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ARTICLE

Lay attitudes toward trust, uncertainty, and the return of pediatric research results in biobanking

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ABSTRACT

Background: Trust plays a role in participants' reactions to clinical residual biobanks. The purpose of this study is to assess whether trust in medical researchers and negative reactions to uncertainty influence the attitudes of parents of pediatric research participants toward the return of genomic research results from biobanking. **Methods**: Focus groups were conducted in collaboration with two community-based organizations. Participants completed a demographic questionnaire and the trust in research and intolerance of uncertainty scales. The focus groups were then conducted according to a thematic focus-group guide; discussions were transcribed and analyzed by two trained coders. **Results**: Emerging themes included the importance of returning research results to both children and parents, sharing results with few limitations based on a child's age, and the desire for results even when researchers had concerns about analytic validity. Negative reactions to uncertainty appear to have influenced only one theme: the paradoxical claim by participants with stronger reactions to uncertainty that they had a "right to information." **Conclusion**: Participants prefer to receive most or all of the results produced by genomic research, and they want their children, within variable age restrictions, to have access to that information as well.

KEYWORDS
return of results; biobanking; trust; uncertainty

Many medical research institutions are instituting large-scale research projects using biorepositories and next-generation sequencing (Henderson et al. 2013). These research projects raise a variety of issues related to consent, return of research results, and the potential for incidental findings (Beskow et al. 2009; Bledsoe et al. 2012; Clayton et al. 2010; Jarvik et al. 2014; Shalowitz and Miller 2005; Wolf et al. 2008). All of these issues become further complicated when biobanking occurs in pediatric settings (American Academy of Pediatrics Committee on Bioethics, Committee on Genetics, and the American College of Medical Genetics and Genomics Social, Ethical, and Legal Issues Committee 2013; Avard et al. 2011; Goldenberg et al. 2009; Hens et al. 2010; Wilfond and Diekema 2012).

In pediatric clinical settings, conflicting recommendations exist for the return of results. One policy statement from the American College of Medical Genetics (ACMG) and the American Academy of Pediatrics (AAP) advises that children should not be tested for adult-onset genetic conditions (AAP Committee on Bioethics, Committee on Genetics, and ACMG Social, Ethical, and Legal Issues Committee 2013), while another ACMG policy statement indicates that when genomic testing occurs in children, a specific set of adult-onset genetic conditions should also be included in the analysis (Green et al. 2013).

Genomic testing in research settings also raises concerns about the propriety of testing for adult-onset conditions and adds further ethical complexities unique to the pediatric research setting. Further empirical research into community attitudes can help guide researchers and institutional review boards (IRBs) as they develop local policies on pediatric return of results (Clayton et al. 2014).

Additionally, a number of statements on the return of results in genomic research and biobanking indicate that "uncertainty" about the information is a key issue for participants, researchers, and IRBs to consider (Bookman et al. 2006; Miller et al. 2008; Shalowitz and Miller 2005; Wolf et al. 2012). For example, genomic tests being developed in a research setting might not have sufficient clinical validity, or the actionability of research results might be unclear or rapidly changing. Uncertainty about these issues might influence the reception of genomic research results.

Finally, in the extant literature (Brothers, Morrison, and Clayton 2011), and in our experience of community engagement around genomics and biobanking, we have seen that trust in medical researchers plays a role in participant attitudes toward biorepository research generally. Being able to assess the degree of trust in a research institution and assessing the degree to which uncertainty of information might be troubling for research participants can contribute to IRB deliberations about local biobanking policy. In this article, we describe a series of focus groups we conducted that explored participants' attitudes toward the potential return of results and participants' trust in medical researchers and their reactions to uncertainty.

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Methods

We combined qualitative focus groups and quantitative survey measures to assess how laypeople respond to the possibility of receiving genomic research results and to see whether scale measures for intolerance of uncertainty and trust in medical researchers could be used to help identify attitudes toward the return of results (ROR) (Creswell 1998; Morgan 1997; Strauss and Corbin 1990).

Participants were recruited through community partner organizations located in neighborhoods that are part of a region-wide community development project. The project is led by our home institutions (i.e., a research university and a pediatric research hospital) and has a substantial public health component. The project oversampled low socioeconomic (SES) and non-white populations, since these groups are likely to have greater mistrust of medical research and are typically underrepresented in studies of the social impact of genomics. Focus groups were conducted at a church and a community center in September and October 2013. This protocol was approved by the IRB at Cincinnati Children's Hospital Medical Center. Participants were given a copy of the informed consent document to read, and a research team member also read through the consent document with each focus group, before participants were asked to sign the informed consent form.

After acquiring informed consent, participants filled out a questionnaire that included demographic questions, the 12-item Intolerance of Uncertainty Scale (IUS) (Carleton, Norton, and Asmundson 2007), the 12-item Trust in Medical Research (TR) scale (Hall et al. 2006), and a series of questions about who should return results to the parents of biorepository participants and to the pediatric biorepository participants themselves. The IUS assesses the degree to which the uncertainty inherent in future events and outcomes creates anxiety and inhibits action (e.g., the degree to which the uncertainty inherent in research results might make participants fearful or alter their health behaviors). The TR scale assesses the degree to which respondents view medical research as an enterprise and individual medical researchers as deserving of their trust. Both the IUS and TR scales ask participants to respond to a series of items on a 5-point Likert scale. Higher scores indicate greater intolerance of uncertainty or greater trust, respectively.

Next, an experienced moderator conducted the focus group using a semistructured moderator guide. Participants were asked about their knowledge of the concept of biobanking and reported their initial reactions. Participants were then given a description of the pediatric biobank and its opt-in consent process and asked to respond. Then participants were asked a series of questions about what results should be returned to parents, what results should only be returned to children, the role children should play in choosing results to receive, whether scientific uncertainty or new scientific developments would change their opinions about ROR, and who should return results to the parents and the child, respectively. The hypothetical context for ROR was a situation where results were not immediately clinically actionable but where the results were clinically valid or the results had minimal or uncertain significance. This broad hypothetical context was used to elicit the range of attitudes, including potentially conflicting attitudes, that motivated participants' ROR preferences.

Missing responses to items on the IUS and TR scales were imputed using the total mean score from all other items. Scores for the IUS and TR scales are produced by summing the total scale score for a range of possible scores on each scale ranging from 12 to 60. Because of the small number of participants, only descriptive statistics are presented. The individual scores on both scales were used for methodological triangulation to help assess individual responses to questions about the return of results.

The focus groups were audio- and videotaped. Transcripts were developed from the audiotape by a professional transcriptionist, and videotape was used to verify the transcript and to assign each speaker a unique identifier. A coding scheme was developed using a combination of deductive codes developed from the questions and inductive codes developed from a close reading of participant responses. Two trained coders independently coded 20% of all transcripts. Intercoder reliability was assessed using Cohen's kappa, with a cutoff of .50 to indicate agreement (Landis and Koch 1977). For codes with reliability less than .50, coders were retrained, and intercoder reliability was reassessed until a minimum of .50 was reached. Final kappa values ranged from .50 to .99. After reliability was established, the remaining transcripts were coded by one of the coders. Coding was performed using NVivo 9.0 qualitative research software. Transcripts and the coding scheme were uploaded into the program. Talk turns were marked with each participant's unique identifier, and the scores for the IUS and TR were linked to those identifiers. This allowed for the use of methodological triangulation to guide analysis of the coding (Denzin 1970).

Results

There were 40 participants in the four focus groups. Participants were overwhelmingly female (37/40 participants; see Table 1); 24 participants were African American, 10 European American, 4 multiracial, and 2 participants identified as Hispanic. Fifty-five percent ($n = 22$) of participants reported

Table 1. Demographics.

	Mean	*n*
Age	32.48 years	35
	Percent	*n*
Gender		
Male	7.5%	3
Female	92.5%	37
Income		
Less than $10,500	59.5%	22
$10,500–13,999	5.4%	2
$14,000–17,599	10.8%	4
$21,200–24,799	10.8%	4
$24,800–29,999	2.7%	1
$30,000–39,999	10.8%	4
Race		
White/Caucasian	26.3%	10
Black/African-American	63.2%	24
Multiracial	10.5%	4

Note. Based on reported/available data.

household incomes of less than $10,500, and 60% percent ($n = 24$) reported that the highest level of education in their household was a high school diploma/GED or less.

Scores on the TR scale ranged from 24 to 56, with a mean score of 40.41, indicating a relatively neutral attitude toward medical researchers. On average, participants were ambivalent in their trust of medical research. Scores on the IUS scale ranged from 19 to 53, with a mean score of 39 indicating relatively neutral attitudes toward uncertainty. Unless otherwise noted in the results, participants had scores on TR and IUS that were in the mid or neutral range (30−47).

Returning results to parents

Participants in all four focus groups indicated that they wanted to receive all results (for an overview of themes, see Table 2). One female European American participant indicated that she wanted "whatever results come out [of the research]" (PH-2-3054). An African American female participant stated, "All of them" (AV-2-2052). During the initial discussion of ROR only one participant, an African American female, indicated any hesitation about returning results: "Well, I think they need to give them back what they can understand" (AV-2-2058).

Most often, participants asserted that parents should receive results of research from a biobank without explaining or justifying their position. When they did offer justifications, participants typically gave one of three broad reasons, emphasizing that it was the right thing for researchers to do. First, some indicated that ROR was necessary to maintain good hospital-community relations. When explaining why the hospital needed to return research results, one African American participant said: "Once you do that, just leave [people] hanging, it makes it even harder [to do] further research because no one wants to do anything when they find out that this was done to them" (AV-2-2058). For this participant, continued support of research and participation in it depend on the institution sharing results with those who participated instead of failing to share (i.e., "just leave [people] hanging").

Second, some individuals emphasized that medically important information should be returned, even when they desired the return of information with clinical significance and information with no clinical significance. One African American participant claimed: "I said it's good to know early because if you do have a cancer gene, you know, you'd be able to, you know, keep an eye on it. Go to the doctor and try to get it detected early" (AV-2-2051). A European American participant stated, "You want to know every possible thing that could be wrong, every possible information *[sic]* you could have" (PH-2-3057). For these participants, the potential clinical utility of information was used to justify the return of results, even when the scope of results they desired to have returned exceeded the bounds of clinical utility.

Third, most participants claimed that parents had a right to information about their children. This was the most prevalent reason given throughout all focus groups. A European American female participant stated, "Everyone should have rights [to this information]" (PH-2-3054). This claim borrows the rights language common to everyday American discourse and applies it to potential return of research results.

Most participants indicated they would still want to receive results even when scientists were uncertain about their results (i.e., scientists were not confident of the test's accuracy or did not know what the results could mean clinically). One African American female participant simply stated, "I would still want to know" (AV-2-2058). Another participant justified receiving the information with the claim "I think it's important" (PH-1-3044), and a European American female participant said, "At least you know what you're facing with your kid" (PH-2-3051).

Some participants in one focus group did indicate concerns about receiving information that medical researchers felt was not completely accurate. The participants who indicated concerns were African American females who had higher scores on the IUS scale. One participant with an IUS score of 50 said, "I mean if they're not accurate, they shouldn't give it to them at all" (AV-1-2045). Another participant with an IUS score of 43 stated, "No. It will make your nerves bad" (AV-1-2043). For these two participants, uncertain information should not be provided because it would aggravate their intolerance of uncertainty and ambiguity, but they were the only participants to say this.

Returning results to children

Most participants initially indicated that children should also receive most or all of the results of research in which they participated, although many subsequently offered restrictions on the return of results based on the maturity of the child. An African American female participant said that children at all ages had a "right to know" the results of research: "I mean [if] something is being done to them, they should have the right to know what's going on" (AV-1-2045). This statement was made just after the group had asserted that concerns about uncertainty or the incompleteness of results should limit the return to parents.

Yet in another focus group, an African American female participant indicated that medically important results should be returned, otherwise "they would never know if something bad happens … it might be too late" (AV-2-2052). The issue of hospital−community relations was not raised when discussing the return of results to children, but participants did raise the issue of reproductive decision making: "When your child turns 18, what if they have children? They need to know" (PH-2-3059).

While many participants indicated children should receive results, there were several who indicated that parents should act as gatekeepers of that information. An African American female participant indicated that she would only pass on information about serious medical conditions to her child: "So it would depend on the situation. If the illness is not that serious

Table 2. Overview of themes.

1. Parents want any and all research results returned to them.
2. Parents feel the return of research results is the proper thing to do.
3. Despite uncertainty, parents want research results.
4. Children should receive results in an age-appropriate fashion and with parents acting as gatekeepers.
5. Parents want to receive results from familiar and trusted sources.

to where they don't need to know, I probably wouldn't tell mine" (PH-1-3041).

Some descriptions of gatekeeping were more complex and emphasized that parents would give meaning and context to information provided by a primary health care provider or medical researcher. A European American participant said, "So you can speak to them in your terms and then when the doctor comes in, he can sit there and explain too" (PH-1-3049). For this participant, the parent would receive the information first and prepare her child to receive that information, before the physician visited the room to share the information with the child.

Overall, participants described a range of ages as appropriate for a child to receive research results. All of the ages identified were justified by appeals to a child's "maturity." Very few participants thought children at ages 10–12 years or younger should be involved, and they emphasized that some complex gatekeeping would be required to make sure the child understood the medical information in an age-appropriate fashion. One African American participant said:

> I think they should sit down and listen and when they go home, the parents can actually tell them what it is really [going] on because the parent is going make it be more like … informed so they know what's going on instead of the doctor or whatever telling them what's going on. (AV-2-2052)

Another African American participant (AV-2-2058) identified reproductive issues as a reason to begin informing children aged 13–15.

Others emphasized that late teen years (16+) were the appropriate time for children to make decisions about receiving research results. A European American participant drew on her own medical experiences: "I think they should get a choice because I know when I was a teenager, I went to the doctors a lot without my mom" (PH-1-3049).

Finally, some participants indicated that even children who had reached the age of consent might not be mature enough to receive results, and the need for reconsent was never raised during the discussions. For the most part, participants did not elaborate on what constituted "maturity." Some did not think legal age of consent was sufficient, but others found that biological maturity and the possibility of the child having his or her own children sufficient.

Who should return results?

Participants were asked who should explain results to parents of biobank participants. The majority identified their primary health care provider as their first choice, with the researcher who identified the result as their second, and a genetic counselor as their third (see Table 3).

Table 3. Who should explain results to parents? ($N = 36$).

	First choice	Second choice	Third choice
Researcher who found result	12	15	9
Primary health care provider	21	11	4
Genetic counselor	3	10	23

When explaining the choice of the primary health care provider as the point of return, several reasons were given. An African American participant indicated the primary health care provider's familiarity with her family was the important factor: "Well, I have a family doctor and they know … about my past and family and whatever" (AV-1-2046). Another multiracial participant with a low TR and a high IUS score emphasized trust in clinical practitioners: "I'd rather it be the physician that I trust with my child" (PH-2-3050). The primary health care provider's presumed knowledge of family health history and the trust participants invest in their health care provider motivated this choice.

Participants who chose the researcher emphasized the researcher's presumed knowledgability: "I feel like a researcher would know the most about it" (PH-1-3048). Participants gave few reasons for why they ranked genetic counselors as they did. The multiracial participant who emphasized her primary health care provider's knowledgability said, "I think they might know the genetics … but they might not necessarily know the research" (PH-1-3047).

Participants were then asked who should explain results to the child participant. Parents were the first choice, followed in order by primary health care provider, researcher, and genetic counselor (see Table 4). Participants explained their choice of parent first by emphasizing their familiarity with their own children. An African American participant said, "I picked myself so I can know how she'll feel. It depends on her first reaction so I can know what I'm dealing with" (PH-1-3047). Again, familiarity more than knowledge about research drove the preferences for who should present the research results.

Discussion

Our findings indicate that all participants, regardless of their trust in medical researchers or their ability to tolerate uncertainty, believe parents of pediatric participants in biorepository-based genomic research should receive research results. This high level of interest in receiving results mirrors the level of interest reported in many different reports, including surveys of clinical genetics professionals (Lemke et al. 2012), surveys of participants in genome research (Fernandez et al. 2014), participants in psychiatric research (Bui et al. 2014), and public attitudes toward pharmacogenetics (Haga et al. 2011a; 2011b).

What is surprising is the level of interest reported in research results when the researchers are not confident in the analytical validity of the test or where the researchers are uncertain of their clinical significance. Only a few of the participants with high scores on the IUS scale reported concerns about

Table 4. Who should explain results to the child? ($N = 36$).

	First choice	Second choice	Third choice	Fourth choice	No answer
Parent	20	10	2	4	0
Researcher who found result	2	7	18	8	1
Primary health care provider	14	14	3	4	1
Genetic counselor	0	4	12	19	1

receiving this category of results, and not all who scored high on the IUS scale agreed with that position. While we cannot rule out some misunderstanding on the part of participants or the possibility that the hypothetical nature of the ROR discussed skews participant attitudes, we believe participants desired the return of even questionable results because of the value attributed to the label "information" in the broader culture and in the historical context of prenatal screening, where the value of screening was articulated in terms of the "information" it provided (Lynch 2011; Press et al. 2011).

While this desire for results with low analytic validity should not necessarily alter how institutions perform ROR, researchers will need to think carefully about how they label and present biorepository research results when communicating with research participants and the general public. Researchers should also develop criteria for returning results at the outset of the study and consider communicating those to participants as well (Knoppers et al. 2014).

Participants offered a diverse range of reasons for ROR. We anticipated that a right to information, the need for clinically actionable results, and the need for results with implications for reproductive decision making would be mentioned. Yet medical and reproductive reasons for ROR only justify more restrictive schemes for ROR than the broad ROR participants indicated (Hens et al. 2013). This might be modified by the perception of some participants that almost any result could have medical or reproductive significance, and it indicates that researchers and IRBs should be cautious and verify that research participants and researchers are employing the same meanings when they use the same or similar words.

Additionally, participants articulated an unanticipated reason of hospital−community relations as the justification for ROR. This, in turn, raises the issue of whether researchers and research institutions have some obligation to research participants and their communities in regards to ROR (Biesecker 2013; Clayton and Ross 2006; Garrett 2013; Gliwa and Berkman 2013; Miller et al. 2008).

Many participants indicated a desire for children to receive results, although more participants indicated qualms about giving results to children than about parents receiving them. There was substantial discussion about the parent's role as a gatekeeper of information for the child. Sometimes gatekeeping was a straightforward yes−no decision about whether a child should hear a specific result, and in other cases it involved a child and parent receiving results simultaneously and having the parent then interpret the results for the child.

Participants indicated that assessments of a child's maturity would influence their belief that a child should or should not be informed of the research results. Furthermore, participants indicated a range of ages at which children would be mature enough to receive results, ranging from 11 to 18 years.

A few participants questioned whether 18-year-olds, who are legally adults, should receive results. It is not clear whether this reflects concerns about their own specific children or perhaps some unconscious concern about broad return of results generally, but it reaffirms the observation that pediatric research involves a three-way relationship among researchers, pediatric participants, and their parents (Avard et al. 2011). This three-way relationship should also lead us to appraise parental desire for all genomic results about their child, including results about adult-onset conditions. Providing all results to parents can threaten the child's right to an open future and the child's right to refuse results.

Our results were equivocal about the value of measurements of trust and intolerance of uncertainty in assessing participants' desires to receive genomic research results. Trust in medical researchers was not associated with any stated preference for ROR or with the reasons expressed for ROR. While trust might be a factor in reactions to biorepository research, it might be more accurately conceptualized as trust in a specific institution rather than trust in medical researchers generally (see Brothers, Morrison, and Clayton 2011). Institution-specific questions of trust might provide a measure that can differentiate degrees of trust in relation to opinions about biorepository research.

A few of the participants who scored higher on the IUS scale (IUS $\geq$ 48) indicated concern about the return of a broad range of results, and participants who scored higher had their talk turns coded more often as "right to information." While "uncertain" information might bother some, it seems that some individuals who do not tolerate uncertainty well still demand access to information, often framing it as a "right." It seems that the assertion of control over information itself is a way of managing the anxiety produced by uncertainty by providing those individuals with a course of action—in this case, to approach researchers and demand information—regardless of the outcome or information received. Whether these individuals would also demand more information or tests from health care providers is unclear. Further research with a larger cohort would be required to establish this relationship and to identify how individuals use the information they have unwaveringly demanded.

Participants indicated a preference for primary health care providers to return research results to parents and for the parents to return the results to their child. Familiarity with the child and the family was the primary reason expressed for both choices. Genetic counselors were listed as the last choice, but this likely reflects a lack of familiarity with genetic counseling rather than an assessment of the practice. Participants' desires to receive results from primary health care providers will be complicated by primary health care providers' lack of knowledge about genomic testing and lack of resources and time for incorporating complex genomic information into their practices (Baars, Henneman, and ten Kate 2005; Goos et al. 2004; Greendale and Pyeritz 2001; Grody, Thompson, and Hudgins 2013).

Limitations

Our study has several limitations. First, because this was a focus-group study, our observations about participants' responses cannot be generalized to entire groups. Also, the responses to the TR and IUS survey measures are only descriptive and do not allow for statistical analysis. Quantitative studies would be needed to assess any hypothesis about IUS and attitudes toward returning results. Second, our sample was overwhelmingly female and African American. It is unclear whether a greater number of male participants or a greater number of non-African American participants would alter our

findings. Third, participants were considering hypothetical ROR. It is possible that being offered actual ROR might elicit different reactions.

Conclusion

Overall, we believe that our findings support existing research showing that members of the public desire the return of genomic research results and that the value inherent in the labeling of results as "information" contributes to that desire despite uncertainty about analytical validity of research results and their ultimate clinical utility.

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Author contributions

John Lynch contributed to the conception, design, data collection, and drafting of the article. Janelle Hines and Sarah Theodore contributed to the data collection and drafting of the article. Monica Mitchell contributed to the design, data collection, and drafting of the article.

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Conflicts of interest

The authors report no conflicts of interest.

Ethical approval

This study was approved by the institutional review board at Cincinnati Children's Hospital Medical Center.

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