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Authors

Arias, Jalayne J
Tyler, Ana M
Beskow, Laura M
et al.

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RESEARCH ARTICLE

Data stewardship in FTLD research: Investigator and research participant views

Jalayne J. Arias[1,2] | Ana M. Tyler[1] | Laura M. Beskow[3] | Maria C. Carillo[4] | Susan Dickinson[5] | Jill Goldman[6] | Mary A. Majumder[7] | Michelle M. Mello[8] | Heather M. Snyder[4] | Jennifer S. Yokoyama[1,9]

[1]Memory and Aging Center, Department of Neurology, Weill Institute for Neurosciences, University of California, San Francisco, San Francisco, California, USA

[2]Department of Health Policy & Behavioral Sciences, School of Public Health, Georgia State University, Atlanta, Georgia, USA

[3]Center for Biomedical Ethics and Society, Vanderbilt University Medical Center, Nashville, Tennessee, USA

[4]Division of Medical & Scientific Relations, Alzheimer's Association, Chicago, Illinois, USA

[5]The Association for Frontotemporal Degeneration, King of Prussia, Pennsylvania, USA

[6]Neurological Institute, Columbia University Irving Medical Center, New York, New York, USA

[7]Center for Medical Ethics and Health Policy, Baylor College of Medicine, Houston, Texas, USA

[8]Stanford Law School and Department of Medicine, Stanford University, Palo Alto, California, USA

[9]Department of Radiology and Biomedical Imaging, University of California, San Francisco, San Francisco, California, USA

Correspondence
Jalayne J. Arias, Department of Health Policy & Behavioral Sciences, School of Public Health, Georgia State University, 140 Decatur Street SE, Atlanta, GA 30303, USA.
Email: jarias@gsu.edu

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Abstract

INTRODUCTION: Federal policies and guidelines have expanded the return of individual results to participants and expectations for data sharing between investigators and through repositories. Here, we report investigators' and study participants' views and experiences with data stewardship practices within frontotemporal lobal degeneration (FTLD) research, which reveal unique ethical challenges.

METHODS: Semi-structured interviews with (1) investigators conducting FTLD research that includes genetic data collection and/or analysis and (2) participants enrolled in a single site longitudinal FTLD study.

RESULTS: Analysis of the interviews identified three meta themes: perspectives on data sharing, experiences with enrollment and participation, and data management and security as mechanisms for participant protections.

DISCUSSION: This study identified a set of preliminary gaps and needs regarding data stewardship within FTLD research. The results offer initial insights on ethical challenges to data stewardship aimed at informing future guidelines and policies.

KEYWORDS
data sharing, ethics, frontotemporal lobar degeneration, policy/law, research

1 | BACKGROUND

Data are one of the most powerful tools researchers have that accelerates scientific progress toward therapeutic discovery. But, as the saying goes, "with great power comes great responsibility".[1] Data stewardship begins the moment data is collected from a research participant and continues through returning individual results to participants and/or sharing data with other investigators. Trends in research have shifted data stewardship expectations and standards. Federal policies and guidelines have expanded the return of individual results to participants[2] and expectations for data sharing between investigators and through repositories.[3]

Data stewardship responsibilities have special salience in frontotemporal lobar degeneration (FTLD) research. While FTLD is a common cause of young-onset dementia,[4] it constitutes a "rare disease" (estimated 2.7-4.1 cases per 100,000).[5] Additionally, a strong familial component of FTLD (~20% of cases due to a single mutation in one of three genes[6]) may increase the desire to learn individual research results, particularly genetic status. Finally, research aimed at accelerating therapeutic discovery has highlighted the importance of increasing power through multiple data sets, triggering an expansion of biorepositories and data-sharing platforms.

Within this context, the National Institutes of Health Data Management and Sharing Policy (NIH DMS)[3] and the 2018 National Academies of Sciences, Engineering, and Medicine (National Academies)[2] report on the return of individual-specific research results present unique ethical challenges for FTLD research. Researchers contend with a tension between making data widely available versus protecting their own interests in publication and guarding participants' privacy in the context of a rare disease. Additionally, researchers must weigh potential risks (stigma, discrimination, loss of confidentiality) against participants' desires to learn individual research results, including genetic status, during clinical trials and other studies. A lack of disease-modifying treatments further complicates data stewardship in FTLD research. Optimal data stewardship practices will assure necessary protections for participants, maximize recruitment and retention efforts, and accelerate research progress toward therapeutic discovery.

RESEARCH IN CONTEXT

1. **Systematic review**: We conducted a systematic search of the existing literature in PubMed and related sources on data stewardship, including the return of individual research results and data-sharing policies. A Scoping Review, part of the original study (reported elsewhere), further supported this review. The existing literature did not address the issues associated with data stewardship specific to frontotemporal lobar degeneration (FTLD) research.
2. **Interpretation**: To address the gap, we conducted a qualitative study of investigators and research participants to understand and evaluate their perspectives and experiences with data stewardship within FTLD research.
3. **Future directions**: This study provides insights on stakeholders' experiences with data stewardship. Ongoing studies use quantitative methods to collect and understand existing trends within Alzheimer's disease and Alzheimer's disease and related dementias (AD/ADRD) research. These future studies will help inform guidelines and best practices aimed at optimal data stewardship practices to advance therapeutic discover, encourage recruitment and retention, and provide protection to research participants.

2 | METHODS

We conducted semi-structured interviews between March and May 2020 with (1) investigators conducting FTLD research that includes genetic data collection and/or analysis and (2) participants enrolled in a single-site longitudinal FTLD study. We used purposeful sampling[7] to identify eligible investigators to interview (e.g., investigators within this study team's professional network, expert referrals, and in-person recruitment at a relevant conference). The study team for a longitudinal study on FTLD ("parent study") assisted with recruitment to identify eligible cognitively healthy participants who were enrolled in the longitudinal study as asymptomatic family members of an individual with a research-confirmed diagnosis. Where possible, we oversampled for historically underrepresented populations within both cohorts. The UCSF Institutional Review Board approved this study.

We used a semi-structured interview guide for each cohort to collect a description of investigator and participant perspectives and experiences with data stewardship (Supplement 1). Interviews were recorded and transcribed. Study team members (JA, AT) applied the Framework Method[8] to analyze data. A Working Group of field experts advised on the prioritization of codes, which were then further analyzed to identify sub-themes and phenomenon embedded within each code.

3 | RESULTS

We conducted 17 participant-interviews and 17 investigator-interviews (sample demographics reported in Tables 1 and 2). Analysis of the interviews identified three meta themes (Table 3): perspectives on data sharing, experiences with enrollment and participation, and data management and security as mechanisms for participant protection.

3.1 | Perspectives on the value and barriers to data sharing

Investigator- and participant-interviewees revealed: (1) the value of data sharing to scientific progress in FTLD as a rare disease, and (2) the

TABLE 1 Demographics: Participants (17).

Parameter	Men	Women
Age (years)		
18-29		1
30-39	1	1
40-49	5	1
50+	3	5
Ethnicity		
Hispanic or Latinx		
Not Hispanic or Latinx	8	8
Unknown/not reported	1	
Race		
White	7	7
Asian	1	
More than one race	1	1
Education		
Some college		1
Bachelor's	4	3
Master's	2	3
Doctoral/professional	3	1
Employment		
Employed	7	6
Retired	2	1
Previously employed, currently unemployed for reasons other than retirement or disability		1
Insurance		
Medicare/Medicaid	2	1
Health insurance, not including Medicare or Medicaid	9	7
Supplemental health insurance	1	1
Long-term care insurance	4	1
Life insurance	4	3
Disability insurance	2	
Genetic status		
Known	6	5
Unknown	3	3
Years as a research participant		
0-2		3
3-5	5	2
6-10	4	2
10+	1	1
Region		
Northeast	1	
Midwest	2	
West	2	7
Pacific Northwest	2	
South	2	1

TABLE 2 Demographics: Investigators (16, 1 did not complete survey).

Parameter	Men	Women
Age (years)		
18-29		
30-39		2
40-49	1	2
50+	7	4
Ethnicity		
Hispanic or Latinx		2
Not Hispanic or Latinx	7	6
Unknown/not reported	1	
Race		
White	8	6
Asian		1
More than one race		1
Training		
Medicine (other than neurology)	1	
PhD—neurosciences		2
Neurology	5	4
PhD—genetics	1	1
Other	1	1
Disclose genetic status to participants		
Yes	6	1
No	2	7
Years postgraduate		
0-10		3
11-20	2	1
20+	6	4
Region		
East	3	3
Midwest	2	1
West	1	3
Northwest		1
South	1	
Non-USA	1	

dynamics between investigators conducting research that generates data and those using the data for secondary analyses. (3) the importance of paticipants experiences with enrollment, Consent, and return of individual research results. Of note, interviews were completed prior to the NIH DMS Policy (2023).

3.1.1 | Data sharing to advance FTLD research

Interviewees from both cohorts reported that data sharing was important to advance research progress. However, the degree of importance

TABLE 3 Major themes.

Enrollment and research participation		Management (security and storage)		Data sharing (From Inv. 1 to Inv. 2)	
Investigator	**Participant**	**Investigator**	**Participant**	**Investigator**	**Participant**
Participant Decision Making	Study Description	Institution	Institution	Data Sharing Process	Data Sharing Process
Disclosure Process	Enrollment	Data Security	Data Security	Researcher Assumptions	Legal Protections
	Unrelated Discrimination	De-Identification Process	Identification	Discrimination	Recommendations
	Participant Trust	Identifiable Data		Importance of Data Sharing	Data Sharing Perspectives
	Status Decision Making	Data Collection and Document		Inv. Trust in Legal/Process	
	Status Disclosure			Oversight/Governance	
	FTD			Researcher Recommendations	
	Discrimination			Gate Keeping	
	Efforts to Avoid Discrimination				

attached to the practice differed. Investigator-interviewees consistently reported data-sharing as "high importance" – often referencing the rarity of FTLD and the need to increase dataset sizes. Participant-interviewees' responses varied in their enthusiasm. Compared to investigators, their views ranged from accepting to encouraging data sharing, with fewer describing data sharing as necessary to advance research in FTLD/FTD.

> *I think it's paramount, right? I mean, I do not think for these complex diseases that we're going to solve problems if people don't share data. (Investigator)*

> All investigators have all their mutation carriers, and we could actually combine the data, probably have a better chance to find some kind of options for treatment, or even early detection, or anything that could help the patient. (Investigator)

> *I would think that's very important, since there is not a cure or a medicine to slow down the symptoms. (Participant)*

> I wouldn't mind because I know that I have an anonymous identification, so I'm fine with that, but obviously I wouldn't want there to [. . .] be a public site that would show my genetic results. But I don't mind [. . .] within this setting, within those who are studying this. (Participant)

3.1.2 | Intra-researcher dynamics affect data sharing practices

Investigator-interviewees provided insights on the significance of dynamics between data generators and data users. A significant subset of investigator-interviewees reported sentiments promoting data sharing.

> *I've been a very strong advocate for data-sharing, as opposed to kind of just holding onto data for one's own purposes. I never really understood that very much. Perhaps I'm very naive or blind to what the exigencies of research may require in this competitive climate. (Investigator)*

Further, some investigators reported that the small community of FTLD researchers promoted data sharing because data requesters were known to the data generator or consortia managing repositories.

> *I also think that at this point, everyone requesting data from us in the FTLD community is more or less a friend, and so it's not as concerning to me. (Investigator)*

Yet, investigator-interviewees also described concerns about withholding (i.e., not sharing data upon request or through a repository) and barriers to data sharing.

> *And my impression is that even with the current policies in place, not everybody is being—there's not good faith sharing across the board [. . .] (Investigator)*

Investigators' perceptions regarding barriers to data sharing may help clarify factors that lead to data withholding. Reported barriers included a lack of control over how data will be used, the potential for being "scooped," and the resources and effort required to prepare data for sharing.

> *[S]haring data is not exactly appealing most of the time because you never know if people are going to use it well or what they'll do with it. (Investigator)*

TABLE 4 Limitations of the consent process.

Trust in the research team due to long-standing relationship with the institution/research team	"Again, we just kind of willfully sign these consents [. . .]. Yeah, we should be reading them, but it's like we just trust you guys implicitly. But I've never really thought about it like that because we just–and we do, like there's–we also have a relationship with [Institution] because my family has been going there for over ten years."
Feeling overwhelmed by the consent process	"Right. Yeah, I mean, I've always felt with every study, there's clear paperwork, communication. I do tend to read before I sign, at the same time, I'm often overwhelmed so I'm like, 'Okay, whatever.' But I've always felt if I ever wanted a copy of some detail, that I could ask for it."
Desperation for advancements in research and disease-modifying therapies	"Right. So I know that with each study, there are documents that we sign, things like that. And I hate to sound so casual about it, but I've sort of–I guess I'm a fairly trusting person and I'm just not all that worried. And the disease is reality for us and so I'm just sort of looking for hope. So yeah, so I haven't stressed over who can see it, who can't. And I know those things matter, so I maybe should be more concerned, but I just haven't been."

> *[T]here is a little bit of frustration in the sense that you do all the QC and the processing of the data, and then you release it, and [. . .] even though you've been involved in all of that, you're basically in the same point as everybody else when it comes to analysis. (Investigator)*

Other investigators reported a need for increased oversight and improved gatekeeping to address concerns (e.g., mechanisms to limit access to data generated through a particular study or stored within a given repository).

> *I would like to see some gatekeeping just to make sure that it isn't some random person who randomly identifies himself as a researcher. Something that legitimizes the researcher's background and ability to handle this data would be important [. . .]. (Investigator)*

3.2 | Experiences with enrollment, consent, and return of individual research results

Interviewees reported intertwining dynamics at the time of enrollment, including factors influencing the decision to enroll (e.g., the option to learn individual research results). Interviewees' responses unveiled challenges in the effectiveness of the consent process.

3.2.1 | Reliance on and limited effectiveness of informed consent

Interviewees' reports on informed consent and enrollment decisions introduced a "conundrum of informed consent" (Table 4). Investigator-interviewees often looked to the consent process as a mechanism to ensure alignment of participants' expectations regarding study procedures, including plans for sharing data. Yet, investigator-interviewees recognized potential limitations of consent practices, including time and effort spent to assure participants comprehend the risks and benefits of study participation.

> *I think that if it's something that somebody consents to, then I think it's great. [. . .] If insufficient time has been spent going over the nuances of data sharing then that might give me some pause. (Investigator)*

The potential limitations of consenting procedures were more evident in participant-interviewees' responses. Participant-interviewees reported a varying understanding of how data would be shared, including who would have access to their data, and the purposes of its use outside of the study site. These variations are particularly notable given that all participants were enrolled in the same study and completed the same consent procedures. Participants were asked who, from their understanding, would receive or have access to their data. The following responses provide examples of differences in participant-interviewees' understanding or interpretation.

> *The staff at [Institution], and I remember there was an assortment of other universities that I can't recall which exactly they were, and I believe the National Institute of Health. (Participant)*

> *From what I understand, it's just [Institution] faculty [. . .] I don't think it's been shared with others. However, we're part of different studies [redacted]. So I don't really know. (Participant)*

While some participants reported to be less concerned about the details of data sharing—others may have more concerns about the breadth of how data might be used. The data here does not provide insights on whether improving participants' understanding of data sharing could influence participants' consent to participate in research. While some participants reported less concern about the details of data sharing, others may have more concerns about the breadth of how data could be used.

> *I don't remember specifically what they said in terms of how the data would be shared other than that there were other institutions who were involved in the study and that the data is made available. My understanding is that at some point, there's a possibility that other groups that are not directly participating in the study as researchers may request access to some of the data to look at it, see how the data has been collected and what data is there. But in terms of that I know, it's just these long forms they read you and you sign them saying, "Yeah, this is fine. This is cool. I know what you're doing. Yeah, blah, blah, blah," and on and on and on and on. So, I'm familiar with that. (Participant)*

Interviewees' responses do not provide insights on whether improving participants' understanding of data sharing could influence participants' consent to participate in research. Yet, we identified three different subthemes that may influence decisions about participation or call into question whether the consent meets requisite standards for comprehension and voluntariness. Participant-interviewees reported (1) trust in the research team due to a long-standing relationship with the institution, (2) feeling overwhelmed by the process, and (3) desperation for research advancements regarding the disease.

> *I know that with each study, there are documents that we sign, things like that. And I hate to sound so casual about it, but I've sort of–I guess I'm a fairly trusting person and I'm just not all that worried. And the disease is reality for us and so I'm just sort of looking for hope. So yeah, so I haven't stressed over who can see it, who can't. And I know those things matter, so I maybe should be more concerned, but I just haven't been.*

Finally, participant-interviewees reported that the opportunity to learn their genetic status was a driver for enrolling in the study.

> *[P]art of the reason for my enrollment in it was—is because of the genetic testing that came out of it and knowing if I was a carrier or not.*

These reported perspectives and views on consent may call into question whether participants are sufficiently "informed," and whether undue influence—even if unintentional—may be present.

3.2.2 | Benefits and challenges of returning individual research results

Although there was some variation between investigator-interviewees and participant-interviewees, both agreed that access to individual research results was beneficial (Table 1). Participant-interviewees who elected to learn their genetic results through research emphasized the potential value of genetic information. Yet, they recognized that this interest was not universal—even within their own families.

> *But I do know that inside my own family, there is the entire spectrum of opinions on that issue. But for me, knowing more is better. (Participant)*

Participant-interviewees recognized that genetic results indicating risk for FTLD could have consequences (e.g., employment, social, and insurance discrimination). These sentiments were similarly emphasized by investigator-interviewees who reported challenges when determining whether to return research results within a study.

> *I think most of it has to do with how people can be using this maliciously, either to take advantage of vulnerable population essentially, how insurance might be able to access information and use it against patients who might be pursuing certain types of disability or if they're doing some sort of long-term financial family planning. I think the concern is around employers and whether or not they're going to have access to this and whether or not there's going to be [. . .] discriminatory practices that might take place that we don't know about. The other piece of it is whether or not a patient wants to share this with anyone else in their social network. I think it has tremendous impact. And especially in a time of a lot of access to social media, I think it's really hard to try to keep this information private. And I feel also there's just too many opportunities for people to take advantage of this type of information in ways that I haven't even thought of. There's also the grave concern about how this knowledge affects the immediate social family environment, families, and friends, right? I mean, it does change, potentially, their perspective on the individuals who may be a carrier. And it also has an impact on the next generation and family planning [. . .] the fact that there is this very real chance of inheriting the genetic alteration. (Participant)*

Investigator-interviewees raised unique logistical challenges in developing disclosure practices in longitudinal studies that recruit multiple family members. The familial-risk characteristics and rare nature of FTLD enhanced investigators' discomfort with disclosing individual research results.

> *The problem with these inherited disorders is information about one family member being released impacts other family members [. . .] some people are prepared to go in front of the cameras and talk to the media, and that's fine. But there are other people who want to preserve their anonymity. So, it's important that we help to protect them.*

Individual family members' preferences create logistical challenges to maintain confidentiality of genetic testing results from other family members: *"One family member knows and yet doesn't want the other family member to know and making sure that we're respecting all of those complicated directives and wishes within families."* These challenges are heightened in the context of a participant who lacks capacity or is deceased.

Resolutions are not immediately evident. While some investigator-interviewees lauded the role of genetic counselors to support disclosure practices broadly, others raised concerns regarding an over reliance on genetic counselors as the solution given their limited availability. *"There are many, many places where you need a genetic test, and you cannot find a genetic counselor."*

3.2.3 | Data management and security as mechanisms for participant protections

A final theme persistent throughout interviews was the reliance on privacy and security measures as tools for protecting participants from potential harm. Participant-interviewees emphasized that de-identification was important to their decision to enroll in research and their comfort with data-sharing practices. De-identification also emerged among investigator-interviewees. Yet, they also referenced re-identification given increasing technical capabilities, small cohort sizes, and rare disease status.

> *It's that when you have rare mutations in families, you want to be particularly careful about not releasing any information that might make somebody identifiable. Especially in that sort of healthcare environment that we live in in the US, you really have to be careful that you never disclose information that would make it apparent. But even a family – [. . .] has an inherited predisposition without identifying an individual [. . .] even though we have GINA, or there should not be genetic information discrimination, I wouldn't want to trust that [. . .]*

4 | DISCUSSION

This study identified a set of preliminary needs that inform future efforts on data stewardship within FTLD research. Investigator-interviewees reported that data sharing is important to advancing research, particularly for a rare disease. Yet, they also recognized barriers to optimal data sharing—including data withholding and concerns regarding the use of data after it is shared. Similarly, participant-interviewees reported a general acceptance with data sharing. Critically, interviewees also reported experiences that challenge effective informed consent. Our results revealed drivers, including the availability of individual research results, that impact participant-interviewees' decisions to enroll in a study and their acceptance of data sharing. Understanding the dynamics affecting participants' decision-making is important in the context of investigators' responsibility to ensure informed and voluntary consent.

The implementation of the NIH DMS policy will continue to alter expectations for data sharing. The new policy, along with existing standards and guidelines (e.g., the FAIR Principles[9] and CAP Principles[10]), establish a foundation for equitable and effective sharing practices. Despite these guidelines, our results highlight barriers and indicate that investigator-interviewees hesitate to share data for a variety of reasons (e.g., time to prepare data, concerns about use after sharing). Reports of data withholding within the field raise concerns about data sharing enforceability. While other studies have previously identified withholding and barriers to data sharing like those reported in our qualitative interviews, here, the unique focus on FTLD further highlights the potential relevance of sub-field culture and professional relationships as a factor affecting practices. Additionally, the field needs evidence on what mechanisms encourage data-sharing practices (e.g., punitive enforcement vs. incentives) with a focus on the role of funders, journals, and academic research institutions to promote policies. Optimal data-sharing practices may require tailoring to address barriers unique to FTLD.

Our data also reveal concerns about participants' understanding of the breadth of data sharing and other factors that might impede an effective consent process to enroll (trust, overwhelm, desperation). Investigators rely on the consent process to ensure that data sharing comports with research participants' expectations, but emotions that influence enrollment in FTLD studies may undercut the degree to which informed consent reflects a deliberate weighing of risks and benefits. However, because we conducted interviews well after participants had originally consented to enrollment in the parent study, we are unable to distinguish between a lack of understanding at the time of consent versus difficulties in recalling details. This may warrant a reconsideration of materials and resources for participants to ensure comprehension across the life of a study. Furthermore, participants' desires to learn their genetic status may drive enrollment, which is consistent with findings from prior studies.[11] The NASEM Report challenges prior policies and ethical standards that discouraged return of individual results,[2] but recognizes that returning results is not an absolute obligation across all studies. The tension between participants' interests in learning their results, and investigators' ethical responsibilities to mitigate potential harms is a particular concern for FTLD given familial risk and lack of disease-modifying treatments.

Our study design equipped our team to capture in-depth insights from key stakeholders (interviewees). Still, some limitations exist. Participant-interviewees are from a single site and study. Their perspectives, particularly regarding returning individual research results, may be biased by their experience within this singular study. Additionally, the investigator and participant-interviewee cohorts lacked diversity. While we made efforts to oversample to increase diversity, our cohorts reflect the population within the parent study and of investigators in the field. The limited diversity in the samples may bias the results, particularly in the participant-interviewee cohort. A quantitative approach that harnesses broader experiences and views across studies with differing consent practices, return of individual research results policies, and approaches to data sharing is needed. Similarly, investigator-interviewees represented diverse experiences from different institutions, but the sample represented experienced investigators. This may result in a bias away from the concerns or considerations of less established investigators. Finally, this study focused on research conducted within the United States. Additional research is needed to understand how variations between countries—including

differences in national policies—may affect data sharing. While data collection pre-dated the publication of the NIH DMS Policy, our analysis did not identify policy changes that would alter the outcome of our results.

We must consider the complex ways in which data security, return of individual results, and data sharing interact and impact other critical aspects of research. Further research to understand the factors that inform investigators and participants' behaviors, and decision-making will promote policies and practices that optimize data management, manage expectations of both researchers and participants, encourage ethical recruitment and retention, and accelerate progress in the field. Here we focus on data stewardship practices relevant to genetic information within FTLD research. However, these issues may have broader salience to other modalities of testing used in research, including imaging and blood-based biomarkers. Our results offer initial insights to help researchers identify challenges to ethical data stewardship and mitigate risks to participants, including considerations during the consenting process. This is an optimal moment in the broader Alzheimer's disease and Alzheimer's disease related-dementias research field to develop clear guidance for investigators and promote gold standard data stewardship.

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CONFLICT OF INTEREST STATEMENT

Drs. Maria Carrillo and Heather Snyder are full-time employees of the Alzheimer's Association. The remaining authors do not have conflicts to disclose. Author disclosures are available in the supporting information. Dr. Jennifer Yokoyama serves on the scientific advisory board for the Epstein Family Alzheimer's Research Collaboration.

CONSENT STATEMENT

All human subjects provided verbal consent prior to enrollment in this study. The UCSF IRB approved a waiver of written consent for this study because participants were exposed to minimal risk and written consent increased the risk of loss of confidentiality.

ORCID

Jalayne J. Arias https://orcid.org/0000-0002-6081-5231

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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