.","author":[{"dropping-particle":"","family":"Rahm","given":"Alanna Kulchak","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Wrenn","given":"Michelle","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Carroll","given":"Nikki M","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Feigelson","given":"Heather Spencer","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Journal of Community Genetics","id":"ITEM-2","issue":"4","issued":{"date-parts":[["2013"]]},"page":"445-

450","title":"Biobanking for research: A survey of patient population attitudes and understanding","type":"article-journal","volume":"4"},"uris":["http://www.mendeley.com/documents/?uuid=7801cd47-477d-329e-98ed-195c7120772c"]},{"id":"ITEM-3","itemData":{"DOI":"10.1159/000276767","ISSN":"1662-4246","abstract":"Research assessing attitudes toward consent processes for high-throughput genomic-wide technologies and widespread sharing of data is limited. In order to develop a better understanding of stakeholder views toward these issues, this cross-sectional study assessed public and biorepository participant attitudes toward research participation and sharing of genetic research data. Forty-nine individuals participated in 6 focus groups; 28 in 3 public focus groups and 21 in 3 NUgene biorepository participant focus groups. In the public focus groups, 75% of participants were women, 75% had some college education or more, 46% were African-American and 29% were Hispanic. In the NUgene focus groups, 67% of participants were women, 95% had some college education or more, and the majority (76%) of participants was Caucasian. Five major themes were identified in the focus group data: (a) a wide spectrum of understanding of genetic research; (b) pros and cons of participation in genetic research; (c) influence of credibility and trust of the research institution; (d) concerns about sharing genetic research data and need for transparency in the Policy for Sharing of Data in National Institutes of Health-Supported or Conducted Genome-Wide Association Studies; (e) a need for more information and education about genetic research. In order to increase public understanding and address potential concerns about genetic research, future efforts should be aimed at involving the public in genetic research policy development and in identifying or developing appropriate educational strategies to meet the public's needs.","author":[{"dropping-particle":"","family":"Lemke","given":"A A","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Wolf","given":"W A","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hebert-Beirne","given":"J","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Smith","given":"M E","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Public Health Genomics","id":"ITEM-3","issue":"6","issued":{"date-parts":[["2010"]]},"language":"eng","page":"368-377","title":"Public and biobank participant attitudes toward genetic research participation and data sharing","type":"article-journal","volume":"13"},"uris":["http://www.mendeley.com/documents/?uuid=7edacfe1-ef8a-3320-9240-3dfd41b37094"]},{"id":"ITEM-4","itemData":{"DOI":"10.1186/s12910-015-0053-5","ISSN":"1472-6939","abstract":"Appropriate information and consent has been one of the most intensely discussed topics within the context of biobank research. In parallel to the normative debate, many socio-empirical studies have been conducted to gather experiences, preferences and views of patients, healthy research participants and further stakeholders. However, there is scarcity of literature which connects the normative debate about justifications for different consent models with findings gained in empirical research. In this paper we discuss findings of a limited review of socio-empirical research on patients’ and healthy research participants’ experiences and views regarding consent to biobank research in light of ethical principles for appropriate information and consent. Review question: Which empirical data are available on research participants’ perceptions and views regarding information and elicitation of consent for biobank research? Search of articles published till March 1st 2014 in Pubmed. Review of abstracts and potentially relevant full text articles by two authors independently. As categories for content analysis we defined (i) understanding or recall of information, (ii) preferences regarding information or consent, and (iii) research participants’ concerns. The search in Pubmed yielded 337 abstracts of which 10 articles were included in this study. Approaches to information and consent varied considerably across the selected studies. The majority of research participants opted for some version of limited consent when being informed about such possibility. Among the factors influencing the type of preferred consent were information about sponsoring of biobank research by pharmaceutical industry and participants’ trade-off between privacy and perceived utility. Studies investigating research participants’ understanding and recall regarding the consent procedure indicated considerable lack of both aspects. Research participants’ perceptions of benefits and harms differ across those studies. The knowledge, perceptions and views of research participants who have undergone a consent procedure within the context of biobank research raise several questions on the issue of how to inform and elicit consent in an ethically acceptable way. In our empirical-ethical analysis we develop suggestions on how the practice of eliciting consent in the biobank context should be improved.","author":[{"dropping-particle":"","family":"D’Abramo","given":"Flavio","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Schildmann","given":"Jan","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Vollmann","given":"Jochen","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"BMC Medical Ethics","id":"ITEM-4","issue":"1","issued":{"date-parts":[["2015","12","9"]]},"page":"60","publisher":"BioMed Central","title":"Research participants’ perceptions and views on consent for biobank research: a review of empirical data and ethical analysis","type":"article-journal","volume":"16"},"uris":["http://www.mendeley.com/documents/?uuid=dbaa1e8d-e81e-3a16-990b-b5757b043e82"]}],"mendeley":{"formattedCitation":"^{5–8}","plainTextFormattedCitation":"5–8","previouslyFormattedCitation":"^{5–8}"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}**}**; how to deliver broad informed consent to enable the sharing of data**{**ADDIN CSL_CITATION

{"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1016/J.AJHG.2017.01.021","ISSN":"0002-9297","abstract":"Individuals participating in biobanks and other large research projects are increasingly asked to provide broad consent for open-ended research use and widespread sharing of their biosamples and data. We assessed willingness to participate in a biobank using different consent and data sharing models, hypothesizing that willingness would be higher under more restrictive scenarios. Perceived benefits, concerns, and information needs were also assessed. In this experimental survey, individuals from 11 US healthcare systems in the Electronic Medical Records and Genomics (eMERGE) Network were randomly allocated to one of three hypothetical scenarios: tiered consent and controlled data sharing; broad consent and controlled data sharing; or broad consent and open data sharing. Of 82,328 eligible individuals, exactly 13,000 (15.8%) completed the survey. Overall, 66% (95% CI: 63%–69%) of population-weighted respondents stated they would be willing to participate in a biobank; willingness and attitudes did not differ between respondents in the three scenarios. Willingness to participate was associated with self-identified white race, higher educational attainment, lower religiosity, perceiving more research benefits, fewer concerns, and fewer information needs. Most (86%, CI: 84%–87%) participants would want to know what would happen if a researcher misused their health information; fewer (51%, CI: 47%–55%) would worry about their privacy. The concern that the use of broad consent and open data sharing could adversely affect participant recruitment is not supported by these findings. Addressing potential participants' concerns and information needs and building trust and relationships with communities may increase acceptance of broad consent and wide data sharing in biobank research.","author":[{"dropping-particle":"","family":"Sanderson","given":"Saskia C.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Brothers","given":"Kyle B.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Mercaldo","given":"Nathaniel D.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Clayton","given":"Ellen Wright","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Antommaria","given":"Armand H. Matheny","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Aufox","given":"Sharon A.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Brilliant","given":"Murray H.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Campos","given":"Diego","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Carrell","given":"David S.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Connolly","given":"John","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Conway","given":"Pat","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Fullerton","given":"Stephanie M.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Garrison","given":"Nanibaa’ A.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Horowitz","given":"Carol R.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Jarvik","given":"Gail P.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Kaufman","given":"David","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Kitchner","given":"Terrie E.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Li","given":"Rongling","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Ludman","given":"Evette J.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"McCarty","given":"Catherine A.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"McCormick","given":"Jennifer B.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"McManus","given":"Valerie D.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Myers","given":"Melanie F.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Scrol","given":"Aaron","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Williams","given":"Janet L.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Shrubsole","given":"Martha J.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Schildcrout","given":"Jonathan S.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Smith","given":"Maureen E.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Holm","given":"Ingrid A.","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"The American Journal of Human Genetics","id":"ITEM-1","issue":"3","issued":{"date-parts":[["2017","3","2"]]},"page":"414-427","publisher":"Cell Press","title":"Public Attitudes toward Consent and Data Sharing in Biobank Research: A Large Multi-site Experimental Survey in the US","type":"article-journal","volume":"100"},"uris":["http://www.mendeley.com/documents/?uuid=5481930d-5886-3767-a242-eed00e1b60a7"]},{"id":"ITEM-2","itemData":{"DOI":"10.1089/bio.2014.0032","abstract":"Purpose: This study examined acceptability of two biobank consent models and evaluated the impact of beliefs about privacy and genetic safeguards on acceptance. Methods: U.S. adults surveyed online in English and Spanish were randomly assigned to one of two scenarios examining acceptance of broad consent (n = 1528), or narrow consent (n = 1533). Results: Overall, willingness to provide broad (76%) and narrow (74%) consents were similar. African Americans were as likely as white non-Hispanics to accept narrow consent (72% vs. 77%, p = 0.35) but significantly less likely to accept broad consent (69% vs. 81%, p = 0.004). Education, insurance, and blood donation history were also related to acceptance. Adjusting for beliefs about privacy and policy protections (Genetic Information Nondiscrimination Act, GINA), the effects of the variables were reduced. Respondents who drew comfort from GINA were more likely to support both consent (both p < 0.001); those who believed it is impossible to maintain privacy were less likely to find both broad (p = 0.04) and narrow models acceptable (p = 0.02). Conclusions: Choice of consent model matters when engaging diverse populations in biobank research. Beliefs underlying concerns about privacy and genetic protections should be considered when constructing biobank protocols.","author":[{"dropping-particle":"","family":"Ewing","given":"Altovise T","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Erby","given":"Lori A H","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Bollinger","given":"Juli","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Tetteyfio","given":"Eva","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Ricks-Santi","given":"Luisel J","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Kaufman","given":"David","non-dropping-particle":"","parse-names":false,"suffix":""}],"id":"ITEM-2","issued":{"date-parts":[["0"]]},"title":"Demographic Differences in Willingness to Provide Broad and Narrow Consent for Biobank Research","type":"article-journal"},"uris":["http://www.mendeley.com/documents/?uuid=8c2fc8fc-fd12-30cc-8e54-c3ace9a751cb"]},{"id":"ITEM-3","itemData":{"DOI":"10.1097/GIM.0b013e3182227589","abstract":"PURPOSE: Despite growing concerns toward maintaining participants' privacy, individual investigators collecting tissue and other biological specimens for genomic analysis are encouraged to obtain informed consent for broad data sharing. Our purpose was to assess the effect on research enrollment and data sharing decisions of three different consent types (traditional, binary, or tiered) with varying levels of control and choices regarding data sharing., METHODS: A single-blinded, randomized controlled trial was conducted with 323 eligible adult participants being recruited into one of six genome studies at Baylor College of Medicine in Houston, Texas, between January 2008 and August 2009. Participants were randomly assigned to one of three experimental consent documents (traditional, n = 110; binary, n = 103; and tiered, n = 110). Debriefing in follow-up visits provided participants a detailed review of all consent types and the chance to change data sharing choices or decline genome study participation., RESULTS: Before debriefing, 83.9% of participants chose public data release. After debriefing, 53.1% chose public data release, 33.1% chose restricted (controlled access database) release, and 13.7% opted out of data sharing. Only one participant declined genome study participation due to data sharing concerns., CONCLUSION: Our findings indicate that most participants are willing to publicly release their genomic data; however, a significant portion prefers restricted release. These results suggest discordance between existing data sharing policies and participants' judgments and desires.","author":[{"dropping-particle":"","family":"McGuire","given":"Amy L","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Oliver","given":"Jill M","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Slashinski","given":"Melody J","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Graves","given":"Jennifer L","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Wang","given":"Tao","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Kelly","given":"P Adam","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Fisher","given":"William","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Lau","given":"Ching C","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Goss","given":"John","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Okcu","given":"Mehmet","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Treadwell-Deering","given":"Diane","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Goldman","given":"Alica M","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Noebels","given":"Jeffrey L","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hilsenbeck","given":"Susan G","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Genetics in medicine : official journal of the American College of Medical Genetics","id":"ITEM-3","issue":"11","issued":{"date-parts":[["2011","11"]]},"page":"948-955","publisher":"McGuire,Amy L. Center for Medical Ethics and Health Policy, Baylor College of Medicine, Houston, TX 77030, USA. amcguire@bcm.edu","publisher-place":"United States","title":"To share or not to share: a randomized trial of consent for data sharing in genome research.","type":"article-journal","volume":"13"},"uris":["http://www.mendeley.com/documents/?uuid=41622375-a686-30c9-9cad-f425c1a24015"]},{"id":"ITEM-4","itemData":{"DOI":"10.1097/GIM.0b013e31815f1e00.DNA","author":[{"dropping-particle":"","family":"McGuire","given":"a.L.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hamilton","given":"J.a.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Lunstroth","given":"R","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"McCullough","given":"L B","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Goldman","given":"a.","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Genetics in medicine: official journal of the American College of Medical Genetics","id":"ITEM-4","issue":"1","issued":{"date-parts":[["2008"]]},"page":"46","title":"DNA data sharing : Research participants' perspectives","type":"article-journal","volume":"10"},"uris":["http://www.mendeley.com/documents/?uuid=95543308-c584-38a5-ae0c-537f5a9e54e9"]}],"mendeley":{"formattedCitation":"<sup>9–

12</sup>","plainTextFormattedCitation":"9–12","previouslyFormattedCitation":"^{9–12}"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}**}**; and patients’ happiness to share their own clinical data for research purposes**{**ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1186/s12910-016-0130-4","ISSN":"1472-6939","abstract":"Personal health information and biospecimens are valuable research resources essential for the advancement of medicine and protected by national standards and provincial statutes. Research ethics and privacy standards attempt to balance individual interests with societal interests. However these standards may not reflect public opinion or preferences. The purpose of this study was to assess the opinions and preferences of patients with kidney disease about the use of their health information and biospecimens for medical research. A 45-item survey was distributed to a convenience sample of patients at an outpatient clinic in a large urban centre. The survey briefly addressed sociodemographic and illness characteristics. Opinions were sought on the research use of health information and biospecimens including consent preferences. Two hundred eleven of 400 distributed surveys were completed (response rate 52.8 %). Respondents were generally supportive of medical research and trusting of researchers. Many respondents supported the use of their information and biospecimens for health research and also preferred consent be sought for use of health information and biospecimens. Some supported the use of their information and biospecimens for research without consent. There were significant differences in the opinions people offered regarding the research use of biospecimens compared to health information. Some respondent perspectives about consent were at odds with current regulatory and legal standards. Clinical health data and biospecimens are valuable research resources, critical to the advancement of medicine. Use of these data for research requires balancing respect for individual autonomy, privacy and the societal interest in the greater good. Incongruence between some respondent perspectives and the regulatory standards suggest both a need for public education and review of legislation to increase understanding and ensure the public’s trust is maintained.","author":[{"dropping-particle":"","family":"Page","given":"Stacey A.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Manhas","given":"Kiran Pohar","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Muruve","given":"Daniel A.","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"BMC Medical Ethics","id":"ITEM-1","issue":"1","issued":{"date-parts":[["2016","12","15"]]},"page":"48","publisher":"BioMed Central","title":"A survey of patient perspectives on the research use of health information and biospecimens","type":"article-journal","volume":"17"},"uris":["http://www.mendeley.com/documents/?uuid=d575c87d-523e-3b86-b647-544a4b8f6768"]},{"id":"ITEM-2","itemData":{"DOI":"10.1016/j.ijmedinf.2015.01.008","ISSN":"1386-5056","abstract":"The development of Electronic Health Records (EHRs) forms an integral part of the information strategy for the National Health Service (NHS) in the UK, with the aim of facilitating health information exchange for patient care and secondary use, including research and healthcare planning. Implementing EHR systems requires an understanding of patient expectations for consent mechanisms and

consideration of public awareness towards information sharing as might be made possible through integrated EHRs across primary and secondary health providers. Objectives: To explore levels of public awareness about EHRs and to examine attitudes towards different consent models with respect to sharing identifiable and de-identified records for healthcare provision, research and planning. Methods: A cross-sectional questionnaire survey was administered to adult patients and members of the public in primary and secondary care clinics in West London, UK in 2011. In total, 5331 individuals participated in the survey, and 3157 were included in the final analysis. Results: The majority (91%) of respondents expected to be explicitly asked for consent for their identifiable records to be accessed for health provision, research or planning. Half the respondents (49%) did not expect to be asked for consent before their de-identified records were accessed. Compared with White British respondents, those from all other ethnic groups were more likely to anticipate their permission would be obtained before their de-identified records were used. Of the study population, 59% reported already being aware of EHRs before the survey. Older respondents and individuals with complex patterns of interaction with healthcare services were more likely to report prior awareness of EHRs. Individuals self-identifying as belonging to ethnic groups other than White British, and those with lower educational qualifications were less likely to report being aware of EHRs than White British respondents and respondents with degree-level education, respectively. Those who reported being aware of EHRs were less likely to say they expected explicit consent to be sought before use of their de-identified record. Conclusions: A large number of patients remain unaware of EHRs, while preference for implicit consent is stronger among those who report previous awareness. Differences in awareness levels and consent expectations between

groups with different socio-demographic characteristics suggest that public education…","author":[{"dropping-particle":"","family":"Riordan","given":"Fiona","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Papoutsi","given":"Chrysanthi","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Reed","given":"Julie E","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Marston","given":"Cicely","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Bell","given":"Derek","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Majeed","given":"Azeem","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"International Journal of Medical Informatics","id":"ITEM-2","issue":"4","issued":{"date-parts":[["2015"]]},"page":"237-247","publisher":"Elsevier Ireland Ltd","title":"Patient and public attitudes towards informed consent models and levels of awareness of Electronic Health Records in the UK","type":"article-journal","volume":"84"},"uris":["http://www.mendeley.com/documents/?uuid=246ff818-1af8-331a-bead-ba5ddd374267"]},{"id":"ITEM-3","itemData":{"DOI":"10.1093/jamia/ocv014","abstract":"New models of healthcare delivery such as accountable care organizations and patient-centered medical homes seek to improve quality, access, and cost. They rely on a robust, secure technology infrastructure provided by health information exchanges (HIEs) and distributed research networks and the willingness of patients to share their data. There are few large, in-depth studies of US consumers' views on privacy, security, and consent in electronic data sharing for healthcare and research together. Objective This paper addresses this gap, reporting on a survey which asks about California consumers' views of data sharing for healthcare and research together. Materials and Methods The survey conducted was a representative, random-digit dial telephone survey of 800 Californians, per-formed in Spanish and English. Results There is a great deal of concern that HIEs will worsen privacy (40.3%) and security (42.5%). Consumers are in favor of electronic data sharing but elements of transparency are important: individual control, who has access, and the purpose for use of data. Respondents were more likely to agree to share deidentified information for research than to share identified information for healthcare (76.2% vs 57.3%, p < .001). Discussion While consumers show willingness to share health information electronically, they value individual control and privacy. Responsiveness to these needs, rather than mere reliance on Health Insurance Portability and Accountability Act (HIPAA), may im-prove support of data networks. Conclusion Responsiveness to the public's concerns regarding their health information is a pre-requisite for patient-centeredness. This is one of the first in-depth studies of attitudes about electronic data sharing that compares attitudes of the same individual to-wards healthcare and research.","author":[{"dropping-particle":"","family":"Kim","given":"Katherine K","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Joseph","given":"Jill G","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Ohno-Machado","given":"Lucila","non-dropping-particle":"","parse-names":false,"suffix":""}],"id":"ITEM-3","issued":{"date-parts":[["0"]]},"title":"Comparison of consumers' views on electronic data sharing for healthcare and research","type":"article-journal"},"uris":["http://www.mendeley.com/documents/?uuid=f6c44b13-061b-31c6-ba0c-dc26a3beb119"]},{"id":"ITEM-4","itemData":{"DOI":"10.1136/bmjopen-2013-003056","ISSN":"2044-6055","PMID":"23929915","abstract":"OBJECTIVE A mixed methods study exploring the UK general public's willingness to donate human biosamples (HBSs) for biomedical research. SETTING Cross-sectional focus groups followed by an online survey. PARTICIPANTS Twelve focus groups (81 participants) selectively sampled to reflect a range of demographic groups; 1110 survey responders recruited through a stratified sampling method with quotas set on sex, age, geographical location, socioeconomic group and ethnicity. MAIN OUTCOME MEASURES (1) Identify participants' willingness to donate HBSs for biomedical research, (2) explore acceptability towards donating different types of HBSs in various settings and (3) explore preferences regarding use and access to HBSs. RESULTS 87% of survey participants thought donation of HBSs was important and 75% wanted to be asked to donate in general. Responders who self-reported having some or good knowledge of the medical research process were significantly more likely to want to donate (p<0.001). Reasons why focus group participants saw donation as important included: it was a good way of reciprocating for the medical treatment received; it was an important way of developing drugs and treatments; residual tissue would otherwise go to waste and they or their family members might benefit. The most controversial types of HBSs to donate included: brain post mortem (29% would donate), eyes post mortem (35%), embryos (44%), spare eggs (48%) and sperm (58%). Regarding the use of samples, there were concerns over animal research (34%), research conducted outside the UK

(35%), and research conducted by pharmaceutical companies (56%), although education and discussion were found to alleviate such concerns. CONCLUSIONS There is a high level of public support and willingness to donate HBSs for biomedical research. Underlying concerns exist regarding the use of certain types of HBSs and conditions under which they are used. Improved education and more controlled forms of consent for sensitive samples may mitigate such concerns.","author":[{"dropping-particle":"","family":"Lewis","given":"Celine","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Clotworthy","given":"Margaret","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hilton","given":"Shona","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Magee","given":"Caroline","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Robertson","given":"Mark J","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Stubbins","given":"Lesley J","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Corfield","given":"Julie","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"BMJ open","id":"ITEM-4","issue":"8","issued":{"date-parts":[["2013","8","7"]]},"page":"e003056","publisher":"British Medical Journal Publishing Group","title":"Public views on the donation and use of human biological samples in biomedical research: a mixed methods study.","type":"article-journal","volume":"3"},"uris":["http://www.mendeley.com/documents/?uuid=635cbb99

-5098-3913-bd95-4b903bd62b3d"]}],"mendeley":{"formattedCitation":"^{13–16}","plainTextFormattedCitation":"13–16","previouslyFormattedCitation":"^{13–16}"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}**}**. Central to these studies has been the need to address privacy and confidentiality of data donors and any fear of data misuse. A balance, therefore, must be struck with permissions to use data for research. As more data is captured including genomic, phenotypic and other health-related data, safeguarding study participants' privacy and confidentiality requires robust governance mechanisms.

Through ethical and legal standpoints, data protection and informed consent policies can support data sharing practice to avoid privacy mishaps. However, current mechanisms most commonly adopted in large consortia (such as a broad consent model) do not go far enough to address individual participants' attitudes and perceptions about data sharing governance and practice**{**ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1159/000324705","ISSN":"1662-8063 (Electronic)\\r1662-4246 (Linking)","abstract":"BACKGROUND: Genomics research data are often widely shared through a variety of mechanisms including publication, meetings and online databases. Re-identification of research participants from sequence data has been shown possible, raising concerns of participants' privacy. METHODS: In 2008-09, we convened 10 focus groups in Durham, N.C. to explore attitudes about how genomic research data were shared amongst the research community, communication of these practices to participants and how different policies might influence participants' likelihood to consent to a genetic/genomic study. Focus groups were audio-recorded and transcripts were complemented by a short anonymous survey. Of 100 participants, 73% were female and 76% African-American, with a median age of 40-49 years. RESULTS: Overall, we found that discussants expressed concerns about privacy and confidentially of data shared through online databases. Although discussants recognized the benefits of data-sharing, they believed it was important to inform research participants of a study's data-sharing plans during the informed consent process. Discussants were significantly more likely to participate in a study that planned to deposit data in a restricted access online database compared to an open access database (p < 0.00001). CONCLUSIONS: The combination of the potential loss of privacy with concerns about data access and identity of the research sponsor warrants disclosure about a study's data-sharing plans during the informed consent process.","author":[{"dropping-particle":"","family":"Haga","given":"S B","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"O'Daniel","given":"J","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Public Health Genomics","id":"ITEM-1","issue":"6","issued":{"date-parts":[["2011"]]},"language":"eng","page":"319-324","title":"Public perspectives regarding data-sharing practices in genomics research","type":"article-journal","volume":"14"},"uris":["http://www.mendeley.com/documents/?uuid=826f9016-064a-3189-80f0-528e470a4d08"]},{"id":"ITEM-2","itemData":{"DOI":"10.1038/ejhg.2012.104","ISSN":"1018-4813","abstract":"Publics and biobanks: Pan-European diversity and the challenge of responsible innovation","author":[{"dropping-particle":"","family":"Gaskell","given":"George","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Gottweis","given":"Herbert","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Starkbaum","given":"Johannes","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Gerber","given":"Monica M","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Broerse","given":"Jacqueline","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Gottweis","given":"Ursula","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hobbs","given":"Abbi","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Helén","given":"Ilpo","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Paschou","given":"Maria","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Snell","given":"Karoliina","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Soulier","given":"Alexandra","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"European Journal of Human Genetics","id":"ITEM-2","issue":"1","issued":{"date-parts":[["2013","1","6"]]},"page":"14-20","publisher":"Nature Publishing Group","title":"Publics and biobanks: Pan-European diversity and the challenge of responsible innovation","type":"article-journal","volume":"21"},"uris":["http://www.mendeley.com/documents/?uuid=5888627e-48e0-395a-9ad4-f6c6945c0674"]},{"id":"ITEM-3","itemData":{"DOI":"10.1097/GIM.0b013e31815f1e00.DNA","author":[{"dropping-particle":"","family":"McGuire","given":"a.L.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hamilton","given":"J.a.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Lunstroth","given":"R","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"McCullough","given":"L B","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Goldman","given":"a.","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Genetics in medicine: official journal of the American College of Medical Genetics","id":"ITEM-3","issue":"1","issued":{"date-parts":[["2008"]]},"page":"46","title":"DNA data sharing : Research participants’ perspectives","type":"article-journal","volume":"10"},"uris":["http://www.mendeley.com/documents/?uuid=95543308-c584-38a5-ae0c-537f5a9e54e9"]},{"id":"ITEM-4","itemData":{"ISSN":"1942-597X","PMID":"25954442","abstract":"We interviewed 70 healthy volunteers to understand their choices about how the information in their health record should be shared for research. Twenty-eight survey questions captured individual preferences of healthy volunteers. The results showed that respondents felt comfortable participating in research if they were given choices about which portions of their medical data would be shared, and with whom those data would be shared. Respondents indicated a strong preference towards controlling access to specific data (83%), and a large proportion (68%) indicated concern about the possibility of their data being used by for-profit entities. The results suggest that transparency in the process of sharing is an important factor in the decision to share clinical data for research.","author":[{"dropping-particle":"","family":"Bell","given":"Elizabeth A","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Ohno-Machado","given":"Lucila","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Grando","given":"M Adela","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"AMIA ... Annual Symposium proceedings. AMIA Symposium","id":"ITEM-4","issued":{"date-parts":[["2014"]]},"page":"1699-708","publisher":"American Medical Informatics Association","title":"Sharing my health data: a survey of data sharing preferences of healthy individuals.","type":"article-journal","volume":"2014"},"uris":["http://www.mendeley.com/documents/?uuid=0b1c4e44-f1ac-39fc-8a40-fe05700ea962"]},{"id":"ITEM-5","itemData":{"DOI":"10.1126/science.1199000","ISSN":"1095-9203","PMID":"21252333","author":[{"dropping-particle":"","family":"Trinidad","given":"S B","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Fullerton","given":"S M","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Ludman","given":"E J","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Jarvik","given":"G P","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Larson","given":"E B","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Burke","given":"W","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Science (New York, N.Y.)","id":"ITEM-5","issue":"6015","issued":{"date-parts":[["2011","1","21"]]},"page":"287-8","publisher":"NIH Public Access","title":"Research ethics. Research practice and participant preferences: the growing gulf.","type":"article-journal","volume":"331"},"uris":["http://www.mendeley.com/documents/?uuid=cdf4eaaa-92af-330a-9d6d-fe8d713144ac"]}],"mendeley":{"formattedCitation":"^{5,12,17–19}","plainTextFormattedCitation":"5,12,17–19","previouslyFormattedCitation":"^{5,12,17–19}"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}**}**. Studies employing broad consent approaches regarding the release of de-identified data for future research may not be sufficiently ethical. There may be inconsistencies in the information provided at the time about how data may be shared and this approach further removes the ability for data donors to have control over what happens to their data after the end of the agreed research period

{ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1093/jamia/ocv014","abstract":"New models of healthcare delivery such as accountable care organizations and patient-centered medical homes seek to improve quality, access, and cost. They rely on a robust, secure technology infrastructure provided by health information exchanges (HIEs) and distributed research networks and the willingness of patients to share their data. There are few large, in-depth studies of US consumers' views on privacy, security, and consent in electronic data sharing for healthcare and research together. Objective This paper addresses this gap, reporting on a survey which asks about California consumers' views of data sharing for healthcare and research together. Materials and Methods The survey conducted was a representative, random-digit dial telephone survey of 800 Californians, per-formed in Spanish and English. Results There is a great deal of concern that HIEs will worsen privacy (40.3%) and security (42.5%). Consumers are in favor of electronic data sharing but elements of transparency are important: individual control, who has access, and the purpose for use of data. Respondents were more likely to agree to share deidentified information for research than to share identified information for healthcare (76.2% vs 57.3%, p < .001). Discussion While consumers show willingness to share health information electronically, they value individual control and privacy. Responsiveness to these needs, rather than mere reliance on Health Insurance Portability and Accountability Act (HIPAA), may im-prove support of data networks. Conclusion Responsiveness to the public's concerns regarding their health information is a pre-requisite for patient-centeredness. This is one of the first in-depth studies of attitudes about electronic data sharing that compares attitudes of the same individual to-wards healthcare and research.","author":[{"dropping-particle":"","family":"Kim","given":"Katherine K","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Joseph","given":"Jill G","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Ohno-Machado","given":"Lucila","non-dropping-particle":"","parse-names":false,"suffix":""}],"id":"ITEM-1","issued":{"date-parts":[["0"]]},"title":"Comparison of consumers' views on electronic data sharing for healthcare and research","type":"article-journal"},"uris":["http://www.mendeley.com/documents/?uuid=f6c44b13-061b-31c6-

ba0c-dc26a3beb119"]},{"id":"ITEM-2","itemData":{"DOI":"10.1097/GIM.0b013e31815f1e00.DNA","author":[{"dropping-particle":"","family":"McGuire","given":"a.L.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hamilton","given":"J.a.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Lunstroth","given":"R","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"McCullough","given":"L B","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Goldman","given":"a.","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Genetics in medicine: official journal of the American College of Medical Genetics","id":"ITEM-2","issue":"1","issued":{"date-parts":[["2008"]]},"page":"46","title":"DNA data sharing : Research participants' perspectives","type":"article-journal","volume":"10"},"uris":["http://www.mendeley.com/documents/?uuid=95543308-c584-38a5-ae0c-537f5a9e54e9"]}],"mendeley":{"formattedCitation":"^{12,15}","plainTextFormattedCitation":"12,15","previouslyFormattedCitation":"^{12,15}"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}**}**.

The question, therefore, arises as to how to design data governance that integrates study participants' preferences, and the first step is to engage with them. Whilst some studies have explored the patient, public, and research participants' perspectives about research consent types, preferences for how and who data should be shared with **{**ADDIN

CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1016/J.AJHG.2017.01.021","ISSN":"0002-9297","abstract":"Individuals participating in biobanks and other large research projects are increasingly asked to provide broad consent for open-ended research use and widespread sharing of their biosamples and data. We assessed willingness to participate in a biobank using different consent and data sharing models, hypothesizing that willingness would be higher under more restrictive scenarios. Perceived benefits, concerns, and information needs were also assessed. In this experimental survey, individuals from 11 US healthcare systems in the Electronic Medical Records and Genomics (eMERGE) Network were randomly allocated to one of three hypothetical scenarios: tiered consent and controlled data sharing; broad consent and controlled data sharing; or broad consent and open data sharing. Of 82,328 eligible individuals, exactly 13,000 (15.8%) completed the survey. Overall, 66% (95% CI: 63%–69%) of population-weighted respondents stated they would be willing to participate in a biobank; willingness and attitudes did not differ between respondents in the three scenarios. Willingness to participate was associated with self-identified white race, higher educational attainment, lower religiosity, perceiving more research benefits, fewer concerns, and fewer information needs. Most (86%, CI: 84%–87%) participants would want to know what would happen if a researcher misused their health information; fewer (51%, CI: 47%–55%) would worry about their privacy. The concern that the use of broad consent and open data sharing could adversely affect participant recruitment is not supported by these findings. Addressing potential participants' concerns and information needs and building trust and relationships with communities may increase acceptance of broad consent and wide data sharing in biobank research.","author":[{"dropping-particle":"","family":"Sanderson","given":"Saskia C.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Brothers","given":"Kyle B.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Mercaldo","given":"Nathaniel D.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Clayton","given":"Ellen Wright","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Antommaria","given":"Armand H. Matheny","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Aufox","given":"Sharon A.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Brilliant","given":"Murray H.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Campos","given":"Diego","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Carrell","given":"David S.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Connolly","given":"John","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Conway","given":"Pat","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Fullerton","given":"Stephanie M.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Garrison","given":"Nanibaa’ A.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Horowitz","given":"Carol R.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Jarvik","given":"Gail P.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Kaufman","given":"David","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Kitchner","given":"Terrie E.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Li","given":"Rongling","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Ludman","given":"Evette J.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"McCarty","given":"Catherine A.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"McCormick","given":"Jennifer B.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"McManus","given":"Valerie D.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Myers","given":"Melanie F.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Scrol","given":"Aaron","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Williams","given":"Janet L.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Shrubsole","given":"Martha J.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Schildcrout","given":"Jonathan S.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Smith","given":"Maureen E.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Holm","given":"Ingrid A.","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"The American Journal of Human Genetics","id":"ITEM-1","issue":"3","issued":{"date-parts":[["2017","3","2"]]},"page":"414-427","publisher":"Cell Press","title":"Public Attitudes toward Consent and Data Sharing in Biobank Research: A Large Multi-site Experimental Survey in the US","type":"article-journal","volume":"100"},"uris":["http://www.mendeley.com/documents/?uuid=5481930d-5886-3767-a242-eed00e1b60a7"]},{"id":"ITEM-2","itemData":{"DOI":"10.1097/GIM.0b013e31815f1e00.DNA","author":[{"dropping-particle":"","family":"McGuire","given":"a.L.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hamilton","given":"J.a.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Lunstroth","given":"R","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"McCullough","given":"L B","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Goldman","given":"a.","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Genetics in medicine: official journal of the American College of Medical Genetics","id":"ITEM-2","issue":"1","issued":{"date-parts":[["2008"]]},"page":"46","title":"DNA data sharing : Research participants’ perspectives","type":"article-journal","volume":"10"},"uris":["http://www.mendeley.com/documents/?uuid=95543308-c584-38a5-ae0c-537f5a9e54e9"]},{"id":"ITEM-

3","itemData":{"DOI":"10.1097/GIM.0b013e3182227589","abstract":"PURPOSE: Despite growing concerns toward maintaining participants' privacy, individual investigators collecting tissue and other biological specimens for genomic analysis are encouraged to obtain informed consent for broad data sharing. Our purpose was to assess the effect on research enrollment and data sharing decisions of three different consent types (traditional, binary, or tiered) with varying levels of control and choices regarding data sharing., METHODS: A single-blinded, randomized controlled trial was conducted with 323 eligible adult participants being recruited into one of six genome studies at Baylor College of Medicine in Houston, Texas, between January 2008 and August 2009. Participants were randomly assigned to one of three experimental consent documents (traditional, n = 110; binary, n = 103; and tiered, n = 110). Debriefing in follow-up visits provided participants a detailed review of all consent types and the chance to change data sharing choices or decline genome study participation., RESULTS: Before debriefing, 83.9% of participants chose public data release. After debriefing, 53.1% chose public data release, 33.1% chose restricted (controlled access database) release, and 13.7% opted out of data sharing. Only one participant declined genome study participation due to data sharing concerns., CONCLUSION: Our findings indicate that most participants are willing to publicly release their genomic data; however, a significant portion prefers restricted release. These results suggest discordance between existing data sharing policies and participants' judgments and desires.","author":[{"dropping-particle":"","family":"McGuire","given":"Amy L","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Oliver","given":"Jill M","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Slashinski","given":"Melody J","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Graves","given":"Jennifer L","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Wang","given":"Tao","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Kelly","given":"P Adam","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Fisher","given":"William","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Lau","given":"Ching C","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Goss","given":"John","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Okcu","given":"Mehmet","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Treadwell-Deering","given":"Diane","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Goldman","given":"Alica M","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Noebels","given":"Jeffrey L","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hilsenbeck","given":"Susan G","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Genetics in medicine : official journal of the American College of Medical Genetics","id":"ITEM-3","issue":"11","issued":{"date-parts":[["2011","11"]]},"page":"948-955","publisher":"McGuire,Amy L. Center for Medical Ethics and Health Policy, Baylor College of Medicine, Houston, TX 77030, USA. amcguire@bcm.edu","publisher-place":"United States","title":"To share or not to share: a randomized trial of consent for data sharing in genome research.","type":"article-journal","volume":"13"},"uris":["http://www.mendeley.com/documents/?uuid=41622375-a686-30c9-9cad-f425c1a24015"]}],"mendeley":{"formattedCitation":"^{9,11,12}","plainTextFormattedCitation":"9,11,12","previouslyFormattedCitation":"^{9,11,12}"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}}, to date, little is known about research participants’ views and preferences about how their biomedical, particularly genetic and phenotypic data from one research project should be shared for future and separate research{ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1038/ejhg.2012.104","ISSN":"1018-4813","abstract":"Publics and biobanks: Pan-European diversity and the challenge of responsible innovation","author":[{"dropping-particle":"","family":"Gaskell","given":"George","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Gottweis","given":"Herbert","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Starkbaum","given":"Johannes","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Gerber","given":"Monica M","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Broerse","given":"Jacqueline","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Gottweis","given":"Ursula","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hobbs","given":"Abbi","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Helén","given":"Ilpo","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Paschou","given":"Maria","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Snell","given":"Karoliina","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Soulier","given":"Alexandra","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"European Journal of Human Genetics","id":"ITEM-1","issue":"1","issued":{"date-parts":[["2013","1","6"]]},"page":"14-20","publisher":"Nature Publishing Group","title":"Publics and biobanks: Pan-European diversity and the challenge of responsible innovation","type":"article-journal","volume":"21"},"uris":["http://www.mendeley.com/documents/?uuid=5888627e-48e0-395a-9ad4-f6c6945c0674"]}],"mendeley":{"formattedCitation":"⁵","plainTextFormattedCitation":"5","previouslyFormattedCitation":"⁵"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}**}**.

While there is increasing recognition to engage and involve research participants in data governance plans in international consortia, studies highlighted here have largely been conducted in North America, with focus on hypothetical data sharing scenarios and improving broad consent at the initial stage of projects. Furthermore, the challenge in engaging research participants about the management of data sharing is compounded when international consortia collect data from people in different countries, where cultural and legal differences can affect readiness and ability to share data

{ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1038/ejhg.2012.104","ISSN":"1018-4813","abstract":"Publics and biobanks: Pan-European diversity and the challenge of responsible innovation","author":[{"dropping-particle":"","family":"Gaskell","given":"George","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Gottweis","given":"Herbert","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Starkbaum","given":"Johannes","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Gerber","given":"Monica M","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Broerse","given":"Jacqueline","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Gottweis","given":"Ursula","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hobbs","given":"Abbi","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Helén","given":"Ilpo","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Paschou","given":"Maria","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Snell","given":"Karoliina","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Soulier","given":"Alexandra","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"European Journal of Human Genetics","id":"ITEM-1","issue":"1","issued":{"date-parts":[["2013","1","6"]]},"page":"14-20","publisher":"Nature Publishing Group","title":"Publics and biobanks: Pan-European diversity and the challenge of responsible innovation","type":"article-journal","volume":"21"},"uris":["http://www.mendeley.com/documents/?uuid=5888627e-48e0-395a-9ad4-f6c6945c0674"]}],"mendeley":{"formattedCitation":"⁵","plainTextFormattedCitation":"5","previouslyFormattedCitation":"⁵"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}**}**. Differences in wanting or having control over data sharing also varies within diverse populations in relation to privacy concerns**{**ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1016/j.ajhg.2009.10.002","abstract":"Concerns about privacy may deter people from participating in genetic research. Recruitment and retention of biobank participants requires understanding the nature and magnitude of these concerns. Potential participants in a proposed biobank were asked about their willingness to participate, their privacy concerns, informed consent, and data sharing. A representative survey of 4659 U.S. adults was conducted. Ninety percent of respondents would be concerned about privacy, 56% would be concerned about researchers having their information, and 37% would worry that study data could be used against them. However, 60% would participate in the biobank if asked. Nearly half (48%) would prefer to provide consent once for all research approved by an oversight panel, whereas 42% would prefer to provide consent for each project separately. Although 92% would allow academic researchers to use study data, 80% and 75%, respectively, would grant access to government and industry researchers. Concern about privacy was related to lower willingness to participate only when respondents were told that they would receive $50 for participation and would not receive individual research results back. Among respondents who were told that they would receive $200 or individual research results, privacy concerns were not related to willingness. Survey respondents valued both privacy and participation in biomedical research. Despite pervasive privacy concerns, 60% would participate in a biobank. Assuring research participants that their privacy will be protected to the best of researchers' abilities may increase participants' acceptance of consent for broad research uses of biobank data by a wide range of researchers.","author":[{"dropping-particle":"","family":"Kaufman","given":"David J","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Murphy-Bollinger","given":"Juli","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Scott","given":"Joan","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hudson","given":"Kathy L","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"American journal of human genetics","id":"ITEM-1","issue":"5","issued":{"date-parts":[["2009","11"]]},"language":"eng","page":"643-654","title":"Public opinion about the importance of privacy in biobank research.","type":"article-journal","volume":"85"},"uris":["http://www.mendeley.com/documents/?uuid=f47d8554-6ed8-3b81-bd8d-6c35ea4fa9d4"]}],"mendeley":{"formattedCitation":"¹","plainTextFormattedCitation":"1","previouslyFormattedCitation":"¹"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}**}**. Participants in large consortia projects are often not consulted about their opinions on how their data should be governed during and after the end of the research project. Differences in attitudes and preferences between culturally dissimilar countries in Europe have been least studied, within the context of future research data sharing**{**ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1038/ejhg.2012.104","ISSN":"1018-4813","abstract":"Publics and biobanks: Pan-European diversity and the challenge of responsible innovation","author":[{"dropping-particle":"","family":"Gaskell","given":"George","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Gottweis","given":"Herbert","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Starkbaum","given":"Johannes","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Gerber","given":"Monica M","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Broerse","given":"Jacqueline","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Gottweis","given":"Ursula","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hobbs","given":"Abbi","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Helén","given":"Ilpo","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Paschou","given":"Maria","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Snell","given":"Karoliina","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Soulier","given":"Alexandra","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"European Journal of Human Genetics","id":"ITEM-1","issue":"1","issued":{"date-parts":[["2013","1","6"]]},"page":"14-20","publisher":"Nature Publishing Group","title":"Publics and biobanks: Pan-European diversity and the challenge of responsible innovation","type":"article-journal","volume":"21"},"uris":["http://www.mendeley.com/documents/?uuid=5888627e-48e0-395a-9ad4-f6c6945c0674"]}],"mendeley":{"formattedCitation":"⁵","plainTextFormattedCitation":"5","previouslyFormattedCitation":"⁵"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}**}**.

Therefore, this study aimed to investigate research participant’s beliefs about the importance of protecting their privacy, advancing research quickly and controlling future data sharing beyond the end of the research project with a subset of participants in four European countries enrolled in the DIRECT (Diabetes Research on Patient Stratification) project.

Materials and Methods

Study population and recruitment

Participants were sampled from a subset of those enrolled in the DIRECT studies. In total 1082 participants attending follow-up appointments for other DIRECT studies at study centres in Denmark, Sweden, The Netherlands, and the UK were invited to complete the cross-sectional survey. The overall DIRECT project participant sample and recruitment is described in detail elsewhere

{ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1007/s00125-014-3216-x","ISSN":"1432-0428","PMID":"24695864","abstract":"AIMS/HYPOTHESIS The DIRECT (Diabetes Research on Patient Stratification) Study is part of a European Union Framework 7 Innovative Medicines Initiative project, a joint undertaking between four industry and 21 academic partners throughout Europe. The Consortium aims to discover and validate biomarkers that: (1) predict the rate of glycaemic deterioration before and after type 2 diabetes onset; (2) predict the response to diabetes therapies; and (3) help stratify type 2 diabetes into clearly definable disease subclasses that can be treated more effectively than without stratification. This paper describes two new prospective cohort studies conducted as part of DIRECT. METHODS Prediabetic participants (target sample size 2,200-2,700) and patients with newly diagnosed type 2 diabetes (target sample size ~1,000) are undergoing detailed metabolic phenotyping at baseline and 18 months and 36 months later. Abdominal, pancreatic and liver fat is assessed using MRI. Insulin secretion and action are assessed using frequently sampled

OGTTs in non-diabetic participants, and frequently sampled mixed-meal tolerance tests in patients with type 2 diabetes. Biosamples include venous blood, faeces, urine and nail clippings, which, among other biochemical analyses, will be characterised at genetic, transcriptomic, metabolomic, proteomic and metagenomic levels. Lifestyle is assessed using high-resolution triaxial accelerometry, 24 h diet record, and food habit questionnaires. CONCLUSIONS/INTERPRETATION DIRECT will yield an unprecedented array of biomaterials and data. This resource, available through managed access to scientists within and outside the Consortium, will facilitate the development of new treatments and therapeutic strategies for the prevention and management of type 2 diabetes.","author":[{"dropping-particle":"","family":"Koivula","given":"Robert W","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Heggie","given":"Alison","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Barnett","given":"Anna","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Cederberg","given":"Henna","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hansen","given":"Tue H","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Koopman","given":"Anitra D","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Ridderstråle","given":"Martin","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Rutters","given":"Femke","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Vestergaard","given":"Henrik","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Gupta","given":"Ramneek","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Herrgård","given":"Sanna","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Heymans","given":"Martijn W","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Perry","given":"Mandy H","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Rauh","given":"Simone","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Siloaho","given":"Maritta","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Teare","given":"Harriet J A","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Thorand","given":"Barbara","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Bell","given":"Jimmy","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Brunak","given":"Søren","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Frost","given":"Gary","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Jablonka","given":"Bernd","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Mari","given":"Andrea","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"McDonald","given":"Tim J","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Dekker","given":"Jacqueline M","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hansen","given":"Torben","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hattersley","given":"Andrew","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Laakso","given":"Markku","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Pedersen","given":"Oluf","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Koivisto","given":"Veikko","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Ruetten","given":"Hartmut","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Walker","given":"Mark","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Pearson","given":"Ewan","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Franks","given":"Paul W","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"DIRECT Consortium","given":"","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Diabetologia","id":"ITEM-1","issue":"6","issued":{"date-parts":[["2014","6"]]},"page":"1132-42","title":"Discovery of biomarkers for glycaemic deterioration before and after the onset of type 2 diabetes: rationale and design of the epidemiological studies within the IMI DIRECT Consortium.","type":"article-journal","volume":"57"},"uris":["http://www.mendeley.com/documents/?uuid=4b482136-e4d4-389d-be26-00a7c79b3d5a"]},{"id":"ITEM-2","itemData":{"DOI":"10.1038/s41436-018-0299-7","ISSN":"1098-3600","abstract":"Biomedical data governance strategies should ensure that data are collected, stored, and used ethically and lawfully. However, research participants' preferences for how data should be governed is least studied. The Diabetes Research on Patient Stratification (DIRECT) project collected substantial amounts of health and genetic information from patients at risk of, and with type II diabetes. We conducted a survey to understand participants' future data governance preferences. Results will inform the postproject data governance strategy. A survey was distributed in Denmark, Sweden, The Netherlands, and the United Kingdom. In total 855 surveys were returned. Ninety-seven percent were supportive of sharing data postproject, and 90% were happy to share data with universities, and 56% with commercial companies. The top three priorities for data sharing were highly secure database, DIRECT researchers to monitor data used by other researchers, and researchers cannot identify participants. 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Engaging participants aids in designing governance to support their choices.","author":[{"dropping-particle":"","family":"Shah","given":"Nisha","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Coathup","given":"Victoria","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Teare","given":"Harriet","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Forgie","given":"Ian","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Giordano","given":"Giuseppe Nicola","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hansen","given":"Tue Haldor","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Groeneveld","given":"Lenka","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hudson","given":"Michelle","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Pearson","given":"Ewan","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Ruetten","given":"Hartmut","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Kaye","given":"Jane","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Genetics in Medicine","id":"ITEM-2","issued":{"date-parts":[["2018","9","28"]]},"page":"1","publisher":"Nature Publishing Group","title":"Sharing data for future research—engaging participants' views about data governance beyond the original project: a DIRECT Study","type":"article-journal"},"uris":["http://www.mendeley.com/documents/?uuid=f98c5948-c4cc-3831-

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. Study participants were eligible to take part in the survey if they were aged 18 years and older, of white European descent and able to consent to participate. This study was approved by institutional review boards in: Denmark (The Secretariat of the Scientific Ethics Committees for the Capital Region Protocol no. H-1-2011-166 Note no. 50965, and H-1-2012-100 Note no. 50694), Sweden (Regional Ethics Examination Board in Lund Dnr 2015/815 and Dnr 2015/843), The Netherlands (Medical Ethics Review Committee Vrije Universiteit Medical Centre Protocol 2012.222), and the UK (Newcastle and North Tynesside 1 Research Ethics Committee 12/NE/0132; East of Scotland Research Ethics Service 11/ES/0046; and 12/ES/0034).

Survey measures

Survey items analysed in this study were selected from a wider patient engagement survey that assessed: DIRECT participants' willingness to participate in medical research; support for data sharing; preferences for control of different types of data; who data are shared with; and, preferences for future data sharing governance. Sociodemographic characteristics and self-reported knowledge of genetics and health status were also collected. The survey was developed with DIRECT diabetes clinicians, participants with Type 2 diabetes (T2D) and consortium researchers through iterative review and adjustment to question items

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Respondents were asked to assert their agreement on four statements measuring beliefs about whether it was important to advance research quickly, whether privacy should be protected, and whether respondents perceived that there were risks or benefits to sharing their genetic information

{ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1159/000334718","ISSN":"1662-8063","abstract":"BACKGROUND: Technological advancements are rapidly propelling the field of genome research forward, while lawmakers attempt to keep apace with the risks these advances bear. Balancing normative concerns of maximizing data utility and protecting human subjects, whose privacy is at risk due to the identifiability of DNA data, are central to policy decisions. Research on genome research participants making real-time data sharing decisions is limited; yet, these perspectives could provide critical information to ongoing deliberations., METHODS: We conducted a randomized trial of 3 consent types affording varying levels of control over data release decisions. After debriefing participants about the randomization process, we invited them to a follow-up interview to assess their attitudes toward genetic research, privacy and data sharing., RESULTS: Participants were more restrictive in their reported data sharing preferences than in their actual data sharing decisions. They saw both benefits and risks associated with sharing their genomic data, but risks were seen as less concrete or happening in the future, and were largely outweighed by purported benefits., CONCLUSION: Policymakers must respect that participants' assessment of the risks and benefits of data sharing and their privacy-utility determinations, which are associated with their final data release decisions, vary. In order to advance the ethical conduct of genome research, proposed policy changes should carefully consider these stakeholder perspectives.Copyright © 2011 S. Karger AG, Basel.","author":[{"dropping-particle":"","family":"Oliver","given":"J M","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Slashinski","given":"M J","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Wang","given":"T","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Kelly","given":"P a.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hilsenbeck","given":"S G","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"McGuire","given":"a. L","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Public Health Genomics","id":"ITEM-1","issue":"2","issued":{"date-parts":[["2012"]]},"page":"106-114","publisher":"Oliver,J M. Center for Medical Ethics and Health Policy, Baylor College of Medicine, Houston, Tex 77030, USA.","publisher-place":"Switzerland","title":"Balancing the Risks and Benefits of Genomic Data Sharing: Genome Research Participants' Perspectives.","type":"article-journal","volume":"15"},"uris":["http://www.mendeley.com/documents/?uuid=d9b5f435-e47f-34e4-bd3a-64b7f5d26fe7"]}],"mendeley":{"formattedCitation":"²²","plainTextFormattedCitation":"22","previouslyFormattedCitation":"²²"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}**}**. The outcome variables assessed participants’ ratings of importance of which data are shared and with whom (importance of control), and were measured by the questions “How important is it that you decide what types of data are shared” and “How important is it that you decide who your data is shared with?”. The survey also measured respondents’ happiness to share different types of data. Similarly, respondents were asked to rate their happiness to share their de-identified data with different research groups. These items were treated as continuous explanatory variables. Participant characteristics were binary or categorical in nature. The explanatory variables were recoded into smaller categorical variables due to low numbers of responses in some categories, except the items measuring happiness to share different types of data and with different research groups, which were treated as continuous. The outcome variables were collapsed into binary variables for ease of interpretation.

Data analysis

Descriptive statistics were calculated as frequencies and percentages, and Chi-Square tests for independence assessed associations between categorical variables. Univariate (see supplementary tables S2 and S3) and multivariate logistic regressions were conducted to assess which explanatory variables predicted the odds of importance for control over 1) types of data shared, and 2) who data are shared with. These outcome variables were binary (important versus not important). The continuous explanatory variables entered into the logistic regressions were the four items measuring beliefs and perceptions about data sharing, happiness to share different types of data and with whom data can be shared. The multivariate logistic regressions were adjusted for by the categorical variables: age, gender, country, education level, self-rated knowledge of genetics, diabetes status, previously worked in health or medicine, and self-reported health (see Tables 4 and 5). Between-country differences were assessed in the multivariate logistic regressions adjusted for by all other variables (see Table 3). All univariate and multivariate models contained complete cases, as not all respondents answered all of the questions and the minimal amount of cases were missing. All analyses were also stratified by country to assess associations within countries and compare findings. The logistic regression results are reported as odds ratios (ORs) with 95% confidence intervals (CI) and significance level $p<0.05$. The reference group in all regression models comparing the countries was the UK due to the largest number of responses received from this participant group. The analyses were performed using SPSS version 22 (SPSS, Inc., Chicago, IL).

Results

Sample characteristics

In total, 1082 DIRECT project participants were approached and 855 participated in the engagement survey from University research centres and Diabetes clinics in the four countries. The combined response rate for all countries was 79%. The majority (73%) of participants were aged 61 and over, 57% were male, 70% had been diagnosed with T2D, 60% had education qualifications above secondary school, and 20% had held a job related to health or medicine at some point in their career (Supplementary Table S1). Sixty-three per cent of 835 respondents rated their health as ‘very good’ or ‘good’ versus 30% rating it as ‘fair’. Forty-five per cent rated their knowledge of genetics as ‘fair’ versus 39% that rated it as either ‘poor’ or ‘very poor’.

Beliefs about research and privacy, and risk-benefit assessments to sharing data

Eighty-nine percent of respondents either strongly agreed or agreed that it is important to advance research as quickly as possible; however, all respondents were already participating in research as they had agreed to enrol onto a study within the DIRECT project. Seventy-seven per cent overall also agreed that protecting privacy was important to them, and this was consistent across all countries when stratified. The perception that there were benefits to sharing their genetic information for research was strongly agreed and agreed by 87% of respondents; in contrast, only 46% agreed that there were risks to sharing their genetic information. There were no other significant differences in respondents' beliefs about privacy or advancing research, and benefits to sharing their data by knowledge of genetics. When stratified, country of origin was significantly associated with all belief statements except the importance of protecting privacy (Table 1), except importance over privacy where there was no significant change in proportions between countries.

Importance of control for participants to share data

Forty-two percent of respondents rated having control of what types of data should be shared as either fairly or extremely important, and when stratified by country the results were: 41% in Denmark, 36% in Sweden, 36% in The Netherlands, and 45% in the UK (Figure 1). However, after adjusting for all variables in the multivariate logistic regressions, none of the countries were significantly more or less likely to want control compared to the UK (see Table 2). Forty-three percent of respondents rated that having control over who their data is shared with was either fairly or extremely important to them, and by country the results were: 42% in Denmark, 44% in Sweden, 46% in The Netherlands, and 42% in the UK (Figure 2). There were no significant differences in the importance of control for deciding who to share data with compared to the UK after adjusting for all other variables (see Table 2).

Examining associations for importance for participants to control types of data shared

In univariate binary logistic regression models (Supplementary Table S2), our findings suggested that questions about: the importance of protecting privacy; beliefs that there are risks to sharing genetic information; and happiness to share: a) genetic information, b) blood test results, c) lifestyle information, and d) personal information, were all significant predictors of the importance of control. There were no significant differences between countries compared to the UK in whether deciding the types of data shared was important vs not important (supplementary TableS2).

The pooled country results (Table 3 and Supplementary Table S4) suggested that agreeing that it is important to protect privacy was significantly associated with beliefs concerning control over which data are shared (OR=1.86, CI (1.38-2.51), p<0.001). Happiness to share lifestyle and personal information were significantly associated with the importance to control which data are shared (OR=0.5, CI (0.29-0.84), p<0.01), and OR=0.64, CI (0.52-0.80), p<0.01) respectively. There were no other significant associations between the covariates and importance for control. When results were stratified by country, similar results were found in the Danish cohort, though results in the UK and Dutch cohorts did not reach significance. The sample size for the Swedish cohort was too small to compute the results for comparison (Supplementary Table S4).

Examining associations for importance for participants to control who data is shared with

Results from univariate logistic regressions found that importance of control was predicted by belief in protecting privacy, agreement that there are benefits to sharing genetic information, happiness to share with commercial companies and charities. This was consistent in the results stratified by country (Supplementary Table S3). The adjusted model showed that there was no significant association between country and importance of respondents to decide who data are shared with (Supplementary Table S5). Table 4 shows that increased importance for protecting privacy resulted in respondents being more likely to indicate that having control over data sharing was important (OR=2.26, CI (1.67-3.1), p<0.001). This was consistent across all countries, except Sweden, which did not yield significant results due to a very small sample (Supplementary Table S5). Respondents in all countries were 1.64 times significantly more likely to also indicate importance of control (data sharing) and believe that there were benefits to sharing their genetic information (p=0.03). Disagreement that there were risks to sharing genetic information was associated with decreased likelihood for rating importance of control (OR=0.74, CI (0.59-0.91), p<0.01). Happiness to share data with commercial companies and charities was significantly associated with rating importance for control (OR=0.43, CI (0.32-0.56), p<0.01) and (OR=0.57, CI (0.39-0.84), p<0.01) respectively. These results were similar across countries, except Sweden where results were not significant.

Discussion

The current study aimed to assess desire for control for sharing data in relation to motivations (measured by attitudes/beliefs) about advancing research and protecting privacy, and willingness to share data. Where previous research has investigated improving informed consent through tiered choices{ADDIN CSL_CITATION

{"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1097/GIM.0b013e31815f1e00.DNA","author":[{"dropping-particle":"","family":"McGuire","given":"a.L.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hamilton","given":"J.a.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Lunstroth","given":"R","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"McCullough","given":"L B","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Goldman","given":"a.","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Genetics in medicine: official journal of the American College of Medical Genetics","id":"ITEM-1","issue":"1","issued":{"date-parts":[["2008"]]},"page":"46","title":"DNA data sharing : Research participants’ perspectives","type":"article-journal","volume":"10"},"uris":["http://www.mendeley.com/documents/?uuid=95543308-c584-38a5-ae0c-537f5a9e54e9"]},{"id":"ITEM-2","itemData":{"DOI":"10.1159/000334718","ISSN":"1662-8063","abstract":"BACKGROUND: Technological advancements are rapidly propelling the field of genome research forward, while lawmakers attempt to keep apace with the risks these advances bear. Balancing normative concerns of maximizing data utility and protecting human subjects, whose privacy is at risk due to the identifiability of DNA data, are central to policy decisions. Research on genome research participants making real-time data sharing decisions is limited; yet, these perspectives could provide critical information to ongoing deliberations., METHODS: We conducted a randomized trial of

3 consent types affording varying levels of control over data release decisions. After debriefing participants about the randomization process, we invited them to a follow-up interview to assess their attitudes toward genetic research, privacy and data sharing., RESULTS: Participants were more restrictive in their reported data sharing preferences than in their actual data sharing decisions. They saw both benefits and risks associated with sharing their genomic data, but risks were seen as less concrete or happening in the future, and were largely outweighed by purported benefits., CONCLUSION: Policymakers must respect that participants' assessment of the risks and benefits of data sharing and their privacy-utility determinations, which are associated with their final data release decisions, vary. In order to advance the ethical conduct of genome research, proposed policy changes should carefully consider these stakeholder perspectives.Copyright © 2011 S. Karger AG, Basel.","author":[{"dropping-particle":"","family":"Oliver","given":"J M","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Slashinski","given":"M J","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Wang","given":"T","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Kelly","given":"P a.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hilsenbeck","given":"S G","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"McGuire","given":"a. L","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Public Health Genomics","id":"ITEM-2","issue":"2","issued":{"date-parts":[["2012"]]},"page":"106-114","publisher":"Oliver,J M. Center for Medical Ethics and Health Policy, Baylor College of Medicine, Houston, Tex 77030, USA.","publisher-

place":"Switzerland","title":"Balancing the Risks and Benefits of Genomic Data Sharing: Genome Research Participants' Perspectives.","type":"article-journal","volume":"15"},"uris":["http://www.mendeley.com/documents/?uuid=d9b5f435-e47f-34e4-bd3a-64b7f5d26fe7"]},{"id":"ITEM-3","itemData":{"DOI":"10.1186/s12910-015-0053-5","ISSN":"1472-6939","abstract":"Appropriate information and consent has been one of the most intensely discussed topics within the context of biobank research. In parallel to the normative debate, many socio-empirical studies have been conducted to gather experiences, preferences and views of patients, healthy research participants and further stakeholders. However, there is scarcity of literature which connects the normative debate about justifications for different consent models with findings gained in empirical research. In this paper we discuss findings of a limited review of socio-empirical research on patients’ and healthy research participants’ experiences and views regarding consent to biobank research in light of ethical principles for appropriate information and consent. Review question: Which empirical data are available on research participants’ perceptions and views regarding information and elicitation of consent for biobank research? Search of articles published till March 1st 2014 in Pubmed. Review of abstracts and potentially relevant full text articles by two authors independently. As categories for content analysis we defined (i) understanding or recall of information, (ii) preferences regarding information or consent, and (iii) research participants’ concerns. The search in Pubmed yielded 337 abstracts of which 10 articles were included in this study. Approaches to information and consent varied considerably across the selected studies. The majority of research participants opted for some version of limited consent when being informed about such possibility. Among the factors influencing the type of preferred consent were information about sponsoring of biobank research by pharmaceutical industry and participants’ trade-off between privacy and perceived utility. Studies investigating research participants’ understanding and recall regarding the consent procedure indicated considerable lack of both aspects. Research participants’ perceptions of benefits and harms differ across those studies. The knowledge, perceptions and views of research participants who have undergone a consent procedure within the context of biobank research raise several questions on the issue of how to inform and elicit consent in an ethically acceptable way. In our empirical-ethical analysis we develop suggestions on how the practice of eliciting consent in the biobank context should be improved.","author":[{"dropping-particle":"","family":"D’Abramo","given":"Flavio","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Schildmann","given":"Jan","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Vollmann","given":"Jochen","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"BMC Medical Ethics","id":"ITEM-3","issue":"1","issued":{"date-parts":[["2015","12","9"]]},"page":"60","publisher":"BioMed Central","title":"Research participants’ perceptions and views on consent for biobank research: a review of empirical data and ethical analysis","type":"article-journal","volume":"16"},"uris":["http://www.mendeley.com/documents/?uuid=dbaa1e8d-e81e-3a16-990b-b5757b043e82"]}],"mendeley":{"formattedCitation":"^{8,12,22}","plainTextFormattedCitation":"8,12,22","previouslyFormattedCitation":"^{8,12,22}"}," properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}**}**, this study sought to obtain a more granular overview of study participants’ judgements about data sharing, and whether there were differences between individual participants across four European countries. When given the choice to have control, less than 50% indicated that having control over what data is shared and with whom was important..

The study findings suggest that control over what data types are shared was less important to respondents than deciding who data are shared with. The importance for control over de-identified data sharing found in this study is consistent with other research, which has highlighted that when data are de-identified, fewer respondents expect the need to have control in sharing of their data**{**ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1016/j.ijmedinf.2015.01.008","ISSN":"1386-5056","abstract":"The development of Electronic Health Records (EHRs) forms an integral part of the information strategy for the National Health Service (NHS) in the UK, with the aim of facilitating health information exchange for patient care and secondary use, including research and healthcare planning. Implementing EHR systems requires an understanding of patient expectations for consent mechanisms and consideration of public awareness towards information sharing as might be made possible through integrated EHRs across primary and secondary health providers. Objectives: To explore levels of public awareness about EHRs and to examine attitudes towards different consent models with respect to sharing identifiable and de-identified records for healthcare provision, research and planning. Methods: A cross-sectional questionnaire survey was administered to adult patients and members of the public in primary and secondary care clinics in West

London, UK in 2011. In total, 5331 individuals participated in the survey, and 3157 were included in the final analysis. Results: The majority (91%) of respondents expected to be explicitly asked for consent for their identifiable records to be accessed for health provision, research or planning. Half the respondents (49%) did not expect to be asked for consent before their de-identified records were accessed. Compared with White British respondents, those from all other ethnic groups were more likely to anticipate their permission would be obtained before their de-identified records were used. Of the study population, 59% reported already being aware of EHRs before the survey. Older respondents and individuals with complex patterns of interaction with healthcare services were more likely to report prior awareness of EHRs. Individuals self-identifying as belonging to ethnic groups other than White British, and those with lower educational qualifications were less likely to report being aware of EHRs than White British respondents and respondents with degree-level education, respectively. Those who reported being aware of EHRs were less likely to say they expected explicit consent to be sought before use of their de-identified record. Conclusions: A large number of patients remain unaware of EHRs, while preference for implicit consent is stronger among those who report previous awareness. Differences in awareness levels and consent expectations between groups with different socio-demographic characteristics suggest that public education…","author":[{"dropping-particle":"","family":"Riordan","given":"Fiona","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Papoutsi","given":"Chrysanthi","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Reed","given":"Julie E","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Marston","given":"Cicely","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Bell","given":"Derek","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Majeed","given":"Azeem","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"International Journal of Medical Informatics","id":"ITEM-1","issue":"4","issued":{"date-parts":[["2015"]]},"page":"237-247","publisher":"Elsevier Ireland Ltd","title":"Patient and public attitudes towards informed consent models and levels of awareness of Electronic Health Records in the UK","type":"article-journal","volume":"84"},"uris":["http://www.mendeley.com/documents/?uuid=246ff818-1af8-331a-bead-ba5ddd374267"]}],"mendeley":{"formattedCitation":"¹⁴","plainTextFormattedCitation":"14","previouslyFormattedCitation":"¹⁴"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}}.

Whilst we found that overall desire for control of de-identified data was moderate (less than 50%), when assessing associations between happiness to share different data types and with research groups, importance for control varied with different options. How participants valued control over data sharing was associated with unhappiness to share data with global universities, commercial companies, and charities that conduct research. A report commissioned by the Wellcome Trust in the UK{ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"abstract":"1 15-029639-01 | PUBLIC |

This work was carried out in accordance with the requirements of the international quality standard for Market Research, ISO 20252:2012, and with the Ipsos MORI Terms and Conditions which can be found at http://www.ipsos-mori.com/terms. 15-029639-01 | PUBLIC | This work was carried out in accordance with the requirements of the international quality standard for Market Research, ISO 20252:2012, and with the Ipsos MORI Terms and Conditions which can be found at http://www.ipsos-mori.com/terms. © Ipsos MORI 2015. 15-029639-01 | PUBLIC | This work was carried out in accordance with the requirements of the international quality standard for Market Research, ISO 20252:2012, and with the Ipsos MORI Terms and Conditions which can be found at http://www.ipsos-mori.com/terms. © Ipsos MORI 2016. The One-Way Mirror: Public attitudes to commercial access to health data | Report prepared for the Wellcome Trust | February 2016 15-029639-01 | PUBLIC | This work was carried out in accordance with the requirements of the international quality standard for Market Research, ISO 20252:2012, and with the Ipsos MORI Terms and Conditions which can be found at http://www.ipsos-mori.com/terms.","author":[{"dropping-particle":"","family":"MORI for Wellcome Trust","given":"Ipsos","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Trust","given":"IPSOS MORI for Wellcome","non-dropping-particle":"","parse-names":false,"suffix":""}],"id":"ITEM-1","issue":"March","issued":{"date-parts":[["2016"]]},"title":"The One-Way Mirror: Public attitudes to commercial access to health data Report prepared for the Wellcome Trust","type":"article-journal"},"uris":["http://www.mendeley.com/documents/?uuid=a67ad777-12e2-4611-8121-27af961cb433"]}],"mendeley":{"formattedCitation":"²³","plainTextForma

ttedCitation":"23","previouslyFormattedCitation":"²³"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}**}**, found that willingness to share data is influenced by trust in the institution and the extent patients are informed about who their data are being shared with, and what aspects, particularly in relation to commercial entities**{**ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"abstract":"1 15-029639-01 | PUBLIC | This work was carried out in accordance with the requirements of the international quality standard for Market Research, ISO 20252:2012, and with the Ipsos MORI Terms and Conditions which can be found at http://www.ipsos-mori.com/terms. 15-029639-01 | PUBLIC | This work was carried out in accordance with the requirements of the international quality standard for Market Research, ISO 20252:2012, and with the Ipsos MORI Terms and Conditions which can be found at http://www.ipsos-mori.com/terms. © Ipsos MORI 2015. 15-029639-01 | PUBLIC | This work was carried out in accordance with the requirements of the international quality standard for Market Research, ISO 20252:2012, and with the Ipsos MORI Terms and Conditions which can be found at http://www.ipsos-mori.com/terms. © Ipsos MORI 2016. The One-Way Mirror: Public attitudes to commercial access to health data | Report prepared for the Wellcome Trust | February 2016 15-029639-01 | PUBLIC | This work was carried out in accordance with the requirements of the international quality standard for Market Research, ISO 20252:2012, and with the Ipsos MORI Terms and Conditions which can be found at http://www.ipsos-mori.com/terms.","author":[{"dropping-particle":"","family":"MORI for Wellcome Trust","given":"Ipsos","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Trust","given":"IPSOS

MORI for Wellcome","non-dropping-particle":"","parse-names":false,"suffix":""}],"id":"ITEM-1","issue":"March","issued":{"date-parts":[["2016"]]},"title":"The One-Way Mirror: Public attitudes to commercial access to health data Report prepared for the Wellcome Trust","type":"article-journal"},"uris":["http://www.mendeley.com/documents/?uuid=a67ad777-12e2-4611-8121-27af961cb433"]}],"mendeley":{"formattedCitation":"²³","plainTextFormattedCitation":"23","previouslyFormattedCitation":"²³"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}}. Therefore, the desire for control over data sharing with particular types of organisations may reflect uncertainty of risks and benefits of sharing data with these groups. We did not investigate trustworthiness of different research groups; however, participants' support for data sharing to advance research in this study is likely to be determined by the actions of researchers and data repositories, who will need to provide rationale for why data may be shared with separate research groups, particularly with the commercial sector{ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1038/ejhg.2012.104","ISSN":"1018-4813","abstract":"Publics and biobanks: Pan-European diversity and the challenge of responsible innovation","author":[{"dropping-particle":"","family":"Gaskell","given":"George","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Gottweis","given":"Herbert","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Starkbaum","given":"Johannes","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Gerber","given":"Monica M","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Broerse","given":"Jacqueline","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Gottweis","given":"Ursula","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hobbs","given":"Abbi","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Helén","given":"Ilpo","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Paschou","given":"Maria","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Snell","given":"Karoliina","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Soulier","given":"Alexandra","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"European Journal of Human Genetics","id":"ITEM-1","issue":"1","issued":{"date-parts":[["2013","1","6"]]},"page":"14-20","publisher":"Nature Publishing Group","title":"Publics and biobanks: Pan-European diversity and the challenge of responsible innovation","type":"article-journal","volume":"21"},"uris":["http://www.mendeley.com/documents/?uuid=5888627e-48e0-395a-9ad4-f6c6945c0674"]}],"mendeley":{"formattedCitation":"⁵","plainTextFormat tedCitation":"5","previouslyFormattedCitation":"⁵"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}**}**. Offering participants the choice about data sharing may develop trust in the research and researchers. D’Abramo et al**{**ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1186/s12910-015-0053-5","ISSN":"1472-6939","abstract":"Appropriate information and consent has been one of the most intensely discussed topics within the context of biobank research. In parallel to the normative debate, many socio-empirical studies have been conducted to gather experiences, preferences and views of patients, healthy research participants and further stakeholders. However, there is scarcity of literature which connects the normative debate about justifications for different consent models with findings gained in empirical research. In this paper we discuss findings of a limited review of socio-empirical research on patients’ and healthy research participants’ experiences and views regarding consent to biobank research in light of ethical principles for appropriate information and consent. Review question: Which empirical data are available on research participants’ perceptions and views regarding information and elicitation of consent for biobank research? Search of articles published till March 1st 2014 in Pubmed. Review of abstracts and potentially relevant full text articles by two authors independently. As categories for content analysis we defined (i) understanding or recall of information, (ii) preferences regarding information or consent, and (iii) research participants’ concerns. The search in Pubmed yielded 337 abstracts of which 10 articles were included in this study. Approaches to information and consent varied considerably across the selected studies. The majority of research participants opted for some version of limited consent when being informed about such possibility. Among the factors influencing the type of preferred consent were information about sponsoring of biobank research by pharmaceutical industry and participants’ trade-off between privacy and perceived utility. Studies investigating research participants’ understanding and recall regarding the consent procedure indicated considerable lack of both aspects. Research participants’ perceptions of benefits and harms differ across those studies. The knowledge, perceptions and views of research participants who have undergone a consent procedure within the context of biobank research raise several questions on the issue of how to inform and elicit consent in an ethically acceptable way. In our empirical-ethical analysis we develop suggestions on how the practice of eliciting consent in the biobank context should be improved.","author":[{"dropping-particle":"","family":"D’Abramo","given":"Flavio","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Schildmann","given":"Jan","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Vollmann","given":"Jochen","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"BMC Medical Ethics","id":"ITEM-1","issue":"1","issued":{"date-parts":[["2015","12","9"]]},"page":"60","publisher":"BioMed Central","title":"Research participants’ perceptions and views on consent for biobank research: a review of empirical data and ethical analysis","type":"article-journal","volume":"16"},"uris":["http://www.mendeley.com/documents/?uuid=dbaa1e8d-e81e-3a16-990b-b5757b043e82"]}],"mendeley":{"formattedCitation":"⁸","plainTextFormattedCitation":"8","previouslyFormattedCitation":"⁸"},"properties":{"noteIn dex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}} found that attitudes to data sharing become restrictive when more information and options are provided about permitting data in biobanks to be publicly available. Similarly, McGuire et al.{ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1097/GIM.0b013e31815f1e00.DNA","author":[{"dropping-particle":"","family":"McGuire","given":"a.L.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hamilton","given":"J.a.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Lunstroth","given":"R","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"McCullough","given":"L B","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Goldman","given":"a.","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Genetics in medicine: official journal of the American College of Medical Genetics","id":"ITEM-1","issue":"1","issued":{"date-parts":[["2008"]]},"page":"46","title":"DNA data sharing : Research participants' perspectives","type":"article-journal","volume":"10"},"uris":["http://www.mendeley.com/documents/?uuid=95543308-c584-38a5-ae0c-537f5a9e54e9"]}],"mendeley":{"formattedCitation":"¹²","plainTextFormattedCitation":"12","previouslyFormattedCitation":"¹²"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}} reported that participants preferred having multiple data sharing options but were less likely to consent to public data release after being given options. Conversely, Bell et al.

{ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"ISSN":"1942-597X","PMID":"25954442","abstract":"We interviewed 70 healthy volunteers to understand their choices about how the information in their health record should be shared for research. Twenty-eight survey questions captured individual preferences of healthy volunteers. The results showed that respondents felt comfortable participating in research if they were given choices about which portions of their medical data would be shared, and with whom those data would be shared. Respondents indicated a strong preference towards controlling access to specific data (83%), and a large proportion (68%) indicated concern about the possibility of their data being used by for-profit entities. The results suggest that transparency in the process of sharing is an important factor in the decision to share clinical data for research.","author":[{"dropping-particle":"","family":"Bell","given":"Elizabeth A","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Ohno-Machado","given":"Lucila","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Grando","given":"M Adela","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"AMIA ... Annual Symposium proceedings. AMIA Symposium","id":"ITEM-1","issued":{"date-parts":[["2014"]]},"page":"1699-708","publisher":"American Medical Informatics Association","title":"Sharing my health data: a survey of data sharing preferences of healthy individuals.","type":"article-journal","volume":"2014"},"uris":["http://www.mendeley.com/documents/?uuid=0b1c4e44-f1ac-39fc-8a40-

fe05700ea962"]}],"mendeley":{"formattedCitation":"¹⁸","plainTextFormattedCitation":"18","previouslyFormattedCitation":"¹⁸"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}}, having surveyed healthy people about hypothetical data sharing preferences, found that participants wanted to have control over uses of their data to varying degrees, including demographics, lab results, and sensitive information, such as mental health and genetic information. Participants were more willing to share data if they were given choices about what they wanted to share, and a high proportion wanted to know more about the public and private researchers requesting access to their data{ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"ISSN":"1942-597X","PMID":"25954442","abstract":"We interviewed 70 healthy volunteers to understand their choices about how the information in their health record should be shared for research. Twenty-eight survey questions captured individual preferences of healthy volunteers. The results showed that respondents felt comfortable participating in research if they were given choices about which portions of their medical data would be shared, and with whom those data would be shared. Respondents indicated a strong preference towards controlling access to specific data (83%), and a large proportion (68%) indicated concern about the possibility of their data being used by for-profit entities. The results suggest that transparency in the process of sharing is an important factor in the decision to share clinical data for research.","author":[{"dropping-particle":"","family":"Bell","given":"Elizabeth A","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Ohno-Machado","given":"Lucila","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Grando","given":"M Adela","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"AMIA ... Annual Symposium proceedings. AMIA Symposium","id":"ITEM-1","issued":{"date-parts":[["2014"]]},"page":"1699-708","publisher":"American Medical Informatics Association","title":"Sharing my health data: a survey of data sharing preferences of healthy individuals.","type":"article-journal","volume":"2014"},"uris":["http://www.mendeley.com/documents/?uuid=0b1c4e44-f1ac-39fc-8a40-fe05700ea962"]}],"mendeley":{"formattedCitation":"¹⁸","plainTextFormattedCitation":"18","previouslyFormattedCitation":"¹⁸"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}**}**. Whether the extent of control participants have over data shared affects future research participation requires further investigation.

Further cultural factors may affect preferences for control. Gaskell et al.**{**ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1038/ejhg.2012.104","ISSN":"1018-4813","abstract":"Publics and biobanks: Pan-European diversity and the challenge of responsible innovation","author":[{"dropping-particle":"","family":"Gaskell","given":"George","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Gottweis","given":"Herbert","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Starkbaum","given":"Johannes","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Gerber","given":"Monica M","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Broerse","given":"Jacqueline","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Gottweis","given":"Ursula","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hobbs","given":"Abbi","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Helén","given":"Ilpo","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Paschou","given":"Maria","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Snell","given":"Karoliina","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Soulier","given":"Alexandra","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"European Journal of Human Genetics","id":"ITEM-1","issue":"1","issued":{"date-parts":[["2013","1","6"]]},"page":"14-20","publisher":"Nature Publishing Group","title":"Publics and biobanks: Pan-European diversity and the challenge of responsible innovation","type":"article-journal","volume":"21"},"uris":["http://www.mendeley.com/documents/?uuid=5888627e-48e0-395a-9ad4-f6c6945c0674"]}],"mendeley":{"formattedCitation":"⁵","plainTextFormattedCitation":"5","previouslyFormattedCitation":"⁵"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-

citation.json"}} found that in their pan-EU study, willingness to participate in biobank research was affected by beliefs in risk of misuse of data, and public in southern European countries were less likely to participate than in north-western countries. The study reported here was specifically situated in Western Europe and involved high-income countries with good health systems and nations that are viewed as socially inclusive. Designing international consortia data governance would benefit from understanding cultural attributes, if research aims to be inclusive of participants in data sharing decision making. As these findings show, some aspects of data sharing are consistently agreed upon, such as importance of privacy, whereas others are not (differences between countries in deciding with whom it is acceptable to share data). Furthermore, differences found between countries in this study show the diversity of perspectives about data sharing in different populations. Danish respondents indicated higher odds of importance to control data types shared, and Dutch respondents showed higher odds of importance to control who data are shared with. This means that large consortia sourcing data from culturally diverse countries may find it challenging to consistently oversee how data are shared and managed for future research.

Maintaining privacy is central for governance of data sharing in research; results from this study show that privacy is key to the likelihood of wanting control over sharing data. However, there may be ambiguity in understanding what privacy means across different populations{ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1159/000336541","ISSN":"1662-8063","PMID":"22722693","abstract":"Most people in Europe do not know what biobanks are. In this study, public perceptions of biobanks and collection of genetic and health data were analyzed in relation to other technologies and digital networks where

personal information is compiled and distributed. In this setting, people contextualized biobanks in line with their daily experiences with other technologies and data streams. The analysis was based on 18 focus group discussions conducted in Austria, Finland and Germany. We examined the ways in which people frame and talk about problems and benefits of information distribution in digital networks and biobanks. People identify many challenges associated with collection of personal data in the information society. The study showed that instead of privacy - which has been the key term of bioethical debates on biobanks - the notions of control and controllability are most essential for people. From the viewpoint of biobanks, issues of controllability pose challenges. In the information society, people have become accustomed to controlling personal data, which is particularly difficult in relation to biobanks. They expressed strong concerns over the controllability of the goals and benefits of biobanks.","author":[{"dropping-particle":"","family":"Snell","given":"K","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Starkbaum","given":"J","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Lauß","given":"G","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Vermeer","given":"A","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Helén","given":"I","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Public health genomics","id":"ITEM-1","issue":"5","issued":{"date-parts":[["2012"]]},"page":"293-302","publisher":"Karger Publishers","title":"From protection of privacy to control of data streams: a focus group study on biobanks in the information society.","type":"article-journal","volume":"15"},"uris":["http://www.mendeley.com/documents/?uuid=eb96d5d

5-c713-32a5-ae5b-c768d7d6828d"]}],"mendeley":{"formattedCitation":"²⁴","plainTextFormattedCitation":"24","previouslyFormattedCitation":"²⁴"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}**}**. The discourse about privacy being important now needs to shift to how it can be facilitated and in what context data donors require control over data sharing. Lemke et al.**{**ADDIN CSL_CITATION

{"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1159/000276767","ISSN":"1662-4246","abstract":"Research assessing attitudes toward consent processes for high-throughput genomic-wide technologies and widespread sharing of data is limited. In order to develop a better understanding of stakeholder views toward these issues, this cross-sectional study assessed public and biorepository participant attitudes toward research participation and sharing of genetic research data. Forty-nine individuals participated in 6 focus groups; 28 in 3 public focus groups and 21 in 3 NUgene biorepository participant focus groups. In the public focus groups, 75% of participants were women, 75% had some college education or more, 46% were African-American and 29% were Hispanic. In the NUgene focus groups, 67% of participants were women, 95% had some college education or more, and the majority (76%) of participants was Caucasian. Five major themes were identified in the focus group data: (a) a wide spectrum of understanding of genetic research; (b) pros and cons of participation in genetic research; (c) influence of credibility and trust of the research institution; (d) concerns about sharing genetic research data and need for transparency in the Policy for Sharing of Data in National Institutes of Health-Supported or Conducted Genome-Wide Association Studies; (e) a need for more information and education about genetic research. In order to increase public understanding and address potential concerns about genetic research, future efforts should be aimed at involving the public in genetic research policy development and in identifying or developing appropriate educational strategies to meet the public's needs.","author":[{"dropping-particle":"","family":"Lemke","given":"A A","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Wolf","given":"W A","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hebert-Beirne","given":"J","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Smith","given":"M E","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Public Health Genomics","id":"ITEM-1","issue":"6","issued":{"date-parts":[["2010"]]},"language":"eng","page":"368-377","title":"Public and biobank participant attitudes toward genetic research participation and data sharing","type":"article-journal","volume":"13"},"uris":["http://www.mendeley.com/documents/?uuid=7edacfe1-ef8a-3320-9240-3dfd41b37094"]}],"mendeley":{"formattedCitation":"⁷","plainTextFormattedCitation":"7","previouslyFormattedCitation":"⁷"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}**}** argued that participants wanted control over release of genetic information and that mechanisms to protect privacy needed to be provided. In consideration of this, it is ethically important to provide research participants with options to control sharing their study data, even after its anonymization. This could be facilitated by having simple mechanisms for choosing preferences, and further research about which (and how many) choices are needed, so as not to overburden participants{ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1097/GIM.0b013e31815f1e00.DNA","author":[{"dropping-particle":"","family":"McGuire","given":"a.L.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hamilton","given":"J.a.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Lunstroth","given":"R","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"McCullough","given":"L B","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Goldman","given":"a.","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Genetics in medicine: official journal of the American College of Medical Genetics","id":"ITEM-1","issue":"1","issued":{"date-parts":[["2008"]]},"page":"46","title":"DNA data sharing : Research participants' perspectives","type":"article-journal","volume":"10"},"uris":["http://www.mendeley.com/documents/?uuid=95543308-c584-38a5-ae0c-537f5a9e54e9"]}],"mendeley":{"formattedCitation":"¹²","plainTextFormattedCitation":"12","previouslyFormattedCitation":"¹²"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}}. With divergences in attitudes to control data sharing, how the availability of control mechanisms is facilitated will require addressing. The first step is for international consortia to communicate and engage with participants to assess preferences for data sharing{ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1038/ejhg.2012.104","ISSN":"1018-4813","abstract":"Publics and biobanks: Pan-European diversity and the challenge of responsible innovation","author":[{"dropping-particle":"","family":"Gaskell","given":"George","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Gottweis","given":"Herbert","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Starkbaum","given":"Johannes","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Gerber","given":"Monica M","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Broerse","given":"Jacqueline","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Gottweis","given":"Ursula","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hobbs","given":"Abbi","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Helén","given":"Ilpo","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Paschou","given":"Maria","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Snell","given":"Karoliina","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Soulier","given":"Alexandra","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"European Journal of Human Genetics","id":"ITEM-1","issue":"1","issued":{"date-parts":[["2013","1","6"]]},"page":"14-20","publisher":"Nature Publishing Group","title":"Publics and biobanks: Pan-European diversity and the challenge of responsible innovation","type":"article-journal","volume":"21"},"uris":["http://www.mendeley.com/documents/?uuid=5888627e-48e0-395a-9ad4-f6c6945c0674"]},{"id":"ITEM-2","itemData":{"DOI":"10.1586/14737159.2014.961917","ISSN":"1473-7159","abstract":"Aim: Introducing data sharing practices into the genomic research arena has challenged the current mechanisms established to protect rights of individuals and triggered policy considerations. To inform such policy deliberations, soliciting public and research participants' attitudes with respect to genomic data sharing is a necessity. Method: The main electronic databases were searched in order to retrieve empirical studies, investigating the attitudes of research participants and the public towards genomic data sharing through public databases. Results: In the 15 included studies, participants' attitudes towards genomic data sharing revealed the influence of a constellation of interrelated factors, including the personal perceptions of controllability and sensitivity of data, potential risks and benefits of data sharing at individual and social level and also governance level considerations. Conclusion: This analysis indicates that future policy responses and recruitment practices should be attentive to a wide variety of concerns in order to promote both responsible and progressive research.","author":[{"dropping-particle":"","family":"Shabani","given":"Mahsa","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Bezuidenhout","given":"Louise","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Borry","given":"Pascal","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Expert review of molecular diagnostics","id":"ITEM-2","issue":"SEPTEMBER","issued":{"date-parts":[["2014"]]},"page":"1-13","title":"Attitudes of research participants and the general public towards genomic data sharing: a systematic literature review.","type":"article-journal"},"uris":["http://www.mendeley.com/documents/?uuid=ea1aa3f4-4ebe-36c1-9b92-8d3077e17819"]},{"id":"ITEM-3","itemData":{"DOI":"10.1097/GIM.0b013e31815f1e00.DNA","author":[{"dropping-particle":"","family":"McGuire","given":"a.L.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hamilton","given":"J.a.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Lunstroth","given":"R","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"McCullough","given":"L B","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Goldman","given":"a.","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Genetics in medicine: official journal of the American College of Medical Genetics","id":"ITEM-3","issue":"1","issued":{"date-parts":[["2008"]]},"page":"46","title":"DNA data sharing : Research participants' perspectives","type":"article-journal","volume":"10"},"uris":["http://www.mendeley.com/documents/?uuid=95543308-c584-38a5-ae0c-537f5a9e54e9"]},{"id":"ITEM-

4","itemData":{"DOI":"10.1097/GIM.0b013e3182227589","abstract":"PURPOSE: Despite growing concerns toward maintaining participants' privacy, individual investigators collecting tissue and other biological specimens for genomic analysis are encouraged to obtain informed consent for broad data sharing. Our purpose was to assess the effect on research enrollment and data sharing decisions of three different consent types (traditional, binary, or tiered) with varying levels of control and choices regarding data sharing., METHODS: A single-blinded, randomized controlled trial was conducted with 323 eligible adult participants being recruited into one of six genome studies at Baylor College of Medicine in Houston, Texas, between January 2008 and August 2009. Participants were randomly assigned to one of three experimental consent documents (traditional, n = 110; binary, n = 103; and tiered, n = 110). Debriefing in follow-up visits provided participants a detailed review of all consent types and the chance to change data sharing choices or decline genome study participation., RESULTS: Before debriefing, 83.9% of participants chose public data release. After debriefing, 53.1% chose public data release, 33.1% chose restricted (controlled access database) release, and 13.7% opted out of data sharing. Only one participant declined genome study participation due to data sharing concerns., CONCLUSION: Our findings indicate that most participants are willing to publicly release their genomic data; however, a significant portion prefers restricted release. These results suggest discordance between existing data sharing policies and participants' judgments and desires.","author":[{"dropping-particle":"","family":"McGuire","given":"Amy L","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Oliver","given":"Jill M","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Slashinski","given":"Melody J","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Graves","given":"Jennifer L","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Wang","given":"Tao","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Kelly","given":"P Adam","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Fisher","given":"William","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Lau","given":"Ching C","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Goss","given":"John","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Okcu","given":"Mehmet","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Treadwell-Deering","given":"Diane","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Goldman","given":"Alica M","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Noebels","given":"Jeffrey L","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hilsenbeck","given":"Susan G","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Genetics in medicine : official journal of the American College of Medical Genetics","id":"ITEM-4","issue":"11","issued":{"date-parts":[["2011","11"]]},"page":"948-955","publisher":"McGuire,Amy L. Center for Medical Ethics and Health Policy, Baylor College of Medicine, Houston, TX 77030, USA. amcguire@bcm.edu","publisher-place":"United States","title":"To share or not to share: a randomized trial of consent for data sharing in genome research.","type":"article-journal","volume":"13"},"uris":["http://www.mendeley.com/documents/?uuid=41622375-a686-30c9-9cad-f425c1a24015"]}],"mendeley":{"formattedCitation":"^{5,11,12,25}","plainTextFormattedCitation":"5,11,12,25","previouslyFormattedCitation":"^{5,11,12,25}"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}}.

The importance to engage and involve study participants in research decisions is very timely. One proposed solution to facilitate participant engagement with future data sharing decisions is Dynamic Consent{ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1038/ejhg.2014.71","author":[{"dropping-particle":"","family":"Kaye","given":"Jane","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Whitley","given":"Edgar A","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Lund","given":"David","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Morrison","given":"Michael","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Teare","given":"Harriet","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Melham","given":"Karen","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"European Journal of Human Genetics","id":"ITEM-1","issue":"2","issued":{"date-parts":[["2015"]]},"page":"141-

146","publisher":"Nature Publishing Group","title":"Dynamic consent : a patient interface for twenty-first century research networks","type":"article-journal","volume":"23"},"uris":["http://www.mendeley.com/documents/?uuid=4d86aed8-f49b-3cec-accf-f80e3445d9dd"]}],"mendeley":{"formattedCitation":"²⁶","plainTextFormattedCitation":"26","previouslyFormattedCitation":"²⁶"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}}. This approach provides participants with an electronic record of their consent decisions, which can be reviewed and updated at any time. This would allow those that wanted greater control to be more directly involved in real-time decision-making, and could potentially provide an infrastructure to support participants beyond the lifetime of a specific project

{ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1186/s12910-016-0162-9","ISSN":"1472-6939","abstract":"Innovations in technology have contributed to rapid changes in the way that modern biomedical research is carried out. Researchers are increasingly required to endorse adaptive and flexible approaches to accommodate these innovations and comply with ethical, legal and regulatory requirements. This paper explores how Dynamic Consent may provide solutions to address challenges encountered when researchers invite individuals to participate in research and follow them up over time in a continuously changing environment. An interdisciplinary workshop jointly organised by the University of Oxford and the COST Action CHIP ME gathered clinicians, researchers, ethicists, lawyers, research participants and patient representatives to discuss experiences of using Dynamic Consent, and how such use may facilitate the conduct of specific research tasks. The data collected during the workshop were analysed using a content analysis approach. Dynamic Consent can provide practical, sustainable and future-proof solutions to challenges related to participant recruitment, the attainment of informed consent, participant retention and consent management, and may bring economic efficiencies. Dynamic Consent offers opportunities for ongoing communication between researchers and research participants that can positively impact research. Dynamic Consent supports inter-sector, cross-border approaches and large scale data-sharing. Whilst it is relatively easy to set up and maintain, its implementation will require that researchers re-consider their relationship with research participants and adopt new procedures.","author":[{"dropping-particle":"","family":"Budin-Ljøsne","given":"Isabelle","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Teare","given":"Harriet J. A.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Kaye","given":"Jane","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Beck","given":"Stephan","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Bentzen","given":"Heidi Beate","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Caenazzo","given":"Luciana","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Collett","given":"Clive","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"D’Abramo","given":"Flavio","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Felzmann","given":"Heike","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Finlay","given":"Teresa","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Javaid","given":"Muhammad Kassim","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Jones","given":"Erica","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Katić","given":"Višnja","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Simpson","given":"Amy","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Mascalzoni","given":"Deborah","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"BMC Medical Ethics","id":"ITEM-1","issue":"1","issued":{"date-parts":[["2017","12","25"]]},"page":"4","publisher":"BioMed Central","title":"Dynamic Consent: a potential solution to some of the challenges of modern biomedical research","type":"article-journal","volume":"18"},"uris":["http://www.mendeley.com/documents/?uuid=438bb51e-b887-3fdd-a05c-aa2803d89aff"]}],"mendeley":{"formattedCitation":"²⁷","plainTextFormattedCitation":"27","previouslyFormattedCitation":"²⁷"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}**}**. Based on the findings of this study, this may be a relevant solution to manage future involvement, and thus would be appropriate to investigate further.

Strengths and Limitations

This was a unique study in that it looked at participants already enrolled in research to engage their views about how their data should be shared for future research. Few studies have investigated data sharing choices of patients from different countries in Europe. However, there are also a number of limitations that must be discussed. Firstly, the results are not generalizable to other patient or healthy populations or countries. Countries included were in the north-western region of Europe, and there may be marked differences in data sharing opinions with other European countries, and between non-white population groups. Due to the socio-demographic and personal characteristics, participation may have been influenced by already being enrolled in DIRECT studies, and data sharing opinions referred to data that would be de-identified. In addition to this, the cross-sectional nature of the study design meant that it was difficult to ascertain whether respondents' views would change over time and with more information about data sharing options, as we did not investigate the level of awareness respondents had about data sharing for future research. Also, collapsing the Likert survey questions from 5 to binary variables removes nuances in opinions of respondents about a given issue. Respondents' views could potentially have been influenced by their level of confidence in the effectiveness of de-identification of their data in protecting privacy

{ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1016/j.ijmedinf.2015.01.008","ISSN":"1386-5056","abstract":"The development of Electronic Health Records (EHRs) forms an integral part of the information strategy for the National Health Service (NHS) in the UK,

with the aim of facilitating health information exchange for patient care and secondary use, including research and healthcare planning. Implementing EHR systems requires an understanding of patient expectations for consent mechanisms and consideration of public awareness towards information sharing as might be made possible through integrated EHRs across primary and secondary health providers. Objectives: To explore levels of public awareness about EHRs and to examine attitudes towards different consent models with respect to sharing identifiable and de-identified records for healthcare provision, research and planning. Methods: A cross-sectional questionnaire survey was administered to adult patients and members of the public in primary and secondary care clinics in West London, UK in 2011. In total, 5331 individuals participated in the survey, and 3157 were included in the final analysis. Results: The majority (91%) of respondents expected to be explicitly asked for consent for their identifiable records to be accessed for health provision, research or planning. Half the respondents (49%) did not expect to be asked for consent before their de-identified records were accessed. Compared with White British respondents, those from all other ethnic groups were more likely to anticipate their permission would be obtained before their de-identified records were used. Of the study population, 59% reported already being aware of EHRs before the survey. Older respondents and individuals with complex patterns of interaction with healthcare services were more likely to report prior awareness of EHRs. Individuals self-identifying as belonging to ethnic groups other than White British, and those with lower educational qualifications were less likely to report being aware of EHRs than White British respondents and respondents with degree-level education, respectively. Those who reported being aware of EHRs were less likely to say they expected explicit consent to be sought before use of their de-identified record. Conclusions: A large number of patients

remain unaware of EHRs, while preference for implicit consent is stronger among those who report previous awareness. Differences in awareness levels and consent expectations between groups with different socio-demographic characteristics suggest that public education…","author":[{"dropping-particle":"","family":"Riordan","given":"Fiona","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Papoutsi","given":"Chrysanthi","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Reed","given":"Julie E","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Marston","given":"Cicely","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Bell","given":"Derek","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Majeed","given":"Azeem","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"International Journal of Medical Informatics","id":"ITEM-1","issue":"4","issued":{"date-parts":[["2015"]]},"page":"237-247","publisher":"Elsevier Ireland Ltd","title":"Patient and public attitudes towards informed consent models and levels of awareness of Electronic Health Records in the UK","type":"article-journal","volume":"84"},"uris":["http://www.mendeley.com/documents/?uuid=246ff818-1af8-331a-bead-ba5ddd374267"]}],"mendeley":{"formattedCitation":"¹⁴","plainTextFormattedCitation":"14","previouslyFormattedCitation":"¹⁴"},"properties":{"no teIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}}.

Conclusions

As it is responsible practice to obtain informed consent from participants to share their data{ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1097/GIM.0b013e31815f1e00.DNA","author":[{"dropping-particle":"","family":"McGuire","given":"a.L.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hamilton","given":"J.a.","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Lunstroth","given":"R","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"McCullough","given":"L B","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Goldman","given":"a.","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Genetics in medicine: official journal of the American College of Medical Genetics","id":"ITEM-1","issue":"1","issued":{"date-parts":[["2008"]]},"page":"46","title":"DNA data sharing : Research participants' perspectives","type":"article-journal","volume":"10"},"uris":["http://www.mendeley.com/documents/?uuid=95543308-c584-38a5-ae0c-537f5a9e54e9"]}],"mendeley":{"formattedCitation":"¹²","plainTextFormattedCitation":"12","previouslyFormattedCitation":"¹²"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}}, it should also be responsible practice to involve participants in decisions about how their data should be governed. Our findings indicate that what research participants expect in terms of control over data sharing needs to be considered and aligned with sharing for future research and re-use of data

{ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1097/GIM.0b013e31818bb3ab","ISSN":"1530-0366 (Electronic)\\n1530-0366 (Linking)","abstract":"PURPOSE: Cohort studies investigating genes, environment, and lifestyle require large study populations. To recruit and retain participants, it is important to understand the relative significance of influences on people's motivation to participate. To this end, 4659 Americans were surveyed about support for and willingness to participate in a proposed large cohort study. METHODS: An online survey of US adults was conducted between December 2007 and January 2008. To measure the influence of study burden, compensation and receipt of individual research results on willingness to participate, respondents were randomized to one of eight different study scenarios. RESULTS: Most respondents (84%) supported the study, and 60% would participate. Returning research results (odds ratio = 1.6, 95% confidence interval 1.3-1.8) and increasing compensation from $50 to $200 (odds ratio = 1.5, 95% confidence interval 1.2-1.7) were associated with increased willingness to participate. Decreasing study burden was less important (odds ratio = 1.2, 95% confidence interval 1.0-1.4). Three in four respondents would be less likely to participate without the return of research results. Support and willingness varied little among demographic groups; variation in influences of the three factors on willingness was observed. CONCLUSION: Widespread support exists in the general public for a large national cohort study. Providing individual research results is a strong motivation to participate; compensating participants $200 may increase participation a similar amount. Incentives, recruitment, and return of results could be tailored to demographics groups' interests.","author":[{"dropping-particle":"","family":"Kaufman","given":"David","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Murphy","given":"Juli","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Scott","given":"Joan","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hudson","given":"Kathy","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Genetics in medicine : official journal of the American College of Medical Genetics","id":"ITEM-1","issue":"11","issued":{"date-parts":[["2008"]]},"page":"831-839","title":"Subjects matter: a survey of public opinions about a large genetic cohort study.","type":"article-journal","volume":"10"},"uris":["http://www.mendeley.com/documents/?uuid=da75dfdd-8e38-367b-9caa-4d2facdb6649"]}],"mendeley":{"formattedCitation":"²","plainTextFormattedCitation":"2","previouslyFormattedCitation":"²"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}**}**. There is a balance to be struck between protecting privacy and benefits to biomedical research from data sharing**{**ADDIN CSL_CITATION {"citationItems":[{"id":"ITEM-1","itemData":{"DOI":"10.1371/journal.pmed.1001937","ISSN":"10.1371/journal.pmed.1001937","abstract":"With the prospect of genomic data becoming ever more easily available, Effy Vayena and Urs Gasser discuss how we could balance making the most of its benefits with reducing its risks to privacy.","author":[{"dropping-particle":"","family":"Vayena","given":"Effy","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Gasser","given":"Urs","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"PLoS Medicine","id":"ITEM-1","issue":"1","issued":{"date-parts":[["2016","1","12"]]},"page":"e1001937","publisher":"Public Library of Science","title":"Between Openness and Privacy in Genomics","type":"article-journal","volume":"13"},"uris":["http://www.mendeley.com/documents/?uuid=7db2733f-dbdb-492c-90bc-a19e64f831fd"]},{"id":"ITEM-2","itemData":{"DOI":"10.1097/GIM.0b013e3182227589","abstract":"PURPOSE: Despite growing concerns toward maintaining participants' privacy, individual investigators collecting tissue and other biological specimens for genomic analysis are encouraged to obtain informed consent for broad data sharing. Our purpose was to assess the effect on research enrollment and data sharing decisions of three different consent types (traditional, binary, or tiered) with varying levels of control and choices regarding data sharing., METHODS: A single-blinded, randomized controlled trial was conducted with 323 eligible adult participants being recruited into one of six genome studies at Baylor College of Medicine in Houston, Texas, between January 2008 and August 2009. Participants were randomly assigned to one of three experimental consent documents (traditional, n = 110; binary, n = 103; and tiered, n = 110). Debriefing in follow-up visits provided participants a detailed review of all consent types and the chance to change data sharing choices or decline genome study participation., RESULTS: Before debriefing, 83.9% of participants chose public data release. After debriefing, 53.1% chose public data release, 33.1% chose restricted (controlled access database) release, and 13.7% opted out of data sharing. Only one participant declined genome study participation due to data sharing concerns., CONCLUSION: Our findings indicate that most participants are willing to publicly release their genomic data; however, a significant portion prefers restricted release. These results suggest discordance between existing data sharing policies and participants' judgments and desires.","author":[{"dropping-particle":"","family":"McGuire","given":"Amy L","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Oliver","given":"Jill M","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Slashinski","given":"Melody J","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Graves","given":"Jennifer L","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Wang","given":"Tao","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Kelly","given":"P Adam","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Fisher","given":"William","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Lau","given":"Ching C","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Goss","given":"John","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Okcu","given":"Mehmet","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Treadwell-Deering","given":"Diane","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Goldman","given":"Alica M","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Noebels","given":"Jeffrey L","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"Hilsenbeck","given":"Susan G","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Genetics in medicine : official journal of the American College of Medical Genetics","id":"ITEM-2","issue":"11","issued":{"date-parts":[["2011","11"]]},"page":"948-955","publisher":"McGuire,Amy L. Center for Medical Ethics and Health Policy, Baylor College of Medicine, Houston, TX 77030, USA. amcguire@bcm.edu","publisher-place":"United States","title":"To share or not to share: a randomized trial of consent for data sharing in genome research.","type":"article-journal","volume":"13"},"uris":["http://www.mendeley.com/documents/?uuid=41622375-a686-30c9-9cad-f425c1a24015"]},{"id":"ITEM-3","itemData":{"DOI":"10.1159/000324705","ISSN":"1662-8063 (Electronic)\\r1662-4246 (Linking)","abstract":"BACKGROUND: Genomics research data are often widely shared through a variety of mechanisms including publication, meetings and online databases. Re-identification of research participants from sequence data has been shown possible, raising concerns of participants' privacy. METHODS: In 2008-09, we convened 10 focus groups in Durham, N.C. to explore attitudes about how genomic research data were shared amongst the research community, communication of these practices to participants and how different policies might influence participants' likelihood to consent to a genetic/genomic study. Focus groups were audio-recorded and transcripts were complemented by a short anonymous survey. Of 100 participants, 73% were female and 76% African-American, with a median age of 40-49 years. RESULTS: Overall, we found that discussants expressed concerns about privacy and confidentially of data shared through online databases. Although discussants recognized the benefits of data-sharing, they believed it was important to inform research participants of a study's data-sharing plans during the informed consent process. Discussants were significantly more likely to participate in a study that planned to deposit data in a restricted access online database compared to an open access database (p < 0.00001). CONCLUSIONS: The combination of the potential loss of privacy with concerns about data access and identity of the research sponsor warrants disclosure about a study's data-sharing plans during the informed consent process.","author":[{"dropping-particle":"","family":"Haga","given":"S B","non-dropping-particle":"","parse-names":false,"suffix":""},{"dropping-particle":"","family":"O'Daniel","given":"J","non-dropping-particle":"","parse-names":false,"suffix":""}],"container-title":"Public Health Genomics","id":"ITEM-3","issue":"6","issued":{"date-parts":[["2011"]]},"language":"eng","page":"319-324","title":"Public perspectives regarding data-sharing practices in genomics research","type":"article-journal","volume":"14"},"uris":["http://www.mendeley.com/documents/?uuid=826f9016-064a-3189-80f0-528e470a4d08"]}],"mendeley":{"formattedCitation":"^{11,17,28}","plainTextFormattedCitation":"11,17,28","previouslyFormattedCitation":"^{11,17,28}"},"properties":{"noteIndex":0},"schema":"https://github.com/citation-style-language/schema/raw/master/csl-citation.json"}**}**. Contributing to the data sharing governance literature, this study argues to move research participants from passive participation in biomedical research to considering their opinions about data sharing and control of de-identified biomedical data. Our findings show that even with de-identified data, respondents prioritise privacy above all else. However, this does not shut data sharing down, this is consistent across all countries investigated. Though some differences between countries in attitudes towards data sharing and need for control were found, it is important not to presume that participants do not wish to be kept informed about study procedures moving forward. These findings will aid the development of future data sharing policy for the DIRECT consortium. While this study was conducted prior to the introduction of the General Data Protection Regulations (GDPR) in Europe, it aligned with the GDPR's emphasis of understanding the preferences of those whose personal data is processed within the lens of privacy by design. While, consortia must adhere to regulatory governance; it can additionally develop specific data governance practices as appropriate through adopting evidence based and well supported engagement and involvement guidelines and policies.

Acknowledgements

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References

{ADDIN Mendeley Bibliography CSL_BIBLIOGRAPHY }

Tables

Table 1 –Beliefs about advancing research and protecting privacy, and risk-benefits assessments to sharing genetic information [a, b]

	UK		**Denmark**		**Sweden**		**The Netherlands**		**Overall**		***p*[c]**
	N	**%**	**N**	**%**	**N**	**%**	**N**	**%**	**N**	**%**	
It is important to me to advance research as quickly as possible [c]											
Strongly disagree and disagree	5	1.2%	4	1.5%	1	1.8%	0	0.0%	10	1.2%	0.04
Neither disagree nor agree	34	8.3%	37	14.0%	1	1.8%	8	7.2%	80	9.5%	
Strongly agree and agree	370	90.5%	223	84.5%	53	96.4%	103	92.8%	749	89.3%	
It is important to me that my privacy is protected [c]											
Strongly disagree and disagree	24	6.0%	18	6.8%	4	7.1%	7	6.3%	53	6.4%	0.495

Neither disagree nor agree	68	16.9%	36	13.7%	10	17.9%	26	23.2%	140	16.8%	
Strongly agree and agree	310	77.1%	209	79.5%	42	75.0%	79	70.5%	640	76.8%	
There are benefits to sharing my genetic information [c]											
Strongly disagree and disagree	5	1.2%	2	.8%	0	0.0%	0	0.0%	7	0.8%	<0.001
Neither disagree nor agree	23	5.6%	43	16.3%	7	13.0%	30	27.5%	103	12.3%	
Strongly agree and agree	382	93.2%	218	82.9%	47	87.0%	79	72.5%	726	86.8%	
There are risks to sharing my genetic information [c]											
Strongly agree and agree	213	53.4%	119	45.4%	21	41.2%	25	23.8%	378	46.3	<0.001

Neither agree nor disagree	111	27.8%	96	36.6%	15	29.4%	57	54.3%	279	34.1	
Strongly disagree and disagree	75	18.8%	47	17.9%	15	29.4%	23	21.9%	160	19.6	

[a] Likert Scale responses collapsed due to small counts in extreme categories.

[b] Not all respondents answered all questions

[c] Pearson chi-square tests assessing association between countries and privacy and research attitudes, and beliefs about risks and benefits to sharing genetic information.

Table 2: Multivariate Logistic regressions – Differences between countries in importance for respondent's to decide what data types are shared and who data is shared with (important versus not important [a, b])

	UK	**Denmark**			**Sweden**			**The Netherlands**		
		OR	CI	p	OR	CI	p	OR	CI	P
How important is it that you decide what types of data from this study are shared? **(N = 749)[c, d]**										
	REF	0.85	0.44-1.63	0.625	0.70	0.35-1.40	0.314	0.64	0.38-1.10	0.106
How important is it that you decide who your data is shared with? **(N = 747)[c, d]**										
	REF	0.93	0.47-1.85	0.838	0.92	0.44-1.93	0.819	0.68	0.38-1.23	0.201

[a] Adjusted odds by: (categorical variables) diabetes diagnosis, gender, age, educational level, country, having ever worked in health or medical related job, self-rated health, and self-rated knowledge; (continuous variables) It is important to advance research quickly, It is important that my privacy is protected, There are benefits to sharing my genetic information, There are risks to sharing my genetic information, Happiness to share Medical history, Happiness to share Genetic information, Happiness to share Blood test results, Happiness to share Lifestyle information, and Happiness to share Personal information.

[b] REF: Not important

[c] Questions collapsed from 5-point Likert Scale (Not at all important (1) to Extremely Important (5)); Respondents stating if they thought control over data sharing was 'Not at all important', 'Fairly unimportant' and 'Neither important nor unimportant' were grouped as 'Not important'; those rating 'Fairly important' and 'Extremely important' were grouped as 'Important'. The 'I don't know' and 'Prefer not to say' options were treated as missing because of minimal or zero counts.

d Complete cases only, as not all respondents answered all questions

Table 3: Multivariate Logistic regression – Importance for respondent's to decide what data types are shared (important versus not important [a, b])

	All Countries N=749[c]		
	OR	CI	P
It is important to advance research quickly (Likert: 1 = Strongly disagree to 5 = Strongly agree)	0.97	0.64-1.467	0.884
It is important that my privacy is protected (Likert: 1 = Strongly disagree to 5 = Strongly agree)	1.86	1.377-2.515	<0.001
There are benefits to sharing my genetic information (Likert: 1 = Strongly disagree to 5 = Strongly agree)	1.26	0.835-1.912	0.269
There are risks to sharing my genetic information (Likert scale: 1 = Strongly agree, 5 = Strongly disagree)	0.82	0.669-1.006	0.057
Happiness to share Medical history (Likert: 1 = Very unhappy to 5 = Very happy)	1.01	0.605-1.671	0.982
Happiness to share Genetic information (Likert: 1 = Very unhappy to 5 = Very happy)	0.92	0.526-1.594	0.756
Happiness to share Blood test results (Likert: 1 = Very unhappy to 5 = Very happy)	1.77	0.850-3.687	0.127
Happiness to share Lifestyle information (Likert: 1 = Very unhappy to 5 = Very happy)	0.5	0.295-0.837	0.009
Happiness to share Personal information (Likert: 1 = Very unhappy to 5 = Very happy)	0.65	0.517-0.804	<0.001

[a] Adjusted odds by: diabetes diagnosis, gender, age, educational level, country, having ever worked in health or medical related job, self-rated health, and self-rated knowledge.

[b] REF: Not important

c Complete cases only, as not all respondents answered all questions.

Table 4: Multivariate Logistic regression – Importance for respondents to decide who data is shared with (important versus not important [a, b])

	All Countries N=747[c]		
	OR	CI	P
It is important to advance research quickly (Likert: 1 = Strongly disagree to 5 = Strongly agree)	1.22	0.772 - 1.915	0.398
It is important that my privacy is protected (Likert: 1 = Strongly disagree to 5 = Strongly agree)	2.26	1.627 - 3.142	<0.001
There are benefits to sharing my genetic information (Likert: 1 = Strongly disagree to 5 = Strongly agree)	1.64	1.058 - 2.555	0.027
There are risks to sharing my genetic information (Likert scale: 1 = Strongly agree, 5 = Strongly disagree)	0.74	0.594 - 0.915	0.005
Happiness to share with Research teams in European universities (Likert: 1 = Very unhappy to 5 = Very happy)	1.88	0.702 - 5.038	0.209
Happiness to share with Research teams in universities around the world (Likert: 1 = Very unhappy to 5 = Very happy)	0.34	0.131 - 0.86	0.023
Happiness to share with Government funded organisations involved with health research (Likert: 1 = Very unhappy to 5 = Very happy)	1.61	0.878 - 2.935	0.124
Happiness to share with Commercial research companies (e.g. drug companies) (Likert: 1 = Very unhappy to 5 = Very happy)	0.43	0.325 - 0.556	<0.001
Happiness to share with Charities involved in research (Likert: 1 = Very unhappy to 5 = Very happy)	0.57	0.388 - 0.84	0.004

Happiness to share with Patient organisations involved in research (Likert: 1 = Very unhappy to 5 = Very happy)	1.55	0.985 - 2.44	0.058

[a] Adjusted odds by: diabetes diagnosis, gender, age, educational level, country, having ever worked in health or medical related job, self-rated health, and self-rated knowledge.

[b] Reference = not important

[c] Complete cases only, as not all respondents answered all questions.

Figure 1. Importance of control over types of data shared from the DIRECT project

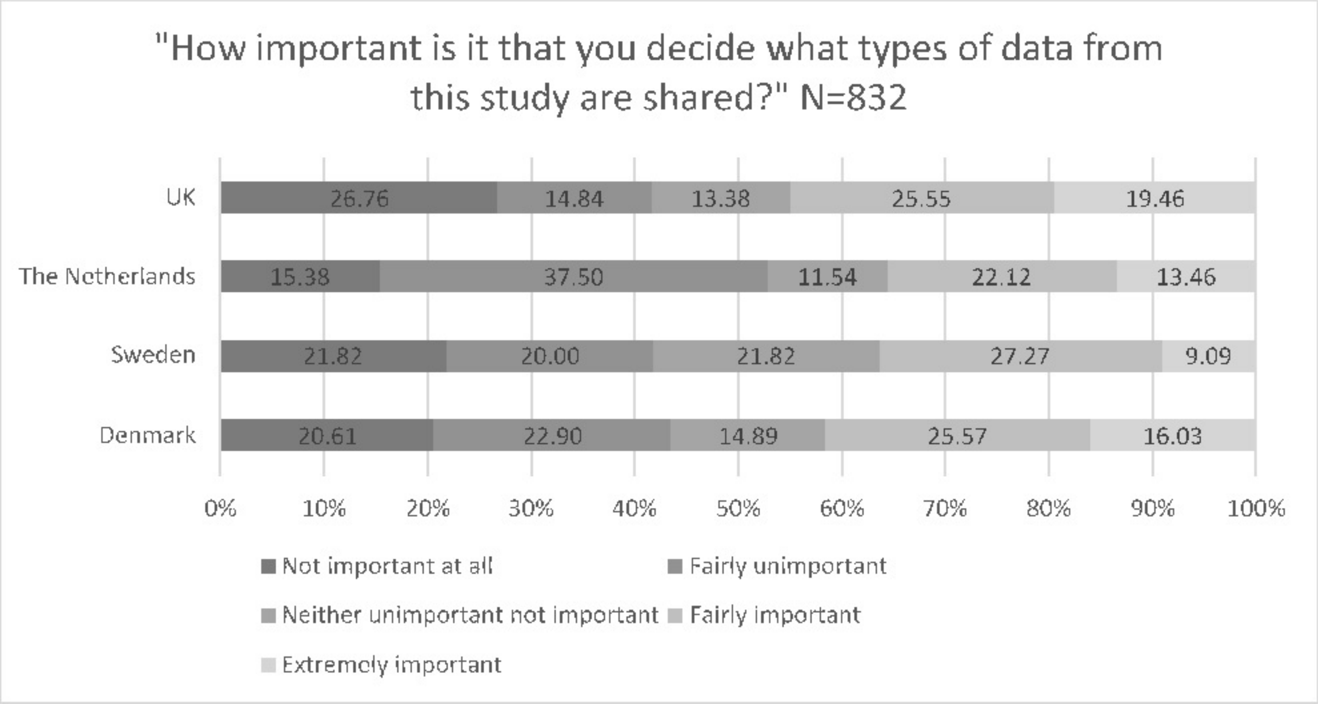

Figure 2. Importance of control over who data is shared with