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Participant Satisfaction With a Preference-Setting Tool for the Return of Individual Research Results in Pediatric Genomic Research

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Abstract

The perceived benefit of return of individual research results (IRRs) in accordance to participants' preferences in genomic biobank research is unclear. We developed an online preference-setting tool for return of IRRs based on the preventability and severity of a condition, which included an opt-out option for IRRs for mental illness, developmental disorders, childhood-onset degenerative conditions, and adult-onset conditions. Parents of patients <18 years of age at Boston Children's Hospital were randomized to the hypothetical scenario that their child was enrolled in one of four biobanks with different policies for IRRs to receive (a) "None," (b) "All," (c) "Binary"—choice to receive all or none, and (d) "Granular"—use the preference-setting tool to choose categories of IRRs. Parents were given a hypothetical IRRs report for their child. The survey was sent to 11,391 parents and completed by 2,718. The Granular group was the most satisfied with the process, biobank, and hypothetical IRRs received. The None group was least satisfied and least likely to agree that the biobank was beneficial ($p < .001$). The response to the statement that the biobank was harmful was not different between groups. Our data suggest that the ability to designate preferences leads to greater satisfaction and may increase biobank participation.

Keywords

participant preferences, pediatrics, return of individual genomic research results, biobank research, preference-setting tool

With dramatic advances in technology and the constant discovery of novel genetic information, individuals are now able to receive individual genetic data that may have meaning to them. In the research setting, there is a growing consensus that return of individual genetic information to participants may be desirable. Not only do participants express the desire to receive research results (E. D. Harris et al., 2012; D. J. Kaufman, Murphy-Bollinger, Scott, & Hudson, 2009; O'Daniel & Haga, 2011; Shalowitz & Miller, 2008), but many argue that research may generate information that is important to participants' health, and that they have a right to such information (Wolf, 2012; Wolf et al., 2012).

Initial guidelines for return of genomic research results emphasized the return of results for severe, potentially life-threatening diseases for which effective treatment and/or prevention was available based on analytic and clinical validity, actionability, and severity of the disease (Fabsitz et al., 2010; National Cancer Institute [NCI], 2010). The American College of Medical Genetics and Genomics (ACMG) recommendations for return of incidental findings in the clinical setting (ACMG, 2014; Green et al., 2013) focused attention on return of genetic information to patients and families. The research community has been struggling with return of individual research results (IRRs; Caulfield et al., 2008; Clayton et al., 2010; Kohane et al., 2007; Kohane & Taylor, 2010; Wolf et al., 2012), in particular whether there is an obligation to return IRRs and the role of participant preferences. Recently the Presidential Commission (Presidential-Commission-for-the-Study-of-Bioethical-Issues, 2013) and a joint Clinical Sequencing Exploratory Research (CSER) Consortium and the

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Electronic Medical Records and Genomics (eMERGE) Network group (Jarvik et al., 2014) put forth guidelines suggesting that unlike the clinical setting, in the research realm there is no duty to return research results, and that if research results are offered, investigators should allow participants to opt out of receiving results (Jarvik et al., 2014; Presidential-Commission-for-the-Study-of-Bioethical-Issues, 2013).

One option for result disclosure is to give participants the option to define, at the time of enrollment, the types of results to receive (Ravitsky & Wilfond, 2006), allowing participants to make selections based on their "personal utility" (Foster, Mulvihill, & Sharp, 2009; Grosse, Kalman, & Khoury, 2010; Grosse, McBride, Evans, & Khoury, 2009; Khoury et al., 2009) and the meaning of genomic information to them (Kohane & Taylor, 2010; Ravitsky & Wilfond, 2006; Rothstein, 2006). In 2007, we proposed the "Informed Cohort" model (Kohane et al., 2007) that reflects this approach, an automated infrastructure for implementing a preference-driven approach to return of results. Our group at Boston Children's Hospital (BCH) had as a goal the implementation of the Informed Cohort model in a pediatric setting as the *Gene Partnership* (GP). We recently published guidelines for return of IRRs from pediatric genomic studies in accordance with parental preferences and considering the developing autonomy of pediatric participants, all in the context of protecting participants from medical and psychosocial harms from result disclosure (Holm et al., 2014; Holm & Taylor, 2012).

A major challenge to incorporating preferences into return of research results is ensuring that it is done in a manner that is both scalable and reflects participants' true desires for information to receive. Although participants may desire to have a choice, does it matter to them how granular their choices are? Or is just having a choice the important factor? In addition, there is a lack of evidence about whether biobank participants truly understand the implications of their choice of IRRs to receive. Given that they are choosing from among hypothetical future events, it is not clear if their stated preferences are stable and reliable.

As our group considered return of IRRs in Gene Partnership, we sought to explore whether degree of satisfaction with enrollment in a hypothetical biobank was associated with having an ability to designate preferences regarding receiving IRRs, considering both the ability to set *any* preferences and the ability to set *nuanced* preferences in comparison with having situations of no choice regarding preferences. Finally, we were interested to see if the opportunity to see hypothetical results returned after exercising nuanced preference setting increased satisfaction with the results received. To answer these questions, we randomized parents to one of four hypothetical child biobank scenarios reflective of different policies for return of research results and gave them a hypothetical research result report for their child. In this analysis, we report on participant satisfaction with the process, biobank, and hypothetical results received. We hypothesized that those in the group with an opportunity to set nuanced preferences for IRRs would have higher satisfaction than those without that opportunity.

Method

This research study was approved by the BCH Institutional Review Board (IRB-P00006896: "Study to Measure the Effectiveness of a Preference-Setting Model for the Return of Individual Research Results").

Development and Testing the Survey

We previously developed a participant-centered preference-setting model through formative research with parents (Bacon et al., 2015). The resultant model allows parents to choose which results to receive based on the severity and preventability of possible conditions. The model also allows parents to opt out of receiving results for specific categories of conditions perceived by many parents in our interviews to be highly sensitive—mental illness, developmental disorders, and childhood-onset degenerative conditions—as well as adult-onset conditions not treatable during childhood.

To assess participant satisfaction with the biobank model to which participants were assigned under our experiment, survey questions were developed by our team of genomic researchers, genetic counselors, behavioral scientists, survey methodologists, and medical geneticists. Baseline survey questions before presenting hypothetical IRRs were adapted primarily from the MedSeq "Expectations/Perceived Utility" Questionnaire (Vassy et al., 2014). Follow-up questions were adapted from quotes from the parent interviews conducted to develop the preference-setting model (Bacon et al., 2015) and from additional literature (DuBenske, Burke Beckjord, Hawkins, & Gustafson, 2009). Cognitive interviews were conducted with parents of inpatients at BCH to test the survey for comprehension and ease of administration. The survey was programmed into REDCap and administered as a web survey (P. A. Harris et al., 2009).

Randomization to Four Hypothetical Biobanks

Prior to enrollment, parents were randomized to one of four hypothetical biobanks with different policies for return of genetic research results models (see Figure 1 for a flowchart of the study): (a) Group 1a received no results (None), (b) Group 1b received all results (All), (c) Group 2 was given a choice to receive all or no results (Binary), and (d) Group 3 used the preference-setting tool to choose categories of results to receive (Granular).

Upon starting the online survey, participants were asked to watch a 5-min educational video about basic genetic

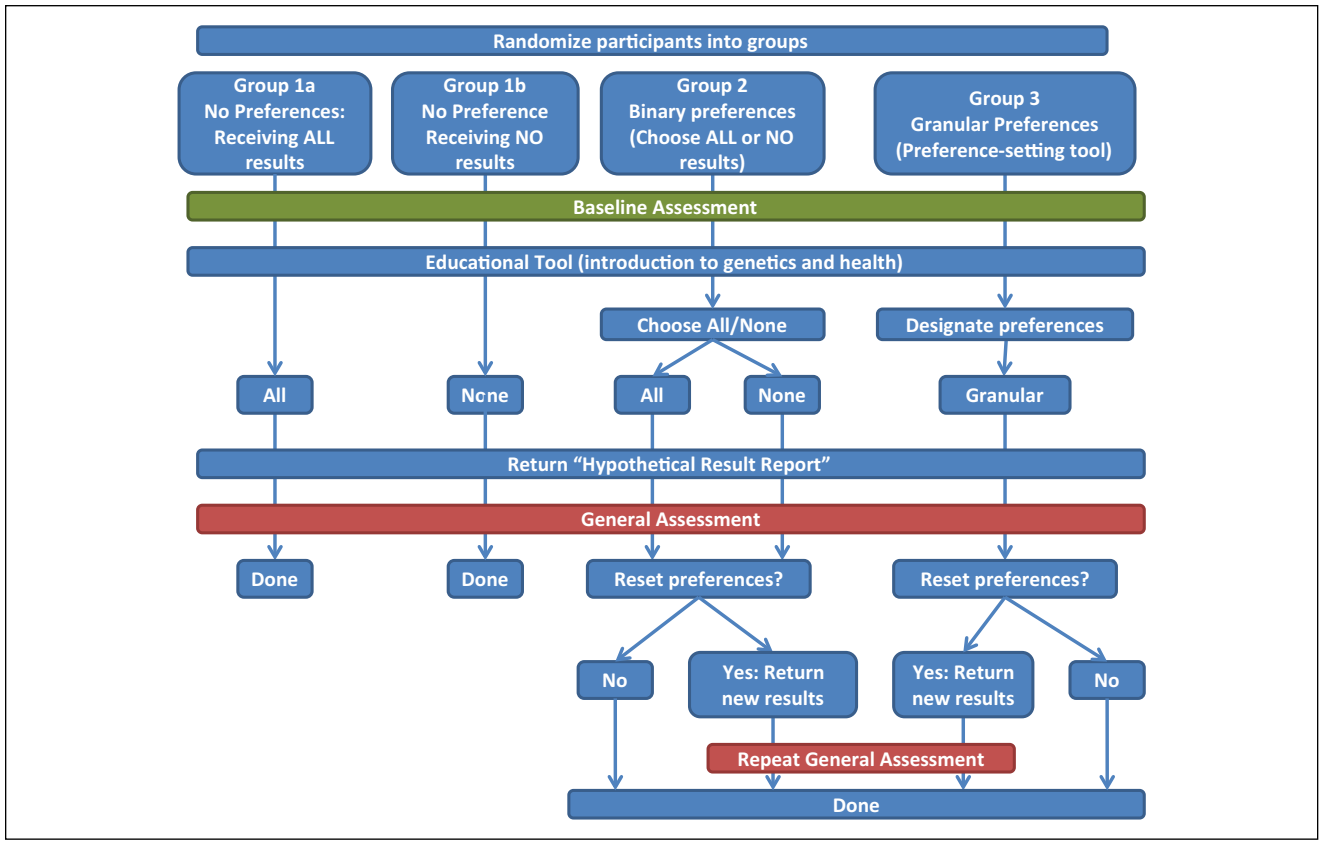

Figure 1. Flowchart of the study.

concepts, including a brief description of genetic biobanks and the potential for return of IRRs. All participants then answered demographic questions and questions about reasons why they might or might not want to receive genetic information about their child. The participants of Groups 1a and 1b (None and All) had no choice regarding receiving IRR and were told they would receive either all genetic research results (Group 1a—All) or no results (Group 1b—None). Group 2 (Binary) was given a choice between receiving all or no results. Group 3 (Granular) was asked to designate preferences with regard to which research results they wanted to receive using a three-step preference-setting process: (a) They were given the option to decide if first they wanted to receive results for disorders that were preventable, non-preventable, both, or neither. (b) Those who chose preventable, non-preventable, or both were given the option to receive results for conditions that were severe, not severe, or both. (c) The participants were offered the option to opt out of receiving results for conditions classified in the following four categories: mental illnesses, developmental and learning disorders, childhood-onset degenerative diseases, and adult-onset conditions not preventable in childhood. The preference-setting process ended for participants who chose to receive neither preventable nor non-preventable results, as all possible results were eliminated with this first decision.

Return of Hypothetical IRRs

All groups were presented with a "Hypothetical Result Report" that showed genetic conditions in a 2 × 2 table according to the criteria of preventability and severity (Figure 2). The conditions had been previously selected and classified into one of the four cells by a group of 20 genetic health care professionals at BCH. Included in each cell were conditions that parents could have opted out of receiving (mental illnesses, developmental and learning disorders, childhood-onset degenerative diseases, or adult-onset conditions). The report was shown to participants in all four groups to highlight research results that they may or may not receive. Conditions would be highlighted as "received" or "withheld" based on which group participants were assigned to. The All group saw all of the conditions highlighted, whereas every condition was crossed out for the None group. The Binary group would receive the appropriate grid that reflected their decision to receive all or no results. The Granular group would be shown the appropriate grid that reflected their decision to receive severe and/or non-severe conditions, preventable and/or non-preventable conditions, mental illnesses, developmental disorders, child-onset degenerative diseases, or adult-onset conditions (see Figure 3 for an example).

Not severe & preventable	Severe & preventable
1. Pet dander (dog) allergy 2. Iron deficiency anemia 3. Kidney stones 4. Lactose intolerance 5. Gastroesophageal reflux disease 6. Reduced response to ibuprofen 7. Chronic mild constipation 8. Delayed response to local anesthetic 9. Increased susceptibility to cavities	1. Alcoholism * (mental health) 2. Asthma 3. Deep vein thrombosis 4. Familial hypercholesterolemia 5. Melanoma 6. Peanut allergy 7. Types II Diabetes 8. Malignant hyperthermia 9. Childhood onset hereditary colon cancer 10. Aortic aneurism
Not severe & non-preventable	**Severe & non-preventable**
1. Attention deficit hyperactivity disorder* (learning disability) 2. Essential tremor 3. Generalized anxiety * (mental health) 4. Hypothyroidism 5. Poor vision 6. Seasonal allergies 7. Turner Syndrome 8. Vitiligo 9. Mitral valve prolapse 10. Obstructive sleep apnea	1. Autism * (developmental and learning disability) 2. Bipolar disorder * (mental health) 3. Duchenne Muscular Dystrophy 4. Juvenile (Type I) Diabetes 5. Juvenile rheumatoid arthritis 6. Polycystic Ovarian Syndrome 7. Rett Syndrome * (Childhood-onset degenerative) 8. Acute lymphoblastic leukemia 9. Batten disease (NCL) * (Childhood-onset degenerative) 10. Alzheimer's disease * (adult-onset) 11. Huntington's disease* (adult-onset)

Figure 2. Hypothetical result report.

Note. This grid was shown to participants in all four survey groups to highlight research results that they may or may not receive. Certain conditions would be highlighted as "received" and others crossed out as "withheld" based on which group participants were assigned to. The All group saw all of the conditions highlighted, whereas every condition was crossed out for the None group. The Binary group would receive the appropriate grid that reflected their decision to receive all or no results. The Granular group would be shown the appropriate grid that reflected their decision to receive severe and/or non-severe conditions, preventable and/or non-preventable conditions, mental illnesses, developmental disorders, child-onset degenerative diseases, or adult-onset conditions.

After participants were presented with their hypothetical result report, they were asked to answer questions assessing their satisfaction with the process of setting preferences and the results. Additional questions assessed their perceived benefits and harms from receiving the hypothetical results they choose. The All and None groups were finished after these sets of questions whereas the Binary and Granular groups were given the option to reset their preferences. If participants indicated that they wanted to change their preferences, they were given the opportunity to reset them and then again answer questions about their satisfaction with the preference-setting process and results.

Sample Design and Participants

The sample for this study was drawn from BCH patient population. Participants were parents or guardians of children who received care at BCH in the 24 months preceding the sample selection date. Eligibility criteria included being 18 years or older and having at least one child less than 18 years at the time of the study. Because a notification about the survey was sent by mail, and the survey was sent by email, the child's medical record had to have an email address and mailing address for the parent. Parents were excluded if either the parents or their child had been, or was, enrolled in the Gene Partnership. Based on our previous experience of a 16% response rate to a previous mailed paper survey that we conducted (Ziniel et al., 2014), we anticipated a slightly better response rate of 20% as the current survey was sent by email. We randomly selected 12,430 individuals to participate and be randomized into one of four groups. To allow more detailed analyses for the Binary and Granular groups, the sample was disproportionately distributed: 15% of the participants were assigned to the All group, 15% to the None group, 30% to the Binary group, and 40% to the Granular group.

Survey Dissemination

A pre-notification letter explaining the study was mailed to the potential parent participant and included the email address to which we were planning to email the survey, and in the letter we encouraged the parent to contact study staff by phone or email to update the email address if needed. The letter also indicated that participants who completed the survey would be entered into a raffle for one US$100 Visa gift cards for every 100 completed surveys. Nine days after the pre-notification letter was mailed, the survey invitation email was sent to parents. The URL link to the survey directed participants to the biobank to which they were

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1. Pet dander (dog) allergy 2. Iron deficiency anemia 3. Kidney stones 4. Lactose intolerance 5. Gastroesophageal reflux disease 6. Reduced response to ibuprofen 7. Chronic mild constipation 8. Delayed response to local anesthetic 9. Increased susceptibility to cavities	1. ~~Alcoholism~~ * 2. Asthma 3. Deep vein thrombosis 4. Familial hypercholesterolemia 5. Melanoma 6. Peanut allergy 7. Types II Diabetes 8. Malignant hyperthermia 9. Childhood onset hereditary colon cancer 10. Aortic aneurism
Not severe & non-preventable	**Severe & non-preventable**
1. ~~Attention deficit hyperactivity disorder~~ * 2. ~~Essential tremor~~ 3. ~~Generalized anxiety~~ * 4. ~~Hypothyroidism~~ 5. ~~Poor vision~~ 6. ~~Seasonal allergies~~ 7. ~~Turner Syndrome~~ 8. ~~Vitiligo~~ 9. ~~Mitral valve prolapse~~ 10. ~~Obstructive sleep apnea~~	1. ~~Autism~~ * 2. ~~Bipolar disorder~~ * 3. ~~Duchenne Muscular Dystrophy~~ 4. ~~Juvenile (Type I) Diabetes~~ 5. ~~Juvenile rheumatoid arthritis~~ 6. ~~Polycystic Ovarian Syndrome~~ 7. ~~Rett Syndrome~~ * 8. ~~Acute lymphoblastic leukemia~~ 9. ~~Batten disease (NCL)~~ * 10. ~~Alzheimer's disease~~ * 11. ~~Huntington's disease~~*

Figure 3. An example of a hypothetical result report for a participant who set his or her preferences to receive the following types of conditions: Preventable only, both severe and non-severe, opt out of mental health conditions.
Note. Conditions with a strikethrough are those that would not be disclosed. Conditions marked with an asterisk are included in the opt-out categories.

randomized. Participants were sent a maximum of three reminder emails.

Survey Pilot

The survey was piloted with 500 participants, randomly selected from the same sampling frame as used in the main study, and minor adjustments were made to the survey based on the results of the pilot study. To determine the number of undeliverable emails in the pilot survey, the final reminder email was sent out from an email account not connected with the web survey program (REDCap) allowing us to see which emails were "undeliverable" (7.4% of these emails). Of the delivered emails, the response rate was 21.6%. Data from the pilot study were not included in the final analyses.

Key Measures

We measured participants' satisfaction with the process and the results themselves after participants were shown their Hypothetical Result Report, which indicated which IRR they would or would not have received. The response scale ranged from 0 to 10, where 0 indicated *very dissatisfied* and 10 indicated *very satisfied*. For those in the Binary and Granular groups who reset their preferences, their satisfaction was assessed again after they saw their second Hypothetical Result Report. Analyses of satisfaction data used the final set of satisfaction ratings from each participant.

Finally, after receiving their Hypothetical Results Report, participants were asked their opinions about the biobank and the preference-setting process they had experienced with regard to each of the following four criteria: "For me, being a part of this type of biobank would be . . . " (a) "a good thing," (b) "a bad thing," (c) "beneficial," and (d) "harmful" (Wade et al., 2012). Each criterion was evaluated on a 5-point Likert-type scale, ranging from *strongly disagree* to *strongly agree*. For ease of presentation for this publication, the two most extreme response options on each side of the bipolar scale (*strongly disagree* and *disagree*, as well as *agree* and *strongly agree*) were collapsed into one as this did not change the results.

Data Management and Analyses

All web survey data were collected using REDCap (P. A. Harris et al., 2009) and analyzed using Stata 12.1 (Stata Statistical Software: Release 12, 2011). Demographic characteristics and background information are shown as percentages or means. Fisher's exact tests and ANOVAs were

Table 1. Demographic and Background Characteristics.

Characteristic	Total *N*[a]	Percentage/*M*					
		All Groups Combined	None	All	Binary	Granular	*p*
Mean age (*SD*)	2,905	42.9 (7.4)	42.9 (7.3)	43.1 (7.5)	42.7 (7.4)	43.0 (7.5)	.830
Female	2,960	91.0%	91.8%	91.3%	90.0%	91.5%	.615
Race*	2874	All Groups	None	All	Binary	Granular	*p*
White		93.2	92.7	92.8	92.7	93.9	.675
Black		2.4	2.1	2.5	3.2	1.8	.267
Asian		3.7	4.9	3.6	3.3	3.5	.500
Native American or Alaskan Native		0.4	0.2	0.2	0.3	0.6	.829
Native Hawaiian or Other Pacific Islander		0.1	0.0	0.2	0.1	0.1	.747
Other		1.6	1.6	2.0	1.3	1.7	.743
Ethnicity	2,889						.314
Hispanic		4.1%	5.1%	4.3%	4.5%	3.3%	
Education	2,896						.857
Eighth grade or less		0.3%	0.2%	0.2%	0.1%	0.5%	
Some high school but did not graduate		0.5%	0.2%	0.7%	0.6%	0.4%	
High school graduate or GED		3.7%	4.4%	3.4%	3.4%	3.9%	
Some college		12.6%	12.6%	11.8%	14.4%	11.4%	
2- or 4-year college graduate		38.0%	37.4%	39.0%	38.0%	37.9%	
More than 4-year college degree		45.0%	45.1%	44.9%	43.5%	46.1%	
Work in health care	2,960	23.6%	24.2%	24.1%	24.0%	22.9%	.904
Prior participation in research	2,959	37.0%	40.1%	38.6%	36.4%	35.6%	.332
Prior genetic testing experience	2,960	40.9%	42.8%	42.2%	39.9%	40.4%	.688
Child diagnosed with genetic disorder	2,960	23.2%	23.7%	23.0%	23.4%	23.0%	.990

[a]Differences in the number (*N*) for each item reflect the fact that not everyone answered every question.
Legend: * multiple selections possible.

used to test for differences between the four groups with regard to these characteristics. The mean satisfaction with the results and the preference-setting process was compared between the groups using Kruskal–Wallis tests because the normal distribution assumption of ANOVA was violated. Participants' opinions about the biobank to which they were assigned were also compared using Fisher's exact tests.

Multiple linear regression was conducted and only variables significant in the bivariate analyses were included in the regression models.

Results

Response Rates and Sample Characteristics

The full survey was conducted in the same manner as the pilot. Sixty-three of the mailed pre-notification letters were not delivered and "returned to sender," and 72 participants asked to be removed from the study after receiving the pre-notification letter and were removed from the survey email list. Ninety-eight participants contacted study staff and provided updated email addresses. We sent 12,295 survey invitation emails and assumed the email undeliverable rate in the full survey was the same as in the pilot survey (7.35%) for an estimated 904 emails not delivered and 11,391 emails reaching respondents. A total of 2,718 respondents completed the survey for a response rate (*Standard Definitions: Final Dispositions of Case Codes and Outcome Rates for Surveys*, 2011; RR3) of 23.9%. The maximum response rate difference between the groups was 2.4% and was not significantly different between groups: None group 24.6% (421/1,709), All group 24.7% (422/1,709), Binary group 24.9% (849/3,414), and Granular group 22.5% (1,026/4,559). In addition, 242 respondents completed at least 25% of the questions presented to them but not the complete survey and this was not significantly different between groups. The inclusion of the partial respondents yielded an overall response rate (RR4) of 26.0% (2,960/11,391). The percentage of partial respondents was lowest in the None group with 4.1% (18/439), followed by the All group with 5.8% (26/448) and the Binary group with 8.4% (78/927), and highest in the Granular group with 10.5% (120/1,146), which was also the longest of the four surveys. All subsequent analyses include partial respondents.

The demographic characteristics of the participants, overall and by group, are shown in Table 1, along with prior experience with research and genetic testing. Participants were on average 42.9 years old and the majority were female and

Caucasian. The four groups did not show significant differences with regard to the demographic and background characteristics (Table 2).

Participant Satisfaction With Preference-Setting Process and Results

Significant differences in satisfaction ratings were observed among groups (Table 2), indicating that participants assigned to the Granular group are the most satisfied with both the preference-setting process and the hypothetical results they had received while the None group was the least satisfied. The Binary and All groups rated their satisfaction very similar and nearly as high as the Granular group.

We then looked at baseline factors associated with satisfaction with the preference-setting process and the hypothetical results they had received (Table 2). None of the demographic factors were associated with satisfaction with the process. Only non-Hispanic ethnicity ($p = .014$) and Caucasian race ($p = .037$) were associated with greater satisfaction with the results. For the overall sample, satisfaction with both process and hypothetical results was associated with perceiving that novel information might (might not) help to (a) prepare for the future, (b) feel more in control over my future, (c) prevent me from worrying, and (d) seek medical treatments for my child ($p < .001$ to $p = .026$). Decreased satisfaction with the process was associated with fear of "finding out something I don't want to know." Being comfortable with the possibility of getting genetic research result about their child was also associated with greater satisfaction with both the process and results.

We then compared satisfaction with the process and results received between the groups (Table 3). The Granular group was more satisfied than all other groups, the Binary was more satisfied than the None group, and the All was more satisfied than the None group (all $p < .001$). The Binary group was equally satisfied as the All group. In a multiple linear regression model, we assessed the simultaneous association of demographics and the baseline variable with satisfaction with the process and with satisfaction with the results (Table 4). For both dependent variables, satisfaction with the process and the results was associated with type of biobank assigned and comfort with the possibility of receiving genetic research results about one's child; satisfaction with the process was associated with the perception that returned results would help a participant feel "more in control over my future."

We then included interactions between the biobank the respondents were randomized to and their answers to the questions why they might or might not want to receive results to assess whether the relationship between the assigned biobank and the satisfaction with the process and the results varies as a function of different baseline preferences with regard to receiving results. Significant interactions were observed among type of biobank and perceiving that IRR would enhance participants' (a) feelings of being in control of one's future and (b) comfort with the possibility of getting genetic research results about their child. We found that although overall those in the None group were less satisfied than those in the other groups, as previously described, satisfaction within the None group was greater for those who agreed with the statement that they feared they might find something that they did not want to know compared with those who disagreed with this statement, whereas within the Binary and All groups satisfaction was greater for those who disagreed with the fear statement compared with those who agreed. A similar pattern emerged for the interaction between biobank randomized to and being comfortable with the possibility of getting genetic research result about their child with the pattern in the None group differing from the other groups. Within the None group, those who were comfortable were less likely to be satisfied compared with those who disagreed with the comfort statement, whereas those within the other three biobanks who agreed with the comfort statement were more likely to be satisfied with the biobank randomized to.

Participants' Opinions Toward Biobanks

When asked to evaluate the biobank they had experienced, participants' opinions were significantly different in three of the four characteristics (Table 5). Proportionately fewer participants in the None group agreed that the biobank to which they were assigned was "a good thing" or "beneficial" compared with the other groups. Proportionately more participants in the None group agreed with the statement that the biobank they experienced was "a bad thing." The All, Binary, and Granular groups showed similar distributions of opinions across all four characteristics. Interestingly, no significant difference could be found between the four groups with regard to the statement that the biobank was harmful.

Discussion

As genetic analyses have become more accessible research tools for gene discovery, questions about if, when, and how to return genomic information to research participants have become more pressing. This is particularly true in large biobank research where thousands of individuals may be enrolled, and extensive and multiple analyses may be performed with potential for myriad findings. Recent recommendations and guidelines have suggested that although returning results to participants in genomic research is not an obligation, it may be desirable (Jarvik et al., 2014; Presidential-Commission-for-the-Study-of-Bioethical-Issues, 2013). One of the biggest limitations to returning

Table 2. Participant Satisfaction With the Process and the Results Received by Group Randomized to, Demographic Characteristics, and Baseline Attitudes.

	Satisfaction with process		Satisfaction with results	
	M (*SD*)	*p*	*M* (*SD*)	*p*
Group randomized to				
Group (*n* = 2,805)				
None	3.17 (2.95)	<.001	3.69 (3.02)	<.001
All	6.50 (2.42)		6.09 (2.47)	
Binary	6.65 (2.51)		6.06 (2.63)	
Granular	7.30 (2.20)		7.00 (2.28)	
Demographic characteristics				
Gender (*n* = 2,805)				
Male	6.29 (2.78)	.753	5.98 (2.70)	.636
Female	6.35 (2.82)		6.07 (2.77)	
Age (*n* = 2,752)				
<35 years	6.46 (2.87)	.675	6.12 (2.89)	.958
35-39 years	6.42 (2.72)		6.05 (2.63)	
40-44 years	6.35 (2.89)		6.09 (2.84)	
45-49 years	6.33 (2.75)		6.05 (2.72)	
50+ years	6.20 (2.89)		5.99 (2.80)	
Education (*n* = 2,749)				
Some grade school	5.88 (2.95)	.745	6.75 (2.19)	.462
Some high school	6.85 (2.27)		7.00 (2.31)	
Graduated high school	6.33 (3.24)		6.25 (2.90)	
Some college	6.45 (2.88)		6.21 (2.81)	
2- or 4-year college graduate	6.41 (2.76)		6.08 (2.70)	
Post-college graduate courses or degree	6.26 (2.81)		5.98 (2.81)	
Hispanic (*n* = 2,742)				
No	6.36 (2.81)	.219	6.09 (2.77)	.014
Yes	6.02 (2.93)		5.42 (2.91)	
Caucasian race (*n* = 2,805)				
No	6.16 (2.76)	.265	5.72 (2.72)	.037
Yes	6.36 (2.83)		6.10 (2.77)	
African American race (*n* = 2,805)				
No	6.34 (2.82)	.881	6.08 (2.76)	.144
Yes	6.29 (2.99)		5.54 (2.99)	
Asian race (*n* = 2,805)				
No	6.36 (2.83)	.218	6.08 (2.76)	.080
Yes	6.00 (2.61)		5.59 (2.82)	
Working in health care (*n* = 2,805)				
No	6.40 (2.77)	.067	6.06 (2.76)	.875
Yes	6.17 (2.96)		6.08 (2.81)	
Baseline attitudes				
Previous research experience (*n* = 2,805)				
No	6.37 (2.75)	.512	6.05 (2.73)	.691
Yes	6.30 (2.94)		6.09 (2.82)	
Previous genetic testing (*n* = 2,805)				
No	6.40 (2.71)	.207	6.09 (2.71)	.533
Yes	6.26 (2.97)		6.03 (2.85)	
Child with genetic diagnosis (*n* = 2,805)				
No	6.33 (2.82)	.602	6.03 (2.76)	.293
Yes	6.39 (2.83)		6.16 (2.80)	
Some reasons I might want information are . . .				
It might help me prepare for the future (*n* = 2,805)				
No[a]	5.66 (2.77)	<.001	5.57 (2.69)	.002
Yes[b]	6.42 (2.82)		6.12 (2.77)	

(continued)

Table 2. (continued)

	Satisfaction with process		Satisfaction with results	
	M (*SD*)	*p*	*M* (*SD*)	*p*
It might help me feel more in control over my future (*n* = 2,805)				
No[a]	5.75 (2.79)	<.001	5.66 (2.64)	<.001
Yes[b]	6.54 (2.80)		6.20 (2.80)	
It might prevent me from worrying (*n* = 2,805)				
No[a]	6.12 (2.76)	<.001	5.86 (2.68)	<.001
Yes[b]	6.55 (2.86)		6.26 (2.83)	
It might help me seek medical treatments for my child (*n* = 2,805)				
No[a]	5.82 (2.93)	.002	5.71 (2.78)	.026
Yes[b]	6.40 (2.80)		6.10 (2.76)	
Some reasons I might not want information are . . .				
I fear that I might find out something I don't want to know (*n* = 2,805)				
No[a]	6.45 (2.91)	.025	6.12 (2.87)	.199
Yes[b]	6.21 (2.69)		5.99 (2.62)	
It might cause me anxiety (*n* = 2,805)				
No[a]	6.42 (2.97)	.237	6.06 (2.96)	.989
Yes[b]	6.29 (2.70)		6.06 (2.61)	
I think it's the doctor's job to deal with health information, not mine (*n* = 2,805)				
No[a]	6.34 (2.82)	.778	6.06 (2.77)	.414
Yes[b]	6.22 (2.61)		6.41 (2.48)	
Sometimes, ignorance is bliss (*n* = 2,805)				
No[a]	6.36 (2.85)	.389	6.07 (2.80)	.835
Yes[b]	6.23 (2.63)		6.04 (2.55)	
I find the information hard to understand (*n* = 2,805)				
No[a]	6.34 (2.84)	.670	6.06 (2.79)	.600
Yes[b]	6.43 (2.52)		6.16 (2.45)	
Comfort				
Comfortable with the possibility of getting genetic research results about your child from a biobank study (*n* = 2,805)				
No[c]	5.75 (2.45)	<.001	5.57 (2.38)	<.001
Yes[d]	6.57 (2.92)		6.25 (2.88)	

Note. The total number of respondents might be different for the rows of this table. They are based on the number of respondents who answered both questions and not every respondent answered every question.
[a]"No" represents the combined response categories "neither agree nor disagree," "disagree," and "strongly disagree."
[b]"Yes" represents the combined response categories "agree" and "strongly agree."
[c]"No" represents the combined response categories "neutral," "somewhat uncomfortable," and "very uncomfortable."
[d]"Yes" represents the combined response categories "somewhat comfortable" and "very comfortable."

Table 3. Post Hoc Multiple Comparisons of Satisfaction With the Preference-Setting Process and the Hypothetical Results Received Across Groups.

		Satisfaction with process		Satisfaction with results	
Group I	Group J	Mean difference (I – J)	*p*	Mean difference (I – J)	*p*
Granular	Binary	0.65	<.001	0.94	<.001
	All	0.79	<.001	0.92	<.001
	None	4.12	<.001	3.31	<.001
Binary	All	−0.15	1.00	−0.03	1.00
	None	3.48	<.001	2.37	<.001
All	None	3.33	<.001	2.39	<.001

genomic research results to participants is feasibility—the model is costly, and adequately educating and guiding participants about types of results they could receive to enable informed choices is challenging. Our goal was to use the return of hypothetical results to evaluate the stability of preferences and satisfaction with process for four biobank

Table 4. Multiple Linear Regression Predicting Satisfaction With the Process and Satisfaction With the Results.

	Satisfaction with the process (n = 2,652)		Satisfaction with the results (n = 2,651)	
	Regression coefficient	p	Regression coefficient	p
Model constant	2.60	<.001	3.21	<.001
All group[a]	3.33	<.001	2.46	<.001
Binary group[a]	3.49	<.001	2.46	<.001
Granular group[a]	4.12	<.001	3.38	<.001
Male	−0.09	.596	−0.09	.608
Age in categories	−0.05	.179	−0.01	.760
Education	−0.05	.442	−0.10	.086
Hispanic	−0.22	.367	−0.35	.177
Caucasian race	0.16	.345	0.32	.073
Child with genetic diagnosis	0.03	.821	0.13	.265
It might help me prepare for the future[b]	0.13	.528	0.13	.532
It might help me feel more in control over my future[b]	0.51	<.001	0.22	.118
It might prevent me from worrying[b]	0.06	.584	0.20	.076
It might help me seek medical treatments for my child[b]	−0.10	.587	−0.12	.541
I fear that I might find out something I don't want to know[b]	−0.12	.227	−0.02	.854
Comfortable with the possibility of getting genetic research results about your child from a biobank study[b]	0.62	<.001	0.58	<.001

[a]The comparison group is the "None group."
[b]Response categories dichotomized into "Yes" (positive categories) and "No" (middle option and negative categories).

Table 5. Participant Opinions About Biobank.

	None (%)	All (%)	Binary (%)	Granular (%)	p
A good thing					
Strongly disagree/disagree	9.50	2.35	2.81	1.85	<.001
Neither agree nor disagree	23.28	16.47	18.71	16.52	
Strongly agree/agree	67.22	81.18	78.48	81.63	
A bad thing					
Strongly disagree/disagree	61.76	72.00	74.30	77.84	<.001
Neither agree nor disagree	27.08	24.24	20.56	18.27	
Strongly agree/agree	11.16	3.76	5.14	3.89	
Beneficial					
Strongly disagree/disagree	8.31	2.35	2.57	2.14	<.001
Neither agree nor disagree	28.50	27.06	22.31	23.62	
Strongly agree/agree	63.18	70.59	75.12	74.25	
Harmful					
Strongly disagree/disagree	64.13	70.82	71.38	72.30	.110
Neither agree nor disagree	29.22	24.24	23.48	22.93	
Strongly agree/agree	6.65	4.94	5.14	4.76	

types, including a biobank that implemented a preference-setting model.

This is the only study that we are aware of where participants are randomized to biobanks with different return of results policies that includes a biobank where the participant designates his or her preferences for return of IRRs. We elucidate specific responses of participants randomized to different biobank conditions including a novel condition that enables specification of granular preferences for IRRs—an issue of high public health and biomedical significance. Our results demonstrate that choice matters with respect to participant satisfaction, with more nuanced choice associated with greatest satisfaction. We found that with the exception of harm, those in None group had the most negative views about the process and the biobank compared with the other groups. When it came to hypothetical results received, those

in the Granular group showed the greatest satisfaction, the None group were the least satisfied, and the All and Binary groups were in between. Thus, the benefits of enrolling in a biobank appear to be perceived as the greatest by those who receive results and those who have choices regarding what to receive. The finding that return of results led to greater satisfaction is not unexpected as others have shown that participants are more likely to enroll in a biobank if there is return of research results (D. Kaufman, Murphy, Scott, & Hudson, 2008; O'Daniel & Haga, 2011). Our results take these findings one step further and examine participant satisfaction once enrolled in a hypothetical biobank, and we show that indeed satisfaction is higher for those enrolled in a biobank where they received hypothetical results.

Our results also show that, although satisfaction was highest for those enrolled in the Granular group, satisfaction was still reasonably high in those who received all results or had a choice between all and none. Thus having a choice, even it is just all or none, or just receiving results, provides more satisfaction than no return of results. The implication is that providing granular preferences may not be needed and that just offering a choice of all or none may be adequately beneficial without requiring an infrastructure to support granular preferences.

There has been much concern about potential harms in return of genomic results. Interestingly, there was no significant difference between the four groups with regard to the statement that the biobank was harmful. Thus, although there is a concern of greater psychological harms with return of results, we did not see any increase in harms in the Granular group. These findings are consistent with the finding of an association with baseline reasons for wanting information, including benefits of preparing for the future, having more control over the future, preventing worry, and seeking medical treatments, with increased satisfaction with the process and results. Thus, it appears that satisfaction is associated with reasons to want information (benefits) and not reasons to NOT want information (harms).

There are limitations to our study. The biobank and the results returned were hypothetical. However, randomizing participants to different biobanks allowed us to compare across groups even though the scenario was hypothetical. The response rate was low, at about 24%, although this was higher than our previous mailed paper survey, and respondents were generally Caucasian, well educated, and female, all of which may affect generalizability. The dropout rate during the survey was highest for the Granular group, which was not unexpected because the Granular survey was longer and more involved.

Return of genomic results from biobank research will be a challenge. In the future, with adequate online education and an opportunity to see what types of results one might receive after setting preferences, may make the ability to set granular preference for return of results a feasibly reality.

Best Practices

Recent recommendations have suggested that returning results to participants in genomic research may be desirable. One of the biggest limitations, however, is feasibility. Our goal was to use the return of hypothetical results to evaluate the stability of preferences and satisfaction with process for four biobank types, including a biobank that implemented a preference-setting model. Our results also show that, although having a choice, even if it is just all or none, or just receiving results, provides more satisfaction than no return of results, a more granular choice is the most desirable. Our findings suggest that biobank researchers may want to consider return of results and some degree of preference setting to enhance enrollment and satisfaction. Our findings also suggest that an online tool, such as the one we developed and implemented, may make preference setting feasible.

Research Agenda

We have now tested a preference-setting tool in an online format with a large cohort of individuals using hypothetical scenarios. The next step is to implement in a biobank where actual results are being returned to participants and to study the outcomes in terms of benefits, harms, and satisfaction.

Educational Implications

We have implemented a model for return of results that provides education for participants and that removes some burden from researchers to return results from large studies on a case-by-case basis. The model teaches participants to consider the potential beneficial and harmful implications of IRRs.

Declaration of Conflicting Interests

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Kurt D. Christensen is a postdoctoral research fellow at Brigham and Women's Hospital and Harvard Medical School. His research focuses on the behavioral and economic impact of emerging genomic technologies in clinical settings, including the use of whole genome and whole exome sequencing in the care of adults and newborns. He assisted in developing the survey, analyzing data, and writing the article.

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