

Moving Beyond Return of Research Results...

To Return of

VALUE

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Overview

- Current state of return of research results
- Why we should rethink return of results
- What is valuable to return to participants

Background: returning results

- Broadly defined as **the process of sharing study results back with study participants**
- May include **individual and/or aggregate** study results
- Increasingly seen as an essential for:
 - Responding to **participants' expectations and interests**,
 - **Recognizing contributions** they make to research,
 - **Engaging** those individuals more deeply in the research process, and
 - Allowing **for integration of results into health and care planning**, when the data is actionable

Challenges: returning results

- A range of **participant preferences** for receiving results
- Variability in participant **literacy**
- **Gaps in researcher expertise** in strategies for returning results
- Challenges in **identifying actionable and/or useful results**
- **Positive and negative impact on participants' perceptions** of research participation
- **Ethical considerations:**
 - Informed consent,
 - privacy
 - sharing of results with provider



Researcher barriers to returning results

- **Which results?**
- **Who discloses?**
- **How long does obligation last?**
- **Challenges with consent**
- **Who pays for associated costs?**
 - Referrals
 - Counseling
 - Education



Challenges to return of results (ROR) in minorities and vulnerable populations

- The proportion of African Americans not interested in ROR was higher as compared to non-African Americans in a study considering sequencing data (Yu et al., Am J Med Genet 2013)
 - May be partly shaped by different expectations about health benefits and how results need to be managed
- Also significant variability in **parental preferences for ROR related to pediatric biobanking and other research**; this is reflected in studies focusing on African American opinions as well (Halverson and Ross, J Med Ethics 2012 & J Community Genet 2012)

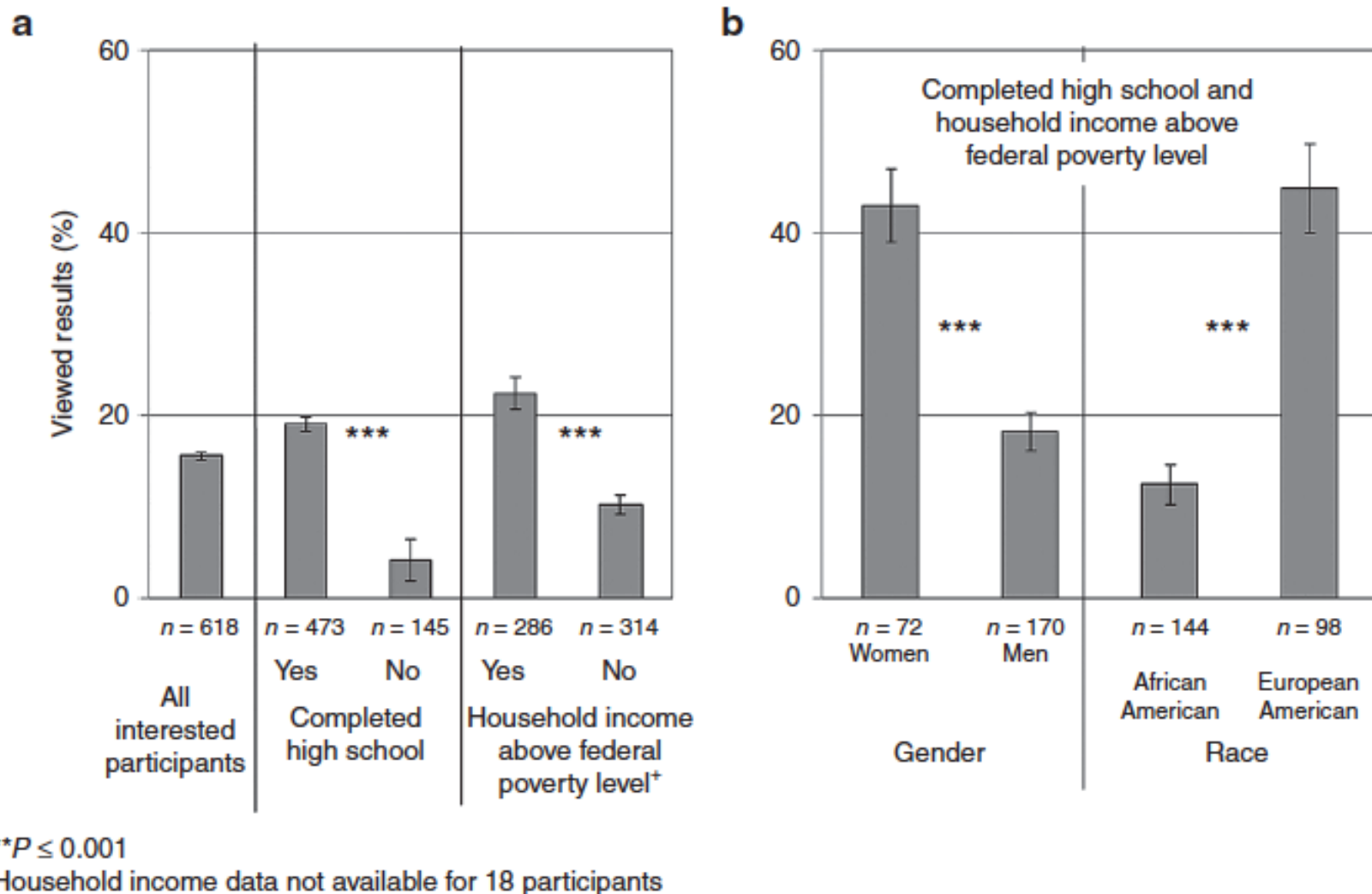


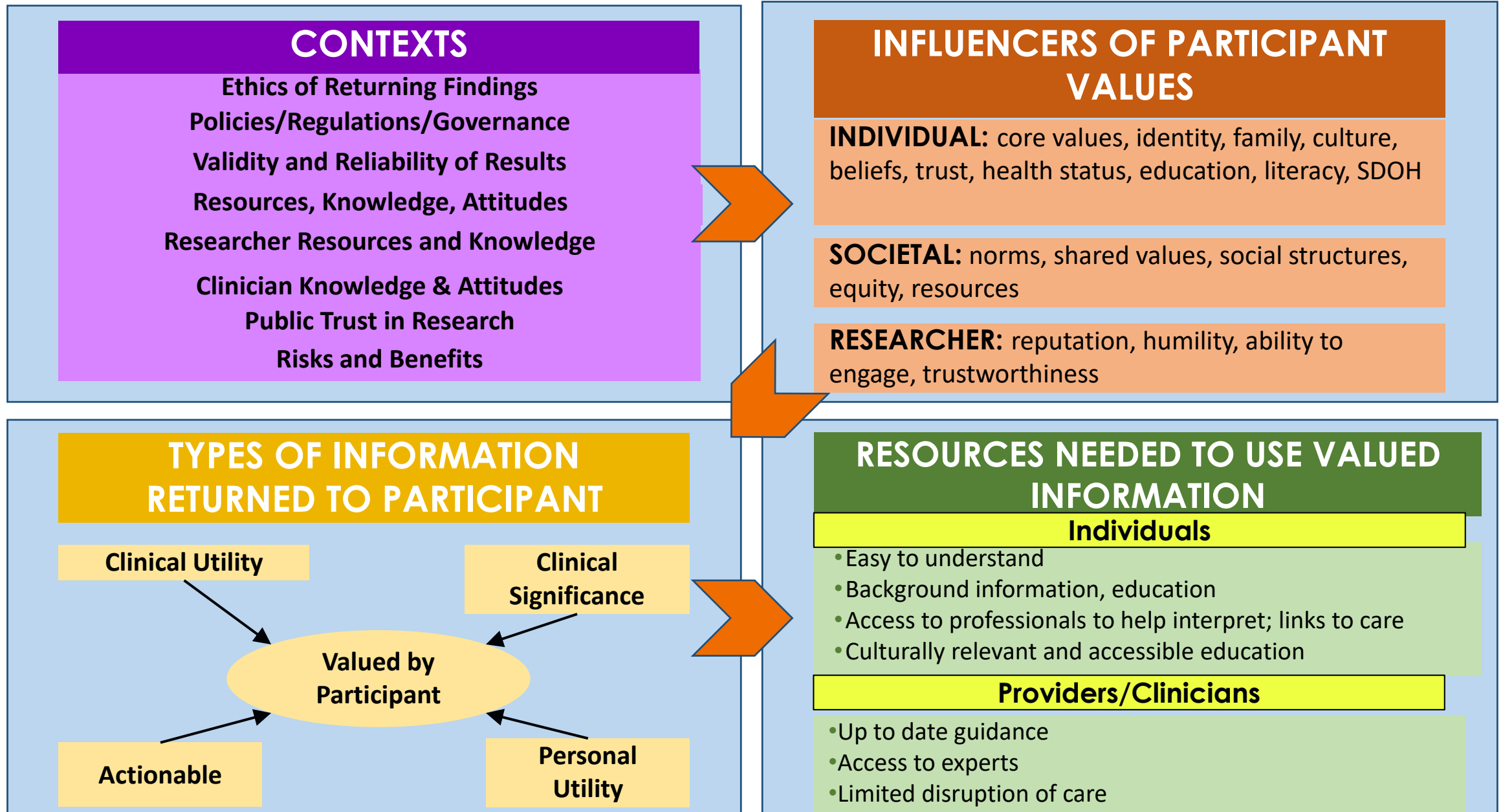
Figure 1 Education, poverty, gender and race impact participant engagement. (a) Viewing genetic ancestry results by participants who reported being "extremely" or "very" interested in viewing results varies by education and income. (b) Viewing genetic ancestry differs across gender and race among interested participants who completed high school and have a household income above the federal poverty level. Error bars represent 95% confidence intervals; *P* values are from logistic regression adjusted for age, gender, and race.



Return of Value

Return of Value Conceptual Framework

Wilkins, Mapes, Jerome, Villalta-Gil, Pulley, Harris. Health Affairs, March 2019



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Wilkins, Mapes, Jerome, Villalta-Gil, Pulley, Harris. Health Affairs, March 2019

CONTEXTS

Ethics of Returning Findings
Policies/Regulations/Governance
Validity and Reliability of Results
Resources, Knowledge, Attitudes
Researcher Resources and Knowledge
Clinician Knowledge & Attitudes
Public Trust in Research
Risks and Benefits

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INFLUENCERS OF PARTICIPANT VALUES

INDIVIDUAL: core values, identity, family, culture, beliefs, trust, health status, education, literacy, SDOH

SOCIETAL: norms, shared values, social structures, equity, resources

RESEARCHER: reputation, humility, ability to engage, trustworthiness

Understanding trust in research

Conceptions of Research

- Perceptions and understandings of research
- Altruism
- **Community benefits**
- Risk-harms

Dimensions of Trust

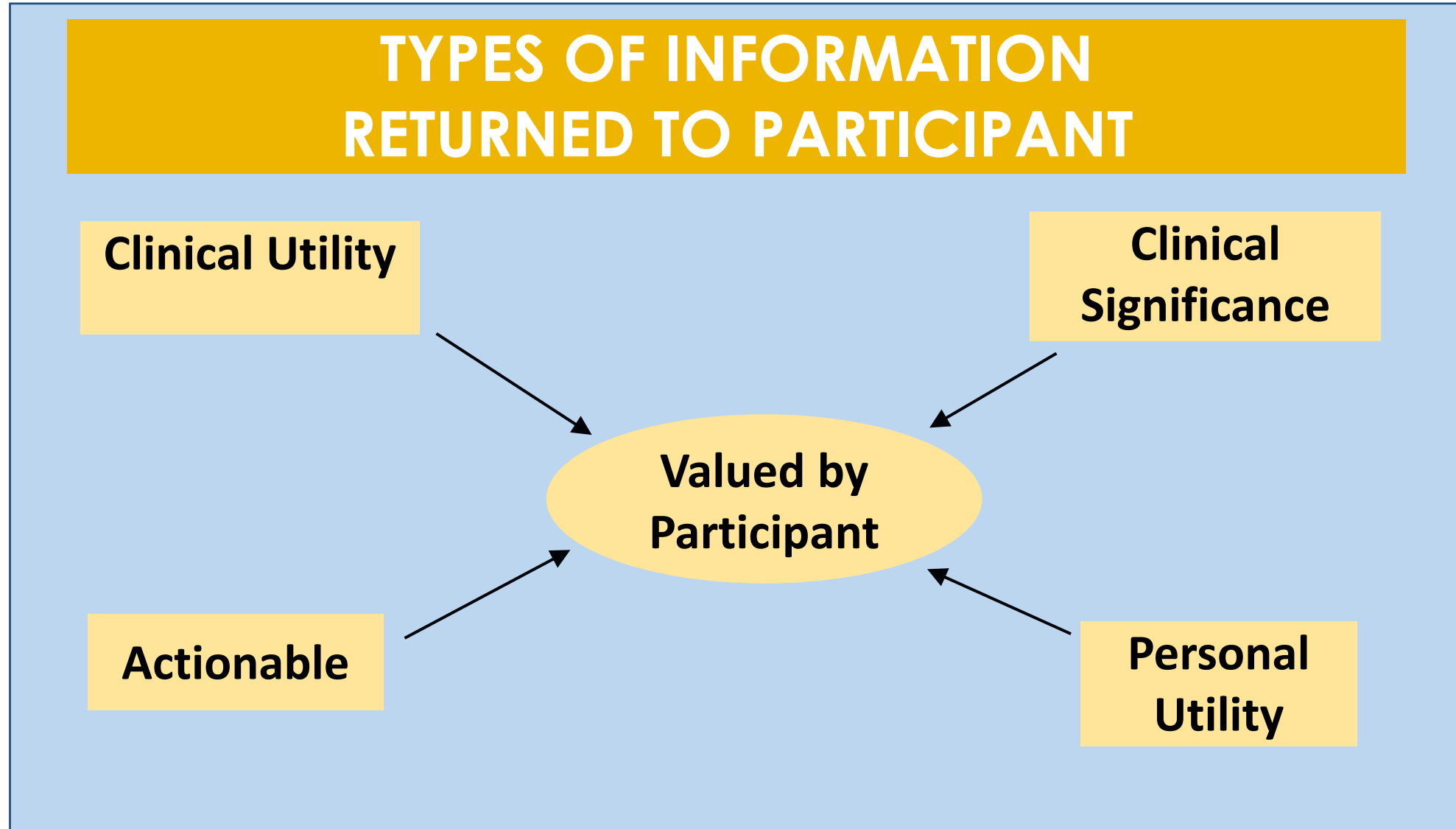
- Communication
- Honesty
- Confidence
- Confidentiality
- **Privacy**
- **Secrecy**

Determinants of Trust

- Trust in clinic/health system
- Trust in researchers
- **Profit-Incentives**
- **Historical Abuse**
- Personal Experiences
- **Trustworthiness of researchers**
- Community involvement in research
- Research participation
- Confidence in research results

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Term	Value Concept	Example
Clinical utility	Direct clinical value	The availability of a proven therapeutic or preventive intervention. ¹¹⁰
Clinical utility	Direct clinical value plus some measure of the other kinds of value	Consider clinical validity, the likelihood of a clinically effective outcome, and the value of the outcome to the individual. ¹¹¹
Clinical significance	Direct clinical value	Results that indicate the need for follow-up clinical consultation; ¹¹² results requiring medical or surgical attention. ¹¹³
Clinical significance	Direct and indirect clinical value	Whether identification of the variant permits an accurate prediction of the presence (or risk) of a clinical condition. ¹¹⁴
Actionability	Direct clinical value	The availability of effective treatment or prevention options; ¹¹⁵ the potential for an improved health outcome. ¹¹⁶
Actionability	Direct clinical value and direct non-clinical value	Getting treatment or prevention, informing family members of risk, making reproductive decisions, working for environmental action or remediation, life and financial planning, and participating in further research. ¹¹⁷
Personal utility	Direct non-clinical value	Information that may have benefits for reproductive decision making or life planning. ¹¹⁸
Personal utility	Direct and indirect nonclinical value	Verifiable results to which participants assign personal value. ¹¹⁹

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RESOURCES NEEDED TO USE INFORMATION

Individuals

- Easy to understand
- Background information, health education
- Access to professionals to help interpret; links to care
- Culturally relevant and accessible information

Providers/Clinicians

- Up to date guidance
- Access to experts
- Limited disruption of care

USING PARTICIPANT PREFERENCES TO INFORM ROUTE, FORMAT, TIMING

- What **kinds of results** confer perceived value by each individual participant?
- What are **preferred formats** for results?
- When is the **ideal time** for return of value for various kinds of information?



EMERGING APPROACHES TO RETURN OF VALUE:

**EXAMPLES OF HOW DATA CAN BE TRANSFORMED INTO
VALUABLE INFORMATION FOR PARTICIPANTS**



If EHR data are captured for research purposes, they can be repurposed and reoriented to participants



Understandable information on a particular **diagnosis** or **risk factor**, including action steps if possible

Research studies focused on my **disease** or **condition**

A list of **diagnoses** and **conditions** I have with links to more information

If vital signs are captured for research purposes, they can be repurposed and reoriented to participants

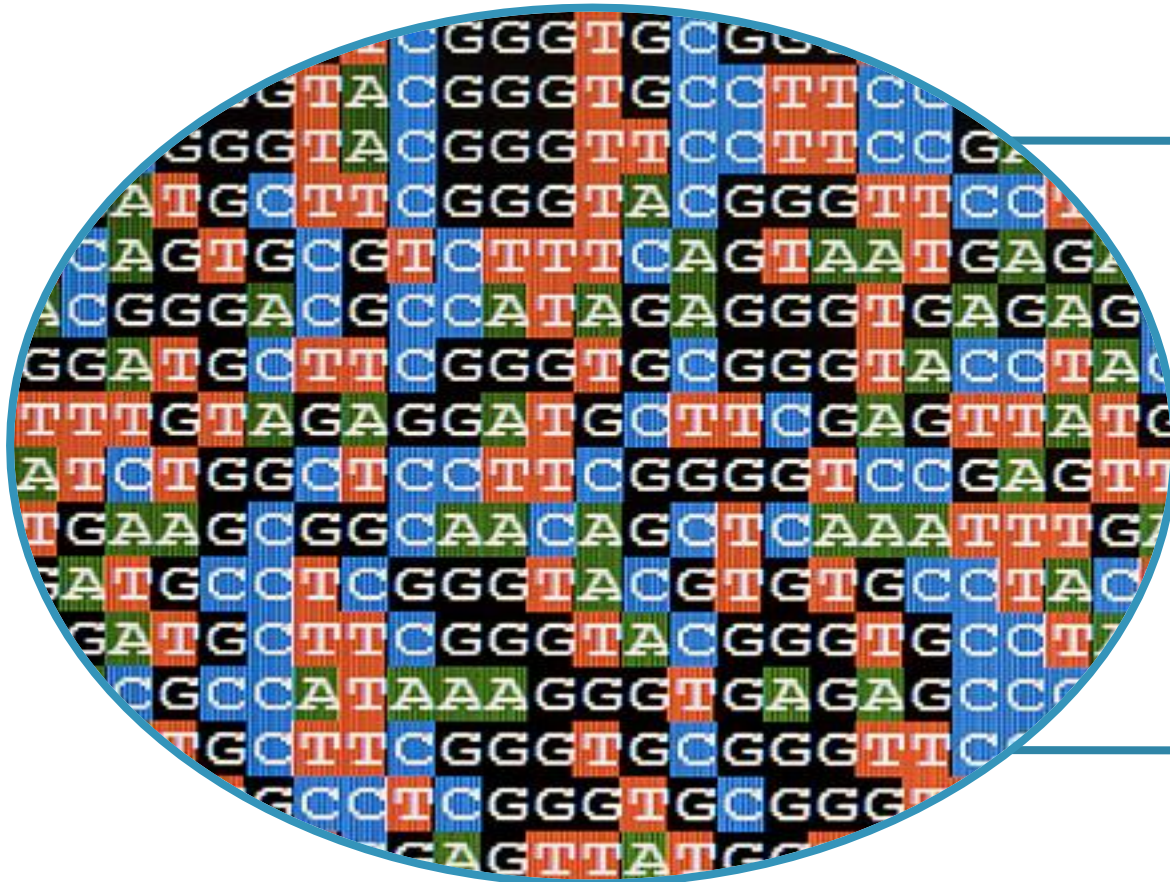


A graph of blood pressure readings **over time**

How my blood pressure **compares to others** similar to me (e.g., by age, gender, race/ethnicity, location, etc.)

Description of **risk profile** represented by my blood pressure trends, including flagging **action steps** if I wish to make a change

If genetic data are captured for research purposes, they can be repurposed and reoriented to participants



A geographical view of my
ancestry

My individual racial
composition

Medications I might not
respond well to



Understanding What Information Is Valued By Research Participants, And Why

Consuelo H. Wilkins, Brandy M. Mapes, Rebecca N. Jerome, Victoria
Villalta-Gil, Jill M. Pulley, and Paul A. Harris

March 2019

doi: 10.1377/hlthaff.2018.05046 HEALTH AFFAIRS 38, NO. 3 (2019): 399–407

HealthAffairs

Demographics of 2,549 participants in Return of Value survey

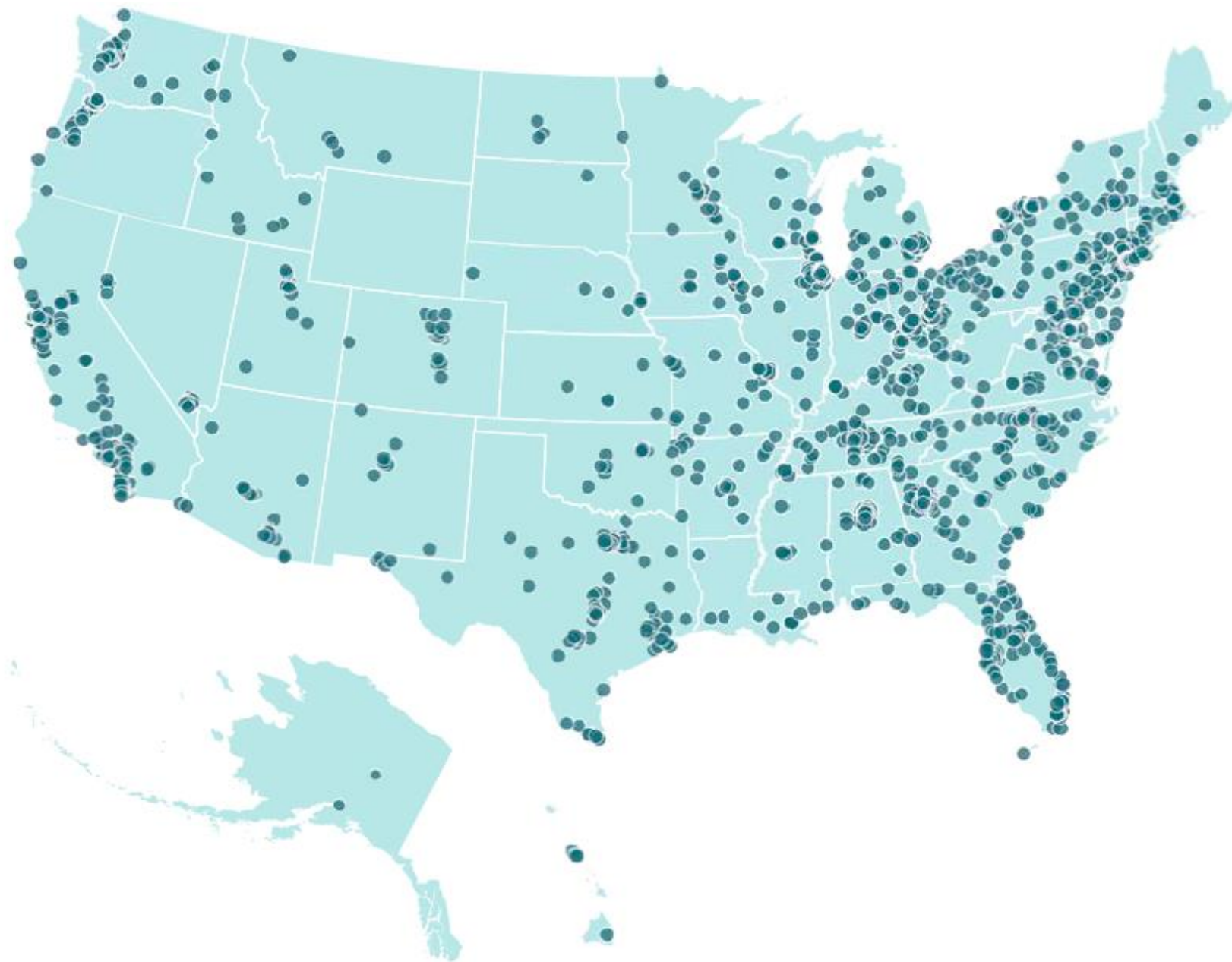
Characteristic	Number	Percent
AGE (YEARS)		
18-29	679	26.6
30-49	875	34.3
50-64	580	22.8
65-74	306	12.0
75 or more	103	4.0
Missing data	6	0.2
GENDER		
Male	980	38.4
Female	1,515	59.4
Other	15	0.6
Neither male nor female	27	1.1
Missing data	12	0.5

Demographics of 2,549 participants in Return of Value survey

	Number	Percent
RACE/ETHNICITY		
American Indian or Alaska Native	42	1.6
Asian or Asian American	307	12.0
Black, African American, or African	696	27.3
Hispanic, Latino, or Spanish	327	12.8
Middle Eastern or North African	22	0.9
Native Hawaiian or other Pacific Islander	10	0.4
White or Caucasian	1,041	40.8
None of these fully describe me	60	2.4
Prefer not to answer	37	1.5
Missing data	7	0.3

Demographics of 2,549 participants in Return of Value survey

	Number	Percent
EDUCATION		
High school or less	569	22.3
Some college	685	26.9
College graduate	688	27.0
Advanced degree	597	23.4
Missing data	10	0.4
ANNUAL INCOME		
Less than \$24,999	514	20.2
\$25,000–\$34,999	302	11.8
\$35,000–\$49,999	357	14.0
\$50,000–\$74,999	462	18.1
\$75,000–\$99,999	276	10.8
\$100,000 or more	424	16.6
Missing data	214	8.4



Wilkins, Mapes, Jerome, Villalta-Gil, Pulley, Harris. Health Affairs, March 2019

SOURCE Authors' analysis of Return of Value survey data from 2018. **NOTES** Data for Puerto Rico are not shown. The map was generated with ArcGIS Pro 2.1.0 using author data. A full list of ZIP codes is available on request.

How valuable are these to participants? (1= not valuable, 7= very valuable)

Item	All participants (N = 2,549)
How I may respond to some medications based on my genetics ^a	6.30
How my genetics affect my risk of getting a medical condition ^a	6.28
How my lifestyle affects my risk of getting a medical condition ^a	5.98
Information about clinical trials near me ^a	5.81
Information about how researchers are using my information ^a	5.77
My ancestry ^a	5.70
Monetary compensation for taking part in the study	5.64
Basic information about me (my lab results, survey responses, height, weight, etc.)	5.39
Information from my medical record ^a	5.35
How my health and behaviors compare to others'	5.31
My genetic traits	5.29
How to connect with others like me in the study ^a	4.08

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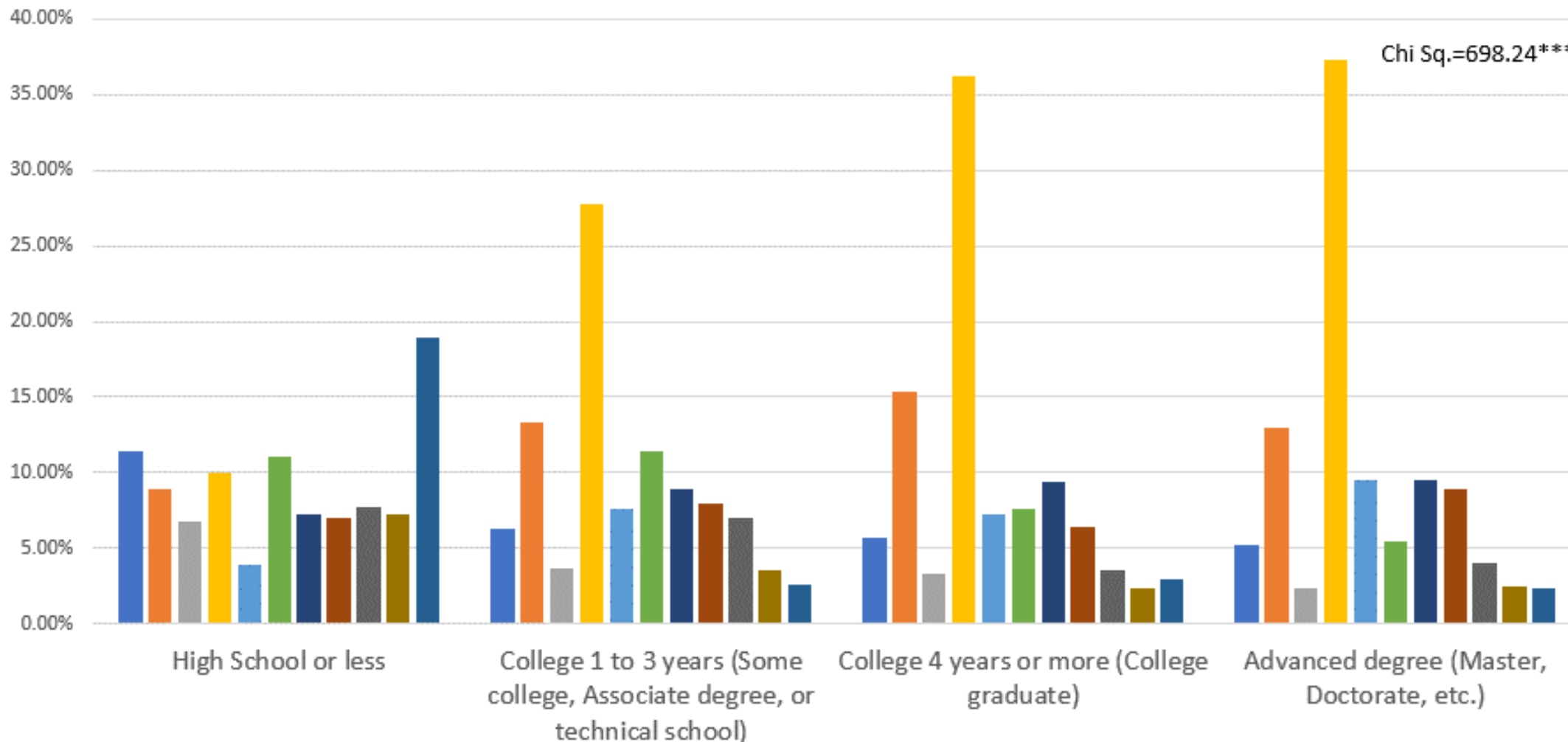
Which would be **most** valuable to you?

(choose only one; compensation an option)

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How my genetics affect my risks of getting a medical

How to connect with others like me.



Which would be **most** valuable to you? (Age)

(choose only one; compensation an option)

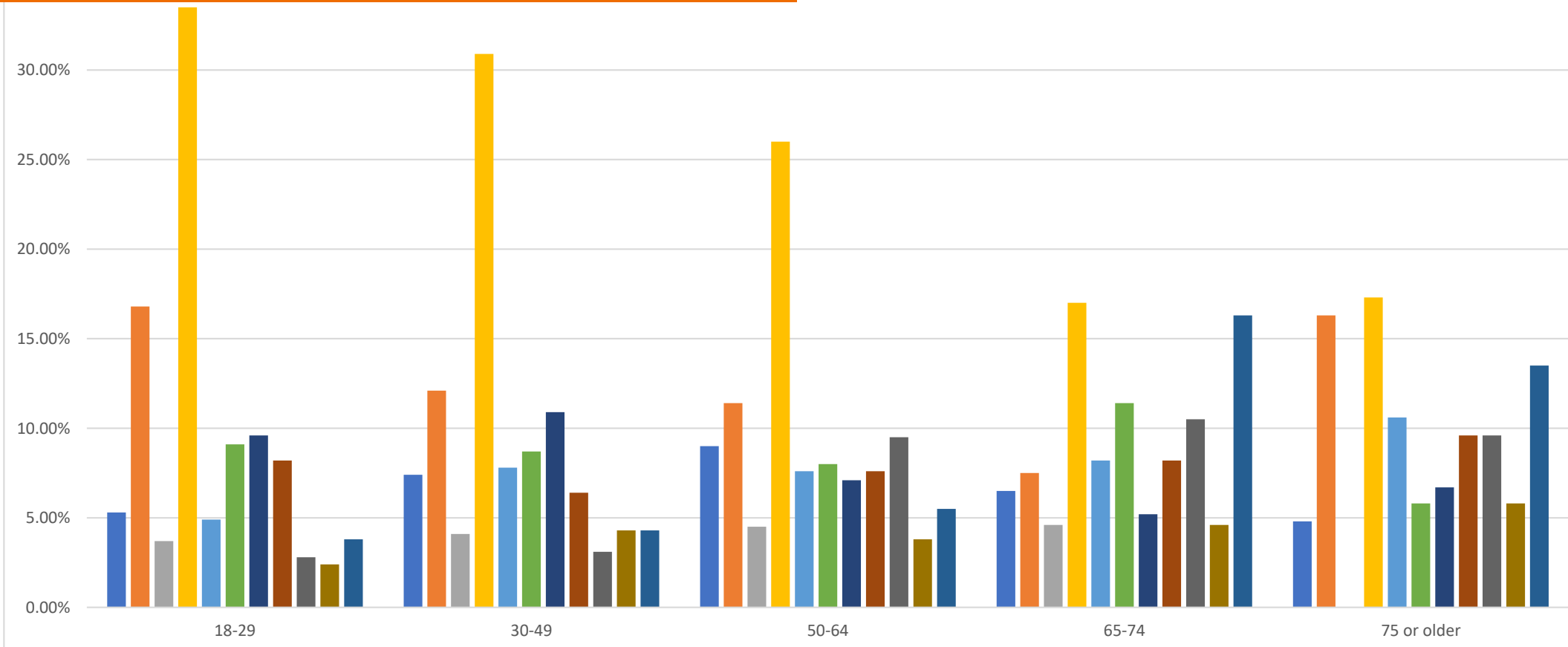
How my genetics affect risks of getting a medical condition?

How to connect with others like me.

How my lifestyle affects risk of a condition.

able to you? (choose only one) BY AGE

Chi Sq. = 524.94***



Which would be **most** valuable to you? (Race/Ethnicity)

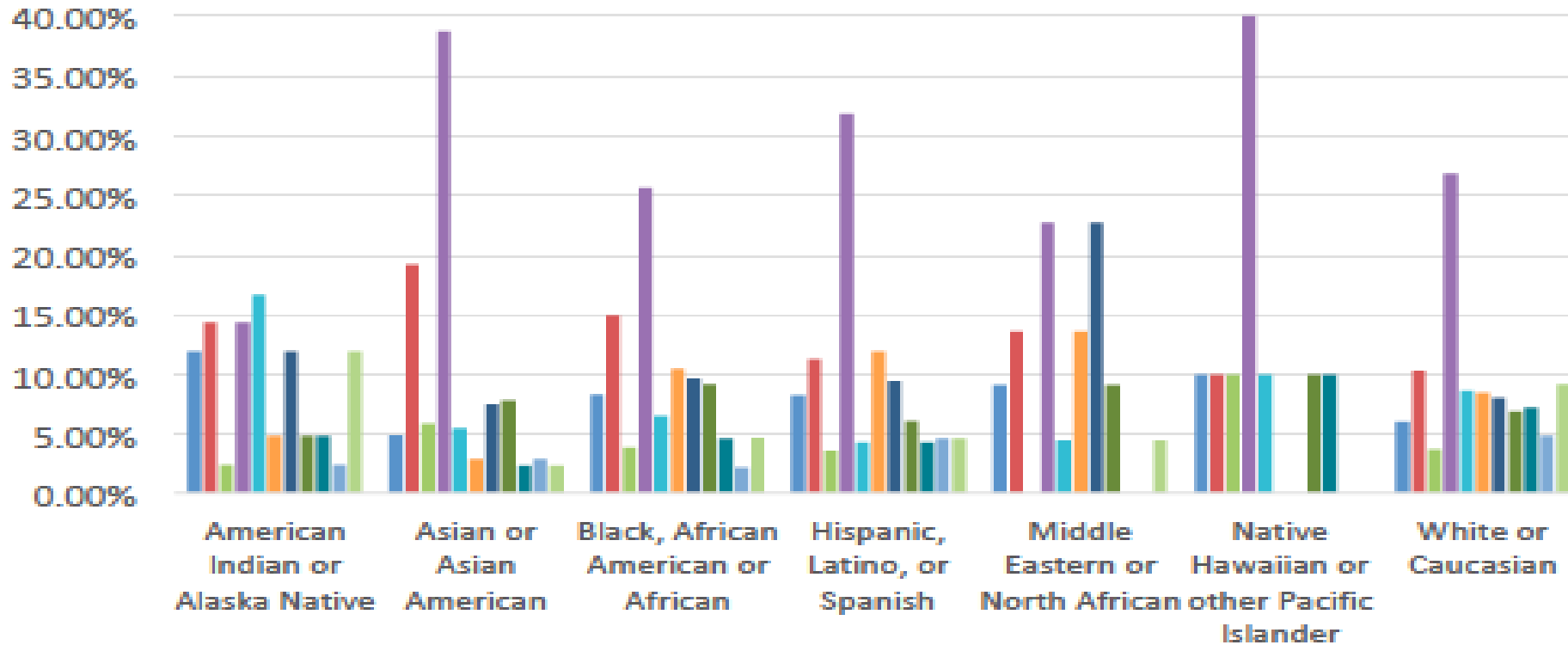
(choose only one; compensation an option)

Genetic risk of disease

Genetic traits

Pharmacogenetics

Chi Sq. = 673.12***



Other notable findings

- Participants more likely to trust research if results returned
- Participants more likely to participate again if results returned
- Value of monetary compensation was variable
 - Age: 30-49 (5.88); 18-29 (5.76); 75+ (4.5)
 - Race/Ethnicity: Blacks (6.01); Asians (5.94); American Indians (4.95)
 - Gender: women (5.75); men (5.5); neither (5.07)
 - Income: <\$24K (5.84); \$50-75K (5.76); >\$100K (5.35)

How is Return of Value Different? Moving beyond “Return of Results”

Return of Results	Return of Value
Sharing overall study results.	Sharing overall study results with added context
Returning data to participants	Returning data prioritized by each participant
Management of individual (incidental) findings by general recommendations.	Management of individual (incidental) findings with specific suggestions for relevant participant actions
Using the same approach across all participants	Informing return of value by soliciting and incorporating participant recommendations and preferences

Thanks!

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COMMUNITY ENGAGEMENT STUDIOS

16 PRIORITY POPULATIONS – CHOSEN TO OPTIMIZE DIVERSITY AND INCLUSIVITY

Priority and hard to reach populations:	
General Population	Native Americans
Older Adults (65+)	Latinos/Hispanics
Parents of children under age 18	Asian Americans
African Americans	Those living in rural areas
Sexual & Gender Minorities	Individuals w/ limited English proficiency
Individuals with limited educational attainment/literacy	Individuals with 3 or more chronic health conditions
Individuals who are deaf or hard of hearing	Individuals who are blind or with limited vision
Individuals with no access the internet	Individuals with limited technical proficiency

Optimizing Diversity and Inclusivity: Giving Voice to Groups Often Underrepresented or Excluded

Sixteen populations chosen to optimize diversity (demographic groups often underrepresented in research) and inclusivity (groups likely unable to participate unless substantial barriers removed)

American Indians/Native Americans	Individuals with limited education or literacy
African Americans	Individuals who are blind or with limited vision
Asians/Asian Americans	Individuals without regular access to Internet
Latinos/Hispanics	Individuals who are deaf or hard of hearing
Sexual & Gender Minorities	Individuals with limited English proficiency
Older Adults (65+)	Individuals with 3 or more chronic health conditions
Children/Parent-Child Dyads	Individuals with limited technical proficiency
People living in rural areas	Community health center patients

Source: Wilkins CH, 02.01.2018

NEJM Catalyst (catalyst.nejm.org) © Massachusetts Medical Society

Optimizing Diversity and Inclusivity: Real People Providing Feedback on Precision Medicine Study Design

Patients of a Health Center in Filipinotown

Caregivers of people with Alzheimer's

People with multiple chronic conditions

Urban African American men

Individuals who are homeless

People who are blind

Transgender men and women

Korean Americans

Individuals who are uninsured

Appalachian people

Latina women in South Florida

Individuals who speak Cantonese

American Indians living in Chicago

People working on their GED

Individuals who use American Sign Language

People from rural South Dakota



Groups who participated in Vanderbilt and Meharry Community Engagement Studios for the Precision Medicine Initiative pilot.

Source: Wilkins CH, 02.01.2018

NEJM Catalyst (catalyst.nejm.org) © Massachusetts Medical Society

Results from Precision Medicine Initiative Community Engagement Studios

Sexual and
Gender
Minorities:
Clinical
Trials

Latinos:
Response to
Medications

- My Health
- Risks
- EHR Access
- Ancestry
Maps

Asian
Americans:
Asking
Experts

Older
Adults:
Overall
Health
Risks