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Allocation of Resources to Communication of Research Result Summaries: Biobank Participant Perspectives

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Abstract

Introduction—Researchers and policymakers recommend communicating summary research results to biobank participants when feasible. To date, however, there have been few explorations of participant preferences for dedicating resources to this activity.

Methods—Fifteen semi-structured interviews were conducted with participants of a genetic medicine biobank. Participants were interviewed by phone about their motivation for participation, and opinions about the allocation of resources to communicating summary results. De-identified transcripts were used for a directed content analysis.

Results—Most biobank participation was altruistic. All participants were interested in receiving summary results, but also expressed a clear preference for allocating limited funds to conducting additional genetic research.

Conclusions—The results suggest that participants have a nuanced view about the allocation of biobank resources to returning summary results and asking their opinion is a valuable exercise. Researchers may benefit from transparency about research goals, and involving biobank participants in decisions about return of summary results.

Keywords

data sharing; biobank; aggregate results; allocation of resources

INTRODUCTION

Researchers and policymakers recommend returning summary results from biobank research to the research participants (Beskow, Burke, Fullerton, & Sharp, 2012; Wolf et al., 2012). In

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contrast to *individual* results, *summary* (or aggregate) research results typically include synthesized data and conclusions drawn from groups of study participants (such as results about the association between certain gene variants and risk of cancer). Return recommendations are founded on a sense that communicating results to participants in long-term projects, such as a biobank, expresses respect and appreciation for participation; and may improve trust in the research institution, as well as in the partnership relationship between researchers and participants (Lemke, Wolf, Hebert-Beirne, & Smith, 2010). Some have taken this one step further, arguing for an ethical “duty” to provide summary results, but others have pushed back, arguing for more for a more careful conceptual development regarding what type of results to provide to participants under what circumstances (Miller, Christensen, Giacomini, & Robert, 2008).

Returning summary results to participants of biobank research may also have downsides, namely cost in time and other resources (Beskow et al., 2012; Partridge & Winer, 2002; VA National Center for Ethics in Health Care, 2009). Researchers have pointed out the distinction between individual and summary results is not always clear to participants and could result in unintended demand for individual results (Beskow et al., 2012; Miller et al., 2008), which is costly and complex to address (Bledsoe et al., 2013). Maintaining the infrastructure of biobanks (sample and data management activities) is also expensive and the availability of public funding for these activities is limited (Baker, 2012). Some researchers have questioned the investment of time and expense associated with the return of aggregate research results (Dixon-Woods, Jackson, Windridge, & Kenyon, 2006; Partridge & Winer, 2002).

Thought leaders have suggested that participants’ perspectives on such issues would be valuable to incorporate into policy and practice (Beskow et al., 2012; Dalal et al., 2010). However, to date there have been few explorations of participant perspectives on receiving summary results, particularly their opinion regarding the allocation of resources to this activity. A better understanding of the value biobank participants attach to summary results, and whether this value is related to their motivation to participate, may help to refine how future studies can best be designed to acknowledge and respect participant preferences.

To explore biobank participant perspectives about the allocation of resources to the communication of summary results, we conducted qualitative interviews with patients who had recently given a DNA sample and provided open-ended consent for researchers to use data available in their medical record for future genetic analyses.

METHODS

Settings and Participants

The study was conducted at Group Health, a large nonprofit health care system that coordinates care and coverage in the Pacific Northwest for approximately 600,000 patients. Participating patients had previously elected to participate in the Northwest Institute for Genetic Medicine [NWIGM] Biobank, enrolling between October 2009 and September 2011. The NWIGM Biobank currently includes genetic and medical record data from 2073 Group Health patients who were age 50–65 and had completed an online health risk

assessment [HRA] at the time of enrollment. The NWIGM biobank was created to link genetic data with self-reported health and behavior data as well as electronic medical record data, so individuals were invited based on their completion the HRA. The age group was chosen because those participants would have a longer enrollment history with Group Health and would be more likely to have chronic conditions of public health impact.

Recruitment

From January to March 2012, a subset of NWIGM Biobank participants were invited by mail to participate in a follow-up phone interview about their participation in the NWIGM Biobank and genetics research in general. Participants were stratified according to sex and minority status and a sample was selected from among the strata. Non-white and male participants were oversampled (n=94) to receive an invitation, to increase the probability of greater racial and gender diversity in the final sample of those who volunteered for the interview (n=15). The invitation letters included an information sheet describing the research project and requested that interested individuals contact the interviewer by phone to schedule an interview. Participants were consented by phone and asked for permission to audiotape and report their responses. Participants received \$20 for participation. All procedures were reviewed and approved by the Group Health Institutional Review Board.

Interview Guide and Analysis

A semi-structured interview guide was developed through discussion and iterative design by the NWIGM biobank Principal Investigator (GJ) and Project Manager (JR), as well as a graduate student (EB) and other researchers whose interests aligned in this area (EL, SF). This analysis focused on a sub-set of interview questions, exploring respondents' motivations for participation in the biobank and attitudes toward the return of summary information. Participants were first asked about their motivation for participation. Next, they were asked about their interest in receiving summary results. Finally, they were questioned about their thoughts about resource allocation for providing biobank participants with summary results versus "scientific work" like genetic analyses. Interviews only focused on the potential return of summary findings and not on the return of individual results, as such return was not anticipated at the time of biobank recruitment. The interviews, which ranged from 8 to 58 minutes in length, were conducted by telephone, digitally recorded, and subsequently transcribed by an in-house transcriptionist.

We conducted a directed content analysis (Hsieh & Shannon 2005) of the transcribed interviews. The transcripts were coded independently by the interviewer (EB) and a second reader (JR). The coding scheme was developed from the questions in the interview guide and expanded to include additional codes identified during the coding process. The coders reviewed and discussed each interview with a third reader (EL) who helped resolve coding discrepancies.

RESULTS

Interview Participants and Motivation for Biobank Participation

We interviewed 10 women and 5 men; 8 self-identified as Hispanic, 3 as White, 2 as Black, 1 as Asian, and 1 as American Indian or Alaska Native. The mean age of interview participants was 61 (range 56–67), consistent with the participants in the NWIGM Biobank's age range of 50–65 years at enrollment.

In response to why they had chosen to participate in the NWIGM Biobank, most stated that they were motivated to participate because they felt that all research was important and genetic research especially. Most interview participants (n=13) stated that they felt participation in research in general was a contribution to a greater good, and that they had no expectation of benefitting personally from the research: *"I have no fears for my own health, no...the only benefit is part of the larger society"* (participant #9). *"I'm very interested in making improvements for the future. I recognize that they won't necessarily help me... anything we can do to help people in the future, I think it's important to do as a society"* (participant #12).

Summary Results & Allocation of Resources

Respondents were next asked whether they felt that they, or other biobank participants, would be interested in receiving summary results, and all participants agreed that they would like to be updated. Many felt that receiving updates would help to provide some meaning to their participation: *"It's like if you donate to a charity or something, you just kind of like to know where your money went, maybe into a pool or something"* (participant #15). Most agreed that aggregate results would give them a sense of accomplishment, and help them to feel that their contribution was valuable. *"It makes you feel like... what you've done is helpful and meaningful"* (participant #12).

Language around participant expectations indicated a desire for what the return of results might represent in terms of respect rather than information. *"I don't think it's a right, I think it's a nicety"* (participant #12). Other participants mentioned that it would be "respectful," "interesting" or "valuable." As one participant put it, *"I just feel, the person, if they participate in a study, they must have some commitment to it... feel like they're vested in it, and if you're vested in something, you'd like to know an outcome"* (participant #2).

However, when questioned about how to best to allocate resources to result return participants uniformly prioritized "research" ahead of receiving summary findings: *"I can't speak for others, but my motivation was to help the future of medicine and treatment of illness...I consider that much more important than keeping me abreast of what's being done with my blood cells"* (participant #8). *"I'm curious, um...and it would be interesting to, you know, to see what the end result is, but you know, if it takes money away from the research then, and time, you know it's not that important to me"* (participant #6). *"I think the research is more important than letting me know. The greater good is better for the whole, rather than just for me in particular (participant #14)."* *"I didn't join this project just so I could be updated about what you're doing. I joined the project so that research, real research could be done"* (participant #9). *"Because I don't think we know enough, so it's a high priority to get*

the knowledge” (participant #8). “If the dollar bill can be spent on research, as opposed to talking to me about it, let’s have it be spent on the research” (participant #3) “Um...because I think it’s important that, um, genetic analyses get done, and understand what budget crunches are like” (participant #13).

When asked if they would be comfortable receiving results less often or even not at all if it meant more research could be done, most participants (14 of 15 interviewed) said yes. *“Absolutely...Because this [biobank] isn’t for me. I hope they’re collecting data that would be useful for the studies...I want them to spend time on the science” (participant #4). Unless there’s something critical that they find that’s related to me, I don’t have a need to find out how they’re using study information (participant #7).* However, one man clarified that while “less often” was acceptable he would not be comfortable with never hearing from researchers. *“When it’s not at all...I’m kind of out of touch...I don’t have any way to get the kind of information I might be seeking” (participant #13).*

The “Right” To Receive Research Results

When probed further some participants disagreed about the notion of the “right” to receive research results. *“Would I have a right? I never considered it a right” (participant #14). “No, it wasn’t specified or promised that they would get that at the time they signed up for the study” (participant #7).* Others were willing to waive that right in order to prioritize research. *“Yeah, theoretically I have a right to know, but I’m not particularly concerned with what information develops from that. I don’t really need to know.” (participant #9). “Oh, I suppose I have the right to know, but do I demand to know? No” (participant #1). “Because the most important thing is to find, um, is the research itself. That’s the most important thing. So, to me, getting updates on what’s going on is a nice to have, but it’s not a must-have” (participant #12).*

Discussion

This study explored biobank participants’ motivations for participation and perspectives on the allocation of resources to providing participants with summary results of biobank research. Most of the individuals interviewed reported participating primarily out of altruism and all participants said they would be interested in receiving summary results of research conducted with biobank samples and data. However, the majority of participants felt that communicating summary results was of lower priority to the conduct of additional research and stated that they would prefer not to receive updates if dedicating resources to this type of communication would compromise resources for further research.

One of the most dominant themes was the question of personal benefit versus altruism and the need to maximize the applications of genetic research. This theme was evident in discussions regarding motivation for participation, as well as discussions of how limited resources would be best allocated. Most participants said they participated in research explicitly to help others, and agreed that the research agenda should be prioritized above the return of summary results. However, many mentioned that while summary results would be of no direct utility, they would be primarily useful in lending a sense of purpose to their contribution as research participants.

A common statement expressed was that the primary value of summary results underscored the value of their own contributions and participation in research. Previous research consistent with these observations (Beskow et al., 2012; Snowdon, Garcia, & Elbourne, 1998; Trinidad et al., 2010), suggest that returning results (especially aggregate results) can provide participants with a sense of accomplishment, and that results play an important role in crafting and maintaining a relationship with participants characterized by transparency and inclusion. It will be up to individual biobanks to decide how best to balance the opportunity costs posed by regularly communicating the fruits of biobank research to participants.

Best Practices

Many studies have examined participant and researcher preferences for return of individual results, and development of new models for individual preference-setting is underway (Bacon et al., 2015). The debate about return of summary results is less contentious, though some researchers have questioned the “duty” to return summary results calling for careful conceptual development of how to carry out this work in practice (Miller et al., 2008), and specifically how to avoid creating unintended demand for individual results of unknown significance (Beskow et al., 2012). Researchers involved in studies of communication strategies with participants have found sustained bi-directional contact with participants may support trust between researchers and participants, building awareness of evolving science and changes in how researchers use biobanks (McCarty et al., 2011; Mester et al., 2015). Going one step further, others have suggested that involving patients as collaborators or biobank representatives could be valuable (Mitchell et al., 2015; O’Doherty et al., 2011).

Decisions about how best to optimize patient participation in biobanking activities and decision making will no doubt vary across the spectrum of different biobanks. Some banks may see themselves as stewards of the data who have ongoing relationships with donors, and therefore have more interest in return of results (either individual or summary). Other banks may act merely as a storage and management resources, and therefore not see their role in returning results, putting that responsibility on the researchers themselves. The results from this study suggest that participants have a nuanced view about the allocation of biobank resources to returning summary results, and asking their opinion is a valuable exercise to help make these types of biobank governance decisions.

Research Agenda

Biobanks are tremendously variable in their size, scope and focus, ranging from small and disease specific, to population-based and genome wide, with collections from patients prospectively recruited and retroactively gathered from various institutions, like academic medical centers, non-profit organizations and for-profit corporations. These differences influence the willingness of patients to participate, as well as how they may feel about the possibility of re-contact (K.E. Ormond, Smith, & Wolf, 2010). Participants in this project may be similar to those recruited for population-based biobanks who agree to provide open-ended consent for researchers to use their DNA and clinical information. However, a sample size of fifteen, recruited from a non-profit member-governed health care organization in the Pacific Northwest, is not large or diverse enough to justify drawing broad-spectrum

conclusions for biobanks. The responses noted here may have been influenced particularly by older age, which is associated with increased risk of having chronic conditions of interest to researchers, but may also influence the perspectives about the trade-off between using resources to return summary findings or conduct research. Health care organization affiliation, as well as other unmeasured demographic and socioeconomic factors may have also influenced responses. However, our interview sample did include diverse racial and ethnic representation, which is important for biorepository-based genetic research to ensure that results are broadly relevant (Fullerton, 2011; Thompson & Hebert, 2014).

Several studies have demonstrated that biobank participants may be overly optimistic about receiving clinically relevant results (Halverson & Ross, 2012; Nobile, Vermeulen, Thys, Bergmann, & Borry, 2013; K. E. Ormond, Cirino, Helenowski, Chisholm, & Wolf, 2009), particularly patients recruited from prior participation in genetic research (Henderson et al., 2008). This phenomenon of therapeutic misconception was not directly addressed in this interview. Though most of the patients interviewed stated they did not expect to directly benefit from biobank participation, the consent form included language about return of “clinically relevant results” and some patients may have participated for this reason. Additional research among a larger biobank population could be useful to address these limitations and extend these preliminary observations.

Educational Implications

The current consensus among researchers and policymakers is that returning summary results to study participants is ethically indicated, demonstrates respect, and gives meaning and relevance to participation. The results of this study support this, but also suggest that, given a choice as to how they would prefer resources be allocated, participants may opt to forego aggregate result return in certain cases. While predating the choice to receive summary findings on limited resources could potentially be considered coercive, it can also be argued that it may be mutually beneficial for researchers to ask biobank participants to help make the decision about returning summary results. If the goal is to demonstrate respect for participants, involve participants in research, and enhance the value of participation, it can be argued that providing full transparency about the researcher’s goals and involving participants in the process of crafting the research agenda fulfills these objectives more comprehensively than the simple provision of summary results.

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References

- Bacon PL, Harris ED, Ziniel SI, Savage SK, Weitzman ER, Green RC, Holm IA. The development of a preference-setting model for the return of individual genomic research results. *J Empir Res Hum Res Ethics*. 2015; 10(2):107–120. doi: 10.1177/1556264615572092 1556264615572092 [pii]. [PubMed: 25742675]
- Baker M. Biorepositories: Building better biobanks. *Nature*. 2012; 486(7401):141–146. doi: 10.1038/486141a 486141a [pii]. [PubMed: 22678297]

- Beskow LM, Burke W, Fullerton SM, Sharp RR. Offering aggregate results to participants in genomic research: opportunities and challenges. *Genet Med*. 2012; 14(4):490–496. doi: 10.1038/gim.2011.62 gim201162 [pii]. [PubMed: 22261761]
- Bledsoe MJ, Clayton EW, McGuire AL, Grizzle WE, O'Rourke PP, Zeps N. Return of research results from genomic biobanks: cost matters. *Genet Med*. 2013; 15(2):103–105. doi: 10.1038/gim.2012.105 gim2012105 [pii]. [PubMed: 22935716]
- Dalal H, Wingham J, Pritchard C, Northey S, Evans P, Taylor RS, Campbell J. Communicating the results of research: how do participants of a cardiac rehabilitation RCT prefer to be informed? *Health Expect*. 2010; 13(3):323–330. doi: 10.1111/j.1369-7625.2009.00580.x HEX580 [pii]. [PubMed: 19906214]
- Dixon-Woods M, Jackson C, Windridge KC, Kenyon S. Receiving a summary of the results of a trial: qualitative study of participants' views. *BMJ*. 2006; 332(7535):206–210. doi: bmj.38675.677963.3A [pii] 10.1136/bmj.38675.677963.3A. [PubMed: 16401631]
- Fullerton, SM. The input-output problem: whose DNA do we study, and why does it matter?. In: Burke, W. Edwards, KA. Goering, S. Holland, S., Trinidad, SB., editors. *Achieving Justice in Genomic Translation: Rethinking the Pathway to Benefit*. New York: Oxford University Press; 2011. p. 59-78.
- Halverson CM, Ross LF. Incidental findings of therapeutic misconception in biobank-based research. *Genet Med*. 2012; 14(6):611–615. doi: 10.1038/gim.2011.50 gim201150 [pii]. [PubMed: 22261760]
- Henderson G, Garrett J, Bussey-Jones J, Moloney ME, Blumenthal C, Corbie-Smith G. Great expectations: views of genetic research participants regarding current and future genetic studies. *Genet Med*. 2008; 10(3):193–200. doi: 10.1097/GIM.0b013e318164e4f5 00125817-200803000-00006 [pii]. [PubMed: 18344709]
- Lemke AA, Wolf WA, Hebert-Beirne J, Smith ME. Public and biobank participant attitudes toward genetic research participation and data sharing. *Public Health Genomics*. 2010; 13(6):368–377. doi: 10.1159/000276767 000276767 [pii]. [PubMed: 20805700]
- McCarty CA, Garber A, Reeser JC, Fost NC, Personalized Medicine Research Project Community Advisory, G., Ethics, & Security Advisory, B. Study newsletters, community and ethics advisory boards, and focus group discussions provide ongoing feedback for a large biobank. *Am J Med Genet A*. 2011; 155A(4):737–741. DOI: 10.1002/ajmg.a.33896 [PubMed: 21572889]
- Mester JL, Mercer M, Goldenberg A, Moore RA, Eng C, Sharp RR. Communicating with biobank participants: preferences for receiving and providing updates to researchers. *Cancer Epidemiol Biomarkers Prev*. 2015; 24(4):708–712. doi: 10.1158/1055-9965.EPI-13-1375 1055-9965.EPI-13-1375 [pii]. [PubMed: 25597748]
- Miller FA, Christensen R, Giacomini M, Robert JS. Duty to disclose what? Querying the putative obligation to return research results to participants. *J Med Ethics*. 2008; 34(3):210–213. doi: 10.1136/jme.2006.020289 34/3/210 [pii]. [PubMed: 18316466]
- Mitchell D, Geissler J, Parry-Jones A, Keulen H, Schmitt D, Vavassori R, Matharoo-Ball B. Biobanking from the patient perspective. *Research Involvement and Engagement*. 2015; 1(4)
- Nobile H, Vermeulen E, Thys K, Bergmann MM, Borry P. Why do participants enroll in population biobank studies? A systematic literature review. *Expert Rev Mol Diagn*. 2013; 13(1):35–47. DOI: 10.1586/erm.12.116 [PubMed: 23256702]
- O'Doherty KC, Burgess MM, Edwards K, Gallagher RP, Hawkins AK, Kaye J, Winickoff DE. From consent to institutions: designing adaptive governance for genomic biobanks. *Soc Sci Med*. 2011; 73(3):367–374. doi: 10.1016/j.socscimed.2011.05.046 S0277-9536(11)00336-4 [pii]. [PubMed: 21726926]
- Ormond KE, Cirino AL, Helenowski IB, Chisholm RL, Wolf WA. Assessing the understanding of biobank participants. *Am J Med Genet A*. 2009; 149A(2):188–198. DOI: 10.1002/ajmg.a.32635 [PubMed: 19161150]
- Ormond KE, Smith ME, Wolf WA. The views of participants in DNA biobanks. *Stanford J Law Sci Policy*. 2010; 1:80–87.
- Partridge AH, Winer EP. Informing clinical trial participants about study results. *JAMA*. 2002; 288(3):363–365. doi: jco20029 [pii]. [PubMed: 12117402]

- Snowdon C, Garcia J, Elbourne D. Reactions of participants to the results of a randomised controlled trial: exploratory study. *BMJ*. 1998; 317(7150):21–26. [PubMed: 9651262]
- Thompson B, Hebert JR. Involving disparate populations in clinical trials and biobanking protocols: experiences from the community network program centers. *Cancer Epidemiol Biomarkers Prev*. 2014; 23(3):370–373. doi: 10.1158/1055-9965.EPI-14-0118 23/3/370 [pii]. [PubMed: 24609845]
- Trinidad SB, Fullerton SM, Bares JM, Jarvik GP, Larson EB, Burke W. Genomic research and wide data sharing: views of prospective participants. *Genet Med*. 2010; 12(8):486–495. DOI: 10.1097/GIM.0b013e3181e38f9e [PubMed: 20535021]
- VA National Center for Ethics in Health Care. Sharing Aggregate Research Results With Participants. InFocus: Topics in Health Care Ethics. 2009. Retrieved May 21, 2015, from http://www.ethics.va.gov/docs/infocus/InFocus_20090330_Sharing_aggregate_research_results.pdf
- Wolf SM, Crock BN, Van Ness B, Lawrenz F, Kahn JP, Beskow LM, Wolf WA. Managing incidental findings and research results in genomic research involving biobanks and archived data sets. *Genet Med*. 2012; 14(4):361–384. doi: 10.1038/gim.2012.23 gim201223 [pii]. [PubMed: 22436882]